

Achievement of Guided Growth in Children With Low-Tone Neuromuscular Early-Onset Scoliosis Using a Segmental Sublaminar Instrumentation Technique

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Abstract

Objectives/Summary of Background Data: Segmental sublaminar spinal instrumentation without fusion results in guided growth and correction of deformity in early-onset scoliosis (EOS). The purpose of this study is to report the outcomes of a series of children with low-tone EOS that were treated surgically with a modification of a Luque trolley technique.

Methods: This study is a retrospective chart and radiographic review of a single-center series of 13 consecutive children who met inclusion criteria with documented progression of scoliosis greater than 25°. All children received surgical treatment with guided growth without fusion using a modified Luque trolley technique. The children's preoperative, postoperative, and most recent radiographs were assessed for Cobb angle, T1–T12 and T1–S1 height, and sagittal alignment including proximal junctional kyphosis. Surgimap spine software was used for calibration and measurement purposes. Complications and need for repeat and/or secondary surgical procedures were recorded.

Results: The mean age at surgery was 7.4 years (4.6–10.5). On average, 15 segments (13–16) were instrumented. None of the children went on to a spontaneous fusion, and the average growth rate per year from T1–T12 and T1–S1 was 0.9 cm/y and 1.5 cm/y, respectively. The mean total growth from T1–T12 and T1–S1 was 22.3 cm (16.6–30.2) and 37.5 cm (30.1–46.4). A total of three additional surgeries were needed in two children to address complications. There were no mortalities.

Conclusions: Sublaminar guided growth is a safe and effective treatment in the Low Tone Neuromuscular subset of EOS. Follow-up studies failed to show signs of auto fusion, and implant failure was not observed in our cohort. All children displayed growth post-operatively without the need for multiple distraction-based surgeries. Guided growth minimizes the risks associated with multiple surgical procedures while maintaining correction and allowing for near-normal rates of spinal growth.

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Keywords: Early-onset scoliosis; Neuromuscular scoliosis; Segmental Sublaminar Spinal Instrumentation

Introduction

Early-onset scoliosis (EOS) is defined as a coronal Cobb angle greater than 20° in a child less than 10 years of age. The EOS classification divides children with spinal

curvatures into Structural/Congenital, Low/High Tone Neuromuscular, Syndromic, and Idiopathic types [1]. Conditions that constitute the Low/High Tone Neuromuscular division of the classification often include myopathies and muscular dystrophies, cerebral palsy, and paralytic conditions.

One such low tone neuromuscular condition is spinal muscular atrophy (SMA). Clinical manifestations of SMA occur secondary to degeneration of the anterior horn cells of the spinal cord leading to symmetrical muscle weakness and atrophy of the trunk and proximal musculature of the shoulder and hip girdle [2]. One of the greatest problems faced by the orthopedic surgeon caring for patients with

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SMA is addressing the spinal deformity associated with this condition [3]. The scoliosis associated with SMA is of the neuromuscular type [4], in which the pelvic obliquity is proportional to the severity of the curve. Scoliosis in SMA has an early onset and progresses rapidly prior to the onset of puberty. Respiratory impairment in the form of restrictive lung disease advances simultaneously with the progression of the spinal curvature. In SMA, survival typically depends on the degree of respiratory involvement with atelectasis and pneumonia as the usual causes of death [5]. The outcomes of orthotic management to slow or halt curve progression have been poor and further contribute to restrictive pulmonary disease [6,7].

Surgical treatment of the spinal deformity associated with SMA and other low/high-tone neuromuscular EOS conditions has become the treatment of choice. For children with EOS, Karol et al. suggested that spinal arthrodesis be performed once thoracic vertebrae 1–12 height is greater than 22 cm to allow for optimal lung volume, otherwise thoracic insufficiency syndrome will likely develop [8]. DiMeglio showed that the spine grows in a bimodal distribution with rapid growth from zero to 5 years and again after 10 years of age and is completed in girls by 13 and boys by 17 [9,10]. Surgical techniques that have been utilized in the EOS population are distraction-based techniques (vertical expandable prosthetic titanium ribs and growing rods), guided growth (Luque trolley and Shilla), compression-based (tether and staples), and early limited arthrodesis [11]. Many of these techniques allow for growth and usually require multiple surgical interventions.

Sublaminar guided growth without fusion has previously been reported as an option to correct spinal deformity while maintaining spinal growth with varied results [12–15]. Recently, efforts to address the scoliotic deformity associated with EOS have focused on various nonfusion and growing techniques [16]. Over the past 15 years a modification of the Luque trolley technique has been performed on a subset of children with EOS at our institution. The purpose of this study is to report the outcomes of a series of children with neuromuscular and syndromic EOS that were treated surgically with sublaminar guided growth without fusion using a modified Luque technique during their years of peak growth.

Material and Methods

Our Institutional Review Board approved a retrospective chart review from 1998 to 2014. Children aged 10 years or younger who carried a diagnosis of scoliosis with progression of their curve greater than 25° and no prior surgery about the spine were included. Only patients who underwent sublaminar guided growth without fusion and had at least two years' follow-up with preoperative, intraoperative, and postoperative radiographs were included. The senior author performed all surgeries.

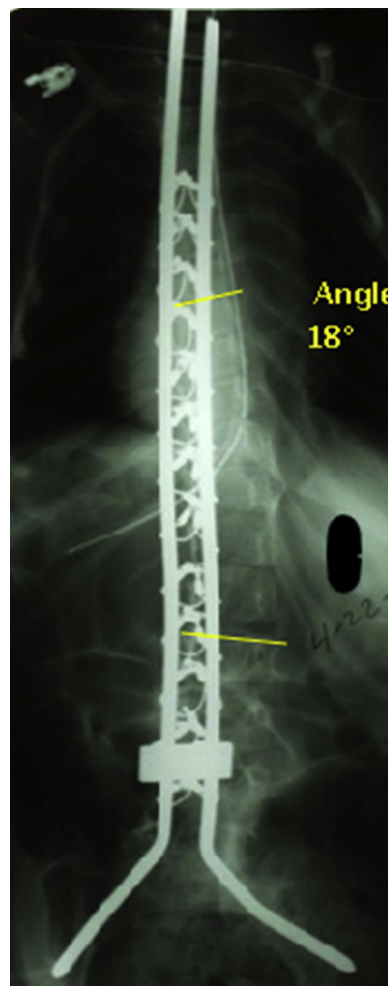


Fig. 1. Child #1 – Intraoperative PA Radiograph, yellow lines represent Cobb angle. Surgimap Spine Software (Nemeric Inc, 306 E 15th Street, New York, NY 10003).

A chart review was conducted and data were collected that included age, sex, length of follow-up, neuromuscular condition, complications, and decline in their pulmonary function studies defined as the need for tracheostomy and/or ventilator dependence post spinal surgery. Pulmonary functions studies were performed every six months for monitoring purposes.

All preoperative, intraoperative, initial postoperative, and most recent follow-up biplanar radiographs were reviewed. Surgimap spine software (Nemeric Inc, New York, NY) was used to measure radiographs. Images were uploaded and the known rod sizes (4.76 mm vs. 5.5 mm) were used for calibration purposes. Radiographic evaluation included coronal plane analysis using the Cobb method and monitoring the cranial end of the construct for sagittal balance. Growth was measured from the anterosuperior endplate of T1 to anteroinferior endplate of T12 and anterosuperior endplate of S1 accordingly via lateral radiographs intraoperatively after instrumentation and then at each visit. The radiographs were assessed for the initial

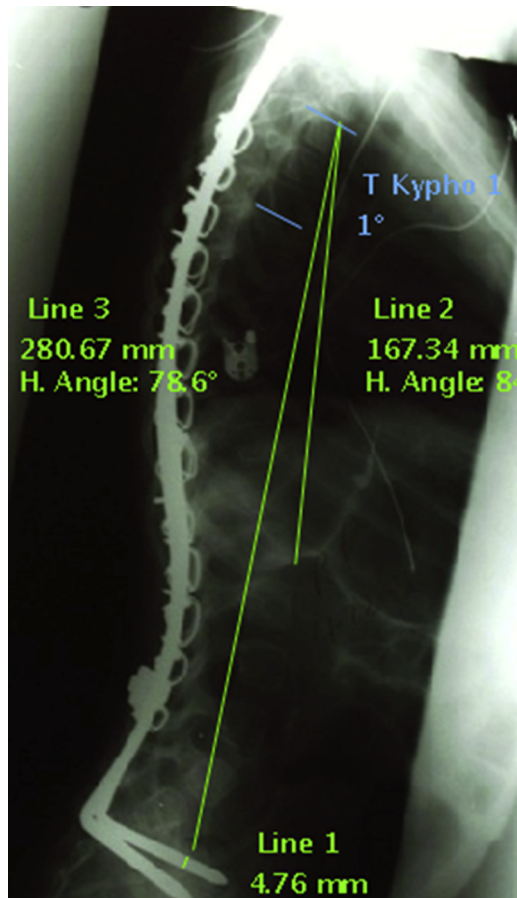


Fig. 2. Child #1 – Intraoperative Lateral Radiograph, green lines depict: calibration, T1–T12 and T1–S1 measurements, blue lines represent proximal junctional kyphosis measurements. Surgimap Spine Software (Nemeric Inc, 306 E 15th Street, New York, NY 10003).

degree of correction, percentage loss of correction at final follow-up, and growth rates from T1–T12 and T1–S1. The duration of follow-up used to calculate growth was ended for each child once the females turned 14 and the males turn 17 in the assumption they reached skeletal maturity or surpassed the period of peak growth. All radiographs pre-operatively, intraoperatively and postoperatively were obtained in either a prone or supine position.

Surgical technique

A posterior midline approach to the spine was utilized. Subperiosteal dissection of the lamina was carried out laterally to the facet joint capsules that were left intact. The interspinous ligament and central portion of the ligamentum flavum were resected at each level to allow passage of sublaminar Songer braided stainless steel cables. The technique employed differs from that originally described by Luque [17]. Fascial incisions were made over the posterior iliac crests bilaterally and the paraspinal musculature was reflected, allowing for pelvic fixation. Two 4.76- or 5.5-mm stainless steel unit rods

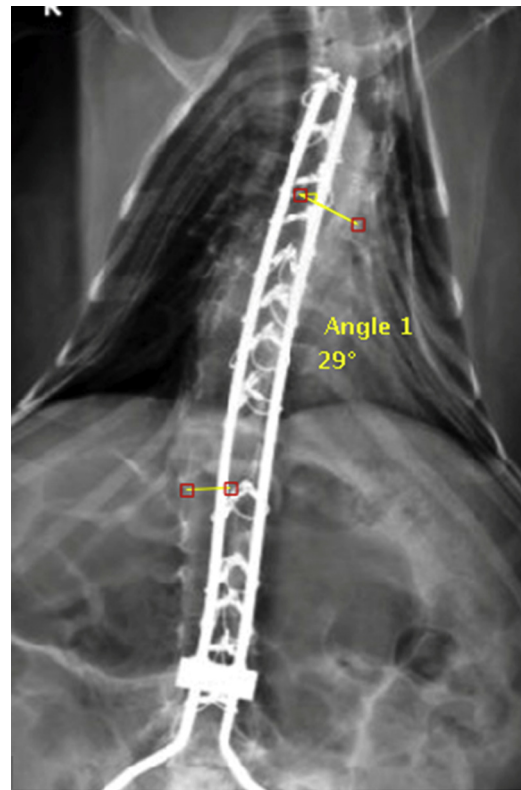


Fig. 3. Child #1 at 156 months, yellow lines represent Cobb angle. Surgimap Spine Software (Nemeric Inc, 306 E 15th Street, New York, NY 10003).

were cut and contoured for maximal curve correction as well as establishment of appropriate lumbar lordosis and thoracic kyphosis. The upper-instrumented level was T2, T3, or T4 depending on the rod prominence so that cervical range of motion was not restricted. The rod position extended to at least T2 to allow for future growth on all of the patients included in the study. The left and right unit rods were placed and sublaminar Songer cables were sequentially tensioned to 40 foot-pounds in a caudal to cephalad direction. A cross link was placed at the caudal end of the construct on all patients who weighed more than 20 kg. There were no techniques employed to obtain an arthrodesis or distraction at the time of the initial surgery, nor was any interposition material used to avoid fusion. The sublaminar guided growth procedure is used as a standalone procedure without future plans for implant removal, distraction procedures, or final arthrodesis surgery. External bracing was not used postoperatively.

Patients were followed two weeks postdischarge from the hospital then at 6 weeks, 3 months, 6 months, 12 months, and annually thereafter. Serial radiographs were followed for spinal growth, implant stability, curve progression, and sagittal balance. Follow-up was facilitated by the specialty clinics that were staffed by the senior author (Figures 1–4).

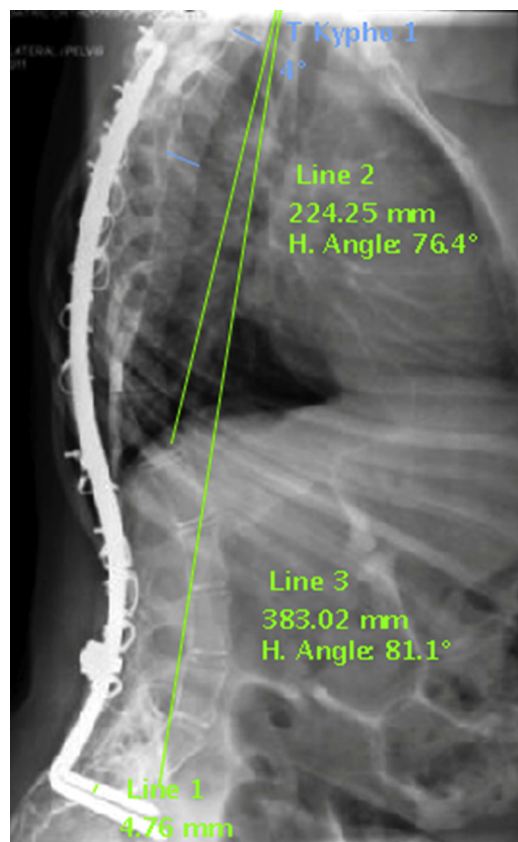


Fig. 4. Child #1 at 156 months, green lines depict: calibration, T1–T12 and T1–S1. Surgimap Spine Software (Nemeric Inc, 306 E 15th Street, New York, NY 10003).

Results

Thirteen consecutive EOS children were identified that met the inclusion criteria (Table 1). The mean age at surgery was 7.4 years (4.6–10.5 years). There were 9 females and 4 males. The mean follow-up was 4.9 years (2.3–13 years). The mean preoperative/postoperative major curve measured 49° (25°–76°) and 17° (2°–39°), respectively. The mean curve correction was 32°. The mean Cobb angle at final follow-up was 23° (2°–57°) with a mean percentage loss of correction of 19%. Proximal junctional kyphosis (PJK) was calculated by measuring the Cobb angle between the superior endplate of the cephalad noninstrumented vertebrae and the inferior endplate of two vertebrae caudal

Table 1
EOS diagnoses.

Condition	No. of children
SMA Type II	5
SMA Type I	4
Cerebral palsy	1
Ulrich muscular dystrophy	1
Rett syndrome	1
Shunted hydrocephalus	1

EOS, early-onset scoliosis; SMA, spinal muscular atrophy.

to the proximal aspect of the construct. The mean PJK at the most recent follow-up was 11° (2°–20°).

On average 15 (13–16) segments were instrumented with the sublaminar Songer cables. The average spinal growth rate was measured from T1–T12 and T1–S1 and was 0.9 cm/y and 1.5 cm/y, respectively. The mean final height at the most recent follow-up from T1–T12 and T1–S1 was 22.3 cm (16.6–30.2 cm) and 37.5 cm (30.1–46.4 cm), respectively. The average total growth when comparing the first postinstrumentation measurement to the most recent follow-up measurement for T1–T12 was 3.8 cm (2–9.1 cm) and T1–S1 was 6.1 cm (1.8–10.8 cm) (Table 2). None of the patients went on to spontaneous fusion prior to the desired T1–T12 goal of 22 cm and many of the children continued to grow until skeletal maturity was reached. The growth was determined by the measured spinal growth via serial radiographic measurements along with the change in the position of the upper instrumented vertebra (UIV) compared to the position of the proximal end of the rod in the initial implant construct versus the most recent radiographs. At this time, 5 of the 13 children had reached the age criteria of 14 years for the females and 17 years for the males.

There were no returns to the operating room for a final fusion procedure on any of the children in this study. None of the children required removal of their entire implant construct. From the initial postoperative period through the latest follow-up there were no mortalities. No patient had a significant decline in their pulmonary function requiring a tracheostomy or ventilator support at any time during their postsurgical follow-up. It must be noted that one of the children was lost to follow-up 52 months after the index surgical procedure. The data for this child were included in the study because it met the inclusion criteria, but long-term complications or revision surgery was unknown. The initial and final postoperative measurements for T1–T12 were 14.2 cm that grew to 16.6 cm, and for T1–S1 were 23.5 cm that grew to 30.5 cm. There were two children who required surgery due to postoperative complications. One

Table 2
Data.

Age at surgery, years	7.4 (4.6–10.5)
Average follow-up, years	4.9 (2.3–13)
Preoperative Cobb	49° (25°–76°)
Postoperative Cobb	17° (2°–39°)
Current Cobb	23° (2°–57°)
Percentage loss of correction	19%
Average proximal junctional kyphosis	11° (2°–20°)
T1–T12 average growth rate, cm/y	0.9
T1–S1 average growth rate, cm/y	1.5
T1–T12 average total growth, cm	3.8 (2–9.1)
T1–S1 average total growth, cm	6.1 (1.8–10.8)
Initial T1–T12 average height, cm	18.5 (14.2–21.7)
Initial T1–S1 average height, cm	31.3 (23.5–26.8)
T1–T12 height at final follow-up, cm	22.3 (16.6–30.2)
T1–S1 height at final follow-up, cm	37.5 (30.1–46.4)

child was found to have a deep infection and required two surgical irrigation and debridement procedures with retained implants and without alteration to the initial implant construct. The second observed complication occurred during the sixth postoperative year in a child that was involved in a motor vehicle accident. The patient presented for interval follow-up with complaints of poorly localized left-sided hip, back, and pelvic pain a year after the injury. Radiographs and subsequent CT scan revealed protrusion of the left unit rod beyond the inner table of the pelvis and the patient underwent surgery to revise the implant construct. The child had resolution of pain and has had an otherwise uncomplicated postoperative course. Another child who has reached skeletal maturity was noted to have worsening of a secondary curve in the cervicothoracic junction but is currently being monitored with annual radiographs.

Discussion

Luque and Cardoso initially described a technique utilizing rigid internal fixation without fusion and bracing in children less than 11 years of age. Their technique employed fixing a unilateral Harrington bar segmentally to each vertebra, with No. 1.22 stainless steel wire loops on either the convex or concave side of the deformity after subperiosteal dissection was performed on the lamina [14].

The use of instrumentation placed in a Luque fashion has been previously reported in the treatment of scoliosis in patients with spinal muscular atrophy [18,19]. Phillips et al. reported on 31 patients with SMA and scoliosis. Of these 31 patients, 22 had been treated nonsurgically and nine treated surgically. The patients treated surgically were found to have improved sitting balance and endurance [20]. In 1983, McAfee et al. reported on the use of sublaminar instrumentation to preserve spinal growth. Through biomechanical testing in immature beagle hounds that underwent sublaminar guided growth using the Luque technique, they provided the experimental basis and showed that progressive longitudinal growth can occur under the instrumented segments without the loss of fixation [21].

Other previously published studies regarding the use of the Luque technique similar to the one used in this study to preserve spinal growth have been less than promising. This may be attributable to the various diagnoses in the cohorts studied. In a review of nine patients who underwent Luque instrumentation without fusion, Mardjetko et al. reported that sublaminar guided growth without fusion was ineffective in controlling spinal deformity. In their study the patients achieved 35% of predicted growth from the instrumented segments and all nine patients went on to a spontaneous fusion and required at least one revision surgery [15]. Rinsky et al. also reported poor results due to rod failure in three of nine patients who underwent sublaminar guided growth without fusion using Luque instrumentation, with a loss of correction of 32% and interval growth of 0.8

cm [22]. Eberle et al. reported implant failure in a group of 19 children with paralytic scoliosis secondary to poliomyelitis treated with guided growth without arthrodesis [12]. In our cohort of children, these unwanted outcomes were not encountered. This may be attributed to the modification employed with our technique in that one rod was placed on each side of the spine where prior studies used dual rods on each side. We feel that this allows for a lower coefficient of friction, which in turn should decrease the risk of growth cessation along the construct.

Our technique utilizes parallel rods with a firm pelvic fixation allowing growth along the entire aspect of the construct in the absence of clinically significant junctional kyphosis except in one patient compared to other techniques. None of the children required revision surgery due to PJK. Low-tone neuromuscular conditions predispose these children to cervical hyperlordosis and scoliosis because of their difficulty supporting the weight of their head. Previous studies utilizing this technique have failed to include the pelvis in the construct, which likely attributed to the higher rates of implant failure. The previous Luque technique used a unilateral bar with sublaminar wire loops, which led to more implant failures. The observed growth demonstrates that these children continued to grow despite the previous concerns with subperiosteal dissection of the lamina. This technique circumvents the need to perform multiple surgeries as the child grows because thoracolumbar containment is maintained.

Ouellet's recently published case series of 5 EOS children showed short-term success with a growing rod technique designed to minimize reoperation rates. They reported that 60% of their patients required reoperation during a follow up period of 4 years; however, their technique was quite different in that they used two pairs of 5 mm titanium rods that were rigidly fixated proximally and distally. Sublaminar wires were placed at the apex of the deformity and looped around the dual rod construct on each side [23].

We found that in our series of 13 patients only one construct revision was needed. Chandran et al. looked at a group of 11 patients with SMA that were treated with a growing rod construct. The patients in their series underwent a total of 45 surgical procedures and maintained curve correction at final follow up [24]. A case-based survey on the use of growing rods for spinal deformity found that for 265 patients, the average lengthening interval was one per 8.6 months, with six to seven lengthening procedures per patient [16,25]. A study by Sankar et al. showed that distraction-based techniques resulted in the law of diminishing returns with each additional lengthening [26]. Furthermore, recent literature has shown the detrimental effect that repeat anesthetics have on the cognitive development in children [27].

This study is not without limitations and is subject to the drawbacks of any retrospective review. Because of the retrospective nature of our review, we were not able to

obtain all of the preoperative and postoperative pulmonary function testing that were performed every 6 months on each of the children. Two-thirds of our cohort carried a diagnosis of SMA and by nature of their disease had altered pulmonary function. Many of the children with SMA had typical chest wall deformities with a bell shaped chest, apical narrowing and pectus excavatum. We did seek to document any decline in pulmonary status, which we determined to be the need for a tracheostomy and/or ventilator dependence post spinal surgery. None of the patients in our cohort exhibited this decline.

Specific measurements of sagittal plane, spinal balance, and pelvic obliquity were not specifically studied secondary to limitations of the radiographs and lack of standardization of the radiographs pre- and postoperatively. This can also be attributed to the fact that only one of our patients was able to stand for erect radiographs and the majority of the films were obtained with the patients in the supine position. One SMA Type 2 patient was independently ambulatory at the time of surgery and has maintained independent ambulation at most recent follow-up. All of the remaining children are nonambulatory. All of the patients had pelvic fixation, and on the supine radiographs there was no significant horizontal plane progression observed.

The observed total growth in our cohort versus that expected as calculated by the reported growth rates made by DiMeglio et al. (0.1 cm/y per vertebrae) was 75% for T1–T12 and 88% for T1–S1. Because of the small numbers in our population, a statistical comparison in observed growth in the SMA and non-SMA population was not performed. We report a mean Cobb angle at final follow-up of 23° (2°–57°) with a mean percentage loss of correction of 19%. Other than coronal Cobb measurement, a formal measure of 3D rotational deformity was not performed. Autofusion and subsequent crankshaft can occur for several reasons, including patient and surgeon factors. In our cohort, we observed consistent rates of growth over a medium-long period (2–15 years) of follow-up during periods of peak adolescent growth.

The mechanical stress on the implants is theoretically less in children with SMA, which makes it a favorable disease entity in which to employ this surgical treatment. We were able to demonstrate success in children with other neuromuscular conditions, but not all EOS conditions were studied. Our modification of the Luque technique may have solved the previously reported mechanical issues, that is, rod breakage and autofusion, which may allow this technique to be used in other EOS children; however, further research will be needed.

In summary, sublaminar guided growth without fusion has served the children included in this analysis very well. It is not possible at this time to recommend this strategy for all children with EOS until a prospective multicenter study using multiple techniques, surgeons, and EOS diagnoses is completed to arrive at this conclusion. The children demonstrated acceptable maintenance of curve correction

and continued spinal growth that were not far from the normal growth rates (75% of the normal growth rate for T1–T12 and 88% of the normal growth rate for T1–S1) for the spine. The data support that these modifications to the Luque technique addressed the concerns for autofusion and implant failure with one standalone procedure without the need for multiple distraction-based surgeries. Our results suggest that sublaminar guided growth without fusion is a safe and effective surgical procedure to address EOS, especially in children with low-tone neuromuscular conditions.

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