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ORIGINAL PAPER

Does a delay of surgery due to a multidisciplinary screening process result in neuromuscular scoliosis curve progression in complex Cerebral Palsy?

Barkha Chhabra[1,2] · Dion Birhiray[1] · Lorenzo Deveza[1,2] · Matthew Gremillion[1] · Grant McHorse[1] · Benny Dahl[1,2] · Frank Gerow[1,2] · Darrell Hanson[1,2] · Brian Smith[1,2]

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Abstract

Purpose While surgical intervention of scoliosis in cerebral palsy (CP) patients has shown notable improvements in quality of life, the high risk of post-operative complications in CP patients necessitates careful preoperative optimization. A preoperative multidisciplinary (Multi-D) pathway at our tertiary pediatric hospital in effect since 2014 led to a significant reduction in mortality at one year. However, such a strategy delays surgery, potentially increasing the risk of curve progression. This study aims to elucidate the impact of the Multi-D screening process on curve progression in neuromuscular scoliosis among complex CP patients.

Methods A retrospective review of all CP patients with scoliosis at a tertiary care center from 2012 to 2020 was conducted. This assessment focused on the progression of the major Cobb angle from the time of the indications conference to surgery of patients who went through Multi-D screening. Patient demographics and perioperative variables were obtained from the electronic medical record (EPIC, Systems Verona, WI).

Results After exclusion criteria were met, there were 85 patients who went through Multi-D, 78 of whom had surgery, and seven who did not. Surgery was delayed an average of 202 days for Multi-D optimization. We found a trend in increasing Cobb angle over time, but this correlation did not reach statistical significance ($p = 0.079$). 45 Multi-D surgery participants had a decrease or no change in Cobb angle and had surgery an average of 5.6 months after indications. 33 Multi-D surgery participants had an increase in Cobb angle and had surgery an average of 8.5 months after indications. Cobb angle progressed an average of 13.4° in the increased group, and −0.4° in the decrease or no change group. There were no associations with change in Cobb angle and GMFCS, starting major curve angle, number of referrals, or intrathecal baclofen pump use according to this analysis.

Conclusions Multi-D optimization resulted in an average delay in surgery of 6.7 months. Patients that did not have a change in Cobb angle had surgery within 5.6 months vs. patients that had an increase in Cobb angle had surgery on average 8.5 months after indicated for surgery, with an average increase of Cobb angle of 13.4°.

Level of evidence Level III, retrospective comparative study.

Keywords Cerebral palsy · Neuromuscular scoliosis · Spine surgery · Cobb Angle · Multi-disciplinary conference

Introduction

Scoliosis, prevalent in up to 74% of spastic quadriplegic patients with Cerebral Palsy (CP) [1], poses unique challenges due to its early onset and association with multiple co-morbidities [2, 3]. While surgical intervention has shown notable improvements in quality of life [3–6], the high risk of post-operative complications (particularly pulmonary, excessive bleeding, and deep infections), in CP patients

✉ Barkha Chhabra
bnchhab2@texas.childrens.org

1 Baylor College of Medicine, Department of Orthopedics, Houston, TX, USA

2 Department of Orthopedics, Texas Children's Hospital, Houston, TX, USA

necessitates careful preoperative optimization [7–11]. Recent findings from the HARMS Study Group [9], a consortium of spine surgeons specializing in pediatric spinal deformity surgery and research, have demonstrated that this risk is associated with both preoperative major curve angle as well as nutritional and pulmonary function [1, 9–11].

Recent publications have further demonstrated benefits to preoperative multidisciplinary (Multi-D) planning and clearance in spine surgery in adults [12]. However, there are limited studies detailing the formal Multi-D protocol and examining these benefits in complex paediatric spine patients [8, 13, 14]. At our institution's tertiary paediatric hospital, five step Multi-D process for all non-idiopathic scoliosis patients indicated for surgery has been in effect since 2014. Results reviewed by the institutional Spine Study Group concluded that a Multi-D process led to a significant reduction in mortality at one year, providing an important safety process to reduce serious complications after scoliosis surgery [14].

However, such a strategy delays surgery, therefore possibly increasing the risk of a significant curve progression. It has been demonstrated that in patients with adolescent idiopathic scoliosis (AIS) a delay of more than six months results in significant progression of the scoliosis [15]. Additionally, a recent multi-centre study concluded that delaying surgery to a curve greater than 90° in CP patients increases the risk of infection, blood loss, and the need for anterior procedures [1].

This study aims to elucidate the delay in timing of surgery and impact the Multi-D screening process has on curve progression in neuromuscular scoliosis among complex CP patients. The research hypothesis was that there would be a statically and clinically significant increase in curve magnitude in patients with CP undergoing Multi-D optimization prior to surgical treatment.

Materials and methods

A retrospective review of all patients with CP indicated for scoliosis surgery from 2012 to 2020 was conducted using our Electronic Medical Record (EMR) registries. Inclusion criteria comprised patients indicated for Posterior Spinal Fusion and Instrumentation to the pelvis, including those requiring pre-operative/intra-operative halo-femoral traction and some with intrathecal baclofen pumps and/or necessitating releases with posterior spinal fusion. Exclusions involved patients with prior surgeries, transfers of care, age < eight years, Spondylolisthesis, and scoliosis types other than neuromuscular (Congenital Scoliosis, Adolescent Idiopathic Scoliosis, and Syndromic Scoliosis) and insufficient x-rays at the designated Multi-D intervals. Surgeons were all fellowship trained Paediatric Orthopaedic or Spine surgeons. A total of 156 individuals were reviewed and of those, 85 patients went through Multi-D planning and met inclusion criteria (78 patients had surgery, 7 patients did not have surgery). The Multi-D planning pathway is illustrated in Fig. 1.

Primary outcomes included the major Cobb angle of the most recent radiograph completed before the first presentation at Indications conference, the major Cobb angle of the most recent radiograph done before the surgery date, and the major Cobb angle at the time from the Indications conference to surgery as well the time between Indications conference to surgery. Scoliosis radiographs were obtained using a standard Posterior Anterior collimation protocol, to superiorly include C7 and inferiorly include the sacral region

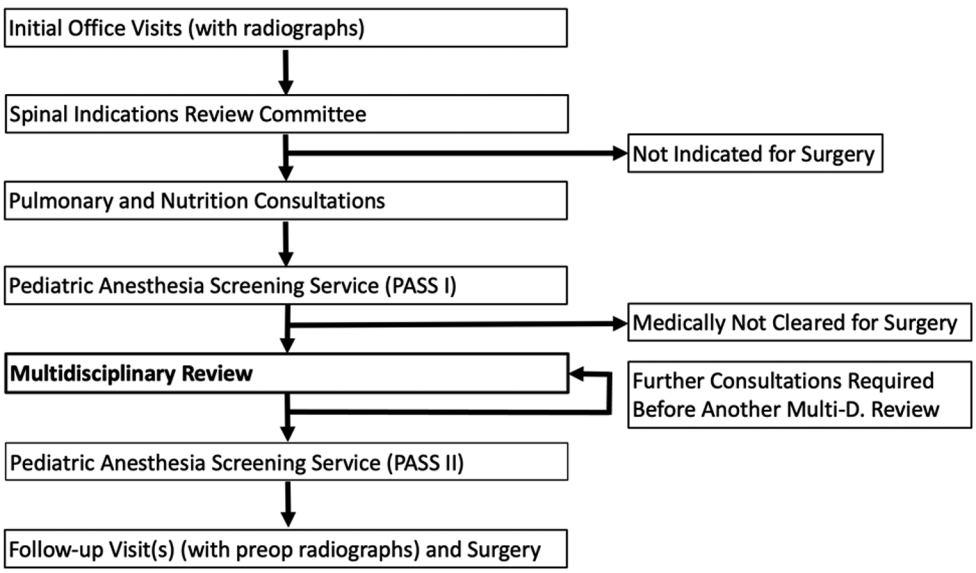

Fig. 1 Multidisciplinary pathway for patients with neuromuscular scoliosis at our tertiary paediatric hospital, including those with cerebral palsy. Completion of this pathway is mandatory for patients to undergo surgical correction of their scoliosis

S1 or the level of the femoral heads with visualization of 3–5 cm of the iliac crests. Radiographs were obtained with arms on the side and patient erect with equal weight on both feet when possible. Secondary outcomes included the number of referrals/consultations requested and GMFCS categorization. Patient demographics and other pre-operative and post-operative outcome variables and characteristics were obtained from the EMR (EPIC, Systems Verona, WI). The institution's IRB approved this research- IRB #H-43,238.

Statistical analysis

The statistical relationship between change in Cobb angle and various variables were reviewed: Pearson correlation for continuous variables, Spearman Rank Correlation for Ordinal variables, and Logistic Regression for Binary variables. Statistical significance was determined for $p < 0.05$. Statistics was performed using Graphpad Prism Software (version 6, San Diego, CA) and RStudio (Boston, MA).

Results

Patients were categorized into those who went through the Multi-D conference and had surgery, and those who went through the Multi-D conference and did not have surgery. Of the 85 patients that went through the Multi-D pathway, 78 patients ended up having surgery and seven patients did not have surgery as of the final data collection date. Demographics for our cohort are summarized in Table 1. As seen in Table 1, for those who went through the Multi-D conference and had surgery, the average time from Indications conference one to the Date of Surgery was found to be 202.1 days. The time from Indications conference one to the last Multi-D conference date was an average of 155.1 days. The time from the completion of the Multi-D screening to the Date of Surgery was an average of 47 days. This was found by subtracting the average time between Indications conference one and the last Multi-D conference date from the average time between Indications conference one and the Date of Surgery.

The primary outcome was to determine if there was a change in Cobb angle over time in patients who underwent a Multi-D conference. Figure 2 demonstrates a trend in increasing Cobb angle over time, but this correlation did not reach statistical significance ($p = 0.079$), likely due to various limitations in our study. Our secondary outcome measures were to determine a correlation between related patient variables and change in Cobb angle. We did not find that time, Cobb angle at indications, # of preoperative referrals, GMFCS, and whether or not the patient had surgery was correlated with a change in Cobb angle. The relationship between our secondary outcome measures and the change in Cobb angle is summarized in Table 2.

Lastly, we sought to compare surgery patients who underwent Multi-D screening and had an increase in Cobb angle to those who underwent Multi-D screening and had a decrease or no change in Cobb angle. Population demographics and our comparative analysis for Cobb angles are captured in Tables 3 and 4, respectively. We found no significant difference in the Pre-Multi-D Cobb angles between groups, but there was a statistically significant difference in the time to surgery. Patients who had no change or decrease in Cobb angle had surgery within 5.6 months, while patients who had an increase in Cobb angle had surgery approximately 8.5 months after they were first indicated for surgery. Patients who had an increase in Cobb angle progressed 13.4° on average, while patients who had a decrease or no change in Cobb angle progressed −0.4° on average. The relationship between Cobb angle increase and Cobb angle decrease, or no change is summarized in Table 4. Additionally, we examined the potential association between intrathecal baclofen pump treatment and Cobb angle progression, as detailed in Table 5, finding no statistical significance in our analysis ($p = 0.65$).

Table 1 Demographics of the operative patients

Number	78
Age (years)	13.5 ± 3.0
BMI	17.9 ± 3.2
GMFCS	5 [3–5]
Number Referrals	3.3 ± 0.7
levels fused (number)	16.3 ± 2.0
Time to Last Multi-D	155.1 ± 151.7
Time to surgery (days)	202.1 ± 160.9

Discussion

The implementation of a formal Multi-D Screening process in the preoperative optimization of CP scoliosis patients was found to delay surgery for patients an average of 6.7 months. This delay in surgery did not always result in curve progression. Over 50% of patients in this study did not have a change in Cobb angle during Multi-D screening. Patients with no change in Cobb angle were likely to have surgery an average of 5.6 months after indicated for surgery. Preoperative Cobb Angle was found to be 90.2° in this cohort. Recent studies in this patient population have found that being proactive (Cobb < 70°) has no advantage in terms of decreasing risks or improving outcomes compared to curves 70°-90° and so our patients that had surgery within 5.6 months were found to be outside this clinically relevant threshold [1].

GMFCS level ranged from 3 to 5 in our study, with the majority of our patients being GMFCS 5 and requiring

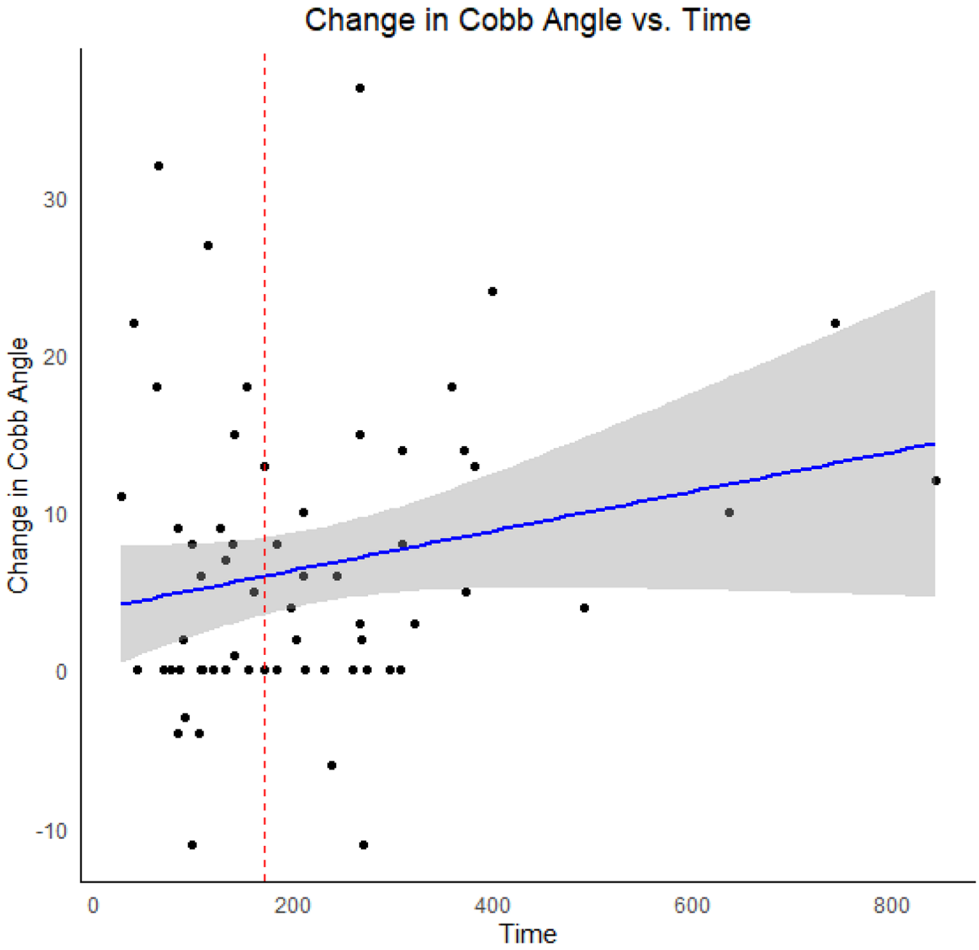

Fig. 2 Change in Cobb angle vs. time. Correlation coefficient = 0.2, P-value = 0.079. The dashed vertical line is the median time to surgery

Table 2 Relationship between change in Cobb angles in various factors

Change in Cobb angle vs.	Correlation coefficient	*p*-value
Time (days)	0.2	0.079
Cobb angle at Indications	-0.185	0.102
Cobb angle at Surgery	0.316	0.005
Referrals	-0.026	0.831
GMFCS	0.051	0.662
Surgery	-0.039	0.22

Table 3 Baseline characteristics and demographics for substudy cohort of patients who underwent Multi-D and had surgery

	No change or decreased	Increased
Population (n)	45	33
Age (years)	13.5 ± 2.8	13.9 ± 2.56
BMI	19.9 ± 3.8	18.9 ± 3.1
GMFCS	4.7 ± 0.53	4.6 ± 0.89
Number Referrals	3.3 ± 0.63	3.3 ± 0.77
levels fused (number)	16.5 ± 0.62	16.7 ± 0.46
Time to first Multi-D	139.2 ± 145.9	183.3 ± 184.6
Time to surgery (days)	171 ± 96.3	257.8 ± 209.9

Table 4 Relationship between variables and increase vs. no change or decrease in Cobb angles when a Cobb angle change is greater than or equal to 5º

	No Change or Decreased	Increased	*p*-value
Number	45	33	
Time (days)	171.0 ± 96.3	257.8 ± 209.9	0.0343
Pre-Multi-D	90.6.8 ± 22.0	86.27 ± 17.6	0.3616
Post Multi-D	90.2 ± 22.9	99.7 ± 18.1	0.0541
Change	-0.4 ± 3.2	13.4 ± 8.03	< 0.0001

Table 5 Relationship between intrathecal baclofen pump treatment and Cobb angle increase

	OR (95% CI)	*p*-value
Baclofen Pump	0.8 (0.30–2.11)	0.65

surgery for sitting, balance and transfers. Higher GMFCS levels often correlate to more medically fragile patients with greater Pulmonary, Gastrointestinal, Nutrition, and Cardiac co-morbidities [3]. However, there was no association with change in major Cobb angle and GMFCS classification in our study group.

Moreover, there was no association between the change in major Cobb angle and the number of preoperative referrals. This may be due to the fact that the majority of the

CP patients required the same referrals (most commonly to Pulmonary and Nutrition). Further, despite referrals and Multi-D coordination, some risk factors are modifiable while others are not. For instance, low BMI or pulmonary conditions may be optimized over time. One study found the high risk of complications, most commonly deep infection, is related to prolonged intubation [9]. They concluded that nutritional and preoperative pulmonary function, as well as preoperative Cobb angle, may be modifiable risk factors [9]. However, risk factors such as Cardiac conditions or an impaired coagulation pathway often can not be changed. Particularly, seizure medications are known to lead to coagulation pathways and yet can not be safely held pre-operatively [7]. Therefore, obtaining a Neurology or Cardiology referral may not result in a significant delay in time but rather involve coordination or planning of post-operative specialized consultations.

However, 42% of the patients in our study were found to have a statistically significant increase in Cobb angle. These patients had surgery an average of 8.5 months after indications with an average change Cobb angle of 13.4°. While the starting Cobb angle was not found to be different between the No Change and the Change in Cobb angle groups, the patients that had an increase in Cobb angle started at 86.3° and ended at 99.7.° Multi-centre studies in this patient population have found that delaying surgery to a curve greater than 90° increases the risk of infection, blood loss, and the need for anterior/posterior procedures [1]. In a different study, combined ASF/PSF in patients with CP accounted for only 9.3% of surgical cases but was associated with the longest hospital stay, highest charges, and increased complications in their group. Ideally, surgery should be recommended for curves less than 90° [1], and delaying surgery for Multi-D optimization may have led to an increased risk of complications in 42% of our patient population.

Recent literature in this high-risk cohort has identified a substantial opportunity for quality improvement in reducing the high perioperative medical and surgical complication rates that lead to lengthy and costly hospital admissions [8, 10, 11]. Furthermore, the U.S. News and World Report ™ now reviews CP NM Scoliosis questions as separate from idiopathic scoliosis surgery, and there is an incentive for institutions caring for these complex patients to optimize patients for surgery and not have them re-admitted for medical reasons within 30 days or re-operation within 90 days. However, there are varied approaches for optimizing the perioperative pulmonary, haematological, and nutritional status as well as reducing wound complications in complex CP scoliosis patients. To our knowledge, no formal optimization process has been employed by multi-centre groups for paediatric CP scoliosis patients [8]. Implementation of Multi-D strategies in the treatment of adult spinal deformity has been found to lead to better patient outcomes [12]. We believe there is a need for further studies of systematic quality improvement strategies to improve safety in pediatric complex CP spinal surgery.

This study examined the possible consequences of curve progression from the implementation of a formal Multi-D pathway that was developed for all non-idiopathic scoliosis patients prior to surgery. It is a five-step process that can take months to years to complete (Fig. 1). The first step was an indications review where the Spine Surgeon Committee agrees that surgery is indicated. Subsequent steps serve to ensure that all necessary consultations have been made prior to surgery and as another mechanism for the intra-operative and post-operative care teams to be notified of upcoming patients so they can anticipate needs and plan accordingly. To our knowledge, this is the first study we are aware of that studies the delay in time and potential curve progression that Multi-D optimization may result in.

This retrospective cohort study comparing the radiographic measurements of major curve angle of patients at multiple time points has several limitations to note. While we were able to see a trend in increasing Cobb angle over time, this correlation did not reach statistical significance ($p = 0.079$). This may be because we were underpowered to find a statistical correlation, or there is none. However, the former is likely true, as many of our CP patients were excluded from the study. New radiographs were not always available at every time point. Several surgeons routinely only obtained bending or traction films preoperatively, and therefore, patients without a new radiograph prior to surgery were excluded from our study, reducing the number of participants for this cohort. Recently, it has also come to light that intrathecal baclofen pumps are associated with an increase in scoliosis progression [16, 17]. It follows that these patients will likely have a more challenging spine fusion. Our assessment of this relationship within our cohort found no significant association between the two; however, this lack of significance may be attributed to our sample population being insufficiently powered. Finally, the data collection was limited by the accuracy of the medical record in documenting all referrals, consultations, and interventions. Despite this, our cohort is one of the largest series, as CP Neuromuscular scoliosis that is indicated for surgery is generally not common.

The accuracy of our research could be improved by conducting a multi-centre study, and further work is needed to see the extent of the cost paid by the inevitable chronologic delay and resultant curve progression. For example, there may be an association between the progression of the major curve angle with intra-operative Spinal surgery complexity. Patients that go through Multi-D may also have increased pelvic obliquity and need pre-operative halo, or

intra-operative femoral-halo traction at a greater rate. Furthermore, the incidence of release and anterior/ posterior spinal surgery may be increased, and an increase in curve complexity may be related to increased blood loss. We also recommend future clinically relevant studies to further investigate how to best decrease the time from indication to surgery would be clinically relevant.

Numerous studies have shown that the surgical treatment of scoliosis in patients with CP results in a significant improvement in health-related quality of life [3–5]; despite this, surgery for these children is a major undertaking. With the Multi-D approach, each patient has the potential to be medically optimized, but nearly 60% of patients may have a clinically and statistically significant increase in their Cobb angle. We recommend that all factors should be considered for each patient individually while attempting to weigh the costs vs. benefits ratio of the delay in surgery that occurs with the Multi-D screening process vs. more prompt surgical intervention at the time surgery is indicated.

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Author contributions All authors contributed to the study conception and design. Material preparation, data collection and analysis were performed by Dr. Barkha Chhabra, Dr. Matthew Gremillion, Dr. Grant McHorse. Statistical analysis and manuscript preparation was performed by Dr. Lorenzo Deveza and Mr. Dion Birhiray. The first draft of the manuscript was written by Dr. Chhabra and Dr. Deveza and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

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Data availability The data supporting the findings of this study can be made available upon reasonable request from the authors.

Declarations

Ethical approval This is a retrospective study authorized by the Institutional Review Board for Baylor College of Medicine and Affiliated Hospitals under reference protocol #h-43,238.

Consent for publication Not applicable.

Informed consent This is a retrospective series of chart review and informed consent was not required.

Competing interests Lorenzo Deveza – Lento Medical: Stock or stock options; Benny Dahl: K2M Educational activities and advisory board. The remaining authors declare that they have no conflict of interest.

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