

REVIEW ARTICLE

Resurgence of Serial Casting in Early-onset Scoliosis: Is It “Old Wine served in a New Bottle”?

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ABSTRACT

Casting as a treatment of scoliosis has been practiced since 1800s. However, the practice fell into disfavor following the serious chest deformities and thoracic constrictions secondary to casting, as well as tremendous improvements in the surgical modalities. Recently, there has been a resurgence of this technique in early-onset scoliosis (EOS) and it has been considered to be a definitive treatment modality and a delaying tactic prior to the inevitable surgery. It carries the triple advantages including correction of the spinal deformity, allowing spinal growth uninterrupted as well as low complication rates. The current article elaborately discusses the role of serial casting in EOS.

Keywords: Early-onset scoliosis, Nonsurgical treatment, Serial casting.

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INTRODUCTION

In the United States, scoliosis is classified as infantile if diagnosed below 4 years of age and juvenile, between 4 and 10 years.¹ In the United Kingdom, any scoliosis diagnosed less than 5 years of age is defined as early-onset deformity.^{1,2} Although these lexical terminologies may differ, the primary purpose of such distinctions is to emphasize upon the gravity of scoliotic deformities occurring at these young ages. The prognosis of such deformities is not encouraging, nevertheless, timely surgical and nonsurgical interventions can potentially ameliorate the outcome.²

The options available to pediatric spine surgeons range from nonsurgical interventions including bracing and casting, to surgical procedures like growth modulating

surgeries and fusion techniques.^{2,3} The need for multiple surgeries, associated surgical complications, and the growing understanding of the concept of “diminishing returns following repeated surgeries” on a growing spine have led to the recent resurgence of the age-old philosophy of serial casting, as a modality to correct deformities as well as delay the inevitable surgery in early-onset scoliosis (EOS).^{2,4} The current article comprehensively reviews the existing literature on the role of serial casting in EOS, and discusses the pros and cons of this modality in comparison with the other management options.

HISTORY OF CASTING

The earliest report on the use of plaster of paris (POP) casting in scoliosis treatment was published in 1877 when Lewis Sayre published his experience with traction and serial casting in correcting spinal deformities.⁵ This was followed by the presentation by Bradford and Brackett in 1893 describing a modified technique involving application of a lateral pressure during casting for achieving deformity correction. In their report of five cases, one patient had a straight spine at the end of 1 year.⁶ Once surgical fusion was introduced, Hibbs described casting as a technique (turnbuckle cast) to achieve preliminary correction preoperatively, as well as to maintain correction postoperatively until fusion was complete.⁷ In 1955, Risser described a detailed technique of cast application on special table frame. The cast had a window cut posteriorly through which the surgical fusion was later performed.⁸ After the advent of segmental spinal instrumentation, the need for postoperative casting became obsolete. The role of casting as the sole modality of management of scoliosis also gradually fell into disfavor with improvements in surgical techniques.⁴

The interest in casting techniques for EOS revived with the strong suggestion by Cotrel and Morel in 1964 that serial casting involving derotational forces on the thoracic cage [elongation, derotation, lateral flexion (EDF)] could be very effective in EOS.⁹ The first formal report on serial casting for idiopathic EOS was presented by Mehta and Cotrel in 1979 at the “Sixth Symposium on Scoliosis”. They reported encouraging outcome in 21 patients with idiopathic EOS with cast application.^{4,10} In 2005, Mehta published her prospective study on the role of casting in infantile scoliosis. She reported complete resolution of the deformity in young

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children with moderate curves and reduction in the magnitude of severe curvature in older children.⁵ Similar later reports have also reconfirmed the role of casting both as a definitive treatment and as a modality to delay definitive surgical intervention in EOS.¹¹⁻¹³

Rationale behind the Resurgence of Casting

The goal of treatment in EOS is to control the progression of spinal deformity, without compromising spinal, thoracic, and pulmonary growth.^{2,14,15} Dimeglio¹⁶ demonstrated that the rate of normal thoracic spine growth is highest until the age of 5 years (1.4 cm/year), which corresponds to 50% of spine growth. Any restriction of growth of spine during this period can potentially jeopardize pulmonary development. Recent literature has indicated that the later a child undergoes growth-sparing surgeries, the fewer the complications encountered and lesser the overall number surgeries until definitive fusion.^{13,17,18} Bess et al¹⁷ reported 13% decrease in complication rates for every year increase in the patient's age at which the lengthening device is initially implanted and 24% additional complication risk for each surgical procedure undergone. Sankar et al¹⁸ purported that the Cobb's angle correction diminished with each lengthening after initial implantation. Growth-sparing surgeries are, therefore, safer and more effective when performed in older children.^{13,17,18} Mehta⁵ explained that the philosophy underlying casting involved guiding the early rapid growth of the initially curved spine, so as to gradually straighten its orientation.

Indications of Casting

Casting may be typically recommended in cases of EOS (age <3 years) with progressive major curvature or rib vertebral angle difference (RVAD) >20° at presentation.¹⁹

Progressive vs Resolving Curves and Thoracic Insufficiency Syndrome

Mehta classified resolving from progressive scoliosis by using the RVAD and rib phase. She suggested that progressive curves carry poor prognosis, with 57% of curves attaining a magnitude of 70° by the age of 5 years.^{5,10,20} Such large curves cause restrictive lung disease or thoracic insufficiency syndrome with reduced thoracic and lung volumes, decreased alveolar development and compromised lung function and ultimately lead to respiratory failure, pulmonary hypertension, cor pulmonale and early death.^{21,22} Early fusion surgery also grossly restricts the thoracic growth and ultimately results in similar and grave pulmonary aftermaths. This is the rationale behind the need for techniques which allow the continued growth of spine and thorax and simultaneously restrict the progression of spinal deformity in EOS.^{23,24}

Casting Table and Patient Positioning

The table typically stabilizes the patient with pelvic and head traction, allowing access to trunk shoulder and pelvic girdles. Risser and Cotrel frames^{8,9} can be used, although they are relatively larger for small children. Risser typically used the technique of "localizer casting" where the frame stabilized the patient using head halter and pelvic tractions, along with a localizer that pushed on the apex of curvature.⁸ Mehta designed a table (marketed by AMIL), which supports the head, arms, and legs, while leaving the trunk free.¹⁹ The Salt Lake Shriners Hospital used a custom table, which would support the child in traction while leaving the body free for casting.¹¹

General anesthesia is required especially in young children. Intubation is also advised as cast molding can increase thoracic pressure and restrict ventilation.¹¹ Head halter and pelvic traction stabilize the child and narrow the body during cast application. Although traction can reduce the curve, the body recoils after the removal of traction unless the cast supports the occiput or mandible too. A mirror may be positioned under the table so as to carefully visualize the posterior cast.

Evolution of Casting Techniques

The most popular technique of casting was developed by Risser,⁸ who advised three-point bending to correct. The correction of angular and translational deformities was thus achieved using pressure over the apex. This resulted in the cast directly pushing the ribs toward the spine and thereby constricted the space available for the lung. The technique developed by Cotrel and Morel⁹ involved the use of broad cloth straps to rotate spine. Here, the posteriorly rotated ribs were rotated anteriorly to recreate normal chest contour and counter rotations were applied through pelvic mold and upper trunk. This technique was termed EDF method. Mehta also used EDF casting technique and advised ambulatory casts to maintain correction of curve.¹⁹

Windows

Sanders recommended an anterior window to relieve the chest and abdomen, while the posterior window on the concave side allows the concave ribs to move posteriorly, so that the curve gets corrected without deforming the chest wall.¹¹

Casting Protocol and Techniques, Weaning and Bracing (Cast Holidays)

Plaster of paris or fibreglass material may be used. The POP has better moldability and also tends to expand while setting, in contrast to fibreglass which contracts. Pelvic portion is considered the foundation of the cast

and is well-molded. Cotrel/Morel and Mehta's techniques used an over-the-shoulder modification of cast.^{9,19} Sanders recommended that curves below T8 could be maintained on underarm casts, whereas higher curves needed mandibular extension.¹¹ Lumbar curves need to be casted with hips in flexion to decrease lumbar lordosis and facilitate correction.¹¹ A silver impregnated shirt or a thin layer of webril with felt over the bony prominences or crape paper (removed after casting) may generally be used as the innermost layer.^{11,19}

Mehta recommended changing of cast under anesthesia every 8 to 16 week, until the curve resolved followed by underarm brace.⁵ She advised weaning of the brace if the correction persisted. Sanders et al¹¹ advised cast change based on child's growth rate: Every 2nd month for those less than 2 years, every 3rd month for those aged 3 years and every 4th month for children aged 4 years and above. When the curves improved to 10° or less, molded brace was applied under anesthesia. They also recommended application of brace in select patients during summer and resumption of casting during fall. When the curve is progressive and approaches 70°, casting may be discontinued in favor of growing rods. Cast discontinuation may also be needed in case of any direct complications related to cast or to manage other medical conditions, such as asthma, abdominal surgeries, and other respiratory illnesses.¹³

Outcomes of Following Serial Cast

Younger age, curves <50°, and idiopathic curves carry better prognosis. In 2008, Tsuji et al^{13,25} documented a mean improvement in Cobb's angle from 55.6 to 22.9° and a mean delay of 2.4 years from initiation of casting to surgery in their cohort of 36 patients (10 idiopathic, 21 syndromic, and 5 congenital).

Fletcher in 2010.^{2,14,15} described a mean improvement in Cobb's angle from 65.7 to 37.9° and 3.1 year average delay time to surgery with serial casting (36 EOS patients – 13 idiopathic and 23 syndromic/congenital). Around 39% of patients in both these series progressed to surgery. In the study by Waldron et al²⁶ involving 16 patients with EOS - 8 idiopathic and 8 syndromic), Cobb's angle improved 73 to 45° and 31% progressed to surgery.

Sanders et al¹¹ published in 2009 that 27% of patients (excluding cases of neuromuscular scoliosis) with cast application resolved completely; 56% showed improvement, although did not entirely correct; 14% stabilized and 3% progressed despite cast application. Around 10% of their patients underwent surgery; however, in the subgroup of patients who had an initial curve larger than 50°, 28% progressed to surgery. Casting delayed surgery in these patients by a mean of 2.7 years.

In a retrospective study by Baulesh et al,¹³ 53 patients underwent serial casting between January 2005 and August 2010. Among the 36 patients who underwent at least 2 serial casts and were followed up for at least 6 months, 19 had idiopathic scoliosis, 14 had syndromic/neuromuscular scoliosis, and 3 had congenital scoliosis. Full resolution of deformity occurred in 17% (6/36 patients), 53% (19/36) were observed in braces, and 31% (11/36) underwent surgery. One patient underwent spinal fusion, while 10 underwent growth-sparing surgeries [(8 vertical expandable prosthetic titanium ribs (VEPTR) and 2 growing rods)]. Surgery was delayed by an average of 2.1 years from the time of initial casting.

Recently, Canavese et al²⁷ reviewed a cohort of 44 patients (36 idiopathic and 8 nonidiopathic) with EOS and reported that serial EDF casting under general anesthesia and neuromuscular blocking drugs were more effective than casting with anesthesia alone or without anesthesia. He suggested that RVAD and apical vertebral rotation were the best measures to follow the deformity. In this series, casting could delay surgery by a mean of 2 years.

Factors Associated with Poor Outcome Following Serial Casting

Mehta reported that casting is more likely to be unsuccessful if started beyond 2 years of age. She observed that 1 year is usually the time needed for casts to result in deformity correction.²⁸ Four patterns of patient phenotype were noted by Mehta: (a) Sturdy phenotype, (b) slender phenotype, (c) named syndromes, and (d) unknown syndromes.⁵ In young patients with smaller curves, all phenotypes had good outcome with serial casts. However, in older children with larger curves, the prognosis significantly worsened across the groups: Sturdy to slender to syndromic phenotypes.¹⁰ In such older and syndromic patients, the primary goal of casting is to delay surgery until spine and lungs have attained sufficient growth.^{13,15}

Casting in Idiopathic Scoliosis vs Nonidiopathic Scoliosis

Baulesh et al¹³ observed that deformity correction occurred in 26% (5/19) of patients with idiopathic scoliosis, compared with 6% (1/17) in nonidiopathic group. This difference was, however, not statistically different. Nonidiopathic patients had a significantly higher surgical rate (9/17), as compared to idiopathic scoliosis.^{2,19} A significantly higher surgery-free survival time was also observed in the idiopathic patients (85% at 48 months), as compared to the nonidiopathic patients (15% at 48 months). In these patients, overall post management mean Cobb's angle decreased by 9° in comparison with

overall mean premanagement angle. Between the groups, idiopathic patients experienced a 22° greater improvement in Cobb's angle in comparison with their nonidiopathic counterparts. In general, it was observed that although correction was achieved in both idiopathic and nonidiopathic groups, loss of correction/deformity progression occurred in nonidiopathic group at the final follow-up. A significant increase in T1-T12 height was observed in both idiopathic and nonidiopathic groups (Overall t-height velocity of 1.56 cm/year, t-height velocity in idiopathic group was 1.59 cm/year and t-height velocity in nonidiopathic group 1.52 cm/year). Demirkiran et al²⁹ concluded that serial EDF was an effective technique in delaying surgery in their cohort of 11 patients with congenital scoliosis (mean delay of 2.1 years).

Complications of Casting

Casting is not essentially a benign procedure and the goals and risks of casting must typically be balanced against the risks of undergoing an early surgery, on a case-by-case basis. Casting can potentially lead to complications including pressure sores, nausea/vomiting, rib and mandibular deformities, thoracic constriction, cast syndrome, and parental dissatisfaction.^{2, 8} Most of these complications are traditionally associated with indiscriminate casting of all types of scoliosis with improper technique.¹¹ Repeated need for use of anesthetics in young children may also predispose to complications.³

Tsuji et al^{13,25} described an overall complication rate of 10% from casting. Fletcher et al^{2,14,15} also described 19% complication rate following serial casting. Baulesh et al¹³ reported a complication rate of 19% (7/36), of whom five patients had nonfatal pulmonary complications requiring discontinuation of casts and two patients had superficial skin irritation.

It is still unclear if the benefits of casting outweigh the risks involved (restriction of chest) in patients with neuromuscular or paralytic scoliosis. Similarly, the problems like sleep apnea, aspiration, and gastroesophageal reflux disease can also be worsened by cast applications.³ Another major risk, which is yet to be evaluated, includes the psychological effect of patients undergoing casting at a very young age.³

Casting vs Bracing

Bracing is the most common nonoperative treatment for EOS. However, there are specific problems related to this technique. Braces are difficult to fit in young children. Unless the braces are sufficiently flexible, repeated application can be hard. Poor compliance may thus be a major concern. The chest wall and ribs are so pliable in young children that using the 3-point bend on apical rib

with braces can lead to chest deformities which ultimately compromise the thoracic volume.^{11,30,31}

Casting compared with Other Techniques (Growth Modulation) and Law of Diminishing Returns

The three major growing rod techniques include growing rods, VEPTR, and growth guidance using Shilla technique.¹⁷ Prospective studies have indicated very high incidence of complications with these techniques. Around 18 to 22 cm gain of thoracic height is possibly the optimal target to be achieved following multiple lengthening procedures, nevertheless, the complications proportionately rise with greater number of lengthening performed.^{11, 32} Spontaneous fusions occur following growth rod application, which negatively affect the outcome.^{1,33} "Law of diminishing returns" is another prime concern with multiple distractions, as the effective lengthening possibly happens not beyond the initial 3 to 4 years.¹⁸ It, therefore, makes complete sense in delaying the growing instrumentations until 6 or 7 years of age through the use of nonsurgical modalities, so that the need for final fusion does not arise at least until 10 years of age.¹¹

Johnston et al,³⁴ in a matched cohort controlled study, showed that casting provides similar control of deformity progression as compared to growing rods, albeit with lesser complication rates. The cast group demonstrated one complication (out of 27 patients), as against 15/27 complications observed in growing rod group. Among the 27 casted patients, 15 eventually were operated after a mean 1.7 years of casting. Unlike surgery, casting also does not interfere with spinal growth.²⁷ McKenzie et al³⁵ reported the incidence of surgical site infections of 0% at growing rod insertion to 29% during the revision lengthening or surgeries. The psychological impact of multiple spine surgeries on a growing child can also not be understated.³⁶ Hemolytic-uremic syndrome casting needs to be considered as an alternative to all other growth sparing-surgical and nonsurgical modalities, whenever possible.

CONCLUSION

Casting is gradually becoming an inevitable component of management of EOS. The EDF technique involving derotation provides the best correction, without causing any thoracic deformation.

The goals of casting typically may include:

- Complete resolution of curve whenever possible
- Substantially reducing the curve magnitude in severe deformities
- Delaying fusion until sufficient pulmonary maturity is attained.

Casting has been clearly beneficial in idiopathic scoliosis, although it definitely has been demonstrated to

delay surgery in nonidiopathic (congenital/syndromic) scoliosis. Casting in neuromuscular scoliosis has not been sufficiently studied and the decision needs to be made on a case-by-case basis.

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