Sagittal spinopelvic alignment in adolescent thoracic scoliosis secondary to Chiari I malformation: a comparison between the left and the right curves

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Abstract

Purpose To investigate whether the sagittal morphology differs between the left and right thoracic curves in patients with Chiari malformation-associated scoliosis (CMS).

Methods Thirty-four patients with a left thoracic curve constituted the CM-L group, whereas 44 patients with a right thoracic curve were assigned into the CM-R group. Another cohort of 90 age- and gender-matched asymptomatic adolescents was enrolled to serve as the control group. Seven sagittal parameters were evaluated on standing lateral radiographs, including thoracic kyphosis (TK), lumbar lordosis (LL), thoracolumbar junctional kyphosis (TJK), sagittal vertical axis (SVA), pelvic incidence (PI), pelvic tilt (PT) and sacral slope (SS).

Results Compared to the normal controls, the TK, LL and SS were significantly greater in the CM-L group, along with a significantly decreased PT. Similar changes in SS and PT were also demonstrated in the CM-R group, while the TK and LL were found to be relatively normal compared with the control group. Concerning CMS patients with different curve directions, significantly increased TK and LL were observed in the CM-L group, whereas all three pelvic parameters were similar for the two groups. In addition, no significant differences were noted between the three groups in PI, TJK or SVA. Moreover, the LL was strongly related to the TK and SS in all three groups, but to the PI only in the control and CM-R groups. A significant

correlation was also noted between TK and SS in the CM-L group.

Conclusions Significant differences in sagittal profiles indeed exist between CMS patients and healthy adolescents, as well as between CMS patients with different curve directions. In CMI patients with a left thoracic curve, compensatory alterations appear to occur in LL in response to the increased TK to maintain a balanced posture.

Keywords Chiari I malformation · Scoliosis · Curve direction · Sagittal spinopelvic alignment · Sagittal balance

Introduction

Sagittal alignment of the spine and pelvis has been increasingly recognized as being an essential aspect to consider in the evaluation and treatment of spinal pathologies [1–3]. Recently, several studies revealed that the classic apical hypokyphosis, as have been invariably demonstrated in adolescent idiopathic thoracic scoliosis (AIS), was absent in most cases with Chiari malformation-associated scoliosis (CMS) [4–7]. To our knowledge, however, no published study has specifically addressed the morphological differences between CMS patients and healthy controls.

In addition, although the so-called “atypical” left thoracic curve pattern has a high frequency of occurrence in CMS [5, 6, 8], there is a paucity of literature focusing on the differences in sagittal profiles between the left and right thoracic curves. Extracted data from the study by Spiegel et al. [5] manifested a trend that CMS patients with a left thoracic curve had larger thoracic kyphosis than those with a right thoracic curve. It appears rather difficult, though, to

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drawn any firm conclusions according to this study, given their small sample size (18 cases) as well as the relatively wide age range (4.7–19.8 years). In an attempt to remedy these limitations and to further clarify their finding, this study aimed to evaluate the sagittal spinal and pelvic morphologies with respect to curve direction on a larger and homogeneous CMS cohort.

The objectives of the current study were threefold: (1) to investigate whether or not the sagittal profiles differ between CMS patients and normal population; (2) to compare the sagittal spinal and pelvic parameters between CMS patients with distinct thoracic curve directions and (3) to analyze the associations between all sagittal parameters.

Methods

Subjects

Under the approval of the institutional review board, a retrospective radiographic study was performed on all patients with scoliosis secondary to Chiari I malformation (CM-I) treated at our scoliosis clinic between December 2004 and December 2007. The inclusion criteria were as follows: (1) age 10–18 years; (2) main thoracic curve pattern with curve apex located between T5 and T11. For a subset of patients with a compensatory thoracolumbar/lumbar (TL/L) curve, the distal curve should be confirmed to be nonstructural (not crossing the midline on standing posteroanterior radiograph or being corrected to less than 25° on side bending film); (3) main curve magnitude between 20° and 70°. Any patients with associated congenital spine deformities or spinal cord malformation, concomitant genetic disorders (e.g., neurofibromatosis) or acquired CM-I anomalies were excluded. A subset of 78 CMS patients, including 43 males and 35 females with an average age of 14.1 ± 2.2 years (range 10–18 years), was finally enrolled for this study. Of note, a total of 35 patients were found to have a nonstructural TL/L curve, among which 26 curves did not cross the midline. In the remaining nine patients, the TL/L curve crossed the midline but was corrected to less than 25° on side bending radiographs. Based on the lateralization of their thoracic curves, CMS patients were further classified into two subgroups: the CM-L group with left thoracic curve ($n = 34$) and the CM-R group with right thoracic curve ($n = 44$).

In accordance with an established protocol [9], another cohort of 90 asymptomatic adolescent volunteers (49 males and 41 females; mean age 13.9 years; age range 10–18 years) was recruited from local secondary schools to serve as the control group. Any volunteers with evidence of spinal disorders (e.g., scoliosis), neurological deficit or leg

length discrepancy were excluded. Informed consent was obtained from the parent of each participant.

Radiographic evaluation

Sagittal alignment of the spine and pelvis was assessed with the Surgimap Spine software (Nemaris, Inc., New York, NY, USA) [10]. In line with previously described methods [2, 5, 9, 11, 12], four spinal and three pelvic parameters were measured on long-cassette standing upright lateral radiographs: thoracic kyphosis (TK): the angle between the superior endplate of the T3 vertebra and the inferior endplate of the T12 vertebra; lumbar lordosis (LL): the angle between the superior endplate of the L1 vertebra and the superior endplate of the S1 [9]; thoracolumbar junctional kyphosis (TJK): the angle between the superior endplate of T10 and the inferior endplate of L2 [12]; sagittal vertical axis (SVA): the distance between the C7 plumb line (C7PL) and the posterosuperior corner of S1 [2]; pelvic incidence (PI): the angle between the line perpendicular to the sacral plate at its midpoint and the line connecting this point to the axis of the femoral heads; pelvic tilt (PT): the angle between the line connecting the

Fig. 1 Identification of the center of the bicoxofemoral axis in pelvic measurement. Using the Surgimap Spine software (Nemaris, Inc., New York, NY, USA), a circle along the border of each femoral head was created first; then the center (c) could be identified automatically

midpoint of the sacral plate to the femoral head axis and the vertical axis; and sacral slope (SS): the angle between the superior plate of S1 and a horizontal line [11]. Figure 1 illustrates the method of identifying the axis of the femoral heads in pelvic measurement.

For all sagittal measurements, the angle was considered to be negative if the curve was lordotic and positive if the curve was kyphotic. SVA was considered to be negative if the C7 plumb line fell behind the sacrum and positive if the C7 plumb line fell anterior to the sacrum [13]. Based on the normative data for sagittal alignment in adolescents [14–16], thoracic alignment was defined as normal (20° – 50°), hyperkyphotic ($>50^{\circ}$) and hypokyphotic ($<20^{\circ}$), and accordingly, lumbar alignment was defined as normal (54° – 74°), hyperlordotic ($>74^{\circ}$) and hypolordotic ($<54^{\circ}$) [5].

In the coronal plane, curve magnitude and apex of the primary curve as well as the apical vertebral rotation (Nash-Moe method [17]) were obtained on standing posteroanterior radiographs. For patients who had prior history of brace treatment or neurosurgical management, only the radiographs at the initiation of bracing or the preoperative radiographs were considered.

Statistical analysis

Data analysis was performed using SPSS version 19.0 (Chicago, IL, USA). Each dependent variable was compared between the three groups (CM-L, CM-R and control groups) utilizing analysis of variance (ANOVA), followed by a Bonferroni's post hoc correction for individual pairwise comparisons when indicated. Comparisons between the three groups of categorical variables were analyzed with the Pearson χ^2 test. The correlations between all parameters were assessed using Pearson coefficients. Statistical significance was considered with $P < 0.05$.

Results

Table 1 summarizes the demographic data of patients in all three groups. Age and gender ratio were similar between the three groups. With respect to the two CMS groups, syrinx was presented in 29 patients in the CM-L group and in 36 cases in the CM-R group. No significant differences were noted between the two groups in terms of the main

Table 1 Patient demographics of the three groups

	CM-L group	CM-R group	Control group	P value
No. of patients	34	44	90	0.749*
Age (year)	13.9 $\pm$ 2.2 (10–18)	14.2 $\pm$ 2.2 (10–18)	13.9 $\pm$ 2.0 (10–18)	0.776*
Gender (male/female)	19/15	24/20	49/41	0.752 [†]
Cobb angle ($^{\circ}$)				
Thoracic	47.7 $\pm$ 13.6 (20–70)	49.2 $\pm$ 14.9 (20–70)	–	0.648
Thoracolumbar/lumbar	29.1 $\pm$ 6.0 (20–41) (<i>n</i> = 16)	30.3 $\pm$ 7.4 (21–48) (<i>n</i> = 19)	–	0.610
Location of curve apex [<i>n</i> (%)]			–	0.585 [§]
T5	2 (5.9)	0 (0)		
T6	3 (8.8)	1 (2.3)		
T7	2 (5.9)	7 (15.9)		
T8	5 (14.7)	15 (34.1)		
T9	5 (14.7)	6 (13.6)		
T10	9 (26.5)	9 (20.5)		
T11	8 (23.5)	6 (13.6)		
Apical vertebral rotation (Nash-Moe) [<i>n</i> (%)]			–	0.428 [§]
0	2 (5.9)	2 (4.5)		
1	21 (61.8)	25 (56.8)		
2	11 (32.3)	17 (38.6)		
Extent of tonsillar descent			–	0.927 [§]
Grade I	18	24		
Grade II	14	17		
Grade III	2	3		
Syrinx size	29 cases	36 cases	–	
Maximal S/C ratio	0.62 $\pm$ 0.17	0.57 $\pm$ 0.21		0.385
Syrinx length (vertebrae)	10.1 $\pm$ 4.4	9.2 $\pm$ 5.5		0.441

CM-L left thoracic scoliosis secondary to Chiari malformation, CM-R right thoracic scoliosis secondary to Chiari malformation, S/C syrinx/cord

* Indicates analysis of variance (ANOVA)

[†] Indicates Pearson χ^2 test

[§] Indicates the Mann–Whitney *U* test, otherwise the student *t* test was used

Table 2 Comparisons of sagittal and pelvic parameters between the CM-L, CM-R and the control groups

Parameters	CM-L group	CM-R group	Control group	P value
Thoracic kyphosis (°)	42.3 ± 11.7 (19 to 70) ^{a,b}	29.6 ± 12.6 (1 to 59) ^a	28.7 ± 11.0 (2 to 63) ^b	<0.001
Lumbar lordosis (°)	−56.7 ± 11.9 (−38 to 89) ^{a,b}	−50.4 ± 12.5 (−25 to 74) ^a	−46.6 ± 9.8 (−25 to 69) ^b	<0.001
TJK (°)	3.5 ± 15.7 (−32 to 45)	1.6 ± 11.3 (−30 to 32)	3.6 ± 9.1 (−15 to 28)	0.607
SVA (mm)	−24.4 ± 23.6 (−72 to 94)	−12.6 ± 19.8 (−45 to 33)	−23.2 ± 29.8 (−102 to 94)	0.061
Pelvic incidence (°)	40.7 ± 11.6 (19 to 70)	40.5 ± 10.3 (20 to 70)	42.6 ± 9.3 (19 to 67)	0.434
Pelvic tilt (°)	3.4 ± 9.3 (−16 to 26) ^b	3.9 ± 6.9 (−14 to 20) ^c	9.9 ± 8.3 (−12 to 31) ^{b,c}	<0.001
Sacral slope (°)	37.3 ± 8.8 (14 to 64) ^b	36.7 ± 8.6 (18 to 54) ^c	32.8 ± 7.1 (19 to 52) ^{b,c}	0.003

CM-L indicates left thoracic scoliosis secondary to Chiari malformation, CM-R right thoracic scoliosis secondary to Chiari malformation, TJK thoracolumbar junctional kyphosis, SVA sagittal vertical axis

^a Indicates significant difference between the CM-L and CM-R groups

^b Indicates significant difference between the CM-L and control groups

^c Indicates significant difference between the CM-R and control groups

curve magnitude, location of the curve apex, apical vertebral rotation, syrinx size or extent of tonsillar descent. Regarding the 35 patients with a nonstructural distal curve (16 in the CM-L group and 19 in the CM-R group), Cobb angle of the TL/L curve did not differ significantly between the two groups.

Comparisons of the sagittal spinal and pelvic parameters between the CMS and control groups are presented in Table 2. The TK, LL and SS were found to be significantly greater in the CMS group, along with a significantly decreased PT ($P < 0.001$). The PI, TJK and SVA did not differ significantly between the two groups ($P > 0.05$).

Sagittal parameters were also compared between CMS patients with different curve directions (Table 2). The TK and LL were found to be significantly greater in the CM-L group when compared with the CM-R or control groups ($P < 0.001$) (Fig. 2), but did not differ significantly between the two latter groups. Compared with the control group, the SS and PT were, respectively, significantly larger and smaller in the CM-L and CM-R groups but were not different between the two CMS groups. In addition, the PI, TJK and SVA did not differ significantly between the three groups ($P > 0.05$).

Based on the definition, 3, 76 and 21 % of patients in the CM-L group were found to have a hypokyphotic, normal and hyperkyphotic thoracic spine, compared to 18, 75 and 7 %, respectively, in the CM-R group ($P = 0.035$). As for the LL, 44 % of patients in the CM-L group were within the normal range, whereas 50 % were hypolordotic and 6 % were hyperlordotic; in the CM-R group, the percentage of normal lordosis, hypolordosis and hyperlordosis was 48, 52 and 0 %, respectively ($P = 0.043$).

Correlations between all sagittal parameters are provided in Table 3. The LL was found to be strongly related to the TK and SS in all three groups and to the PI only in the control and CM-R groups. The PI was found to be

Table 3 Correlation coefficients (r) between the sagittal parameters for all three groups

Parameters	CM-L group	CM-R group	Control group
TK-LL	−0.529**	−0.406**	−0.297**
TK-TJK	0.006	0.351*	0.408**
TK-PI	0.177	0.026	−0.066
TK-PT	−0.117	−0.127	−0.020
TK-SS	0.358*	0.131	−0.062
LL-TJK	−0.122	−0.041	0.092
LL-PI	−0.112	−0.538**	−0.234**
LL-PT	0.339	0.108	0.273
LL-SS	−0.772**	−0.735**	−0.626**
TJK-PI	−0.225	−0.234	−0.175
TJK-PT	−0.229	0.040	0.003
TJK-SS	−0.052	−0.319*	−0.133

CM-L indicates left thoracic curve, CM-R right thoracic curve, TK thoracic kyphosis, LL lumbar lordosis, TJK thoracolumbar junctional kyphosis, PI pelvic incidence, PT pelvic tilt, SS sacral slope, SVA sagittal vertical axis

** $P < 0.01$; * $P < 0.05$

significantly related to PT and SS in all three groups. A significant correlation was also noted between TK and SS in the CM-L group.

Discussion

Currently, spinopelvic alignment and sagittal balance are considered as the major contributors in maintaining an energy-efficient upright posture in both normal and diseased states [1–3, 18]. By the same token and for the sake of achieving promising clinical outcomes, the restoration or maintenance of physiological sagittal plane contour appears to be mandatory during reconstructive spinal

Fig. 2 a A 15-year-old male with left thoracic scoliosis (55°, **a1**) secondary to Chiari malformation/syringomyelia. The standing lateral radiograph (**a2**) demonstrates the thoracic hyperkyphosis (52°) and the lumbar hyperlordosis (82°). **b** A 16-year-old male with Chiari

malformation-associated right thoracic scoliosis (53°, **b1**). Despite the presence of a hypokyphotic thoracic component (kyphosis: 19°, **b2**), the lumbar lordosis was within the normal range (45°, **b2**)

procedures. Recently, sagittal alignment in spinal disorders such as adolescent idiopathic scoliosis, Scheuermann's kyphosis and spondylolisthesis has received extensive investigation [1, 2, 12, 19]. While for patients with scoliosis secondary to Chiari I malformation, the sagittal profiles and, in particular, the sacropelvic alignment still remains incompletely understood. Moreover, despite the high incidence of left thoracic curve in CMS [5, 6, 8], only two previous studies differentially evaluated the sagittal

alignment with respect to curve direction and yielded contradictory results [5, 20]. Hence, by comparing CMS patients with age- and gender-matched asymptomatic adolescents, we attempt to elucidate the subtle differences in sagittal spinopelvic parameters. To our knowledge, the present study involves the largest cohort so far dedicated to the evaluation of sagittal alignment in CMS patients, with particular attention paid to the curve direction-related disparities.

By far, differences in sagittal spinal geometries between CMS and AIS patients have been well documented [4–6]. Ouellet et al. [21] reported that the apical lordosis (the typical feature of AIS) was absent in the majority of patients with scoliosis secondary to syringomyelia. Likewise, with historical normative values of TK as reference, Qiu et al. [6] reported that thoracic hyperkyphosis was found in 24 of 87 (28 %) CMS patients. Propst-Proctor and Bleck [22], however, found no significant difference in TK between 114 patients with scoliosis and 104 control subjects, although the etiology or curve pattern of the scoliosis was not clarified.

In the cohort presented herein, various sagittal parameters of thoracic, pelvic and lumbar segments were found to be significantly different between CMS patients and normal subjects (Table 2). Mirroring the trend that has been previously observed [5, 6], the average values of TK and LL were both significantly higher in CMS patients. However, when patients with distinct thoracic curve directions were analyzed separately, those differences existed only in the CM-L group. That is, the majority of patients with right thoracic curves were found to have normal sagittal spinal alignment. In addition, it is noteworthy that the values of TK and LL are smaller compared with Caucasian CMS patients, which again, certifies the role of ethnicity in affecting the sagittal morphology and spinopelvic anatomy [23]. Consequently, in dealing with the growing spine of CMS patients, the impacts of race and the coronal spinal deformity upon the sagittal morphology must be considered in efforts to explicate the standing sagittal posture.

As a key factor defining pelvic morphology, the PI was found to be smaller in CMS patients compared with normal controls though not reaching statistical significance. Regarding pelvic orientation, the average SS and PT were, respectively, larger and smaller in both CMS groups. At first glance, anteversion of the sacrum appears to be a compensatory mechanism in response to the increased TK and LL observed in the CM-L group. However, given that patients with right thoracic curves also had similar changes in SS and PT in spite of normal TK and LL (Table 2), this specific pattern of pelvic orientation seems more likely to be an intrinsic characteristic of CMS patients than being associated with the direction of the thoracic curves.

Currently, there remains scant and conflicting evidence surrounding the influence of curve direction on sagittal geometries of the spine and pelvis in CMS patients. Spiegel et al. [5], in 18 patients with thoracic scoliosis secondary to Chiari I malformation, found the TK to be larger for the 6 patients with a left curve ($46^\circ \pm 9^\circ$ versus $39^\circ \pm 16^\circ$). Recently, however, a retrospective review from Bradley et al. [20] revealed that the average kyphosis was significantly smaller in the three CMS patients with a left thoracic curve than in the nine patients with a right thoracic curve

($33^\circ \pm 3^\circ$ versus $50^\circ \pm 14^\circ$). Apparently, the limited sample sizes restrained the validity of their results and primarily contributed to this inconsistency. Besides, bearing in mind the well-documented correlation between age and sagittal spinal parameters [18, 24, 25], confounding biases might be potentially induced by the wide age ranges in both studies. Therefore, analyses in the current study were restricted to adolescent CMS patients with main thoracic curves. With the purpose of avoiding the potential influence of severe scoliosis on the sagittal alignment, patients with curve magnitude larger than 70° were also excluded.

In concordance with the findings of Spiegel et al. [5], our results showed that the TK and LL were significantly larger in the CM-L group than in the CM-R group (Table 2), as well as significantly increased percentages of hyperkyphosis and hyperlordosis. Considering the asymmetrical distribution of thoracic organs, we speculate that the occupied effect of anterior heart may potentially contribute to the hyperkyphosis of spine in the CM-L group. With respect to the increment of LL, although no explicit explanation can be obtained from the results of this study, it seems plausible that compensatory alteration occurs in the LL as a response to the increased TK to maintain a balanced posture. The strong correlation between TK and LL in the CM-L group, as shown in Table 3, partly supported this hypothesis. Therefore, when addressing the sagittal plane deformities of CMS patients, surgeons should take into account the possible chain relation of these parameters. Theoretically, adequate restoration of physiological thoracic kyphosis may provide an opportunity for spontaneous postoperative reciprocal improvement in the lumbar segments, as previously observed in patients with AIS [26]. Moreover, in spite of larger TK and LL in CMS patients with a left thoracic curve, the global sagittal balance (as defined by SVA) was overall well maintained. In this context, one crucial question remains as follows: are these patients at higher risk for postoperative sagittal plane decompensation (proximal or distal junctional kyphosis) following the restoration of normal kyphosis? In addition, no significant differences were found in the pelvic parameters, indicating that the lateralization of the thoracic curve in CMS patients is not associated with a specific pattern of sagittal morphology and balance. Furthermore, the apical vertebral rotation, as evaluated by the Nash-Moe method, did not differ significantly between the two groups. Added to the fact that the rotation was confirmed to be mild to moderate ($\leq$ grade 2) in all patients, it seems unlikely that the observed differences between CMS with different thoracic curve directions are attributed to the axial rotation.

The relationships between parameters were also investigated for the CMS and control groups. As shown in Table 3, the LL was significantly correlated with PI and SS in all cases except for patients in the CM-L group,

confirming the interdependence between the contour of the lumbar spine and pelvis in the maintenance of an efficient balanced posture [27]. With regard to the CM-L group, the lack of correlation between PI and LL might be attributed to the remarkably increased LL. Moreover, the LL was strongly associated with TK in all three groups, which, although does not definitely imply a causal relationship, indeed supports the concept that a modification in the orientation parameter of a given anatomical region will affect the adjacent anatomical segments [18].

To summarize, sagittal alignment of the spine and pelvis was found to be different between CMS patients and healthy adolescents, and between CMS patients with different curve directions. The presence of thoracic hyperkyphosis and hypokyphosis in a certain proportion of CMS patients highlights the necessity of sagittal plane assessment for surgical decision-making. In addition, no interaction was demonstrated between the pelvic parameters and the direction of the thoracic curves.

The lack of accurate axial plane evaluation is a recognized limitation of this study. Although the Nash-Moe method was used to estimate the rotation of the curve apex, it has been reported to be subjected to variability and overestimate the actual vertebral rotation [28]. Ideally, using three-dimensional spine parameters measured on CT or MR images would help to better understand the significance of sagittal alignment in the evaluation and treatment of CMS. In addition, as the inherent shortcoming of a retrospective and transverse study, the long-term implications of the radiographic findings are difficult to evaluate and need to be explored by future studies.

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

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