Long-term observation and management of resolving infantile idiopathic scoliosis

A 25-YEAR FOLLOW-UP

From the University of Bonn, Germany

Of 42 patients with resolving infantile idiopathic scoliosis, 34 were followed up for more than 25 years. Twenty had been primarily treated in a plaster bed and 14 by physiotherapy. The mean angle of the curve at presentation was 17° and at follow-up it was 5°. No patient had significant progression of the scoliosis during the growth spurt. When adults few had back pain or an increased disability score and there was no interference with work or social activities. The rib-vertebra angle difference proved to be an essential radiological sign when distinguishing resolving from progressive infantile idiopathic scoliosis. There was no advantage of plaster over physiotherapy with regard to either the time to resolution or the functional outcome. Treatment of resolving infantile idiopathic scoliosis in a plaster bed is therefore now outdated.

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Infantile idiopathic scoliosis is a lateral curvature of the spine with apical rotation and wedging, which may present before the age of three years. In the 1930s, Harrenstein recognised that the course of infantile scoliosis can be very variable. Some curves progress and cause cardiopulmonary pathology while others resolve or disappear spontaneously within the first years of life. James described this separate resolving type of infantile scoliosis. Two theories have been proposed to explain its cause, intrauterine moulding and postnatal external pressure on the spine. The observation that plagiocephaly, the convex side of a curve, and hip dysplasia all occur on the same side is evidence for the pressure-moulding theory. The postnatal pressure theory is supported by those cases in which scoliosis is not evident at birth. Some authors have divided resolving scoliosis into two types, resolving and postural. The cause of resolving infantile idiopathic scoliosis, however, remains unknown.

The review by Nachmenson and other long-term follow-up studies during the last 30 years have proposed different treatments for patients with idiopathic scoliosis. In order to achieve sufficient numbers of patients many of these studies had poorly-defined patient selection. Early-onset idiopathic scoliosis and resolving idiopathic scoliosis have a different natural history from adolescent-onset scoliosis, often making the groups of patients which were investigated inhomogeneous. Varying forms of treatment within these groups and the paucity of information do not allow conclusions to be drawn. As yet, only short- and medium-term or minor long-term observations concerning resolving infantile scoliosis are available. In contrast to progressive infantile, juvenile and adolescent idiopathic scoliosis, little is known about its natural history at and beyond skeletal maturity. While there are guidelines concerning the treatment of progressive idiopathic scoliosis which have been verified in many clinical studies, there is not a standardised concept for the management of resolving idiopathic scoliosis. Treatment may be by a plaster shell in an overcorrected position or by physiotherapy, and some authors claim that no specific treatment is required. The differing forms of treatment which are recommended probably reflect the fact that it is difficult to differentiate between resolving and progressive curves before the age of five years.

Our aim was to describe the long-term outcome in patients with resolving infantile idiopathic scoliosis, and particularly to consider whether there is recurrence or progression of the deformity in adolescence. We have also compared the correction which may be achieved by either a
plaster shell in the overcorrected position or by physiotherapy.

**Patients and Methods**

Our study was based on a retrospective review of 73 patients with infantile idiopathic scoliosis, who attended special clinics between 1964 and 1974 in two university hospitals. All the children had developed a curvature before the age of three years. The mean age at diagnosis was 15 months (2 to 36) Since differentiation of resolving from progressive curves in the first years of life is difficult, we classified the curves retrospectively according to the radiological course during the first six years of life. The focus of the study was the long-term observation and management of resolving infantile idiopathic scoliosis, which was diagnosed in 42 of these patients. Thirty-four (15 women and 19 men) were followed up for at least 25 years. Three had been lost to follow-up and for five the radiographs were incomplete. These were therefore excluded from the study. The mean follow-up was 28 years (25 to 34). Table I gives the details of the patients.

**Radiological evaluation.** At the last follow-up, all 34 patients underwent radiological evaluation (Fig. 1) with a

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![Fig. 1a](image1.png) ![Fig. 1b](image2.png) ![Fig. 1c](image3.png) ![Fig. 1d](image4.png)

Radiographs of a boy with resolving right thoracic infantile idiopathic scoliosis. At the age of three months the Cobb angle was 28° (a) with a small rib-vertebra angle difference (RVAD) of 14°. He was treated by physiotherapy alone. The curve regressed to 17° after six months (RVAD 12°) (b) and 4° after 1.5 years (RVAD 5°) (c). At the final follow-up at 26 years there was normal alignment (d).

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**Table I.** Details of the patients with resolving infantile idiopathic scoliosis treated in a plaster bed in an overcorrected position (group I) and by early physiotherapy (group II)

<table>
<thead>
<tr>
<th></th>
<th>Group I</th>
<th>Group II</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number</td>
<td>20</td>
<td>14</td>
<td>34</td>
</tr>
<tr>
<td>Female:Male</td>
<td>9:11</td>
<td>6:8</td>
<td>15:19</td>
</tr>
<tr>
<td>Single curves</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Thoracic</td>
<td>7</td>
<td>6</td>
<td>13</td>
</tr>
<tr>
<td>Thoracolumbar</td>
<td>13</td>
<td>8</td>
<td>21</td>
</tr>
<tr>
<td>Right:left side</td>
<td>10:10</td>
<td>5:9</td>
<td>15:19</td>
</tr>
<tr>
<td>Mean age (range) at presentation in months</td>
<td>7 (2 to 18)</td>
<td>6 (2 to 16)</td>
<td>7 (2 to 18)</td>
</tr>
<tr>
<td>Mean age (range) at final follow-up in years</td>
<td>28 (25 to 34)</td>
<td>27 (25 to 32)</td>
<td>28 (25 to 34)</td>
</tr>
<tr>
<td>Mean (±SD; range) primary curve in degrees</td>
<td>17° ± 7.6 (5° to 34°)</td>
<td>17° ± 9.5 (6° to 36°)</td>
<td>17° ± 8.3 (5° to 36°)</td>
</tr>
<tr>
<td>First seen</td>
<td>17° ± 7.6 (5° to 34°)</td>
<td>17° ± 9.5 (6° to 36°)</td>
<td>17° ± 8.3 (5° to 36°)</td>
</tr>
<tr>
<td>Final follow-up</td>
<td>5° ± 3.6 (0° to 13°)</td>
<td>4° ± 3.2 (0° to 12°)</td>
<td>5° ± 3.6 (0° to 13°)</td>
</tr>
</tbody>
</table>
standing anteroposterior (AP) view taken on a 30 × 90 cm film. Observations and measurements were based on all full-length erect AP films of the spine made at the first visit and at the final follow-up and any intervening radiographs. These were interpreted and measurements made by one author (OD). The study considered primary curves only. Lateral radiographs were often not available and thus kyphosis, lordosis, and sacral inclination were not studied.

The Cobb angle was measured, and we compared the course of short and long primary curves, setting the limit of differentiation at nine spinal segments.

We also determined the rib-vertebra angle as described by Mehta, the rib-vertebra angle difference (RVAD) at the apex of the primary curve and the relationship between the rib head and the vertebral body (phase-I or phase-II rib). In order to assess the predictive value of the RVAD in the diagnosis of a resolving or progressive curve we retrospectively measured it on the first available radiograph. Two of 31 patients with progressive curves had incomplete radiographs and were excluded.

Methods of treatment. After the initial diagnosis of resolving infantile scoliosis which was made at a mean age of seven months, 20 patients were treated in a plaster bed with the child supine and the curve overcorrected in lateral flexion as was first described by Harrenstein. It was retained day and night for at least 12 weeks. As the children grew older or with improvement of the curve, it was used only at night and physiotherapy was started.

Fourteen patients with infantile scoliosis were treated by physiotherapy alone. Parents were advised to place the children in a prone sleeping position. At that time the higher incidence of sudden infant death syndrome in the prone position was not known.

Back pain and work activity. The subjective clinical status was assessed at final follow-up using two scores. We used a scheme described by Weinstein et al which differentiates between the frequency and incidence of spinal symptoms and whether hospitalisation was required. The assessment of subjective disability was based on the Oswestry Disability Index (ODI) version 1.0.

Occupational activity was divided into three categories, heavy (construction workers, farmers), medium (including household tasks) and light work (office job).

Statistical analysis. The independent t-test was used to analyse the differences in the radiological data between the groups with p < 0.05 being taken as significant. The chi-squared test was used to compare the clinical results and work activity between the groups.

Results

Resolving scoliosis was diagnosed slightly more often in males (56%) and was predominantly left-convex (56%) on the initial radiographs. All 34 patients had single curves. The residual curve at final follow-up was <5° in 20 patients (59%), between 5° and 10° in 11 (32%) and >10° in three (9%), with an overall mean of 5° (0 to 13). None had significant progression of the scoliosis during the adolescent growth spurt (Fig. 2). Treatment by surgery or with a brace was not required. Although the curve was not clinically apparent radiography had been performed in 16 patients (47%) during adolescent growth.

Rib-vertebra angle difference (RVAD). Figure 3 shows the RVAD. In cases in which it was ≥20° sensitivity was found to be 90% and specificity 85% in the diagnosis of a progressive scoliosis. In all cases of resolving scoliosis the RVAD decreased with skeletal growth. In none of these patients were the ribs found to be in phase II. However, in 17 of the 29 patients (59%) with a progressive curve there was an overlap of the head of the
Resolving scoliosis

Number of cases

<table>
<thead>
<tr>
<th>Number of cases</th>
<th>Resolving scoliosis</th>
<th>Progressive scoliosis</th>
</tr>
</thead>
<tbody>
<tr>
<td>30</td>
<td>90%</td>
<td>90%</td>
</tr>
<tr>
<td>40</td>
<td>85%</td>
<td>85%</td>
</tr>
<tr>
<td>15%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>10%</td>
<td></td>
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</tbody>
</table>

RVAD less 20° RVAD 20° or more

Progressive scoliosis

AISControlPhysiotherapy

ODI

Plaster bed

20

25

15

5

0

0

10

20

30

Fig. 3

Histogram showing rib-vertebra angle difference (RVAD) at the manifestation of resolving or progressive infantile idiopathic scoliosis.

Fig. 4

Histogram showing the outcome of the Oswestry Disability Index (ODI) for resolving curves at the final follow-up compared with normative data of healthy control subjects and patients with adolescent idiopathic scoliosis (AIS).

The apical rib on the convex side on the upper corner of the corresponding vertebral body on the initial radiograph. In a further seven progressive cases (24%) the rib-vertebra relationship converted from phase I to phase II during the following six months.

Pattern of curves. Patients with resolving infantile idiopathic scoliosis had either a C-type thoracic or a thoracolumbar single curve at the initial presentation. In six infants flexion radiographs taken within the first year after birth were available; five showed a fixed lateral curve. In distinguishing between short and long C-shaped curves we set the limit at nine spinal segments, measured from the apical to the distal vertebral body of the major curve. Only four patients had short major curves.

Degree of initial curve. The mean angle of the curve at presentation for those with resolving scoliosis was 17° (5 to 36); 26 patients (77%) had a curve of <20°, five (15%) a curve of between 20° and 30° and three (9%) a curve of >30°. At the final follow-up the mean decrease was 12° (-29 to 3). In five of eight patients, in whom the initial curve was >20°, there was a virtually complete (<5°; Fig. 1) correction at the final follow-up. The persisting degree of curvature at the final follow-up was not related to its initial severity (p = 0.40). Patients with an initial resolving curvature of ≤20° had a mean persistent angulation at the final follow-up of 5° (0 to 13) whereas in those with a mean initial angulation of >20° it was 4° (0 to 8).

Methods of treatment. The median treatment time for a resolving deformity was three years (nine months to 15 years). Both groups showed most improvement during the first four years of life (Fig. 2). There was no significant difference in the time to resolution between the two treatment groups. At the final follow-up the lateral curve had decreased by a mean of 11° (-26 to +4; SD 8.0) in group I and by a mean of 13° (-29 to 0; SD 9.9) in group II. There was no significant difference between the groups with regard to either the decrease in the curve during the period of observation or the persistent curve at the time of follow-up. A comparison of the two groups showed a statistical power <0.80 for the parameters of the spinal profile.

There was conversion from single C-curves to double S-curves in three of the 20 children, who had been treated in a plaster bed (15%). In group I we also found a change in the direction of the primary curve with a slight permanent overcorrection in four of the patients (20%). No similar complications were found in group II.

Back pain and work activity. At the final follow-up 13 patients (38%) claimed not to have suffered from back pain. One had been hospitalised and six (18%) had visited a physician with back pain at some time (Table II). Figure 4 shows the ODI for resolving curves at the final follow-up and compares it with normative data of healthy control subjects and patients with adolescent idiopathic scoliosis.

There are no significant differences between patients treated in a plaster bed (group I) and those treated by physiotherapy (p = 0.46).

At the final follow-up only two patients (6%) were unemployed. None had retired prematurely or was unable to work because of spinal symptoms. Twelve patients (35%) assessed their occupation as light work, 14 (41%) as medium strenuous and eight (24%) as heavy work. Only two patients (6%) claimed that they were restricted in their occupational or recreational activities because of spinal symptoms. The occupations were not significantly different between the treatment groups.

Discussion

The reported incidence of resolving infantile idiopathic scoliosis varies between 17% and 92% of all patients with infantile idiopathic scoliosis. The true incidence is clearly unknown since many minor and resolving curves are not diagnosed; 58% of our patients had resolving curves.
There are few studies which deal with early-onset resolving scoliosis from the time of diagnosis to skeletal maturity. Most authors observe patients only until or just after the adolescent growth phase.\textsuperscript{5,9,21,30,38} At the time of the final follow-up most of our patients had a residual curve of <10°. We found, as did Sommer\textsuperscript{5,9,\textsuperscript{30,38}} and Thompson and Bentley,\textsuperscript{21} that the prepubertal and pubertal growth spurt were not associated with recurrence or progression of the curves. We found no patient with an initial resolving curve which later progressed to an adolescent idiopathic curve.

**Radiological findings**

*Rib-vertebra angle difference (RVAD).* Our results underline previous studies\textsuperscript{2,5,7,10,21} which have shown that the RVAD is an essential radiological sign in distinguishing between resolving and progressive curves. In all resolving cases the RVAD decreased during the period of observation. In none of these patients were the ribs found to be in phase II. This confirms the conclusions of Mehta\textsuperscript{10} that the diagnosis of the rib head in phase II is a definitive sign of a progressive scoliosis. When the rib head is in phase I, the RVAD is even more important since it determines the prognosis. This diagnostic tool has a marked influence on the treatment of infantile scoliosis.

**Pattern of curves.** The most common pattern of resolving infantile idiopathic scoliosis is the long single C-type lateral thoracic or thoracolumbar curve with a slight rib hump on the convex side. In contrast to progressive infantile and juvenile idiopathic scoliosis, double primary curves are rare.\textsuperscript{10} Five of six patients who had lateral flexion radiographs within the first year had a fixed lateral curve. Although statistically not significant this suggests that resolving infantile idiopathic scoliosis can be associated with structural changes which may be identified radiologically.

**Degree of the initial curve.** In our study most resolving infantile idiopathic curves had a Cobb angle of <20° at the initial presentation although some curves had an angle of >30°. The most pronounced curve which resolved was of 36°. Curves of 40° have been shown to resolve spontaneously.\textsuperscript{7,10,21} The Cobb angle at the final follow-up was not related to the severity of the curve at presentation.

**Back pain and work activity.** According to the ODI patients with a resolving infantile idiopathic scoliosis grad-

<table>
<thead>
<tr>
<th>Back pain</th>
<th>Group I</th>
<th>Group II</th>
<th>Total</th>
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<tbody>
<tr>
<td>None</td>
<td>8</td>
<td>5</td>
<td>13</td>
</tr>
<tr>
<td>Rarely (1 to 5 times in entire life)</td>
<td>5</td>
<td>3</td>
<td>8</td>
</tr>
<tr>
<td>Occasionally (few days a year)</td>
<td>3</td>
<td>4</td>
<td>7</td>
</tr>
<tr>
<td>Frequently (few days a month)</td>
<td>4</td>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>Daily</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Visited doctor for back pain</td>
<td>4</td>
<td>2</td>
<td>6</td>
</tr>
<tr>
<td>Hospitalised for back pain</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Mean (±SD; range) Oswestry Disability Index</td>
<td>4.3 ± 4.1 (0 to 14)</td>
<td>4.9 ± 7.6 (0 to 27)</td>
<td>4.6 ± 5.7 (0 to 27)</td>
</tr>
</tbody>
</table>

-ed their disability as minimal. Few patients sought advice for back pain and only one required hospitalisation. Almost identical ODI values were found for the normal population in the meta-analysis of Roland and Fairbank,\textsuperscript{37} who also showed that patients with adolescent idiopathic scoliosis had slightly higher values. Patients with a resolving infantile idiopathic scoliosis, when compared with the healthy population, do not have increased back pain or disability scores, and the variety of occupations pursued is not limited. The treatment of resolving infantile idiopathic scoliosis had no effect on disability at skeletal maturity.

**Treatment.** The treatment of early-onset resolving scoliosis by means of a plaster bed which was common practice until recently, now seems incomprehensible. Although it was accepted that infantile idiopathic scoliosis may resolve spontaneously, the fear of missing the progressive form led surgeons to advise this form of treatment for all infantile idiopathic curves. More recently, numerous authors have proposed that resolving idiopathic scoliosis should be treated by early physiotherapy.\textsuperscript{2,5,12,21,23,33} We believe that once a resolving idiopathic curve has been identified no further treatment is required.

**Critical aspects.** Although the importance of long-term reviews for a better understanding of the natural history of spinal deformities is well established,\textsuperscript{1,10,16,19,38} the design of retrospective studies remains controversial. The inevitable initial selection of patients, flaws resulting from irretrievable patients and insufficient documentation justify some scepticism when interpreting the results. Our observations may not represent the normal incidence found in an unselected group of patients. However, there will be few prospective studies with a follow-up of more than 25 years.

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

**References**