Original article

Detorsion night-time bracing for the treatment of early onset idiopathic scoliosis

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ABSTRACT

Background: Management for early onset scoliosis has recently changed, with the development of new surgical procedures. However, multiple surgeries are often required and high complication rates are still reported. Conservative management remains an alternative, serial casting achieving excellent results in young children. Better compliance and improvement over natural history have been reported with night-time bracing in adolescent idiopathic scoliosis (AIS), but this treatment has never been reported in early onset idiopathic scoliosis (EOS).

Methods: All patients treated for progressive EOS by detorsion night-time bracing (DNB), and meeting the Scoliosis Research Society (SRS) criteria for brace studies were reviewed. Recommendations were given to wear the DNB 8 h/night and no restriction was given regarding sports activities. Radiological parameters were compared between referral and latest follow-up. Based on the SRS criteria defined for AIS, a similar classification was used as follows to analyze the course of the curves: success group: patients with a progression of 5° or less; unsuccessful group (progression or failure): patients with a progression > 5°, patients with curves exceeding 45° at maturity, or who have had recommendation for/undergone surgery, or patients who changed orthopaedic treatment, or who were lost to follow-up.

Results: Thirty-three patients were included (21 girls and 12 boys), with a median Cobb angle of 31° (Q1–Q3: 22–40). Age at brace initiation averaged 50 months (Q1–Q3: 25–60). Median follow-up was 102 months (Q1–Q3: 63–125). Fifteen patients (45.5%) had reached skeletal maturity at last follow-up. The success rate was 67% (22 patients), with a median Cobb angle reduction of 15° (P < 0.001). Four patients stopped DNB due to an important regression. Eleven patients were in the unsuccessful group (33%). Only one had surgery. All patients remained balanced in the frontal plane and normokyphotic. Initial curve magnitude and age at brace initiation appeared to be important prognostic factors.

Conclusions: DNB is an effective conservative treatment, which can be considered a delaying tactic in the management of EOS. This brace offers potential psychosocial and compliance benefits, and allows unconstrained spinal and chest wall growth, resulting in normokyphosis at maturity.

Level of evidence: Therapeutic study (retrospective consecutive case series): Level IV.

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1. Introduction

In 1954, James et al. [1] defined three types of idiopathic scoliosis based on the age of onset: infantile scoliosis (IIS, 0 to 3 years of age), juvenile scoliosis (JJS, 4 to 9 years), and adolescent scoliosis (AIS, >10 years). Early onset idiopathic scoliosis (EOS) has been defined as a significant spinal deformity beginning before the age of five and remains a challenging condition to treat [2]. The age of onset is an important prognostic factor, since major thoracic curves before the age of five are more likely to be associated with thoracic insufficiency syndrome and increased mortality [3]. Treatment can be life saving, and its goal is to control the deformity, allow spinal and chest wall growth, lung development and improve pulmonary function. The role of surgery in the management of early onset scoliosis has increased in the past decade, with the development of growing rods, Vertical Expandable Prosthetic Titanium Ribs (VEPTR, Synthes Spine, West Chester, PA, USA) and growth guidance (Shilla) procedures [4,5]. However, multiple surgeries are required and high complication rates, above 50%, have been reported [6].

Initial non-operative management remains an alternative option, since excellent results have been reported in very young children with curves less than 60°, with serial casting to maintain correction [7]. The Milwaukee brace has also been proposed, but results remain controversial, with high impact on childhood activities, and worsening of sagittal curves has been reported [8]. Recently, the effectiveness of part-time bracing, such as Charleston and Cheneau braces, has been emphasized in AIS and JJS [9,10] with an improvement of quality of life and compliance [11]. The purpose of this study was to determine the effectiveness of detorsion night-time bracing (DNB) in the treatment of EOS.

2. Materials and methods

2.1. Patients

All patients with progressive EOS, treated at our institution by DNB between 1996 and 2008, were included. Data were included prospectively. The clinics follow-up schedule was every six months and two weeks after each brace replacement. Two surgeons reviewed all patients. They all met the following SRS criteria for brace studies [12]: curves greater than 25°, or greater than 20° but with progression of more than 5° in the last six months, Risser 0–2, no prior treatment, and a minimum 2-year follow-up. Patients were screened to ensure that there were no confounding congenital anomalies or comorbidities which may have contributed to the spinal deformity. All non-idiopathic early onset scoliosis were excluded.

2.2. Detorsion night-time brace

All braces were made by the same orthotist (TechnicOrtho, Paris, France), using cast molding before 2001, and optic molding and computer-aided design after 2001. The underarm braces were made of polyethylene, and worn at night for a minimum of 8 h in the supine position (Fig. 1). Correction in the frontal plane was based upon the bending principles that were described for the Charleston brace, and pads were placed at the apex of the curves to obtain maximal correction. Attention was paid to correct the ilolumbar angle by placement of one pad against the opposing hip and a second pad at the apex of the lumbar curve. Maximum correction obtainable with the brace was used, and inversion was obtained in the most flexible curves. A posterior slit on the concave side permitted correction by gravity in the sagittal plane, and allowed unconstrained development of the thoracic spine. Vertebral detorsion was sought by applying orthogonal forces on the chest wall.

Supine radiographs were performed while the patient was wearing the brace and verified by the surgeon during the first fitting visit. Parents were educated and implicated in the treatment. No sensor was used to control compliance. Replacement of the brace was indicated on a 12-months basis, or if the patient’s trunk height had increased by more than 5 cm. No restriction was imposed regarding sports activities.

2.3. Radiological analysis

Patients were evaluated every six months using standing posteroanterior and lateral radiographs, performed after 3 nights out of the brace. All films were digitized, then analyzed by the same investigator using previously validated software (SpineBalance, Surgiview, Paris, France) [13].

The following parameters were measured in the coronal and axial planes:

- Cobb angles of the different curves;
- T1 tilt angle (angle between a horizontal line and the upper endplate of T1, with positive value when the left side of the endplate is higher than the right);
- shoulder tilt (angle between the tangent to the superior edge of the clavicles and a horizontal line, with positive values when the left shoulder is higher than the right);
- global coronal balance, distance between the center of T1 and the center sacral vertical line (CSVL);
- iliolumbar angle (angle between the upper endplate of L4 and a line joining the lower extremities of the sacroiliac joints);
- apical vertebral rotation (AVR), according to Nash and Moe [14].

In the sagittal plane, the following parameters were measured [15,16]:

- thoracic kyphosis, measured from the upper endplate of T4 to the lower endplate of T12;
- lumbar lordosis, measured from the upper endplate of L1 to the upper endplate of S1;
- sagittal vertical axis (SVA), distance between the C7 plumbline and the posterosuperior corner of the sacrum.

Due to lack of criteria defined for EOS, based on the SRS criteria [12] defined for AIS, a similar classification was used as follows to analyze the course of the curves:

- success group: patients with a progression of 5° or less;
- unsuccessful group (progression or failure): patients with a progression > 5°. patients with curves exceeding 45° at maturity, patients who have had recommendation for/undergone surgery, patients who changed orthopedic treatment, or patients who were lost to follow-up.

For patients with curves exceeding 45° at initial examination, a regression was not a success if curves at maturity was still above 45°.

2.4. Statistical analysis

Two-tail paired t-tests were used to compare initial and latest radiological measurements. Unpaired t-tests and Mann–Whitney U-tests were used to compare groups. A P value < 0.05 was considered to be significant. All statistical analyses were conducted using the software Statview (SAS Institute Inc, Cary, NC, USA).

3. Results

3.1. Patients

Thirty-three patients were included (21 girls and 12 boys), with a median age at brace initiation of 4 years and 2 months (median in months: 50; Q1–Q3: 27–60, range: 20–72) and with a median Cobb angle of 31° (Q1–Q3: 22–40; range: 20–60). There were 21 thoracic curves, 5 thoracolumbar curves, 4 lumbar curves, and 3 double major curves. Median follow-up was 102 months (Q1–Q3: 63–125). At latest examination, 15 (45.5%) had reached skeletal maturity (Risser 3 or more), and 14 patients were still wearing a brace.

3.2. Frontal and axial planes

Comparison between radiological parameters at brace initiation and last follow-up is reported in Table 1. Median Cobb angle significantly decreased (P = 0.013) at final follow-up in the overall
population, and the global coronal balance was significantly improved \(P<0.01\).

According to criteria defined above, the success rate was 67\% (22 patients), with a median Cobb angle reduction of 15° (3–27) \(P<0.001\). Eleven patients were in the unsuccessful group (33\%). Six had an elongation–derotation–flexion cast due to poor compliance (2 patients) or for a progression >10° in a six-month period (4 patients). One underwent arthrodesis at maturity for a thora- columbar curve of 50°. Four were still with DNB with a median progression of 16°. Three patients (9\%), exhibiting a main Cobb angle >50° at final follow-up, had an important Cobb angle at initial examination (40°, 40°, 60°).

The unsuccessful group was older (58 months vs 42 months) and had more important main Cobb angle (35° vs 28°) at the initial examination (Table 2). These differences are not statistically significant. There is no difference in sex ratio and type of curve between the two groups.

From 15 patients who had reached skeletal maturity (Risser 3 or more), 4 were in the unsuccessful group, but only one had surgery.

From 18 patients who had not reached skeletal maturity, 12 were in the success group. Due to important regression, 4 were even under observation.

3.3. Sagittal plane

Sagittal plane measurements are summarized in Table 3. Thoracic kyphosis at referral averaged 23° (13–29) with 22% hypokyphotic patients according to Lenke’s classification (i.e. <10°) [17]. At latest follow-up, thoracic kyphosis had significantly improved \(P=0.008\), and all patients were normokyphotic (i.e. between 10° and 40°). Median lordosis also increased significantly, from 48° (Q1–Q3: 41–50) to 58° (Q1–Q3: 50–63) \(P=0.007\). Global sagittal axis, evaluated by SVA, remained unchanged at latest follow-up.

4. Discussion

From a case series of 33 EOS patients, at an average of 10 years follow-up, DNB shows a very good efficacy with 2 patients with a progression <5° and 19 with a regression of the main Cobb angle (Fig. 2). Initial curve magnitude and age at brace initiation appeared to be important prognostic factors.

4.1. Surgical treatment

Management for early onset scoliosis has changed in the past decade, with the development of spinal growing rods and chest wall-based surgical procedures. Fletcher et al. [16] showed that new operative techniques have rapidly been adopted by POSNA members, with an equal number of surgeons using growing rods and casting to manage all types of early onset scoliosis, and with more than one-third using chest wall expansion devices. Nevertheless, most of the surgeries concerned non-idiopathic scoliosis. However, the variability in treatment preference and the lack of consensus, even among surgeons with much experience in this field, have recently been highlighted [18]. The aim of the DNB is

Table 1
Frontal and axial planes radiological parameters of the 33 patients. CSVL: Center Sacral Vertical Line, RVA: axial rotation of the apical vertebra.

<table>
<thead>
<tr>
<th></th>
<th>Brace initiation Median (Q1–Q3)</th>
<th>Latest follow-up Median (Q1–Q3)</th>
<th>(P)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Main Cobb angle (in degrees)</td>
<td>31 (22–40)</td>
<td>27 (10–40)</td>
<td>0.013</td>
</tr>
<tr>
<td>T1 tilt (in degrees)</td>
<td>4.6 (2–8)</td>
<td>4 (1–7)</td>
<td>NS</td>
</tr>
<tr>
<td>Shoulder tilt (in degrees)</td>
<td>2.5 (2–5)</td>
<td>2 (0–3)</td>
<td>NS</td>
</tr>
<tr>
<td>T1-CSVL (mm)</td>
<td>19 (7–18)</td>
<td>15 (9–20)</td>
<td>0.01</td>
</tr>
<tr>
<td>Llilolumbar angle (in degrees)</td>
<td>5 (2–5)</td>
<td>2 (1–7.5)</td>
<td>NS</td>
</tr>
<tr>
<td>RVA (in degrees)</td>
<td>12 (5–9)</td>
<td>11 (3–11)</td>
<td>NS</td>
</tr>
</tbody>
</table>

Table 2
Characteristics of success group or unsuccessful group (Fisher test).

<table>
<thead>
<tr>
<th></th>
<th>Success (67%) n=22</th>
<th>Non-success (33%) n=11</th>
<th>(P)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at DNB initiation (months)</td>
<td>42 (25–60)</td>
<td>58 (35–70)</td>
<td>NS</td>
</tr>
<tr>
<td>Median (Q1–Q3)</td>
<td>28 (20–38)</td>
<td>35 (28–40)</td>
<td>0.2</td>
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</table>

In axial plane, rotation of the apical vertebra (RVA) did not progress during follow-up.

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to avoid several surgical procedures and to improve the compliance and quality of life of young patients. The results of this study prove the effectiveness of conservative treatment in EOS.

In a survey regarding 265 patients from the Growing Spine Study Group database, Yang et al. reported that surgical management was recommended after failure of conservative treatment, and for curves over 60° in patients under 10 years of age [5]. Indeed, the age at surgery was quite young in that series, averaging 6 ± 2.5 years, confirming a shift away from conservative management and the corresponding increase in surgical indications. Drawbacks of surgical techniques include high complications rates with unplanned procedures, need for multiple surgeries, progressive diminution of length achieved (law of diminishing returns) at each lengthening, autofusion of the spine and increased burden of care on the family [19].

4.2. Conservative treatment

EOS treatment objectives should include the maximization of pulmonary function, spine length, and residual mobility, while minimizing hospitalizations and psychological consequences. Serial casting with subsequent bracing has proven to be a satisfactory conservative option in young patients with idiopathic curves less than 60°, allowing curve resolution in patients younger than 20 months. Unfortunately, this method was less effective in older children (mean age of 30 months), with 35% requiring spinal fusion at a mean age of 10 years [7]. With a median age at referral of 50 months and only 3 patients braced before 25 months, results of the current DNB series are encouraging, with a 67% success rate and only 3 patients (9%) exhibiting a main Cobb angle > 50° at final follow-up. As expected, rotation of the apical vertebra also seemed to be correlated with deformity progression [20]. These promising results must be tempered by the fact that only 45.5% of the patients had reached skeletal maturity at last follow-up.

Mener et al. reported their experience with full-time Milwaukee brace in 75 children under 7 years of age, and found that deterioration was often seen at puberty, resulting in many indications for spine fusion [8]. Similarly, Mannherz et al. [21] reported curve progression in 84% of a series of JIS patients treated by full-time bracing, with 42% of the patients requiring surgery at skeletal maturity. They also pointed out a 20% rate of thoracic hypokyphosis at latest examination. In our series, the DNB was open posteriorly on the concave side and did not use any distraction corrective force. It therefore allowed for unconstrained spinal and chest wall growth, which probably explains why normokyphosis was found in all of the patients at most recent follow-up (Fig. 3). During growth, both thoracic kyphosis and lumbar lordosis significantly increased (P = 0.008 and P = 0.007, respectively), but all patients remained balanced in the sagittal plane. The global sagittal balance, remained unchanged at latest follow-up, with 86% of the patients considered balanced according to Lee et al. [15].

This advantage, essential for pulmonary function and cosmetic appearance, had never been reported with previous conservative treatments.

The other important advantage of DNB was improved compliance, with only 2 patients (6%) refusing the treatment and changing to another conservative option. Compliance with full-time bracing has always been a problem in AIS, with reported consequences on self-esteem, social well-being and quality of life [22]. Since the introduction of the rigid Milwaukee brace, alternatives, such as low-profile braces and night-time only bracing have been tried, with excellent tolerance, and improvement on natural history already documented in AIS and JIS [9–11,23]. Results of the current series confirm the effectiveness of DNB in EOS, especially in young patients with moderate curves.

4.3. Limitations

The main limitations of this study are its retrospective nature, and the lack of a control group. The sample size was relatively small, but EOS remains a rare spinal deformity and the current series offers a mean 10-year follow-up. However, only 45.5% of the patients had reached skeletal maturity at latest examination, and results need further confirmation. Moreover, the skeletal maturity was evaluated by using only the Risser classification. This study was radiological, and no clinical or functional outcome was assessed, such as a quality-of-life questionnaire [24] or trunk height gain. Finally, the compliance might have been overestimated, because it was based upon patients’ and/or patients’ responses, no adherence sensor being used [25]. However, skin marks could usually be seen in compliant patients and were reported in the charts.

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In conclusion, results of the current study show that non-operative management of moderate idiopathic curvatures remains a realistic option in early onset scoliosis, and can be proposed as a “delaying tactic” for surgery until adolescent years. DNB is as effective as other bracing or casting treatment, but it might offer potential psychosocial and compliance benefits, given the longer course of treatment compared to AIS. Serial casting could maintain evaluated curves upper than before going into a surgical option [26]. Age at referral and magnitude of the curve appeared to be prognostic factors. In addition, DNB allows unconstrained spinal and chest wall growth, resulting in normokypnosis at maturity. Nevertheless, education of parents and patients, orthotist skills and the surgeon’s implication in bracing observation remain essential.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References