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Trunk Growth in Early-Onset Idiopathic Scoliosis Measured With Biplanar Radiography

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Abstract

Study Design: Cross-sectional and longitudinal retrospective study.

Objectives: To measure thoracic dimensions and volume during growth in early-onset idiopathic scoliosis (EOIS) patients and to compare them to a population of asymptomatic adults and to the previous literature.

Summary of Background Data: Data on trunk growth for scoliotic children between 6 and 14 years of age is sparse in the literature.

Methods: Thirty-six patients (29 girls and 7 boys, between 3 and 14 years old, average Cobb angle $33^{\circ} \pm 15^{\circ}$) were included, all with a minimum two-year follow-up. Sixty-one asymptomatic girls and 54 asymptomatic adults were included as control groups. All subjects underwent biplanar radiography and 3D reconstruction of the spine, pelvis, and rib cage. EOIS patients repeated their radiologic examination every six months. Cobb angle, rib cage volume, anteroposterior and transverse diameters, thoracic index, thoracic perimeter, pelvic incidence, and T1–T12 and T1–S1 distance were calculated. Reproducibility of measurement was assessed.

Results: Measurement reliability in such young patients was comparable to previous studies in adolescents and adults. Geometrical parameters of EOIS patients increased linearly with age. For instance, rib cage volume in girls with EOIS increased from 2200 cm³ at six to seven years of age to 4100 cm³ at 13–14 years (65% of adult values, 294 cm³/y). Comparison with asymptomatic girls showed that EOIS could affect growth spurt. Longitudinal analysis on a cohort of six girls who had a follow-up of six years confirmed the cross-sectional data.

Conclusions: In this longitudinal and cross-sectional study, trunk growth between 3 and 14 years of age was characterized, for the first time, with biplanar radiography and 3D reconstruction. The results can be useful to estimate patient growth and thus have potential application in the surgical planning of EOIS patients.

Level of Evidence: Level II, retrospective study.

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Keywords: Three-dimensional thoracic growth; Trunk size; 3D reconstruction; Spine; Surgery

Introduction

According to the Scoliosis Research Society, early-onset idiopathic scoliosis (EOIS) is a three-dimensional

deformity of the spine that starts before 10 years of age. Severe deformities can affect growth of the rib cage and lead to a decrease in pulmonary volume and to the development of a restrictive respiratory syndrome [1,2].

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IRB approval: Data collection was approved by the local ethical committee at the Robert Debré hospital, Paris, France.

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Respiratory involvement is mainly due to anatomic changes of the chest wall, which cause impaired movement and reduced compliance [3]. Thoracic insufficiency syndrome is difficult to predict or diagnose, especially in younger patients who cannot perform pulmonary function testing [4]. Nevertheless, such syndrome is strictly related to an alteration of thoracic growth.

Treatment of EOIS presents specific difficulties: conservative approaches can have a significant impact on slowing down or halting the progression of the deformity [5,6], but surgical approach is still needed for patients with aggressive progression to prevent respiratory impairment [7]. Surgery in such young patients must take into account potential growth of the thorax in such young patients. Fusionless surgery are now the standard surgical procedure to correct EOIS [8-10]; single- or dual-rod assemblies, with conventional or magnetic rod lengthening, both at some point require external actions to adapt rod length to the patient's growth, and the rate of these adjustment sessions must be planned by the surgeon. Therefore, normative values of EOIS thorax growth are important for the surgical management of EOIS.

Clinical and morphologic changes of the scoliotic spine and rib cage during growth have been described by Dimeglio [11,12]. This work represents the main reference for our knowledge on scoliotic trunk growth, but it is based on cross-sectional study rather than a longitudinal one, and it does not thoroughly describe the employed measurement methodology and the included cohort of subjects, which limits the potential exploitation of the data.

Few other studies assessed the correlation between spinal deformity and rib cage parameters specifically over the growth period, and most of them were based on conventional 2D radiography [13]. However, scoliosis is a three-dimensional deformity of the trunk, and 2D analyses are therefore limited. Optical surface acquisition is the only type of 3D reconstruction method that has been used to measure thoracic parameters in this specific population [14,15]. Although this method has some advantages for screening as it is completely noninvasive, it is an external examination that does not give reliable information on bone geometry.

The recent development and diffusion of biplanar stereoradiography gave access to high-quality radiographs without distortion while being 8–10 times less irradiating than conventional radiographs and 800 times less than computed tomography. 3D reconstruction methods have been developed, which can provide quantitative assessment of the trunk in the standing position [16-19]. Nevertheless, these methods were validated for adolescent idiopathic scoliosis patients as young as 10 years. It is still unclear if 3D reconstruction could maintain the same reliability with younger patients, given their uncomplete mineralization of the bony structures.

The main objectives of this study were to validate the reliability of spine and rib cage 3D reconstructions from

EOS biplanar stereoradiography in EOIS patients and to compute 3D thoracic dimensions of these patients during growth, in order to assess the influence of EOIS on these parameters when compared with asymptomatic children and adults and the existing literature.

Materials and Methods

EOIS cohort

After IRB approval, 36 patients (29 girls, 7 boys) diagnosed with EOIS and a minimal radiographic follow-up of two years were retrospectively included. Only thoracic and thoracolumbar scoliosis (apical vertebrae above the L1–L2 disc, Cobb angle $> 10^\circ$) were included. All patients were treated conservatively with brace, and none had previous spinal surgery. All patients underwent clinical and radiographic examination every six months, as detailed below, during normal follow-up.

Control groups

Sixty-one asymptomatic girls (11.2 ± 1.8 years old, between 8 and 14 years old) were retrospectively included to compare their growth to the EOIS cohort. Furthermore, a control group of 54 asymptomatic adults (27 females and 27 males, without any history of spinal deformity) was retrospectively included in order to estimate EOIS spine and rib cage parameters in terms of percentage of adult size. All control subjects were free of musculoskeletal pathologies and they were radiologically examined to exclude scoliosis. Mean age was 25 ± 5 years and 26 ± 6 years respectively for male and female young adult control groups. All subjects were classified Risser 5 [20].

Imaging and 3D reconstructions

Biplanar radiographs were acquired with subjects in weight-bearing free-standing position, hands on clavicle or on cheeks to avoid superimposition with the spine, with EOS device (EOS Imaging, Paris, France). 3D reconstructions of the spine, pelvis, and the rib cage were performed using previously validated methods [17,21].

The following parameters were calculated for each patient: rib cage volume (cm^3), T1–T12 vertical distance (mm), rib cage maximum perimeter (cm), maximum width (mm) and maximum thickness (mm), thoracic index (maximum thickness to maximum width ratio), and pelvic incidence ($^\circ$). Rib cage volume was calculated from the surface enclosing ribs from 1st to 10th [17]. Rib cage perimeter was calculated from the cross section of an elliptical cylinder tangent to the most external ribs (Fig. 1).

Reliability

Reliability of spine and rib cage 3D reconstruction was evaluated on a subcohort of 12 patients between 6 and 10

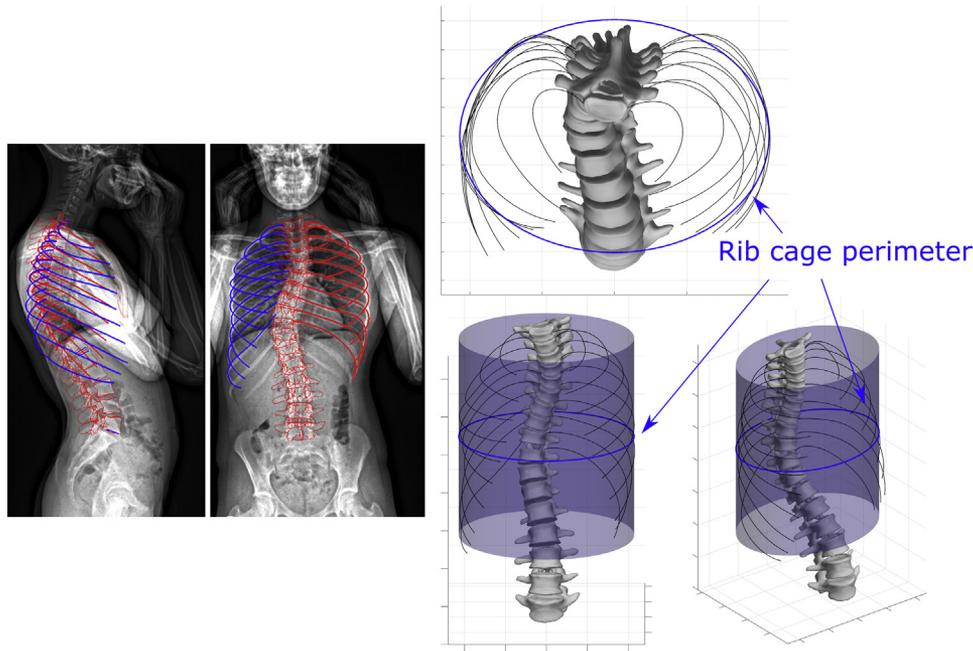


Fig. 1. Estimation of rib cage perimeter: a virtual oval cylinder was built tangent and external to the rib cage main axis, and its perimeters were calculated to approximate rib cage perimeter.

years of age. Two trained operators performed two 3D reconstructions for each patient. Inter- and intraoperator uncertainty were calculated according to ISO 5725 standard in terms of standard deviation, and intraclass correlation coefficients (ICCs) were calculated for all clinical parameters.

Statistical analysis

A cross-sectional analysis was conducted on the cohort (Table 1) by dividing radiographic examinations into age groups from 6–7 to 13–14 years of age. For each parameter and group, the mean and standard deviation were compared with the adult control group in terms of percentage of the adult values. The same analysis was applied to the control group of asymptomatic young girls.

For six patients, radiographic examinations were available from 6 to 13 years of age: a longitudinal analysis was

performed on the variation of clinical parameters of these patients with age.

Spearman rank test was used to analyze variations of the different parameters over growth period and correlations. The average growth speed of EOIS patients was also determined over this period for each parameter, as the slope of a linear regression between the parameter and age. Differences between EOIS and control young girls were analyzed at each age with Mann-Whitney tests. Analyses were performed with Matlab 2016b (The Mathworks Inc., Natick).

Results

Age at diagnosis was 7.5 ± 1 years (minimum 3 years old). All patients were classified Risser 0 at diagnosis. Mean Cobb angle at diagnosis was $33^\circ \pm 15^\circ$ (range: 12° – 80°). There were 30 main thoracic curves, 4 main thoracolumbar curves, and 2 double curves. The mean follow-up was 4.7 ± 1.4 years. A total of 283 3D reconstructions of the spine and rib cage were performed (144 of 36 EOIS patients at different ages, 54 young asymptomatic adults, 61 asymptomatic girls, and 12 reconstructions repeated twice for the reproducibility study).

Reliability

ICC and interoperator reproducibility values for the main clinical parameters are reported in Table 2. ICC was excellent (>0.8) for all parameters but T1–T12 kyphosis, for which it was good (0.72). Reproducibility was the same

Table 1
Number of 3D reconstructions for cross-sectional analysis.

Age group	Early-onset idiopathic scoliosis		Healthy girls
	Girls	Boys	
6- to 7-y-old	10	1	—
7- to 8-y-old	12	3	—
8- to 9-y-old	13	4	7
9- to 10-y-old	18	4	18
10- to 11-y-old	20	3	7
11- to 12-y-old	21	3	9
12- to 13-y-old	15	4	15
13- to 14-y-old	9	4	5

3D, three-dimensional.

Table 2

Intraclass correlation coefficient (ICC) and interoperator reproducibility of clinical parameters.

	Present study		Interoperator reproducibility in the literature				
	ICC	Interoperator reproducibility (2 SD)	Pietton et al. (2018)	Vergari et al. (2018)	Aubert et al. (2016)	Ilharreborde et al. (2011)	Humbert et al. (2009)
Spine and pelvis							
Cobb angle (°)	0.98	6.6				6.2	
T1–T12 kyphosis (°)	0.72	9.7				7	5.5
T1–T12 vertical distance (mm)	1.00	2.1					
Pelvic incidence (°)	0.89	5.6				4.7	3.4
Rib cage							
Maximum width (mm)	0.98	4.8	4.4	3.8	3.2		
Maximum thickness (mm)	0.84	8	14.7	9.7	9.3		
Maximum rib hump (°)	0.87	3.9	3.5	2.3	5		
Volume (cm ³)	0.97	197.9	387.3	395	294		

SD, standard deviation.

order of magnitude of previous studies. Rib cage volume reproducibility was better than in Aubert et al [17], but the average volume in this reproducibility cohort (2670 ± 406 cm³) was smaller than in the previous study (4528 ± 825 cm³), given the younger age of the present cohort.

Spinal and pelvic clinical parameters in EOIS patients

The average Cobb angle for girls in the first age group was $36^\circ \pm 19^\circ$ and $59^\circ \pm 12^\circ$ in the last one. Progression of the curves was monotonic during this period at the average speed of 3.3° per year. The increase of Cobb angle with age in the boys group was not significant ($p > .05$).

The average vertical growth speed for the T1–T12 segment was 0.7 cm/y for both girls and boys (Spearman rho = 0.71 and 0.76, respectively, $p < .001$; Fig. 2). In the 13- to 14-year-old girls group, T1–T12 vertical distance

was already almost 90% of the adult average distance, whereas it was at 80% in the boys group.

The average vertical growth speed for the T1–S1 segment was 1.2 cm/y for girls and 1.3 cm/y for boys (Spearman rho = 0.8, $p < .001$; Fig. 2).

Pelvic incidence tended to significantly increase by 1.4° /y in girls (Spearman rho = 0.37, $p < .001$; Fig. 3). No significant change was observed in boys, who remained at about 75% of the adult values.

The Cobb angle was not correlated to T1–T12 or T1–S1 distances ($p > .05$).

Rib cage in EOIS patients

Rib cage volume in girls with EOIS reached 50% of the adult value at age 10 years and at age 11 years for boys. The average growth speed for rib cage volume was 294 and 319 cm³/y for girls and boys, respectively.

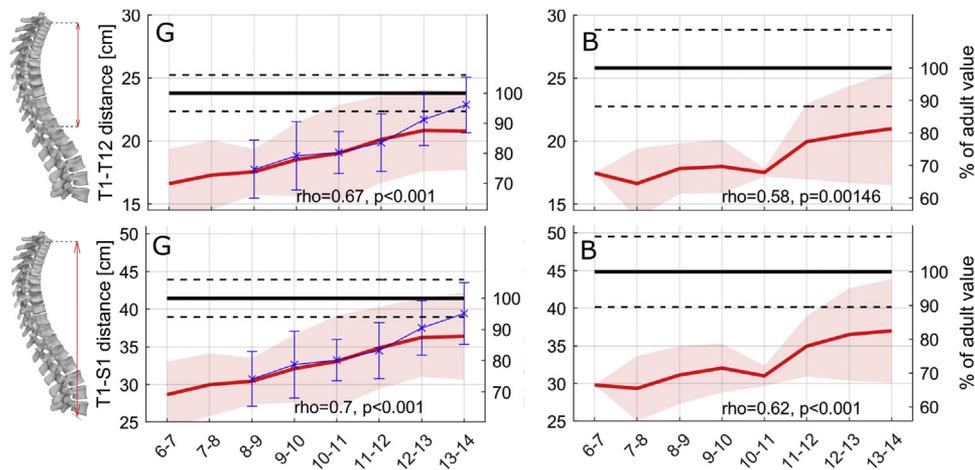


Fig. 2. T1–T12 and T1–S1 vertical distances in girls (G) and boys (B, average values \pm 2 SD shaded area) with early-onset scoliosis compared with asymptomatic girls (blue bars) and adults (average \pm 2 SD black lines). Spearman rank test rho and p value for the correlation between parameters in patients and age are reported.

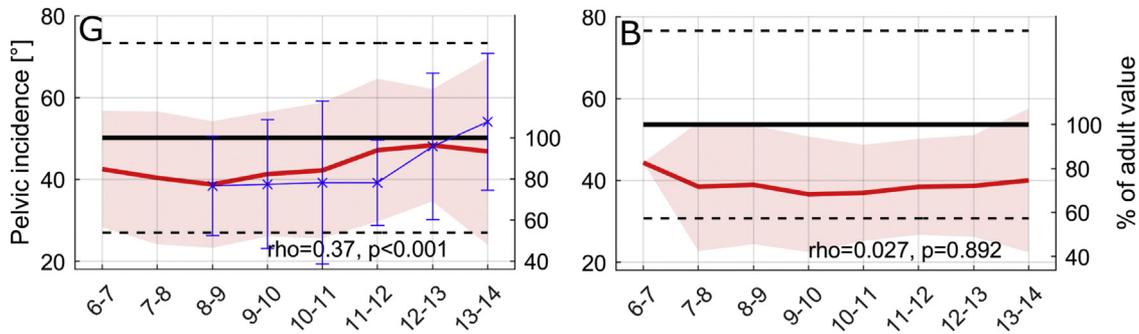


Fig. 3. Pelvic incidence in girls (G) and boys (B, average values \pm 2 SD red-shaded area) with early-onset scoliosis compared with asymptomatic girls (blue bars) and adults (average \pm 2 SD black lines). Spearman rank test rho and p value for the correlation between parameters in patients and age are reported.

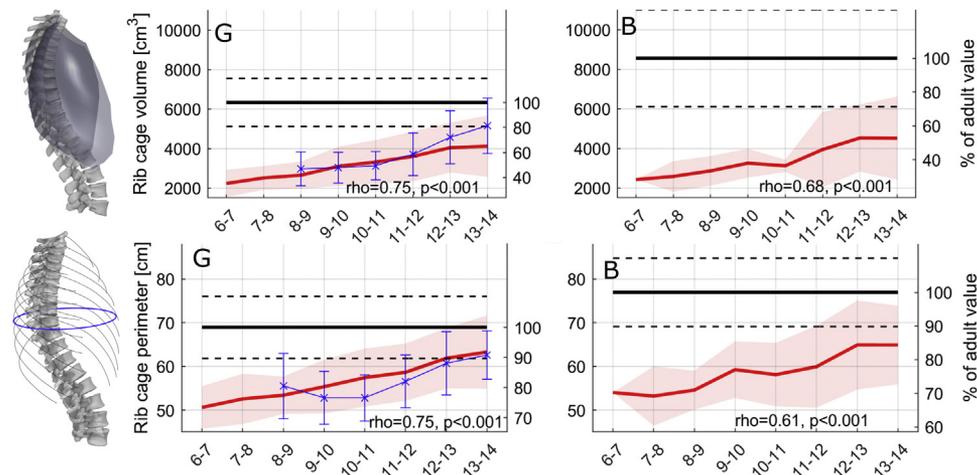


Fig. 4. Rib cage volume and perimeter in girls (G) and boys (B, average values \pm 2 SD red-shaded area) with early-onset scoliosis compared with asymptomatic girls (blue bars) and adults (average \pm 2 SD black lines). Spearman rank test rho and p value for the correlation between parameters in patients and age are reported.

Rib cage perimeter increased from 51 to 63 cm in girls with EOIS (75% and 92% of adult values; Fig. 4) and from 54 to 65 cm in boys with EOIS (54% to 90% of adult values). The average growth speed was 1.8 cm/y for girls and 2 cm/y for boys (Spearman rho = 0.8, $p < .001$).

Growth speed for rib cage thickness was 0.4 cm/y both for girls and boys (Fig. 5), whereas the rib cage width increased by 0.7 cm/y (Fig. 5). The thoracic index did not change significantly (Fig. 5).

The Cobb angle was correlated to rib cage volume (Spearman rho = 0.22, $p = .007$), width (rho = 0.28, $p = .0006$), thickness (rho = 0.34, $p < .0001$), and perimeter (rho = 0.49, $p < .0001$).

Longitudinal analysis in EOIS patients

Figure 6 shows the change of trunk parameters for five girls and one boy with EOIS who were followed for six years, compared with the cohort of girls with EOIS.

Longitudinal data confirms the cross-sectional analysis. Moreover, it can be noticed that those patients who had smaller-than-average parameters at 6 years of age tended to remain below average at 14 years (and the opposite is also true).

Comparison between EOIS and asymptomatic girls

Rib cage perimeter, width and thickness, rib cage index, and pelvis incidence were similar between asymptomatic and girls with EOIS at all ages ($p > .05$; Figs. 2–5). However, asymptomatic girls showed signs of growth spurt at 12–13 years of age, when rib cage volume, T1–T12, and T1–S1 distances suddenly started increasing faster (Figs. 2 and 4). In particular, rib cage volume between 12 and 14 years of age was significantly higher in asymptomatic girls than the girls with EOIS ($p < .05$), whereas T1–T12 ($p = .006$) and T1–S1 distances ($p = .03$) were higher in the 13–14 years range.

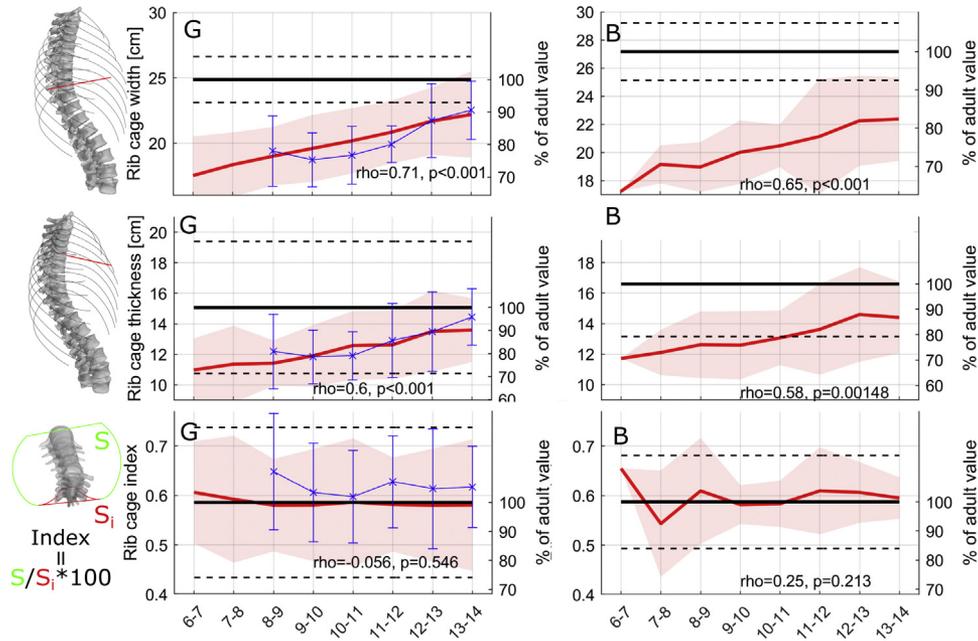


Fig. 5. Rib cage width, thickness, and index (maximum thickness to maximum width ratio) in girls (G) and boys (B, average values \pm 2 SD red-shaded area) with early-onset scoliosis compared with asymptomatic girls (blue bars) and adults (average \pm 2 SD black lines). Spearman rank test rho and p value for the correlation between parameters in patients and age are reported.

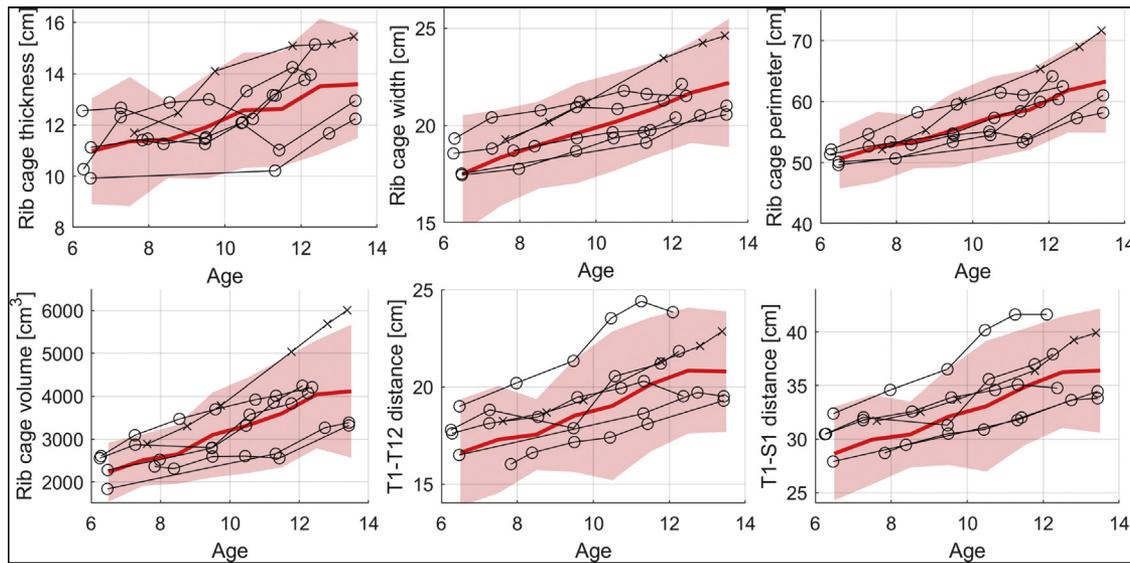


Fig. 6. Longitudinal analysis of five early onset idiopathic scoliosis girls (circles) and one boy (crosses), compared with early-onset idiopathic scoliosis girls (average values \pm 2 SD red-shaded area).

Discussion

In the present work, thorax sizes in young EOIS patients (6–14 years old) were quantified through low-dose biplanar radiography and 3D analysis. Thoracic growth was compared with a population of adults, finding that rib cage and spine tend to grow linearly in time, and that only a few parameters are close to the adult values at 14 years. Girls with EOIS were also compared with a cohort of age-matched asymptomatic girls: growth appeared similar until 12 years of age. However,

girls with EOIS did not show signs of growth spurt, whereas asymptomatic girls did. Moreover, longitudinal analysis over 6 years of follow-up showed that potential growth in EOIS patients can be estimated at early stages. These normative data can help the surgeon when planning the rate of interventions to adjust rod length in fusionless surgery.

Rib cage 3D reconstructions from biplanar radiographs have not previously been validated for subjects younger than 10 years; the reproducibility study showed that the method was as reproducible as in adolescents and adults

(Table 2). Reproducibility of pelvic incidence and T1–T12 kyphosis was slightly lower than in previous works, possibly because of the incomplete calcification of the pelvis and vertebrae.

In their prospective studies using optical trunk surface acquisition, Charles et al. [14,15] did not find any significant difference between asymptomatic children and mild or severe scoliosis patients, regardless of the severity, except for the anteroposterior diameter, which decreased significantly as the Cobb angle increased. In the present study, asymptomatic girls showed larger rib cage volume and longer T1–S1 and T1–T12 segments than girls with EOIS after 12 years of age.

The main limitation of this study is that the cross-sectional cohort was relatively small; the limited number of patients in each age subgroup did not allow statistical analysis between groups, but only on the global growth trends. Post hoc analysis showed that statistical comparison of EOIS and asymptomatic girls at 13–14 years of age had a power of 0.8 for volume comparison, and of 1 for vertical distances.

All patients had at least two years' follow-up, which is more than can be found in the literature, and which amounted to a total of 156 3D reconstructions (ie, 156 data points). Although six years' longitudinal data were only available for six patients, including one boy, this longitudinal study corroborated the cross-sectional analysis. Moreover, control cohorts were composed of young adults and asymptomatic girls; no asymptomatic age-matched boys were included. The same inclusion bias that often leads to include more girls than boys in studies on scoliosis also had an effect on the inclusion of asymptomatic boys.

Dimeglio et al. [11,12] found that trunk growth followed a linear pattern between the 5 and 10 years of age, and it was followed by an increased growth rate coinciding with the growth spurt. In this study, all those parameters that tended to increase did so linearly in EOIS patients. For instance, the average vertical growth speed for the T1–T12 segment was 0.7 cm/y for both girls and boys with EOIS, which is consistent with Dimeglio et al. [22]. However, those authors found that growth speed increased to 1.1 cm/y thereafter. Two hypotheses could explain such discrepancy: first, a statistical bias due to the cross-sectional analysis, which tends to flatten the growth curve because of different times of growth spurt. However, none of the six patients included in the long-term longitudinal analysis showed signs of growth spurt (Fig. 6). Second, the radiologic follow-up might be too short to observe the growth spurt usually linked with puberty at the age of 12 years for girls and 14 years for boys. Nevertheless, an increase in growth speed could be observed in the cross-sectional control group of asymptomatic girls. This suggests that growth-spurt onset could be more variable or less pronounced in EOIS patients.

The average rib cage volume reached 50% of adult value at 10 years of age in girls and one year later in boys, consistent with the results of Dimeglio et al. [12]. In both sexes, thoracic volume almost doubled between 6 and 14 years, but it was still far from normal adult values (65% in girls and 60% in boys), suggesting either a decrease of thoracic volume or a late development of the thorax in this population. Indeed, asymptomatic 14-year-old girls already had a rib cage volume of 80% of the expected value at adulthood.

At 13–14 years, the T1–T12 segment was 21 ± 1 cm in girls with EOIS and 23 ± 1 cm (96% of adult values) in asymptomatic girls. The T1–T12 segment reached 18 cm at approximately 10 years both for girls and boys with EOIS. Dimeglio et al. [11,12] reported this length for five-year-old children. However, these authors also reported that T1–T12 segment in adults was 27 cm, against 24 ± 0.7 cm in the present study, without providing any methodologic detail to reproduce their measurements.

According to those authors, the thoracic perimeter attains a mean value of 89.2 cm and 85.4 cm for boys and girls, respectively, against 72.1 cm and 69.2 cm in this study. However, the latter measurements did not include soft tissues, so lower values were expected. The advantage of this method is that it neglects differences in body mass between subjects, which does not directly affect the respiratory capacity.

It might be surprising that rib cage volume at 14 years of age was 65% of adult value, whereas the perimeter and T1–T12 distances were already 90% (at least in girls with EOIS). However, this is expected because the relationship between the volume, height, and perimeter are not linear: a 40% volume reduction and 10% height reduction correspond to only a 10% reduction in perimeter. Calculation details are provided in Appendix 1.

Pelvic incidence tended to slightly increase with age in girls with EOIS (Spearman $\rho = 0.37, p < .001$), reaching adult values at 11–12 years of age; a similar pattern was observed in asymptomatic girls (Fig. 3). Pelvic incidence remained lower than 80% of the adult reference values in boys with EOIS. Previous studies showed that changes in the pelvis occur during the acquisition of lumbar lordosis and bipedalism in infancy [23,24].

In conclusion, 3D reconstruction methods of spine and rib cage, which were previously validated in adults and adolescents, were also reliable in patients as young as six years. The growth of bony trunk regions was measured in 3D, and it showed that growth can be affected by EOIS.

Such an approach could be useful in clinical routine to estimate growth when planning fusion surgery, and in particular to decide when to implant growing rods. Moreover, thanks to the low radiation dose of biplanar radiography, the measurement does not pose an additional risk for the patient. Longitudinal data confirmed the cross-

sectional analysis and showed that the growth potential could be estimated from early assessment; this could have an impact on surgical planning, especially concerning growth-friendly approaches. Further data collection would be needed to follow the growth of these children to skeletal maturity and to confirm the presented results on a larger cohort.

Key points

- Trunk growth in early-onset scoliosis patients was measured with low-dose biplanar radiography and 3D reconstructions.
- Rib cage volume, perimeter, width, and thickness increased linearly between 6 and 14 years of age.
- This growth data can be useful to estimate patient potential growth when planning fusionless surgery.

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Appendix 1

In this appendix, we will highlight the relationship between rib cage volume, perimeter, and height, to show that a large reduction in volume can correspond to a small reduction in perimeter. If for simplicity we consider a perfectly cylindrical rib cage of radius $R = 10$ cm (half the width or depth of women's ribcage; Fig. 3) and height $H = 24$ cm (as the adult T1–T12 distance; Fig. 6), we obtain an approximated volume $V = \pi \cdot H \cdot R^2 = 7500$ cm³ and perimeter $p = 2 \cdot \pi \cdot R = 62$ cm, which are

consistent with the values for women shown in Figure 2. A 40% reduction of volume yields $V_r = 7500 \cdot 0.6$ cm³ = 4500 cm³, whereas a 10% reduction in the T1–T12 distance yields $H_r = 24 \cdot 0.6$ cm = 21.6 cm (Figs. 2 and 6). From this reduced volume, we can estimate the new radius as $R_r = (V_r/H_r/\pi)^{0.5} = 8.2$ cm, and therefore a new perimeter of $P_r = 2 \cdot \pi \cdot R_r = 51.3$ cm. Hence, an 18% reduction of the rib cage perimeter corresponds to a 40% reduction of rib cage volume and 10% reduction of height.