Health-Related Quality of Life in Early-Onset Scoliosis Patients Treated Surgically: EOSQ scores in Traditional Growing Rod vs. Magnetically-Controlled Growing Rods

Michael E. Doany, BS 1, Z. Deniz Olgun, MD 2, Gizem Irem Kinikli, PT 3, Senol Bekmez, MD 4, Aykut Kocyigit, MD 5, Gokhan Demirkiran, MD 6, A. Ergun Karaagaoglu, PhD 7, Muharrem Yazici, MD 8

Corresponding Author:
Muharrem Yazici, MD
Hacettepe University
Department of Orthopaedics and Traumatology,
Sihhiye, Ankara, Turkey

1 New York Medical College – Valhalla, NY
2 Attending Orthopaedic Surgeon, Children’s Hospital of Pittsburgh of UPMC – Pittsburgh, PA
3 Assistant Professor, Hacettepe University, School of Physical Therapy and Rehabilitation, Ankara, Turkey
4 Attending Orthopaedic Surgeon, Cankaya Hospital, Ankara, Turkey
5 Resident, Hacettepe University, Department of Orthopaedics and Traumatology, Ankara, Turkey
6 Associate Professor, Hacettepe University, Department of Orthopaedics and Traumatology, Ankara, Turkey
7 Professor, Hacettepe University, Department of Biostatistics, Ankara, Turkey
8 Professor, Hacettepe University, Department of Orthopaedics and Traumatology, Ankara, Turkey
Email: mimyazici@gmail.com
Phone: +90-505-619-1351
Fax: +90-312-310-0580

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STRUCTURED ABSTRACT

STUDY DESIGN: Cross-sectional study.

OBJECTIVE: To compare quality of life and caregiver burden in TGR and MCGR patients.

SUMMARY OF BACKGROUND DATA: Magnetic controlled growing rods (MCGR) decrease surgical sessions associated with treatment of early onset scoliosis (EOS), hoping to minimize the burdens seen with repetitive invasive surgeries in traditional growing rod (TGR) treatment. Although the clinical indications for these treatments have largely been agreed upon, there is a lack of understanding of their impact on patients’ and families’ quality of life.

METHODS: Inclusion criteria: ≤ 10 years of age at index procedure, major curve ≥ 30º, no previous spine surgery, minimum 1-year postoperative follow-up. The previously validated 24-Item Early Onset Scoliosis Questionnaire (EOSQ-24) was utilized to assess quality of life. Statistic methods were applied to compare domain scores between TGR and MCGR patients.

RESULTS: 44 children with EOS were enrolled; 25 TGR and 19 MCGR. Groups were similar in gender and age at index surgery. Age at time of questionnaire and mean length of follow-up were significantly different; patients were older (14.0 vs 8.8 years) and had longer follow-up (101.3 vs 34.3 months) in TGR (p<0.01). Deformity correction and complication rates were similar between groups. At the time of questionnaire, scores of economic burden and overall satisfaction in MCGR were significantly superior to those in TGR by univariate analysis. When controlled for duration of follow-up, some domain scores trended towards statistical significance, some remained stable, and others regressed to non-significance.

CONCLUSIONS: Health related quality of life data reveals superior outcomes in overall satisfaction and financial burden domains in the MCGR group. However, the positive effects of MCGR decrease when controlled for length of follow up, indicating that the MCGR is not yet a magic fix-all, and that the TGR remains an option in the treatment of EOS.

KEYWORDS: Early Onset Scoliosis (EOS), Traditional Growing Rod (TGR), Magnetic Controlled Growing Rod (MCGR), Health Related Quality of Life (HRQoL), Early Onset Scoliosis Questionnaire (EOSQ-24), Scoliosis, Pediatric Spine Deformity, Spinal Surgery, Patient Satisfaction, Family Burden, Financial Burden

LEVEL OF EVIDENCE: 3
INTRODUCTION

The management of early-onset scoliosis (EOS) is a major concern for the pediatric orthopedic surgeon due to the disorder’s progressive nature and severe complications, such as cardiopulmonary derangements, functional disability, and poor cosmesis.\textsuperscript{1-4} After the determination that early and extensive spinal fusion causes unacceptable stunting of pulmonary development in children,\textsuperscript{5,6} growth-sparing treatment techniques were developed. Goals of current treatment of EOS include controlling curve progression, preserving spinal balance and improving cardiopulmonary function while remaining respectful of continued spinal growth, and ultimately, improving quality of life for the patient and family.

‘Growth-sparing’ techniques, originally described in the 1960s by Harrington,\textsuperscript{7} and popularized by Akbarnia\textsuperscript{8, 9} and others, are the current gold standard in treatment of EOS.\textsuperscript{10, 11} Traditional growing rod (TGR) implants are placed in the spine during an index procedure where usually only two anchor levels proximal and distal undergo subperiosteal dissection and fusion. The goal is to leave intervening levels intact, so as to continue stimulating growth by recurrent distractions. This is performed with open surgery under general anesthesia on average every 6-9 months as the child grows, in an attempt to maintain normal thoracic spinal growth, and thus, pulmonary development.\textsuperscript{5, 6, 12} Unfortunately, recurrent surgery during childhood has been correlated with negative long-term sequelae from exposure to anesthesia,\textsuperscript{13, 14} complications of the actual operation,\textsuperscript{15} and psychological distress caused by recurrent hospital visits.\textsuperscript{16, 17} The novel magnetically-controlled growing rod (MCRG) was developed in an attempt to decrease surgical sessions associated with TGR, and achieve more natural spinal growth due to frequent non-
invasive distractions. In traditional growing rods, lengthening is done in bursts at each surgical procedure, and therefore does not resemble natural growth which is more continuous. MCGRs, on the other hand, allow the surgeon to mimic biology better by more frequent and more controlled lengthenings. Furthermore, the MCGR allows the clinician to provide periodic distractions in the office without general anesthesia or surgical intervention. The technology was first proved effective in animal models, and has since been shown to be safe and effective in children. While curve correction, thoracic lengthening, and surgical complication rates have been evaluated in support of the use of the MCGR, no studies to date have focused on one of the ultimate treatment goals: improving quality of life.

Health-related quality of life (HRQoL) outcomes measures have become popular in recent literature, and selection of an evaluation instrument specific to the target patient population is critical. The 24-Item Early-Onset Scoliosis Questionnaire (EOSQ-24) was developed and validated as a specific instrument to measure HRQoL in EOS. The final version contains 24 questions spanning 8 domains of HRQoL (General Health, Pain, Pulmonary Function, Transfer, Physical Function, Daily Living, Fatigue, Emotion), Family Satisfaction, and Family Burden. The EOSQ-24 has been validated by several groups and shown excellent inter-patient reproducibility, consistency within etiologic groups, and the ability to track post-surgical changes.
The current study is intended to evaluate quality of life, family satisfaction, and family burden in MCGR and TGR patients using a previously validated version of EOSQ-24. **We hypothesized that the MCGR would be superior to the TGR in most domains.**

**MATERIALS AND METHODS**

Approval for this cross-sectional study was obtained from the Institutional Review Board prior to initiation. At this institution, a university hospital that serves as a reference center for spinal deformity, consecutive patients with EOS undergoing surgical treatment between 2005 and 2015 were enrolled in a prospective database. Those meeting the following inclusion criteria were included in the study: (1) EOS of any etiology, (2) age less than 10 years at index surgery, (3) major curve greater than or equal to 30°, (4) no history of spine surgery prior to growing rod insertion, and (5) a minimum of 12-month postoperative follow-up. Patients were approached during a routine follow-up and the questionnaire offered to those who chose to participate in the study. Parents completed the questionnaire in the presence of either a nurse or resident physician of the orthopaedic department. Demographic data including age at index surgery, diagnosis, radiographic information, and lengthening count was extracted from the aforementioned database after enrollment into study.

Growing rods at this institution are inserted utilizing standard surgical technique as described by Akbarnia et al. Subperiosteal dissection is limited to proximal and distal anchor levels and intervening levels left intact. Lengthenings are performed in the operating room every 6-9 months as outpatient procedures for patients in the TGR group. Magnetic rod implantation is performed
with similar technique, and lengthenings performed every 6-12 weeks in the office. Radiographs are taken before and after every lengthening procedure in both groups. Complications are classified according to the need for unplanned surgical procedures.

Statistical analysis was performed with the aid of SPSS ver. 18 (Chicago, IL, 2009). Differences in demographic, radiologic and surgical data between groups were analyzed by using student’s t-test, Pearson chi-square, Kolmogrof-Smirnof test and/or Fisher’s exact test. Differences in HRQoL scores were analyzed either by student’s t-test or by Mann-Whitney U test. During preliminary statistical analysis, length of follow-up was found to be significantly different between the groups. The effect of follow-up was controlled on quality of life domains by using covariance analysis. 32

RESULTS

Forty-four patients who met the inclusion criteria were approached. All parents and children agreed to participate in the study. The MCGR group included 19 patients (13 female, 6 male) with a mean age at time of questionnaire of 8.8 years (range 5-14), and at index surgery of 6.7 years (range 4-12). Mean follow-up was 34.3 months (range 14-76) and mean number of lengthenings 8.3 (range 3-22). The average pre-operative major curve Cobb angle for the group was 58.5° (range 43-88°), which became 32.5° (range 8-62°) at the time of last follow-up. The TGR group included 25 patients (11 female, 14 male) with a mean age at time of questionnaire of 14.0 years (range 9-18) and at index surgery of 6.1 years (range 3-11). Mean follow-up for this group was 101.3 months (range 56-142) and mean number of lengthenings 9.2 (range 4-15). The
average pre-operative major curve Cobb angle for the group was 42.6° (range 40-104°), which became 42.0° (range 11-77°) at the time of last follow-up. Statistical analysis of demographic data of the two groups revealed a significant difference (p<0.01) in the age at questionnaire and the duration of follow-up. Otherwise, no statistically significant difference was found between the two groups in regards to age at index surgery, pre-index surgery/final follow-up Cobb values, number of lengthenings or unplanned procedures per patient. Cobb angle distributions were tested between the two groups found to be normally distributed according to the Kolmogrof-Smirnoff test (p=0.69 pre-operative, p=0.61 post-operative). These findings are summarized in Table 1.

Statistical analysis revealed two domains to be significantly superior in the MCGR group at the time of questionnaire: Financial Burden and Satisfaction. When EOSQ-24 results were controlled for length of follow-up utilizing univariate analysis of covariance, the following was noted: the projected scores of the MCGR group in the domains of General Health, Pain/Discomfort, and Physical Function approached statistical superiority but did not achieve it, those in the domains of Pulmonary Function, Transfer, Daily Activities, Fatigue, Emotion, Parental Burden and Average Tool Score remained stable and statistically insignificant, and the statistical significance of the scores in the Financial Burden and Satisfaction domains noted at the time of questionnaire became statistically insignificant. Average EOSQ-24 results for the two groups, both at time of questionnaire and when statistically controlled for time of follow-up, are listed in Table 2.
DISCUSSION

Treatment of EOS is a complex problem for the pediatric orthopaedic surgeon. Management goals include correction of spinal deformity, maintenance of normal thoracic growth, improvement of pulmonary function, and ultimately, positively impacting quality of life. The traditional growing rod (TGR) has been the staple of the surgical treatment of EOS since the successful modification of distraction-based fusionless instrumentation by Akbarnia et al. in the late 90s. It has proved itself over the intervening two decades with consistently good correction rates and acceptable complication rates. However, the price of offsetting the disastrous effects of early fusion on an immature thorax and the preservation of growth has been repetitive lengthenings in the operative suite every 6-9 months, resulting in a commitment of 6-18 scheduled surgical procedures at the outset of treatment. The implant-related answer to this has been the development of the magnetically-controlled growing rod (MCGR). The MCGR, in mainstream use for the past perhaps 2-3 years, promises a similar insertion procedure, followed by non-invasive office lengthening procedures every 6-12 weeks, with hopes to achieve less invasiveness and more natural growth. Recent reports with on average 2 years follow-up have been promising, and more studies are ongoing at the moment, many of them comparing the TGR with the new MCGR from a clinical point of view. The socioeconomic and psychological impact of recurrent surgery during childhood is commonly reported as a supporting argument for the use of MCGRs; however, to date there have been no published studies on HRQoL outcomes for patients undergoing MCGR. Our study intends to begin the discussion of how the MCGR, a relatively new implant compared to the more established TGR, affects the patient and the family as a whole. We report on the EOSQ-24 scores of our MCGR patients as compared with our TGR
patients utilizing statistical projection to normalize length of follow-up, which is necessarily
different due to the recent introduction of the MCGR.

The necessity of committing to a great number of surgical procedures brings with itself a number
of problems, most quiteobvious, and some subtle. Operative complications are a glaring
consequence of repetitive surgical procedures. For TGR, as there are planned surgical procedures
twice a year where certain non-urgent complications can be addressed, the term ‘unplanned
surgery’ has been coined to denote problems that cannot wait. Most of these are hardware
related, although high rates of infection have also been reported in TGR. The MCGR, although
it promises fewer planned operations, has been shown to have its share of unexpected trips to the
operating room, or ‘unplanned surgery,’ as well. In a 2014 study by Akbarnia et al., 17 MCGR
patients were matched with TGR patients and outcomes analyzed. No differences in major curve
correction or growth were noted. MCGR patients had 57 fewer operations compared to TGR.
However, the incidence of unplanned surgical revisions as a result of complications was similar
between groups. Several authors have reported similar findings. Reported unplanned surgery
rates in TGRs range from 29-53%, and in MCGRs range from 12-70%. Bess et al. reported on 140 patients with TGRs, and at final follow up the patients underwent an average
of 6.4 procedures, including 74 unplanned procedures. The MCGR complication rates are
derived from smaller cohort studies with far fewer patients, likely contributing to the large range
in reported complications. However, despite the similar or even higher rate of ‘unplanned
surgery’ with the MCGR, average total surgery counts for patients with MCGR range from 1.1-
1.7, while with TGRs the average number of procedures ranges from 6-10 per
patient. All authors reported similar Cobb angle correction and T1-S1 height gain between
TGR and MCGR groups. Our findings of similar unplanned surgery count and Cobb angle corrections in both groups are consistent with previous studies.

The subtler aspect of repetitive surgery is the impact twice-yearly surgical procedures have on the child and family as a whole. Several studies have shown correlations between recurrent surgery during childhood and significant psychosocial pathology. Flynn et al. followed 12 children who underwent an average of six procedures for the management of EOS with TGRs. Twenty-five percent of patients displayed “clinically significant” neurobehavioral dysfunction, 33% scored “at-risk,” and the remaining 42% scored in the normal range. Internalizing problems, including anxiety, depression, and somatization, was the most commonly seen pathology. Similarly, Matsumoto et al. reported an association between recurrent surgery for EOS and increased incidence of aggression and rule breaking. In both cases, two variables were significantly correlated with increased risk of psychological dysfunction: (1) younger age at index surgery and, (2) greater total number of surgeries. Similar psychiatric sequelae have also been demonstrated at a high rate in children undergoing multiple surgeries for orthopaedic trauma, cancer, liver transplantation, and cardiac surgery. Recurrent hospitalization for surgery every six months also places a major financial burden on families and the healthcare system. It is in these areas that the MCGR shows the most promise of improvement. While the index implantation procedure is unavoidable, non-invasive office-based lengthening promises to decrease both the psychological burden of multiple hospitalizations and the financial one upon the families.
Health-related quality of life has been scarcely researched in early-onset scoliosis. The 24-Item Early-Onset Scoliosis Questionnaire has been validated in multiple languages as a reliable and reproducible tool for measuring health status for patients with EOS and the burden on their caretakers.26, 29-31 We report on the EOSQ-24 scores in two groups of our growing-rod patients, 19 of whom were treated with MCGRs and 25 with TGRs. These scores were broken down and compared between groups according to domains, and revealed statistically superior outcomes in the domains of Patient Satisfaction and Financial Burden in the MCGR group. This is understandable, as the MCGR involves fewer trips to the surgical suite. However, as the MCGR is a relatively new implant, it must be remembered that the time of follow-up between the groups is significantly different: 34 vs. 101 months. We theorized that the length of follow-up would affect scores, and with the aid of statistical projection, simulated group scores based on the condition that length of follow-up was statistically similar. Several changes were seen to take place with normalization of follow-up. First of all, Financial Burden and Overall Satisfaction scores in the MCGR group decreased to no longer be statistically superior to the TGR. It is our opinion that this can be explained in that although the MCGR does decrease the number of trips to the surgical suite itself, it does not remove the dependency of the child or the family on the hospital or medical office. For many families, any lengthening procedure, be it in the operating room or in their doctor’s office, includes a lengthy commute to the facility, waiting times, lost productivity, and disruption of daily routines. For most small children, any trip to the physician is wrought with anxiety even if the procedure is benign, and more frequent trips may remind them more strongly of the fact that they are different from their peers, resulting in a further decrease in satisfaction scores. Secondly, scores in the domains of Pulmonary Function, Transfer, Daily Activities, Fatigue, Emotion, Parental Burden and Overall Averages did not
change with length of follow-up. These domains are most likely mainly affected by the condition itself and less by the method of treatment. Thirdly, the MCGR group’s scores in the domains of General Health, Pain/Discomfort and Physical Function increased, approaching but not quite achieving statistical significance. We believe that this phenomenon does reflect the positive impact of significantly lower number of surgical procedures, which would result in less scar tissue, and consequently, better physical function.

The limitations of our study are obvious. The length of follow-up between the two groups is significantly different due to the recent introduction of the MCGR as an implant, and further follow-up with periodic applications of the questionnaire during the course of treatment will be required to ascertain whether the statistical projections we utilized are reflective of the group’s actual progress. Furthermore, patients in the MCGR group were younger at the time of survey administration than those in the TGR group, which may have affected patients’ and families’ outlook on treatment and opinions, skewing results.

In conclusion, this first study comparing HRQoL in TGR and MCGR patients reveals that the MCGR, with its significantly shorter follow-up, has superior domain scores in Financial Burden and Overall Satisfaction, a phenomenon that loses statistical significance when controlled for follow-up. Furthermore, unplanned surgical procedures were seen to be similar in both groups. The MCGR does promise better outcomes in certain domains of the HRQoL as the consequence of fewer planned surgical procedures. However, we believe that many of the questionnaire outcomes are primarily affected by the underlying condition, and despite the
MCGR’s certain inherent advantages, it is not enough to remove the patients’ and caretakers’
dependence on and recurrent trips to a medical facility, the associated stress and anxiety on the
patients, or the loss of productivity on part of the caretakers. Based on these findings, we conclude
that despite the MCGR’s numerous advantages, the TGR is not obsolete yet, and remains an
acceptable choice for the treatment of EOS, at least in the near future.
REFERENCES


Rolton D, Thakar C, Wilson-MacDonald J, Nnadi C. Radiological and clinical assessment of


Table 1. Demographics

<table>
<thead>
<tr>
<th></th>
<th>MCGR (n=19)</th>
<th>TGR (n=25)</th>
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<tbody>
<tr>
<td>Gender</td>
<td>13 F, 6 M</td>
<td>11 F, 14 M</td>
</tr>
<tr>
<td>Age at questionnaire (yr)*</td>
<td>8.8</td>
<td>14.0</td>
</tr>
<tr>
<td>Age at index operation (yr)</td>
<td>6.7</td>
<td>6.1</td>
</tr>
<tr>
<td>Follow-up duration (mo)*</td>
<td>34.3</td>
<td>101.3</td>
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<tr>
<td>Lengthening count (per patient)</td>
<td>8.3</td>
<td>9.2</td>
</tr>
<tr>
<td>Pre-index Cobb (°)</td>
<td>58.5</td>
<td>42.6</td>
</tr>
<tr>
<td>Final follow-up Cobb (°)</td>
<td>32.5</td>
<td>42.0</td>
</tr>
<tr>
<td>Unplanned surgery (per patient)</td>
<td>0.37</td>
<td>0.76</td>
</tr>
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</table>

*indicates statistically different (p<0.01).
MCGR| magnetically-controlled growing rod; TGR| traditional growing rod.
Table 2: Questionnaire results, adjusted for follow-up.

<table>
<thead>
<tr>
<th>Domain</th>
<th>Questionnaire Results (mean)</th>
<th>Adjusted for follow-up (means; 95% confidence interval)</th>
<th>Approach significance</th>
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</thead>
<tbody>
<tr>
<td></td>
<td>MCGR</td>
<td>TGR</td>
<td>p</td>
</tr>
<tr>
<td>General Health</td>
<td>59.9</td>
<td>58</td>
<td>0.703</td>
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<tr>
<td>Pain/Discomfort</td>
<td>71.1</td>
<td>77</td>
<td>0.642</td>
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<td>Physical Function</td>
<td>72.4</td>
<td>57</td>
<td>0.075</td>
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<tr>
<td>Pulmonary Function</td>
<td>86.2</td>
<td>87</td>
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<tr>
<td>Transfer</td>
<td>65.8</td>
<td>51</td>
<td>0.16</td>
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<tr>
<td>Daily Living</td>
<td>50</td>
<td>61.5</td>
<td>0.287</td>
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<tr>
<td>Fatigue/Energy Level</td>
<td>71.1</td>
<td>77</td>
<td>0.421</td>
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<td>Emotion</td>
<td>61.2</td>
<td>52</td>
<td>0.219</td>
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<tr>
<td>Parental Burden</td>
<td>53.4</td>
<td>46</td>
<td>0.308</td>
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<tr>
<td>Financial Burden</td>
<td>61.8</td>
<td>38</td>
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<tr>
<td>Overall Satisfaction</td>
<td>82.9</td>
<td>67.5</td>
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<tr>
<td>Average</td>
<td>66.9</td>
<td>61.1</td>
<td>0.194</td>
</tr>
</tbody>
</table>
Numbers in bold face indicate statistical significance ($p<0.05$).

MCGR| magnetic controlled growing rod; TGR| traditional growing rod.