Infantile Idiopathic Scoliosis: Outcomes of Brace Treatment until Skeletal Maturity or Spinal Fusion

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Abstract

Background: Serial casting under general anesthesia, which is considered as a gold standard of treatment for patients with infantile idiopathic scoliosis (IIS), can lead to significant negative neurodevelopmental effects. Therefore, the appropriateness of this type of treatment is controversial. Brace treatment is one alternative method of treatment for IIS patients. However, long-term studies have not yet verified its effectiveness. Thus, the present study aimed to evaluate the effectiveness of brace treatment in patients with IIS until skeletal maturity or spinal fusion.

Methods: The medical records of all IIS patients with the referral age of 0-3 years who received brace treatment from June 1986 to November 2013 were reviewed. Those patients with pre-brace Cobb angle > 20° were included and followed up to skeletal maturity or the time of spinal fusion. The Cobb angle was recorded at the time of diagnosis before the initiation of bracing, weaning time, brace discontinuation, and final follow-up. In addition, the maximum in-brace curve correction was measured.

Results: Out of 87 patients with IIS, a total of 29 cases (19 males and 10 females) with the average curve magnitude of 35.62° at the time of diagnosis were included in the study. The average best in-brace correction was 57.32% for successfully treated patients and 36.97% for progression/surgery patients. Based on the results, brace treatment failed for a total of 20 patients (69%), with a scoliosis curvature progress ≥ 45°. Of these patients, 12 cases (60%) reached spinal fusion. Finally, four patients (13%) in the surgery-treated group underwent surgery before the age of 10.

Conclusion: The results revealed that bracing was successful for more than two-thirds of patients with IIS curves, preventing surgery before the age of 10.

Level of evidence: IV

Keywords: Brace treatment, Infantile idiopathic scoliosis, In-brace correction, Skeletal maturity, Spinal fusion

Introduction

Infantile idiopathic scoliosis (IIS) is described as scoliosis of unknown causes appearing in infants aged 0-3 years (1) and includes 0.25-12.8% of all types of idiopathic scoliosis (2). According to natural history studies, 70-92% of infantile curves are resolvable, especially among patients younger than one and in those with milder curves at the time of diagnosis, while the scoliosis is presumably progressive for the patients older than one and those with double major curves (1, 3, 4). If left untreated, patients with progressive IIS are exposed to pulmonary compromise due to deformity in the chest wall. Therefore, treatment should be initiated immediately after diagnosis. Serial elongation derotation flexion (EDF) casting and bracing are the most common methods of non-operative treatment for IIS cases, and serial casting is known as the standard
of care (5). The highest success rate of serial casting was reported in the study of Mehta, where the infantile curves of 100% of patients were treated before age two years (6). Casting is typically performed under general anesthesia and should be changed every 6-12 weeks (1). However, repeated anesthesia can have a negative effect on neurodevelopment in early childhood. In December 2016, the Food and Drug Administration issued a “Drug Safety” announcement warning that general anesthesia and sedation drugs in children under three years of age “may affect the development of the child’s brain” (7). In addition, various studies have emphasized the toxicity of general anesthesia to children’s nervous system (8-10). Therefore, the usefulness of serial casting in treating children with IIS is controversial.

The early use of a brace (with the ability to make frequent adjustments) can eliminate the need for repeated anesthesia. Accordingly, bracing is recommended for those patients with progressive scoliosis curvatures of larger than 20° (11). However, to the best of our knowledge, no study has evaluated the long-term efficacy of brace treatment for IIS patients. In a short-term study, Thometz and Liu revealed that bracing can potentially be successful in controlling IIS curves. However, the long-term effect of bracing, until skeletal maturity, was not reported (12). Therefore, the present study aimed to evaluate the outcomes of brace treatment in IIS until skeletal maturity or spinal fusion.

Materials and Methods

Patients

The present case series, the retrospective chart review study regarding the effect of brace treatment on the curve progression of IIS patients, was approved by the Ethics Committee of Iran University of Medical Sciences (case no.1398.381). To this end, from June 1986 to November 2013, the medical records of all patients with IIS who were treated with a brace were reviewed to obtain data. The inclusion criteria were the referral age of 0-3, Cobb angle more than 20 degrees at the initiation of brace treatment, following treatment up to the end of skeletal maturity (Risser sign > 4) or spinal fusion. On the other hand, all patients with missing brace treatment data and the follow-up, those who discontinued their treatment before reaching skeletal maturity, and those who were still under treatment were excluded from the study.

The brace treatment for patients with IIS mainly seeks to control the curve progression and prevent spinal fusion before the age of 10 (13). Therefore, the average scoliosis Cobb angle at the initiation of brace treatment was considered the primary goal of bracing for patients with IIS curves > 45°.

Brace treatment

During the chart review, different data including the patients’ sex, age (at the time of diagnosis, before the initiation of bracing, and before surgery), and the Cobb angle (at the time of diagnosis, before the initiation of bracing, best in-brace, the initiation of weaning, brace discontinuation, final follow-up, and before surgery), brace wearing time, follow-up duration, curve pattern, and the brace type were extracted. The maximum in-brace curve correction was evaluated based on Jarvis et al. (14). The physician recorded brace compliance by questioning the patient’s parents and the appearance of the brace in every routine visit of the patients (with four- to six-month intervals). To evaluate the results of brace treatment based on the curve severity, the Cobb angle before the initiation of bracing was divided into two groups of ≤ 30° and ≥ 31°. For all patients, the largest curve at the initiation of brace treatment was considered for statistical analysis.

Based on the criteria of the Scoliosis Research Society, brace treatment is considered successful if the curve severity at the final follow-up was measured < 46° (15). The patients were divided into “success” or “failure” groups with regard to treatment outcome.

Statistical analysis

Statistical analyses were conducted using SPSS software, version 17 (SPSS Inc., Chicago IL, USA). Descriptive statistics were reported as mean, standard deviation, and range. Additionally, the chi-squared test and the independent-sample t-test were performed for nominal and continuous variables, respectively. Eventually, the analyses were conducted between the success and failure groups. A p-value < 0.05 was considered for statistically significant differences.

Results

Patients

Amon the 87 cases with IIS, 58 were excluded from the study. Of these, 16 cases were still under treatment while 42 cases had lost the final follow-up [Figure 1]. Finally, a sample of 29 patients (10 girls and 19 boys) with an average age of 18.58 ± 8.90 months at the time of diagnosis met the inclusion criteria (male to female ratio of 2:1). At the initiation of brace treatment, a custom-made body-jacket was prescribed for all of the patients. Further, adequate space in the anterior chest wall of the orthosis was considered to maintain lung function and allow the expansion of the chest during breathing. A perineal strap was further added to prevent the superior migration of the orthosis on a patient’s body. After ensuring the patient’s active involvement in the treatment process, the physician prescribed a Thoracolumbosacral Orthosis (TLSO) for scoliosis curves with apex vertebra up to T8 or Milwaukee brace for main thoracic curves with an apex above T8. In general, 21 (72%) and eight (18%) patients were treated with TLSO and Milwaukee brace, respectively. The average scoliosis Cobb angle was 35.6° ± 11.6° (range 15°–60°) and 37.7° ± 11.1° (range 20°–60°) at the initial visit and at the initiation of brace treatment, respectively. Overall, 16 patients (55.1%) had left thoracic curve pattern and three cases (10.4%) had a right thoracic curve pattern. Furthermore, seven patients (24.1%) had a right thoracolumbar curve pattern and three (10.4%) had left thoracolumbar curve patterns. Table 1 presents a summary of the patients’ characteristics from the initial
Figure 1. Flow diagram illustrating the included and excluded patients in the present study.

<table>
<thead>
<tr>
<th>Patient no.</th>
<th>Gender</th>
<th>Curve pattern</th>
<th>Age</th>
<th>Cobb angle</th>
<th>Wear- ing time (yr)</th>
<th>Follow- up duration</th>
<th>Brace type</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>2</td>
<td>Left Th</td>
<td>26</td>
<td>33</td>
<td>28</td>
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<td>Left Th</td>
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<td>19</td>
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<tr>
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<td>23</td>
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<td>TLSO</td>
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<tr>
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<td>Left Th</td>
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<td>30</td>
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<tr>
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<tr>
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<tr>
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<td>TLSO</td>
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<td>Left Th</td>
<td>3</td>
<td>11</td>
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<td>17</td>
<td>Milwaukee</td>
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<td>Right Th</td>
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<td>1</td>
<td>Left Th</td>
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<td>Left Th</td>
<td>15</td>
<td>18</td>
<td>28</td>
<td>15</td>
<td>TLSO</td>
</tr>
</tbody>
</table>

Th indicates thoracic; T/L, thoracolumbar; DC, discontinue; TLSO, Thoracolumbosacral orthosis.
visit to the final follow-up or spinal fusion. At the baseline examination, five patients (17%) demonstrated an asymmetric or diminished lower extremity/abdominal reflexes. However, the following magnetic resonance imaging revealed no remarkable intraspinal pathology.

**Result of brace treatment**

In general, the scoliosis curve progressed in 20 patients (69%), 12 of which (60%) reached spinal fusion. According to the Scoliosis Research Society, the effectiveness of bracing for the samples of the present study was 31%. As shown in Table 2, the mean scoliosis Cobb angle at the initial visit, the initiation of brace treatment, and the weaning time was significantly lower in the success group compared to the failure group (P<0.05). In fact, no significant differences were observed at the initial visit and initiation of treatment between the two groups in terms of the patient’s age (P>0.05). Based on the results, in-brace correction was significantly higher (P<0.05) in the success group compared to the failure group (57% and 37%, respectively). Table 3 provides the characteristics of patients in the success group at different stages of treatment.

Among the studied patients, 41% (12 of 29) of cases underwent spinal fusion, who had a mean age of 11.50 ± 4.48 (within the range of 3-18 years) and the mean curve magnitude of 65.5° ± 8.8° (ranging from 52° to 81°) at the time of surgery. Regarding surgery patients, four cases (13%) underwent spinal fusion before the age of 10 years. These patients had a curve magnitude of 55° to 70° at the time of surgery. Considering the factors such as the patient’s age, severity of the curve, functional status, and parent’s choice, the decision for the operative treatment of a patient with IIS may be difficult and challenging. However, the existence of a curve size > 50° is considered as the routine recommendation for spinal fusion in IIS.

Table 2. The mean age and scoliosis Cobb angles at different stages of brace treatment between the success/stable and progression/surgery groups (n=29).

<table>
<thead>
<tr>
<th>Variables</th>
<th>Success (n=9)</th>
<th>Failure (n=20)</th>
<th>P</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at baseline (month)</td>
<td>21.33 ± 7.51</td>
<td>17.35 ± 9.37</td>
<td>0.30</td>
</tr>
<tr>
<td>Age at initiation of bracing (month)</td>
<td>27.33 ± 7.41</td>
<td>24.60 ± 7.69</td>
<td>0.37</td>
</tr>
<tr>
<td>Cobb angle at baseline (°)*</td>
<td>28.77 ± 8.46</td>
<td>38.70 ± 11.76</td>
<td>0.03</td>
</tr>
<tr>
<td>In-brace curve correction (%)</td>
<td>57.32 ± 13.56</td>
<td>36.97 ± 18.94</td>
<td>0.007</td>
</tr>
<tr>
<td>Cobb angle at initiation of bracing (°)</td>
<td>30.00 ± 7.03</td>
<td>41.25 ± 11.00</td>
<td>0.01</td>
</tr>
<tr>
<td>Cobb angle at brace discontinue (°)**</td>
<td>28.33 ± 6.30</td>
<td>57.45 ± 12.04</td>
<td>&lt;0.001</td>
</tr>
</tbody>
</table>

*Largest curve was measured.
**For surgery group, preoperative Cobb angle was considered.

Table 3. Results of the patients with stable/improved curve (n=9)

<table>
<thead>
<tr>
<th>Variables</th>
<th>Mean (SD)</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td>Baseline (month)</td>
<td>21.33 (7.51)</td>
</tr>
<tr>
<td></td>
<td>Initiation of bracing (month)</td>
<td>23.73 (7.41)</td>
</tr>
<tr>
<td></td>
<td>Menarche (yrs.)</td>
<td>12.00 (1.15)</td>
</tr>
<tr>
<td></td>
<td>Weaning (yrs.)</td>
<td>14.00 (1.00)</td>
</tr>
<tr>
<td></td>
<td>Skeletal maturity (yrs.)</td>
<td>15.55 (0.72)</td>
</tr>
<tr>
<td></td>
<td>Cobb angle (°)</td>
<td>Baseline</td>
</tr>
<tr>
<td></td>
<td>Initiation of bracing</td>
<td>30.00 (7.03)</td>
</tr>
<tr>
<td></td>
<td>Initiation of weaning</td>
<td>23.88 (6.17)</td>
</tr>
<tr>
<td></td>
<td>Brace discontinue</td>
<td>28.33 (6.30)</td>
</tr>
<tr>
<td></td>
<td>Final follow-up</td>
<td>31.66 (6.36)</td>
</tr>
<tr>
<td></td>
<td>Brace wearing time (yrs.)</td>
<td>13.55 (1.33)</td>
</tr>
<tr>
<td></td>
<td>Follow-up duration (month)</td>
<td>22.88 (4.56)</td>
</tr>
</tbody>
</table>

SD indicates standard deviation.

Table 4. Impact of pre-brace curve type and curve magnitude on effectiveness of bracing

<table>
<thead>
<tr>
<th>Variables</th>
<th>Success n/N (%)</th>
<th>Failure n/N (%)</th>
<th>P</th>
</tr>
</thead>
<tbody>
<tr>
<td>Curve pattern</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Thoracic</td>
<td>8/19 (42%)</td>
<td>11/19 (58%)</td>
<td>0.49</td>
</tr>
<tr>
<td>Thoracolumbar</td>
<td>1/10 (10%)</td>
<td>9/10 (90%)</td>
<td>0.01</td>
</tr>
<tr>
<td>Curve magnitude</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>≤30°</td>
<td>5/9 (55%)</td>
<td>4/9 (45%)</td>
<td>0.73</td>
</tr>
<tr>
<td>≥31°</td>
<td>4/20 (20%)</td>
<td>16/20 (80%)</td>
<td>0.007</td>
</tr>
</tbody>
</table>
with a thoracolumbar curve pattern compared to those with a thoracic curve pattern (44.8° and 38.2°, respectively). Figure 2 displays the related data.

As shown in Table 4, nine (31%) and 20 (69%) of patients were observed with the pre-brace Cobb angle ≤ 30° and the Cobb angle ≥31°, respectively. The progression rate was significantly higher in the patients with a major Cobb angle of ≥ 31° compared to those with a Cobb angle ≤ 30° (80%). Therefore, it is noteworthy that the success rate is only 20% if an infantile curve magnitude is more than ≥ 31° at the initiation of brace treatment.

Discussion

In the non-operative treatment of IIS, the outcomes are considered excellent if the patients never needed spinal fusion. However, the outcomes are considered good if the patients need surgery during adolescence (13). The present study evaluated the effectiveness of bracing in IIS patients until skeletal maturity or spinal fusion. The results demonstrated that the success rate of brace treatment was good to excellent for 77% of the patients. In addition, 13% of the patients underwent surgery before the age of 10 years.

Serial casting, which is performed under general anesthesia, is recognized as the most effective non-operative treatment of IIS. One of the advantages of casting is that the patient or his/her parents cannot remove it at any time. Thus, its compliance rate represents an increase (12). However, serial casting has several disadvantages. For instance, the cast should be replaced at the interval of 8 to 16 weeks, which should be performed under general anesthesia to apply the optimum corrective forces to the trunk (6). Further, repeated anesthesia can have negative effects on the neurodevelopmental status of the infants and their parents' quality of life (8). Furthermore, the sensitive area cannot be readily inspected when pressure or friction sores occur, since no verbal communication exists between the parents and the infants. A decrease in physical contact between the infant and parents, which can affect the maternal or paternal attachment attitudes, is considered as another concern about prolonged serial casting which can affect the quality of life among parents and infants. The lack of physical contact raises concerns which may have a negative impact on the social development of growing children in the long term (12). Therefore, the usefulness of serial castings for the treatment of IIS is questionable. The early use of a brace (with the ability to make frequent adjustments) can eliminate the need for repeated anesthesia, increase the daily inspection of the infant’s body, and the adjustments to the areas of excessive pressure or friction, and allow cleaning the inside of the brace. Additionally, it helps the parents to embrace and touch their child more easily, which can improve the quality of life of parents and infant for a short time (12). Other benefits of early bracing over casting include low weight, affordable, further convenience, and possibility of its doffing for bathing the baby.

Some studies evaluated the effectiveness of brace treatment in the IIS. However, the follow-up period in these studies was short and the patients were not evaluated until skeletal maturity. For example, Smith et al., in a study on 17 infants with IIS, found that 47% of patients were successfully treated with the brace (11). In addition, Thometz and Liu evaluated the effect of serial CAD/CAM bracing on nine patients with IIS and reported that the curve magnitude of four patients reached less than 10 degrees, and the average Cobb angle improved from 57° to 21° for the other five patients (12). However, the results of this study were only reported for a two-year period while not describing the long-term outcomes of bracing (until skeletal maturity). To the best of our knowledge, the present study is the first one in which patients were reviewed until skeletal maturity or spinal fusion. The results demonstrated that the average in-brace curve correction was 49% and 40% in patients with the Cobb angles of ≤ 30° and ≥ 31°, respectively. In the study of Thometz and Liu, the rate of in-brace curve correction for patients with Cobb angles of < 30° was 81%, which was significantly higher than the obtained value in the present study (12). On the other hand, the results of the present study regarding the Cobb angles of ≥ 31° are in line with those of the above-mentioned study (40%). However, considering the long duration of the treatment process for IIS patients, the long-term results of wearing a brace highly differs from the short-term results.

The findings of different studies on idiopathic scoliosis revealed that some parameters such as age at the initiation of bracing, the pre-brace Cobb angle, the curve pattern, and the value of in-brace curve correction can influence the outcomes of the treatment. In the present study, the severity of the scoliosis curve at the initiation of bracing was significantly lower in the success group compared to the failure group. Further, the in-brace curve correction was significantly higher in the success group compared to the failure group. Based on the results of the previous studies on adolescents with idiopathic scoliosis, the thoracic curve pattern is considered a potential risk
factor for the failure of brace treatment. The results of
the present study indicated that 58% of patients with
thoracic curve patterns experienced curve progression.
However, the value of curve progression in patients with
thoracolumbar curve pattern was 90%. Furthermore,
the results represented that the curve magnitude at the
initiation of bracing was significantly higher in patients
with a thoracolumbar curve pattern compared to those
with thoracic curve patterns (44° vs. 38°). Therefore, the
magnitude of a scoliotic curve at the initiation of brace
treatment is regarded as a major risk factor for further
curve progression in patients with IIS. In addition,
the results demonstrated that the maximum in-brace
correction occurs during the first year after initiating the
brace treatment, which may be related to the viscoelastic
changes in the curve over time. Thus, the value of
immediate in-brace correction (i.e., curve correction
that occurs immediately after wearing the brace) is
not an important predictor for the positive outcome of
bracing in IIS patients. These results corroborate those of
Welborn et al. (13).

The patient’s age at the initiation of treatment is
regarded as another important predictor of success in
the non-operative treatment of IIS. Mehta found that the
scoliosis curves were resolved for 100% of cases among
those who had a mean age of 19 months at the initiation
of casting (6). However, for those patients with a mean
age of two years and six months at the initiation of
treatment, casting could only control the progression of
the curve, among which 35.7% finally underwent spinal
fusional. In another study, Iorio et al. concluded that the
earlier age of the child at the initiation of treatment leads
to a higher success rate of treatment with casting
(16). On the other hand, Welborn et al. found no
relationship between the age at the initiation of casting
and the success rate of treatment (13). The results of
the present study are consistent with those of Welborn
et al. In the present study, the mean age at the initiation
of treatment was 21 and 17 months in success and
failure groups, respectively. Considering the differences
in the pre-brace curve magnitude between failure and
success groups (38° vs. 28°), the curve magnitude is a
major risk factor for curve progression in IIS compared
to the patient’s age at the initiation of brace treatment.
Furthermore, the finding was confirmed by Iorio et al.
and Sanders et al. (16, 17). However, Welborn et al.
reported that the curve magnitude at the initiation of
treatment is not an important predictor for the success
rate of serial casting (13).

The present study had several important limitations.
This study was a retrospective review of a 27-year period.

Thus, the retrospective nature of the study inherently
limits the quality and quantity of data in patients’ clinical
records. Brace compliance is another factor affecting the
success of brace treatment in idiopathic scoliosis, which
was emphasized in various studies (18-20). Subjective
and objective methods are used to evaluate brace
compliance. Regarding the objective method in which a
reliable thermal or pressure data logger is used, more
accurate information is obtained from the daily wearing
hours of the brace. Lavelle first introduced the use of
these sensors in 1996 (21). The measurement of objective
compliance was impossible due to the retrospective
nature of the present study in which the patient’s medical
records were evaluated from 1986 to 2013. However,
at each follow-up during brace treatment, the treating
physician recorded the average hours of brace wearing
by evaluating the appearance of the brace and asking the
patients and parents. In addition, comparing the results
with those of the casting method was impossible since all
patients were treated with a brace. Finally, considering
the retrospective nature of the study, the statistics were
limited to the data available in the clinical records of the
patients although the evaluation and documentation of
the information of all cases were consistently conducted
by a treating physician (M. S. G.).

In conclusion, Bracing is a good strategy to preclude the
curve progression during infantile and juvenile years.
The long-term success rate of brace treatment for IIS was
31% when considering final curve magnitude of ≤ 45°.

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