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ORIGINAL ARTICLE

Proximal junctional kyphosis in thoracic adolescent idiopathic scoliosis: risk factors and compensatory mechanisms in a multicenter national cohort

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Abstract

Introduction Proximal junctional kyphosis (PJK) is a frequent complication, up to 46%, in adolescent idiopathic scoliosis surgical treatment (AIS). Several risk factors have been evoked but remain controversial. The purpose of this study was to analyze the incidence of PJK in a multicenter cohort of AIS patient and to determine risk factor for PJK.

Materials and methods Lenke I and II AIS patients operated between 2011 and 2015 (minimum of 2-years follow-up) were included. On fullspine X-rays, coronal and sagittal radiographic parameters were measured preoperatively, postoperatively and at final follow-up. Occurrence of radiological PJK corresponding to a 10° increase in the sagittal Cobb angle, measured between the upper instrumented vertebra (UIV) and UIV + 2, between postoperative and 2-years follow-up X-rays, was reported.

Results Among the 365 patients included, 15.6% ($n = 57$) developed a PJK and only 10 patients required a revision surgery. Preoperatively, PJK patients had significantly larger pelvic incidence (57° ± 13° vs. 51° ± 12°), larger lumbar lordosis (LL) (63° ± 12° vs. 57° ± 11°) and bigger C7 slope. Postoperatively (3 months), in the non-PJK group, thoracic kyphosis (TK) was increased and LL was not significantly different. However, postoperatively, in the PJK group, no significant change was observed in TK, whereas C7 slope decreased and LL significantly increased. There was also a postoperative change in inflection point which was located at a more proximal level in the PJK group. Between postoperative time and final follow-up, TK and LL significantly increased in the PJK group.

Conclusion PJK is a frequent complication in thoracic AIS, occurring 16%, but remains often asymptomatic (less than 3% of revisions in the entire cohort). An interesting finding is that patients with high pelvic incidence and consequently large LL and TK were more at risk of PJK. As demonstrated in ASD, one of the causes of PJK might be postoperative posterior imbalance that can be due to increased LL, insufficient TK or inflection point shift during surgery.

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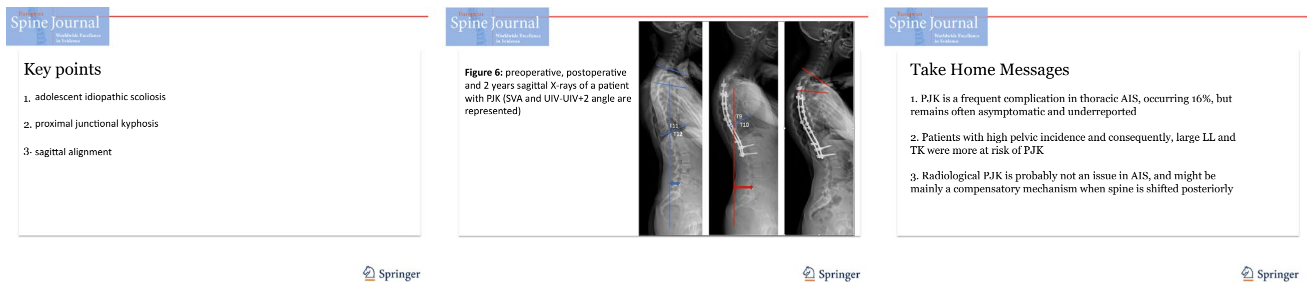
Extended author information available on the last page of the article

Graphical abstract These slides can be retrieved under Electronic Supplementary Material.



Keywords Adolescent idiopathic scoliosis · Proximal junctional kyphosis · Sagittal alignment

Introduction

Proximal junctional kyphosis (PJK) is one of the most frequent iatrogenic sagittal complications in adolescent idiopathic scoliosis surgery (AIS), with a reported incidence ranging from 7 to 46% [1, 2]. The high variability of PJK rates can be partly explained by the use of different definitions and the poor visibility of the upper thoracic spine on lateral radiographs. The definition in AIS was radiological and corresponded to a 10° increase in the sagittal Cobb angle, measured between the upper instrumented vertebra (UIV) and UIV + 2 [3, 4]. Multiple risk factors have been suggested, not always modifiable, such as fusion levels selection, correction technique and approach, type of instrumentation at the UIV, thoracoplasty, high preoperative thoracic kyphosis and finally postoperative flattened sagittal alignment [1, 4, 5, 7–9]. However, in the literature, cohorts are often heterogeneous and few studies have distinguished and reported PJK occurrence among the different Lenke types [1, 10]. In Lenke 5 curves, a mismatch between pelvic incidence (PI) and postoperative lumbar lordosis (LL) can lead to a posterior shift of the fusion mass and therefore be responsible for an adaptive PJK. However, the pathogenesis seems more complex in thoracic curves (Lenke 1 and 2). In particular, whether pediatric PJK mostly occur in "at-risk" patients, or whether they are more iatrogenic due to surgical technique and/or postoperative change in sagittal alignment is still controversial. Although PJK is not necessarily associated to poor patients outcomes and to revision surgery, a better knowledge of risk factors remains necessary [4–8]. The primary aim of this study was, therefore, to analyze the incidence and risk factors for PJK in a large multicenter cohort of thoracic AIS patients. The second objective was to describe the most frequent subsequent compensatory mechanisms in the sagittal plane.

Materials and methods

Patients

After IRB approval, a multicenter database of AIS patients (8 orthopedic departments), operated between January 2011 and February 2015 for thoracic structural curves (Lenke 1 or 2), was retrospectively analyzed. Minimum 2-year follow-up was required. All patients underwent coronal and sagittal full spine standing radiographs preoperatively, postoperatively and at latest examination. Patients with previous spine surgery or anterior release were excluded.

Parameters

Data were consecutively collected in medical charts. Demographic and surgical data were reported. Radiological parameters included pelvic parameters (pelvic incidence, sacral slope and pelvic tilt), spinal parameters (maximal thoracic kyphosis, TK, maximal lumbar lordosis, LL and cervical lordosis, CL), C7 slope (angle between superior endplate of C7 and the horizontal) in the sagittal plane, and Cobb angle of the main curve, proximal and distal contra curves, shoulder tilt (angle between the coracoid processes and horizontal) and global coronal C7 tilt (angle between the line joining the center of C7 and the middle of the sacral endplate with the vertical) in the coronal plane (Figs. 1, 2). The location of the inflection point, defined as the transitional vertebra between TK an LL, was also recorded [11]. Global sagittal alignment was assessed using C7 tilt (Fig. 1). Radiological PJK was defined by an increase of 10° or more of the sagittal Cobb between the superior endplates of the upper instrumented vertebra (UIV) and UIV + 2, between the first postoperative images and the last follow-up, as described by Kim et al. [4] (Fig. 3).

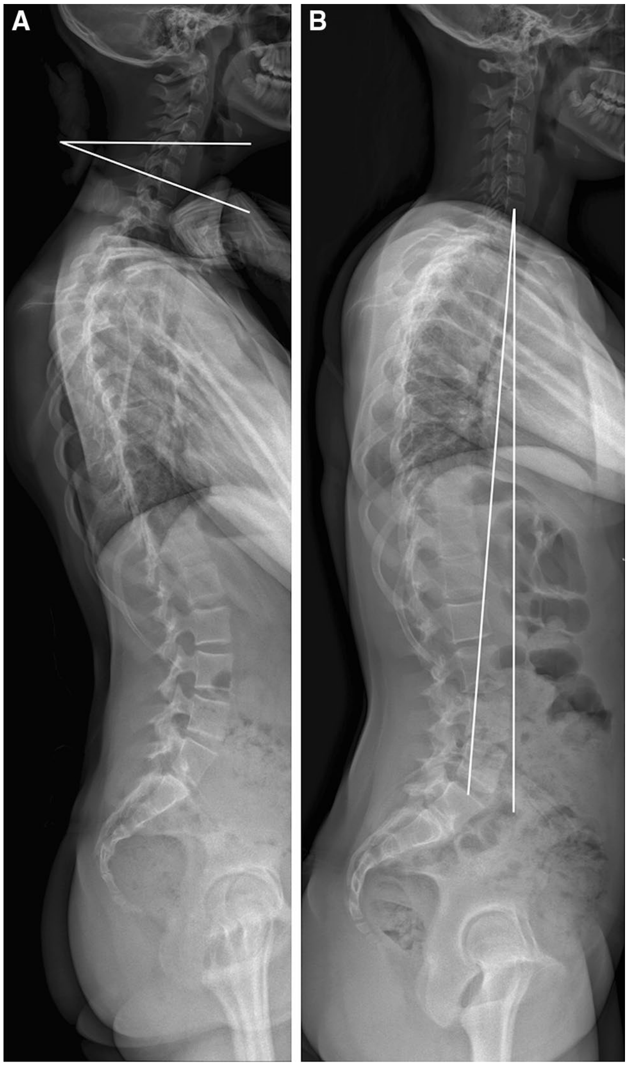


Fig. 1 Sagittal X-rays with C7 slope (on the left) and C7 tilt (on the right) measurements

A single trained operator performed all 2D radiographic measurements using Keops Software (SMAIO, Lyon, France), which is an update of the previously validated Optispine software (Paris, France) (SMAIO, Lyon, France) [12].

Statistical analysis

A descriptive analysis of the entire cohort was first performed for demographic, radiographic and surgical data. The incidence of PJK was firstly determined based on treating surgeon's report. Then, the rate of radiological PJK was compared to the one reported in charts. Second, patients with (PJK) and without radiological PJK (Non-PJK) were compared, using Student T test or X^2 as appropriate, and we tried to identify patients at risk of PJK. Third, influence of surgical parameters on occurrence of PJK was assessed. Finally, compensatory mechanisms in case of PJK were analyzed on sagittal X-rays at follow-up. Results are presented as mean ± standard deviation for continuous variables and as percentage for categorical variables. Statistical significance was defined as $p < 0.05$. Statistical analysis was performed with Stata version 13.0 (Statacorp LP, Lakeway Drive, College Station, Texas).

Results

Descriptive analysis of the cohort

Three hundred and sixty-five patients were included, with a mean age of 15 ± 2.6 years. There was a majority of females (85%), and Lenke 1 curves were the most frequent (81% of the patients), with 46% of Lenke 1A (Table 1). Males were more likely to present Lenke 2 curves (20% of the patients with Lenke 2 were males, whereas only 11% of the patients with Lenke 1 were males; $p = 0.04$). No significant difference was observed between Lenke 1 and Lenke 2 subgroups regarding age (14.9 ± 2.4 vs. 14.4 ± 2.3, $p > 0.05$) and BMI (19.6 ± 3.2 vs. 19.8 ± 2.9, $p > 0.05$).

Incidence of PJK

Twenty cases (6%) of PJK were prospectively reported in the medical reports by the treating surgeon. However, 57 cases (15.6%) of radiographic PJK were retrospectively diagnosed at latest follow-up (average 2.5 ± 0.4 year). In 10 cases (17% of the radiological PJK), a revision surgery was performed (in 7 cases due to proximal implants loosening or prominence). These 10 cases were among the 20 patients detected by the surgeons.

Non-modifiable demographic and radiological risk factors

The 57 PJK patients were not significantly different from controls (non-PJK patients) regarding age, gender or BMI (Table 2). The rate of PJK was not significantly different between Lenke 1 and 2 curves (17 vs. 11%; $p = 0.26$). Preoperative shoulder balance, coronal C7 tilt and Cobb angle of main curve, proximal and distal contra-curve were not associated to higher PJK occurrence ($p > 0.1$) (Table 3). The only risk factors identified were preoperative pelvic incidence ($p = 0.004$), lumbar lordosis ($p = 0.0002$) and sagittal C7 slope ($p = 0.01$). However, preoperative thoracic kyphosis, cervical lordosis and sagittal C7 tilt were not significantly different between the two groups (Table 3).

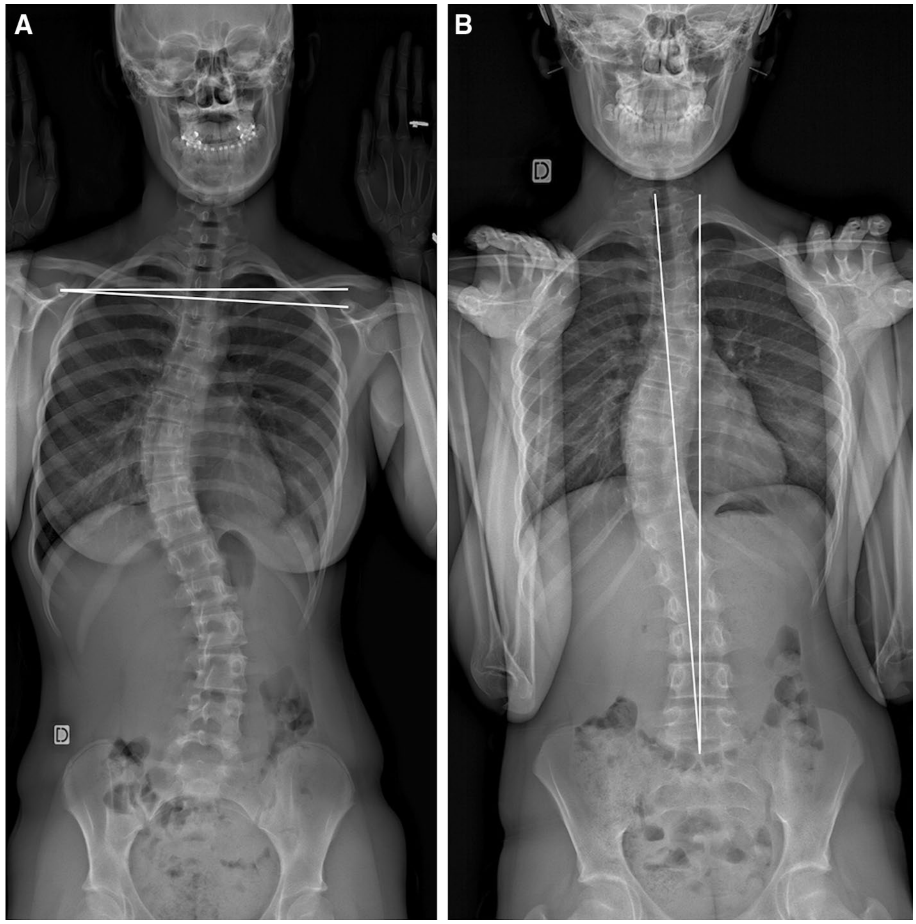


Fig. 2 Shoulders tilt (on the left) and global coronal C7 tilt (on the right)

Modifiable surgery-related risk factors

The location of either UIV or LIV (lower instrumented vertebra) was not associated to higher risk of PJK (Figs. 4, 5). Thoracoplasty was not more frequent in the PJK group ($p=0.23$), and the type of implant (hook or screw) selected for proximal fixation was not significantly associated with PJK incidence ($p=0.31$) (Table 4). The only surgery-related factor found in the analysis was the type of rod alloy, with significantly higher risk with cobalt chromium (28%), compared to titanium and stainless steel (12%) ($p<0.0001$).

Some postoperative (3 months) radiographic changes were also associated with higher PJK incidence. Postoperative C7 slope was significantly lower in PJK patients than non-PJK patients, while other coronal and sagittal parameters were not significantly different (Table 5). In the non-PJK group, thoracic kyphosis increased postoperatively from 27° ± 18° to 34° ± 16° ($p<0.0001$) whereas no changes occurred in the PJK group (32° ± 16° to 33° ± 14°; $p=0.7$). C7 slopes increased from 16° ± 10° to 22° ± 10° ($p=0.002$) in the non-PJK patients, while C7 slope decreased in the PJK patients (20° ± 9° to 14° ± 18°; $p=0.003$). Lumbar lordosis was not significantly modified in the non-PJK group (56° ± 11° to 57° ± 11°; $p=0.5$), but lumbar lordosis decreased in the PJK group (63° ± 12° to 57° ± 11°; $p<0.0001$), while remaining adapted to pelvic incidence in each group (Table 6).

Number of vertebrae included in LL was not significantly different between pre- and postoperative measurements in the PJK group (5.2 ± 1.8 vs. 5.1 ± 1.4; $p=0.6$). Nevertheless, postoperative lumbar lordosis was longer than preoperative LL in 39 patients of the PJK group (68%), with a more proximal inflection point (2 levels higher on average) (Fig. 6).

Compensatory mechanisms

During the follow-up, TK significantly increased by 12° ± 9° in PJK patients. This increase in TK was associated with a significant increase in PJK angle. A trend toward C7 slope increase between the immediate postoperative period and latest examination was also observed, but the difference did not reach significance. Another mechanism to restore alignment was the increase in LL during follow-up in the PJK group to compensate for the final increase in TK (Table 7). Nevertheless, final lumbar lordosis of 65° ± 12° remained adapted to the pelvic incidence (57° ± 13°). However, no

Fig. 3 Sagittal X-rays: early postoperative (on the left) and at 2 years with a PJK (on the right). PJK measurements

Table 1 Demographic data and Lenke type of the entire cohort

	n	Mean ± SD (or %)
Sex	365	
Female	312	85
Male	53	15
Lenke type	365	
Lenke I	296	81
IA	166	46
IB	71	19
IC	59	16
Lenke II	69	19
IIA	38	10
IIB	20	6
IIC	11	3

Table 2 Demographic data and Lenke type: comparison between PJK and non-PJK patients

	Non-PJK (n = 308)	PJK (n = 57)	p
Age (years)	15 ± 2.4	14.7 ± 2.5	0.3
Sex			0.3
Female	262	50	
Male	46	7	
BMI	19.6 ± 3.2	19.9 ± 3	0.6
Lenke type			0.3
Lenke I	245	49	
Lenke II	63	8	

change was reported in global sagittal alignment between postoperative and follow-up (Table 7). No loss of coronal correction was observed in each group at follow-up.

Discussion

The current series is one of the largest of the literatures dedicated to thoracic AIS. Numerous risk factors were identified, but distinction was made between the non-modifiable ones, related to the patient and its preoperative alignment (pelvic incidence, preoperative lumbar lordosis and sagittal C7 slope), and the modifiable ones, more related to the surgical procedure (rods alloy, postoperative hypokyphosis and change in inflection point location).

Incidence of PJK

PJK rate in this series was 15.6%, and stood in the lower range of the literature for similar patients (26–35%) [3–5, 9]. The high rate of PJK in some cohorts can be explained by a different definition. For some authors, PJK is defined by an increase of 5° or more of the sagittal Cobb between the superior endplates of the UIV and UIV + 2 [13]. Thus, PJK rate might be higher when using this definition. Therefore, a unique definition of radiological PJK seems necessary to compare studies and improve patient treatment [13]. In addition, most series included all Lenke types and were inhomogeneous. Looking specifically at Lenke 1 and 2, the incidence of PJK was similar to the literature; however, Lonner et al. [1] in his series did not find any difference in PJK incidence between Lenke 1 and 2.

One important finding was the difference between the incidence of PJK reported by surgeons in their report (6%) and the rate (15.6%, 3 times higher) retrospectively determined by independent observers on radiographs. This difference highlights an underestimated problem in many studies, since the visibility of anatomical structures is altered in the upper thoracic spine, while most sagittal Cobb angles need

Table 3 Preoperative radiographic parameters: comparison between PJK and non-PJK patients (mean ± SD)

	Entire cohort ($n = 365$)	Non-PJK ($n = 308$)	PJK ($n = 57$)	p
Sagittal parameters				
Pelvic incidence	52° ± 12°	51° ± 12°	57° ± 13°	0.004
Pelvic tilt	10° ± 8°	9° ± 8°	10.9° ± 9°	0.16
Maximal lumbar lordosis	58° ± 12°	57° ± 11°	63° ± 12°	0.0002
Maximal thoracic kyphosis	28° ± 18°	27° ± 18°	32° ± 16°	0.06
Cervical lordosis	10° ± 19°	11° ± 20°	7° ± 17°	0.20
C7 sagittal tilt	3.1° ± 2.4°	3.2° ± 2.4°	2.8° ± 2.4°	0.28
C7 slope	17° ± 10°	16° ± 10°	20° ± 9°	0.01
PJK angle	4° ± 8°	4° ± 10°	2° ± 10°	0.20
Coronal parameters				
Main curve	56° ± 13°	55° ± 13°	58° ± 13°	0.15
Proximal contra-curve	30° ± 10°	30° ± 10°	31° ± 12°	0.80
Distal contra-curve	29° ± 10°	29° ± 11°	29° ± 10°	0.85
Shoulders tilt	2° ± 2°	2° ± 2°	3° ± 2°	0.13
C7 coronal tilt	1.8° ± 1.5°	1.8° ± 1.5°	2° ± 1.8°	0.31

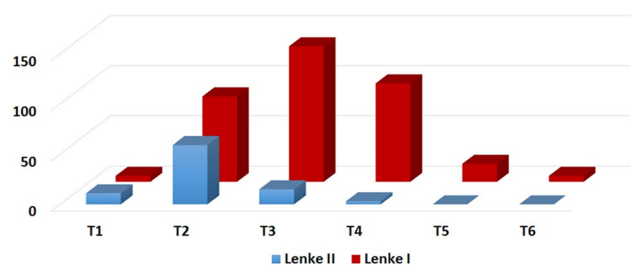


Fig. 4 UIV distribution in Lenke I and II

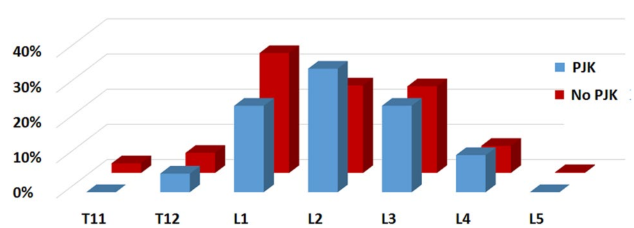


Fig. 5 LIV distribution between PJK and non-PJK patients

Table 4 Surgical data: comparison between PJK and non-PJK patients

	Entire cohort ($n = 365$)	Non-PJK ($n = 308$)	PJK ($n = 57$)	p
Thoracoplasty	87	77	10	0.23
Proximal implants				
Hooks	55	45 (15%)	10 (18%)	0.31
Claw	272	227 (74%)	45 (79%)	
Screw	1	1 (0%)	0 (0%)	
Screw + hook	37	35 (11%)	2 (3%)	

to be calculated between T1 and T4. Moreover, given that PJK is rarely symptomatic in AIS, it is underreported in medical reports. Consequently, to improve epidemiological studies on PJK, a consensual definition is needed as well as an independent measurement and accurate analysis.

Risk factors

Many risk factors have been reported in the literature. But the distinction between the ones modifiable and non-modifiable is rarely made. Surgeons need to know both who the patients "at risk" are preoperatively, but also focusing on the main mistakes to avoid during surgery.

In the current study, none of the demographic data was found as a risk factor. In particular, patients with higher BMI were not found to develop more PJK, as previously reported [7]. Preoperative PI and associated LL were significantly greater in the PJK group (PI: 57° ± 13° vs. 51° ± 12°, LL: 63° ± 12° vs. 57° ± 11°). This result was not reported in the literature. Higher C7 sagittal slope (− 20° ± 9° vs. − 16° ± 10°) corresponding to a higher proximal thoracic kyphosis was also observed. As previously described, patients with higher risk of PJK had a tendency to have greater TK (32° ± 16° vs. 27° ± 18°; $p = 0.06$) [4, 5].

Concerning factors related to surgery, there was no difference in PJK rate between hooks and screws in this study, whereas for Wang et al. [7] and Helgeson et al. [9] proximal screws were significantly associated with PJK. As a matter of fact, proximal implants and disruption of the interspinous ligamentum or exposure of adjacent joint did not seem to have consequences on the occurrence of PJK. Another modifiable parameter debated in the literature is the choice of UIV level, which was not associated with PJK in the current study, since no difference was observed for UIV

Table 5 Postoperative radiographic parameters: comparison between PJK and non-PJK patients

	Cohort ($n=365$)	Non-PJK ($n=308$)	PJK ($n=57$)	p
Sagittal parameters				
Pelvic tilt	12° ± 8°	11° ± 8°	12° ± 8°	0.51
Maximal lumbar lordosis	56° ± 11°	56° ± 11°	57° ± 11°	0.49
Maximal thoracic kyphosis	34° ± 15°	34° ± 16°	33° ± 14	0.47
Cervical lordosis	3° ± 17°	3° ± 17°	3° ± 17°	0.88
C7 sagittal tilt	3° ± 2.4°	3° ± 2.5°	3.1° ± 2.1°	0.83
C7 slope	21° ± 17°	22° ± 10°	14° ± 18°	0.003
Coronal parameters				
Main curve	19° ± 11°	19° ± 11°	20° ± 10°	0.70
Proximal contra-curve	10° ± 8°	10° ± 8°	10° ± 9°	0.97
Distal contra-curve	18° ± 10°	17° ± 10°	18° ± 9°	0.79
Shoulders tilt	3° ± 2°	3° ± 2°	4° ± 2°	0.44
C7 coronal tilt	1.6° ± 1.4°	1.6° ± 1.4°	1.7° ± 1.4°	0.40

Table 6 Pre- and postoperative comparison in PJK and non-PJK groups

	PJK group			Non-PJK group		
	Preoperative	Postoperative (< 3 months)	p	Preoperative	Postoperative (< 3 months)	p
Sagittal parameters						
Pelvic tilt	11° ± 9°	12° ± 8°	0.28	9° ± 8°	11° ± 8°	0.12
Maximal thoracic kyphosis	32° ± 16°	33° ± 14°	0.70	27° ± 18°	34° ± 16°	< 0.0001
Cervical lordosis	7° ± 17°	3° ± 17°	0.12	10° ± 20°	3° ± 17°	0.07
C7 sagittal tilt	2.8° ± 2.4°	3.1° ± 2.1°	0.43	3.2° ± 2.4°	3° ± 2.5°	0.89
C7 slope	20° ± 9°	14° ± 18°	0.003	16° ± 10°	22° ± 10°	0.002
Coronal parameters						
Main curve	58° ± 13°	20° ± 10°	< 0.0001	55° ± 13°	19° ± 11°	< 0.0001
Proximal contra-curve	31° ± 12°	10° ± 9°	< 0.0001	30° ± 10°	10° ± 8°	< 0.0001
Distal contra-curve	29° ± 10°	18° ± 9°	< 0.0001	29° ± 11°	17° ± 10°	< 0.0001
Shoulders tilt	3° ± 2°	4° ± 2°	0.46	2° ± 2°	3° ± 2°	0.57
C7 coronal tilt	2° ± 1.8°	1.7° ± 1.4°	0.77	1.8° ± 1.5°	1.6° ± 1.4°	0.86

level between groups (Figs. 4, 5). An interesting finding in this study was the influence of rods alloy. CoCr rods were at higher risk of PJK with 28% rate versus 12% for other rods alloy. Although this result is controverted, it might be explained by the biomechanical properties of this alloy. CoCr was associated with a better hypokyphosis correction [14]. Indeed, CoCr rods stiffness is more important; thus, it allows for a better correction but it induces more constraints on the junctional level. Consequently, it might be responsible for bone and soft tissue failure, leading to PJK [15]. Concerning deformity correction, Cobb angle correction was not reported as risk factor for PJK, contrary to Wang et al. [9]. Another difference between PJK and controls was the change regarding the location of the inflection point in 68% of the PJK group, which was shifted proximally by 2 vertebral levels on average (Fig. 6). This finding had never been reported before in the literature, and might explain some misunderstood cases of PJK in which LL and TK (anatomical measures) remained unchanged postoperatively, but with posterior global alignment after surgery. Then, changes in the location of inflection point could induce a posterior shift with a compensatory hyperlordosis and a subsequent adaptating proximal kyphosis. Thoracoplasty had been previously reported as an independent risk factor for PJK, but no correlation was found in our results in opposition to Kim et al. [4] and Wang et al. [9].

Compensatory mechanisms

PJK might not be a complication but a compensatory mechanism due to a change in spinal balance. Results showed that the increase in global maximal TK was lower in the PJK group than control. Since PJK was responsible for an increase at the non-instrumented upper junction, it may

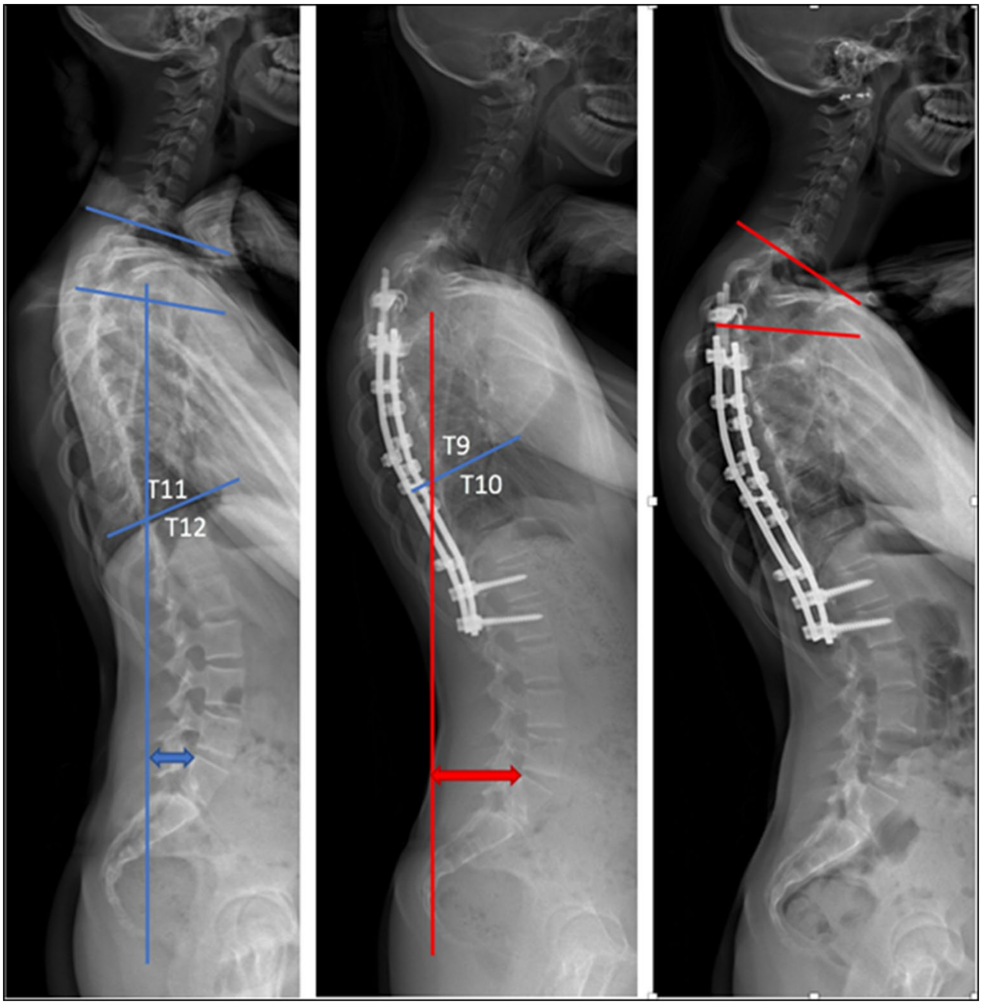


Fig. 6 Preoperative, postoperative and 2 years sagittal X-rays of a patient with PJK (SVA and UIV-UIV + 2 angle are represented)

Table 7 Comparison of sagittal parameters between postoperative and final follow-up in PJK group

	Postoperative (< 3 months)	Final follow-up	*p*
Pelvic tilt	12° ± 8°	11° ± 8°	0.08
Maximal lumbar lordosis	57° ± 11°	65° ± 12°	< 0.0001
Maximal thoracic kyphosis	33° ± 14°	45° ± 16°	< 0.0001
Cervical lordosis	− 3° ± 17°	1° ± 17°	0.04
C7 sagittal tilt	3.1° ± 2.1°	3.1° ± 2.3°	0.96
C7 slope	14° ± 28°	21° ± 9°	0.25
PJK angle	1° ± 7°	17° ± 11°	< 0.0001

be associated with a reduced TK in the instrumented part. Therefore, to maintain global alignment, there was a flattening of postoperative lumbar lordosis to be adapted to the lack of TK (Table 7) [16]. At follow-up, PJK patients increased their proximal kyphosis above UIV to compensate for the posterior shift due to postoperative decrease in instrumented TK and to balance head over the pelvis. This is associated with an increase in C7 slope. Consequently, LL increased at follow-up to keep a balanced spine. Nevertheless, static radiographic measurements of spinal alignment might not be sufficient, as balance is a dynamic process. Indeed, good alignment is necessary for a good balance but insufficient, and role of central nervous system is questionable. As matter of fact, radiological PJK is probably not an issue in AIS, and might be mainly a compensatory mechanism when spine is shifted posteriorly.

Limitations

One of the limitations of this study was its retrospective nature. However, our series is one of the largest dedicated to Lenke 1 and 2 pediatric curves and the high number of cases permitted to smooth some bias. Second, the cervico-thoracic junction is often fastidious to evaluate with current X-rays due to the superimposition with arms but stereoradiographic system provided better definition [17]. Third, no difference was observed between the types of proximal implants; it could be explained by the very small number of subject in the all-screw group. Indeed, most surgeons prefer supralaminar or transverse hooks for the UIV, to avoid adjacent joint opening, potentially destabilizing and reduce neurological risk. Moreover, mechanism of PJK in AIS remains partially understood. Some interesting parameters in PJK cases are difficult to evaluate such as the posterior shift of the fusion mass. Further 3D studies with stereoradiographic images could permit a better analysis of the posterior shift of the UIV and T1, and better understood what the correct postoperative sagittal alignment to avoid PJK is.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest except for the authors B. Ilharreborde: consulting for EOS imaging, Medtronic, Implanet, Zimmer Biomet. K. Mazda: consulting for Implanet, royalties from Zimmer.

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