Congenital scoliosis: A frontal plane evaluation of 251 operated patients 14 years old or older at follow-up


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KEYWORDS
Congenital scoliosis; Spinal deformity; Scoliosis; Spine; Hemivertebra

Summary
Introduction: Congenital scoliosis, carrying an incidence between 0.5 and 1 per 1000 births, raise the problem of their evolutive potential.
Hypothesis: Some predictive factors for the evolution of scoliotic curvature due to congenital vertebral malformation (CVM) can be found.
Material and methods: This was a retrospective multicenter study of 251 patients, at least 14 years old when evaluated at end of follow-up, with CVM and spinal deformity predominating in the frontal plane.
Introduction

Congenital scoliosis is defined as a spinal deformity secondary to a congenital vertebral malformation (CVM). CVM may induce spinal deformity in all three planes; the convention is to use “congenital scoliosis” to refer to predominantly frontal deformities. Incidence of congenital scoliosis is estimated at between 0.5 and 1 per 1000 births [1]. Discovery of CVM in a patient raises many questions, including that of evolutive potential. Clinical and imaging analysis of CVM and any associated abnormalities is essential to the assessment of the evolutive potential of the induced spinal deformities [2]. The severity of the congenital scoliosis is dependent on CVM type, location and number, but also on the patient’s age [3,4].

The present study looked for predictive factors for the evolution of scoliotic curvature induced by CVM, so as to offer management recommendations according to CVM type and location and patient age at diagnosis.

Material and methods

Population

This was a retrospective multicenter study of CVM patients with spinal deformity predominating in the frontal plane. Clinical and radiology records for 340 patients (178 female, 162 male) were reviewed.

Exclusion criteria

Exclusion criteria were age less than 14 years at end of follow-up, incomplete clinical and/or radiological records, and predominantly sagittal spinal deformity. Patients with kyphosis angle greater than scoliosis angle were thus systematically excluded.

Data collection form

A computerized data collection form was created. Each patient’s general data and CVM type, number, location and laterality were thus precisely inventoried.

Any associated neurologic, visceral and orthopedic malformations and rib malformations were listed.

The form specified scoliotic and kyphotic curvature parameters: number, location, laterality and any associated compensatory curves. Scoliotic and kyphotic curve Cobb angles, whether malformative or compensatory, were measured at diagnosis, preoperatively, postoperatively and at end of follow-up.

Location

CVMs and spinal deformities were located according to three main spinal segments and three junction segments. The main segments were cervical (C), thoracic (T) and lumbar (L); the junction segments were defined as cervicothoracic (CT: C6 (6th cervical vertebra) to T2 (2nd thoracic vertebra)), thoracolumbar (TL: T11 (11th thoracic vertebra) to L2 (2nd lumbar vertebra)) and lumbosacral (LS: L4 (4th lumbar vertebra) to S1 (1st sacral vertebra)).

X-ray assessment

Each patient underwent radiologic analysis on posteroanterior and lateral whole-spine views, taken in standing posture.

Results: 38.8% of patients showed associated neurologic, visceral or orthopedic abnormalities. CVM was single in 60.6%, double in 20.3%, triple in 6.4% and multiple in 12.7% of cases. 34.1% of CVMs were thoracic. Congenital scoliosis curvature was single in 88.8% of patients, double in 10% and triple in 1.2%. Mean curvature angle was 31.7° at diagnosis (range, 0—105°) and 41.3° preoperatively (range, 10—105°). Sixty-one patients showed associated kyphosis. Mean change in postoperative curvature angle over follow-up was 1.6° (range, −20° to 38°) in the 73 patients managed by arthrodesis, −0.4° (−24° to 30°) in the 64 managed by epiphysiodesis, and 0.4° (−18° to 35°) in the 49 managed by hemivertebra (HV) resection. Results were found to correlate significantly with age at surgery for patients managed by epiphysiodesis, but not for those managed by HV resection or arthrodesis.

Discussion: More than 30% of congenital scolioses involve associated intraspinal abnormality. All CVM patients should therefore undergo medullary and spinal MRI to assess the CVM in all three planes, and the medullary canal and its content. The evolution of scoliotic curvature induced by CVM is hard to predict. Several factors are to be taken into account: CVM type, number and location, and patient age. Curvature progression may be slow or very fast. It accelerates during the peak of puberty, stabilizing with bone maturity. Surgery is mandatory in evolutive scoliosis. Four procedures may be recommended, according to type of CVM and especially to patient age: arthrodesis, convex epiphysiodesis, HV resection or rib distraction. Surgery seeks to correct the spinal deformity induced by the CVM and prevent compensatory curvature and neurologic complications, while conserving sagittal and frontal spinal balance and sparing as many levels as possible. In case of HV involvement, the procedure of choice is CVM resection, which provides 87.5% good results in this indication; the procedure is relatively safe, conservative of spinal levels, and without age limit.

Level of evidence: Level IV. Retrospective study.
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Congenital scoliosis except when scoliosis was diagnosed before walking age. Measurement used a graduated goniometer, following Cobb [5].

Curvature evolutivity was assessed by comparing Cobb angles at diagnosis and at last follow-up in patients managed nonsurgically and postoperatively and at last follow-up in patients managed by surgery.

Curvature evolutivity was classified in one of three groups: improved, stabilized or worsened. Curvature was considered stabilized if the difference between postoperative and last-FU Cobb angles was between $-5\degree$ and $+5\degree$, worsened if strictly greater than $+5\degree$ and improved if strictly less than $-5\degree$.

Statistics

Correlations between Cobb angle evolution and CVM type and location, type of treatment and patient age at treatment were analyzed using SPSS version 15.0 software for Windows®. The significance threshold was set at $p < 0.05$.

Results

Population

After exclusion of patients less than 14 years old at end of follow-up, 251 of the 340 patients were included for analysis: 139 female and 112 male. CVM was diagnosed before 1 year of age in a quarter of patients, before 3 years of age in half and before 9 years of age in three-quarters. Mean age at end of FU was 18.5 years (range, 14—68 years).

38.8% of the series as a whole showed associated neurologic, visceral or orthopedic abnormality. In 4.8%, these constituted a multiple malformation syndrome: VACTERL ($n = 4$), Klippel Feil ($n = 2$), Goldenhar ($n = 2$), Jarcho-Levine ($n = 1$), Noonan ($n = 1$) or Pierre Robin ($n = 1$). Abnormalities were urodigestive (18%), cardiac (8%), neurologic (7.6%) or orthopedic (6.4%).

CVM characteristics

CVM was single in 60.6% of patients, double in 20.3%, triple in 6.4% and multiple ($\geq 4$) in 12.7% des patients (Table 1).

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Congenital vertebral malformation according to type and location.</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>C</td>
</tr>
<tr>
<td>HV</td>
<td>1</td>
</tr>
<tr>
<td>B</td>
<td>4</td>
</tr>
<tr>
<td>Bi</td>
<td>0</td>
</tr>
<tr>
<td>P</td>
<td>5</td>
</tr>
<tr>
<td>T</td>
<td>5</td>
</tr>
<tr>
<td>Total</td>
<td>15</td>
</tr>
</tbody>
</table>

Location: cervical segment (C); cervicothoracic junction segment (CT); thoracic segment (T); thoracolumbar junction segment (TL); lumbar segment (L); lumbar-sacral junction segment (LS). Type: hemivertebra (HV); bar (B); binucleate vertebra (Bi); puzzle (P); trapezoid vertebra (T).

Figure 1 Clinical case. Child with free HV in T11. A: At diagnosis, and preoperatively at 9 years of age, 28\degree malformation curve. B: Postoperatively, after T9-L1 arthrodesis on double approach, 12\degree malformation curve. C: At 7 years’ FU, 16\degree malformation curve.

One hundred and sixty-one of the 251 patients presented with single or multiple hemivertebra (HV) (Fig. 1) without other associated CVM. 55.2% of the 235 HVs were free, 30.5% semi-fused, and 14.3% fused.

There were 32 rib abnormalities: 27 synostoses and five agenesis. Eight patients showed severe restrictive syndrome, requiring invasive ventilation in two cases and tracheostomy in one. These eight patients had CVM localized in the T vertebrae, and only two had associated rib abnormality; CVM was a bar in three cases, binucleate vertebra in two, free T5 HV in two, and fused HV with two associated contralateral bars in 1. Restrictive syndrome correlated significantly with a T vertebral location of the CVM ($p < 0.01$), but not with type of CVM ($p > 0.05$) or associated rib abnormality ($p > 0.05$).

Malformation curve characteristics

Congenital scoliosis curve was single in 88.8% of cases, double in 10% and triple in 1.2%. One hundred and five patients (41.8% of the series as a whole) showed associated compensatory curvature (Tables 2 and 3). Compensatory curvature did not correlate significantly with number ($p > 0.05$) or type of CVM ($p > 0.05$).

Sixty-one patients (24.3%) had associated kyphosis, with a smaller angle than the scoliosis curve. Kyphosis was located in TL in 56.3% of cases, T in 28.1%, L in 9.4% and LS in 6.2%, and did not correlate significantly with number ($p > 0.05$) or type of CVM ($p > 0.05$).

Sixty-five patients were managed nonsurgically, and 186 surgically. Seventy-three of the 186 had arthrodesis, 64 epi-physiodesis and 49 HV resection.

69.2% of the patients managed nonsurgically presented with HV, 23.1% with bars, and 7.7% with puzzles. Mean mal-
Table 2  Malformation, compensation and kyphosis curve angles, at diagnosis, preoperatively, postoperatively and at end of follow-up.

<table>
<thead>
<tr>
<th>Curve</th>
<th>N</th>
<th>Diagnosis</th>
<th>Preop</th>
<th>Postop</th>
<th>End FU</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>M</td>
<td>Min</td>
<td>Max</td>
<td>M</td>
</tr>
<tr>
<td>Malformation</td>
<td>281</td>
<td>31.7</td>
<td>0</td>
<td>105</td>
<td>41.3</td>
</tr>
<tr>
<td>Compensation</td>
<td>105</td>
<td>19.1</td>
<td>0</td>
<td>50</td>
<td>29</td>
</tr>
<tr>
<td>Kyphosis</td>
<td>61</td>
<td>29</td>
<td>0</td>
<td>80</td>
<td>34.6</td>
</tr>
</tbody>
</table>

M: mean angle (°); Min: minimum angle; Max: maximum angle; N: number of curves.

Table 3  Evolution of malformation curves according to type of treatment.

<table>
<thead>
<tr>
<th>Number</th>
<th>Age at diagnosis (yrs)</th>
<th>Preop/diagnosis evolution</th>
<th>Age at surgery (yrs)</th>
<th>Preop/postop evolution</th>
<th>FU (yrs)</th>
<th>Evolution of last correction/immediate postoperatory</th>
</tr>
</thead>
<tbody>
<tr>
<td>Arthrodesis</td>
<td>73</td>
<td>6.8</td>
<td>9.6° (range, 0 to 60°)</td>
<td>13</td>
<td>−11.9° (20.7% correction)</td>
<td>7.2</td>
</tr>
<tr>
<td>Epiphysiodesis</td>
<td>64</td>
<td>2.8</td>
<td>6.6° (range, 0 to 45°)</td>
<td>5.8</td>
<td>−4.5° (10.5% correction)</td>
<td>11.5</td>
</tr>
<tr>
<td>HV resection</td>
<td>49</td>
<td>5.4</td>
<td>5.6° (range, 0 to 35°)</td>
<td>8.7</td>
<td>−16.8° (46.7% correction)</td>
<td>8.8</td>
</tr>
</tbody>
</table>

formation curve angle at diagnosis was 24.5° (range, 0° to 80°), three-quarters being less than 32°. Mean evolution between diagnosis and last follow-up (at a mean 13.4 years) was 6.4° (range, 35° improvement to 59° worsening), and less than 12° in three-quarters. Mean compensatory curvature at diagnosis, found in 24 of the 65 patients managed nonsurgically, was 18.6° (range, 0° to 50°), with mean evolution between diagnosis and last follow-up of 11.1° (range, 25° improvement to 40° worsening), and less than 21° in three-quarters.

Three-quarters of the 73 patients managed by arthrodesis (Fig. 1) showed less than 5° evolution in malformation curve angle between postoperative and end-of-FU values. At end of follow-up, 62.5% had stable results, 16.7% showed improvement and 21.8% deterioration. No correlation was found between these results and age at surgery (p > 0.05).

Three-quarters of the 64 patients managed by epiphysiodesis showed less than 8° evolution in malformation curve angle between postoperative and end-of-FU values. At end of follow-up, 37.6% had stable results, 31.2% showed improvement and 31.2% deterioration. There was a significant correlation with age at surgery (p = 0.011); results were better (stable or improved) in case of early surgery.

Three-quarters of the 49 patients managed by HV resection (Fig. 2) showed less than 3° evolution in malformation curve angle between postoperative and end-of-FU values. At end of follow-up, 66.7% had stable results, 20.8% showed improvement and 12.5% deterioration. No correlation was found between these results and age at surgery (p > 0.05).

Mean evolution in kyphosis curve angle between postoperative and end-of-FU values was −1° (range, 52° improvement to 28° worsening). 9.6% of the 61 patients with associated kyphosis showed 5 to 10° deterioration and only 2.6% more than 10°. There was no correlation between these results and type of CVM (p > 0.05), type of treatment

(p > 0.05) or CVM location (p > 0.05), although there was a trend for worsening of kyphosis to be associated with T (33%) or TL (30.7%) locations.

Incomplete data on instrumentation levels prevented statistical analysis of this parameter.
Surgical techniques

The surgical approach was generally posterior in case of arthrodesis (64.4%) and combined anterior/posterior in epiphysiodesis (92.9%) and HV resection (73.5%). Surgery was instrumented for 108 patients: for 78% of arthrodeses, 83% of HV resections, and 16% of epiphysiodeses. An autologous graft (mainly iliac costal and/or vertebral: spinous process or resected CVM) was used in 96% of cases.

Evoked potentials were monitored peroperatively in 64 patients.

Postoperatively, 152 patients wore a corset (plaster and/or Plexidur®) for at least 2 months: < 6 months for 67.8% and < 12 months for 75%.

Complications

There were eight immediate postoperative complications, of which five were neurological (one [postoperative para-plegia] was severe, and four cases of radiculopathy, two infections [one venous catheter infection and one respiratory infection] and one case of non-cicatrization. The one severe neurologic complication followed arthrodesis for a bar detected on CT, and showed favorable evolution after immediate removal of the arthrodial material.

Secondary complications comprised five cases of neurologic complication, two of infection, two if disassembly (hooks) and one of material protrusion. Over and above the cases of worsened malformation, compensation or kyphosis curvature, late complications also comprised two cases of infection, two of nonunion and one of disassembly. The secondary neurologic complications all showed spontaneous regression over follow-up.

Six patients underwent instrumentation removal: three due to neurologic complication (including the case of postoperative paraplegia), one due to infection, one to disassembly and one to residual pain.

Thirty-two of the 186 patients managed surgically underwent revision. These were mainly HV patients (59.3%) with T-located CVM (46.8%). There were two neurologic complications, two nonunions and one disassembly; the other surgical revisions were for secondary worsening of malformation and/or compensation curvature.

Discussion

Spinal embryology is essential to the understanding of CVM, which forms during the embryonic phase in the first month of pregnancy [6,7]. CVM results from defective embryonic somite formation and/or segmentation, and may be acquired, secondary to aggression, although a genetic component cannot be excluded [1,8]. In pregnant rats, induced CVMs were specific to the type of aggression: by varying the degree, duration and timing of in utero anoxia, Rivard et al. were able to produce all of the different types of CVM found in human embryos [9]. Other chemical, thermal, nutritional, traumatic and infectious factors are also implicated in CVM formation [10]. In most cases, genetic and environmental factors interact in what must be seen as a multifactorial etiology [1].

Several classifications have been made of CVM, all including the same specific abnormalities but differently classified, whether as defects of formation or segmentation or both [11—13] classified deformities in terms of three basic groups of CVM: HVs, bars and anterior or binucleate trapezoid aplastic vertebrae; this was the classification used in the present study.

CVM was diagnosed in the first year of life in only a quarter of the patients in the present series: mean age at diagnosis was 5 years, ranging from in utero to 25 years. These figures are comparable to those of the literature [14]. The development of antenatal ultrasound screening techniques should enable earlier diagnosis and prevent delayed treatment.

The present 38.8% incidence of neurologic, visceral or orthopedic malformation is in line with literature data, which vary from 26 to 60% [15,16].

The most frequent congenital malformations associated with congenital scoliosis involve the urogenital tract, since, embryologically, the same undifferentiated mesenchyma differentiates into vertebrae and mesonephros, the latter going on to form the kidneys and urinary tract [1]. Eighteen percent of the present series presented with associated gastrointestinal or urogenital abnormality; in the literature, the rate of urogenital tract abnormality associated with congenital scoliosis ranges from 10 to 40% [1,6,16]. All CVM patients should therefore have a urinary system US scan to check for any associated abnormality.

The second type of malformation to look out for is cardiac, associated in 7 to 15% of cases in the literature [11—17], and in 8% in the present series.

More than 30% of congenital scoliosis have associated intraspinal abnormality, notably diastematomyelia and syringomyelia [2,6,15,18,19]. In the present series, 14 patients (5.6%) showed abnormalities nosologically classifiable as closed spinal dysraphism [13]. All CVM patients should therefore undergo medullary and spinal MRI to assess the CVM in all three planes and the medullary canal and its contents [6].

Natural history is essential in CVM, as it determines prognosis and treatment. The progression of the scoliotic curve induced by CVM is hard to predict. Several factors are to be taken into account: CVM type, number and location, and patient age [3,4,14]. Progression may be slow, or very fast. Vertebral growth is very active during the first months of life, slowing down after 6—7 years until the peak of puberty [20], when it accelerates and then stabilizes at bone maturity [4].

Several studies have demonstrated that most congenital scoliosis are evolutive, except in only 10 to 25% of cases [4]. McMaster and Ohtsuka [4] reported 11% nonevolutive curves, 14% showing low-level progression, and 75% with significant evolution. Similarly, in the present series, 19.4% of patients showed little (< 12°) or no curve evolution without surgery. Some scoliotic curves are stable throughout evolution; others worsen significantly and quickly, by 5° to 15° per year, requiring rapid surgical intervention [21]. The third and most frequent pattern of evolution is deterioration between birth and 5 years of age, stabilization between the ages of 5 and 10, followed by renewed aggravation at puberty. Treatment options are relatively straightforward in case of stable curvature and rapid deterioration, but the third pattern is more tricky to manage. The low level of evolution between the
ages of 5 and 10 years may encourage an attitude of simple surveillance, which, however, may come to be regretted when evolution surges at puberty. Early surgery could be limited to a restricted segment, whereas much more extensive arthrodesis may be required in the same patient in a later intervention.

According to McMaster and Ohtsuka [4], curve evolution rate depends on the spinal region involved and the type of CVM. T and TL curves show the greatest evolutivity, and are thus associated with the poorest prognosis [16]. All the T curves in Winter’s series [16] worsened with growth, 70% by more than 30°. CT curves have the greatest esthetic impact, causing lateral inclination of the neck and head and asymmetry of the shoulders [16]. The most strongly evolutive type of CVM is a concave bar associated with one or more contralateral convex HVs [2,22]; then, in descending order of poor prognosis, come concave bars, double convex HV, free HV and block vertebrae [4]. The present study was unable to determine statistically the predictive factors for evolution in CVM-induced scoliotic curves according to CVM type or location.

The choice of treatment and age at surgery was specific to the individual center. Some scheduled surgery at a very early age for minor deformities, indicating either epiphysiodesis or CVM resection; others exclusively performed arthrodesis at the peak of puberty. The series was thus too heterogeneous to determine a recommended procedure according to age at diagnosis, curvature angle or CVM type or location.

Surgery is mandatory for evolutive curves. The four possible procedures, according to CVM type and especially to patient age, are arthrodesis, convex epiphysiodesis [23], CVM resection in case of HV, and rib distraction [20,24]. In the present series, patients managed by rib distraction happened to be excluded, all being under 14 years of age at end of follow-up.

Isolated posterior arthrodesis for lateral or posterolateral HV conserves HV anterior growth potential. Continued anterior growth while the spine is blocked posteriorly aggravates vertebral rotation by a bit-brace-like mechanism. Combined anterior and posterior surgery is thus required to prevent progression of the deformity. Winter et al. [25] reported 40 cases (13.8%) involving >10° postoperative increase in curvature angle following isolated posterior arthrodesis, which could be considered as a bit-brace effect. The extent of the arthrodesis depends on patient age and curvature angle. The mean angle correction obtained in congenital scoliosis managed by straightening arthrodesis was 11.9° in the present series, much less than in idiopathic scoliosis. Winter et al. [25], in a series of 290 patients managed by arthrodesis, reported a 14% rate of complications, including 20 nonunions, six septic complications and six neurologic complications, two of which were severe: postoperative paraplegia following correction of deformity located in T and TL, exactly as in the case in the present series. He found no difference between instrumented and non-instrumented arthrodesis in terms of correction, subsequent angle loss or occurrence of nonunion [25]: instrumentation reduced corset immobilization time, but without otherwise improving results.

Age at diagnosis and at surgery was lower in case of epiphysiodesis than in CVM resection (mean, 2.8 vs. 5.4 and 5.8 vs. 6.8 years, respectively). Choice of treatment may have been influenced by patient age at CVM diagnosis. At end of follow-up, CVM resection cases showed a higher percentage of stable or improved curvature than those managed by epiphysiodesis (66.7% stable and 20.8% improved vs. 37.6% and 31.2%, respectively): these findings may be explained by the fact that only results on epiphysiodesis correlated with age at surgery.

The aim of surgery is to correct the spinal deformity induced by CVM, and to prevent the development of compensation curvature or of neurologic complications, while conserving sagittal and frontal balance and conserving as many levels of the spine as possible. The procedure of choice is therefore CVM resection where possible, as it provides 87.5% good results. HV resection provided a mean 46.7% angular correction in the present series, comparable to the 58.5% reported by Bollini et al. [24]. The procedure is low-risk, conservative of spinal levels, and without age limit.

Postoperative anterior or combined anterior/posterior approach morbidity and pain can be limited by using thoracoscopy: several reports of HV resection, anterior release and epiphysiodesis under thoracoscopy have found satisfactory results [26].

Conclusion

In antenatal screening for spinal malformation, it is essential to have full information not only on CVM as such but also on the associated prognosis, so as to inform families and decide on treatment options. Only 50% of CVMs in the present series were diagnosed before the age of 3 years; very early diagnosis is needed, in order to avoid delayed management.

The poor degree of correction obtained by straightening arthrodesis in congenital scoliosis operated on near the age of bone maturity makes it clear that earlier treatment would enable better results in terms of curvature angle and less extensive fusion. HV resection and epiphysiodesis provide identical long-term results, but only the former enables immediate (and durable) angular correction of nearly 50%.

The present study was not able to highlight predictive factors for curve evolutivity; it thus remains to be determined which curves will be evolutive, to enable early surgery with minimal curvature angle.

Conflict of interest statement

None.

References


