

LONG-TERM RESULTS OF SURGICAL CORRECTION OF SCHEUERMANN'S KYPHOSIS

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Objective. To analyze long-term results of surgical correction of kyphosis due to Scheuermann's disease.

Material and Methods. Design: retrospective cohort study. The study group included 43 patients (m/f ratio, 34/9). The mean age was 19.1 (14–32) years; the mean postoperative follow-up was 6 ± 10 (5–20) years. Two-stage surgery including discectomy and interbody fusion followed by posterior correction and fusion was conducted in 35 cases (Group A). Eight patients (Group B) underwent only posterior correction and spinal fusion. The following parameters were determined for each patient: thoracic kyphosis (TK), lumbar lordosis (LL) (scoliotic deformity of the thoracic/thoracolumbar spine, if the curve magnitude was > 5°), sagittal vertical axis (SVA), sagittal stable vertebra (SSV), first lordotic vertebra (FLV), proximal junctional angle (PJA) and distal junctional angle (DJA). All measurements were performed immediately before surgery, one week after surgery, and at the end of the follow-up period. All patients answered the SRS-24 questionnaire after surgery and at end of the follow-up period.

Results. Groups were comparable in terms of age and gender of patients, body mass index and initial Cobb angle (p < 0.05). The curve decreased from 77.8° to 40.7° in Group A and from 81.7° to 41.6° in Group B. The loss of correction was 9.1° and 6.0° in groups A and B, respectively. The parameters of lumbar lordosis remained normal during the follow-up period. At implant density less than 1.2, deformity correction and correction loss were 44.5° (54.7 %) and 3.9°, respectively (p < 0.05). Proximal junctional kyphosis (PJK) was detected in 21 out of 43 patients (48.8 %). The frequency of PJK was 45.4 % among patients whose upper end vertebra was included in the fusion and 60 % among those whose upper end vertebra was not included. PJK developed in eight (47.8 %) out of 17 patients with kyphosis correction \geq 50 % and in 13 (50.0 %) of those with correction < 50 %. The rate of DJK development was 39.5 %. The lower instrumented vertebra (LIV) was located proximal to the sagittal stable vertebra in 16 cases, with 12 (75 %) of them being diagnosed with DJK. In 27 patients, LIV was located either at the SSV level or distal to it, the number of DJK cases was 5 (18.5 %); p < 0.05. Only two patients with complications required unplanned interventions. According to the patient questionnaires, the surgical outcome score increases between the immediate and long-term postoperative periods for all domains and from 88.4 to 91.4 in total. The same applies to answer to the question about consent to surgical treatment on the same conditions: positive answers increased from 82 to 86 %.

Conclusions. Two-stage surgery, as a more difficult and prolonged one, has no advantages over one-stage operation in terms of correction magnitude and stability of the achieved effect. Surgical treatment improves the quality of life of patients with Scheuermann's disease, and the improvement continues in the long-term postoperative period.

Key Words: Scheuermann's disease, surgical correction, long-term results.

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Many years have passed since Scheuermann's disease was recognized as an independent pathology with specific clinical and radiological manifestations, as well as course patterns [1–3]. The first results of surgery for severe progressive forms of the disease turned out to be quite successful to inspire orthopedists in many countries to continue this study [4]. The number of publications on this topic has been increasing over the years. However, there are a number of issues, which are, apparently, too far from being

solved. It was originally believed that two-stage intervention, which includes discectomy, interbody fusion, and deformity correction using spinal instrumentation, provides optimal correction with its minimal loss [5–8]. Incorporation of pedicle fixation into the medical practice and its common use have allowed many surgeons to abandon the anterior treatment stage [9–12]. However, no final consensus has been reached yet. Another issue is development of proximal and distal junctional deformities, a certain

percentage of which require unplanned reoperations. The collective experience of many surgeons has made it possible to determine a number of risk factors for the development of these deformities; however, even these recommendations are not always applicable [13–18]. The data of assessing the quality of life of patients after surgery using various questionnaires provides an ambiguous picture [19–22]. In addition, studies on the longterm (over five years) results of surgical treatment are quite rare in contrast to a

large number of works on surgical correction of juvenile kyphosis [23–26].

The main aim of the current study is to analyze results of surgical correction of kyphosis due to Scheuermann's disease.

Study Design: Retrospective cohort study.

Material and Methods

Patients

A total of 213 patients with kyphosis due to Scheuermann's disease have been operated on at the Clinic for Pediatric and Adolescent Vertebrology of Novosibirsk Research Institute of Traumatology and Orthopaedics n.a. Ya.L. Tsivyan starting from 1997. Among them, 152 individuals were operated on before October 2015, which means that the actual period of postoperative follow-up was more than five years. The complete survey dataset was available for 43 patients, who comprised the study group. There were 34 boys and nine girls in the group. The mean age was 19.1 (14-32) years. The mean postoperative follow-up was 6 years 10 months (5–20 years). The examination revealed a comorbidity in nine patients: chronic gastritis, bullous disease, atopic dermatitis, Henoch -Schönlein purpura, bronchial asthma, corpus luteum cyst, and maxillary cyst. Two patients had been operated on for anterior abdominal hernia before admission to the clinic.

A total of 28 patients complained of pain (either persistent or intermittent) in the thoracic and lumbar spine upon admission to the clinic; all participants were unsatisfied with their appearance. Two individuals showed mild symptoms of pyramidal insufficiency during overhead traction.

Techniques

Radiological examination. Radiography of the thoracic and lumbar spine including the pelvis and the femoral heads was performed in an upright position. All measurements were conducted by experienced radiographers not involved in the clinical management of the patients. The following parameters were determined for each patient:

- Thoracic kyphosis (TK), which is the angle formed between the cranial endplate of the most tilted cranial vertebra and the caudal endplate of the most tilted caudal vertebra;
- Lumbar lordosis (LL), which is the angle between the cranial endplates of L1 and S1 vertebrae;
- Scoliotic deformity of thoracic/thoracolumbar spine (in case of 5°);
- Sagittal Vertical Axis (SVA), namely the distance between the plumb line drawn from the center of the C7 vertebral body and the posterior-superior corner of the sacrum;
- Sagittal Stable Vertebra (SSV), which is the vertebral body most evenly divided in half by the vertical line passing through the posterior-superior angle of S1 (SSV-B, where B stands for bisected) [27];
- Proximal Junctional Angle (PJA), namely the angle between the caudal endplate of the upper instrumented vertebra (UIV) and the cranial endplate of vertebra located two levels caudal to the UIV:
- Distal Junctional Angle (DJA), which is the angle between the cranial endplate of the lower instrumented vertebra (LIV) and the caudal endplate of the inferior vertebra.

All measurements were taken immediately before the surgery, one week after it, and at the end of the follow-up period. All patients answered the SRS-24 questionnaire after surgery and at the end of the follow-up.

Surgical technique. All patients were operated on using modern segmental instrumentation. The correction stage was preceded by the ventral procedure, namely discectomy and interbody fusion with autologous bone graft in 35 cases (Group A). The remaining eight individuals were operated on using posterior approach only (Group B). All the operations mentioned in this study were performed by three surgeons, each of whom has more than 25 years of experience in spinal surgery.

The two-stage intervention was initiated with anterior release. The patient was placed on the operating table on his left side (except for cases with left-sided

scoliotic curve). Transthoracic approach was carried out through the base of the rib corresponding to the spinal segment located two levels cranial to the kyphotic apex. Dissection of the parietal pleura, wound extension, and lung collapsing were followed by dissection of the visceral pleura and the prevertebral fascia. A total of 3–5 discs at the kyphotic apex including the endplates of adjacent vertebrae up to the posterior parts of the annulus fibrosus were resected using osteotomes and forceps. Intervertebral defects were filled with bone chips from the rib resected during the approach. The wound was sutured in layers. The patient was then turned to the prone position. The posterior parts of the vertebrae included in the fusion were exposed at the required length through the posterior medial approach. Fixation elements, namely hooks and/or pedicle screws, were implanted in accordance with the preoperative planning. Thus, one of the three types of instrumentation was used: either hook, hybrid or pedicle screw fixation. The type of instrumentation was selected based on the surgical approach used in the clinic during a specific period. For instance, hooks (24 patients) were incorporated into the practice with the advent of CDI in the late 1990s; hybrid constructs (14 patients) were used at the end of the 2000s; all-pedicle-screw constructs (5 patients) were utilized since 2013. Hybrid instrumentation involved placement of pedicle screws in the lumbar and lower thoracic regions, while pedicle constructs were implanted at all instrumented levels, except for the cranial region. The latter consisted of hooks only in all cases. The ligamentum flavum remained intact at all levels, since only pedicle and transverse hooks were used. Ponte osteotomy [28] was usually performed at three levels at the kyphotic apex. The rods were contoured to the physiological curvatures of the thoracic and lumbar spine and attached to the heads of fixation elements at the cranial end of the instrumentation first. Further correction was carried out using lever reduction and apical compression. Heads of the fixation elements were tightened. The cortical bone was accurately removed from the posterior vertebral elements along the fusion area, and spinal fusion with autologous bone was conducted. No external immobilization was used. The patients were allowed out of bed two days after the intervention. The area of spinal fusion was 11.6 (10–13) vertebral motion segments. The duration of one-stage and two-stage interventions was 206 (90–395) and 272 (160–480) min, respectively. The mean blood loss was 788 (150–2,050) and 1,009 (350–3,250) mL in posterior and anterior–posterior approaches, respectively.

Statistical methods

Descriptive characteristics for continuous data are presented as median [first quartile (Q1); third quartile (Q3)], arithmetic mean \pm standard deviation. The distributions of parameter values in the groups were compared using the unpaired Mann - Whitney U test with calculating the distribution bias and the 95 % confidence interval (CI) for the bias. Complications are presented as quantity, % [95 % CI of the percent calculated using the Wilson formula]. Complications were compared using the twotailed Fisher's exact test with calculating the odds ratio (OR) of complications and constructing a 95 % CI for OR. Statistical hypotheses were tested at the critical level of significance of p = 0.05, i.e., the difference was considered statistically significant at p < 0.05.

All statistical calculations were carried out using the RStudio software package (version 1.3.959 − © 2009−2020 RStudio, Boston, USA, URL https://www.rstudio.com) in the R language (versions 4.0.2 (2020-06-22), URL https://www.R-project.org).

The study was approved by the Ethical Committee of the Institute; all patients gave their informed consent to participate in the study.

Results

The distribution of patients between groups depending on the surgical strategy applied is presented in Table 1. The groups were comparable in age, sex, BMI, and initial Cobb angle.

The achieved correction significantly exceeds the preoperative mobility in the general group (Table 2). The resulting correction was 48.6 % of initial kyphosis and maintained at 86.3 % of the achieved level during the entire follow-up. The two subgroups (A and B) demonstrate practically the same pattern. The only difference is that Group B patients initially had greater segmental mobility (41.8 % vs 30.0 %). The initial Cobb angle in Group A was less than that in Group B. The achieved correction was also slightly smaller in Group A; the postoperative loss of correction was greater in both absolute and percentage values in Group A. One can state that the anterior release (Group A) does not provide any advantages in terms of both the correction rate and its maintenance.

As for lumbar lordosis, all parameters remained normal during the follow-up period (Table 3). However, lordosis increased by 10.5° in Group A and by 1.6° in Group B in the postoperative period.

Thoracic scoliosis of 5° was diagnosed in 18 patients. It averaged 13.3° (6-32°) before surgery, 6.0° (1-15°) immediately after the intervention, and 10.3° (5-22°) at the end of the follow-up.

According to our data, implant density (ID), which is defined as the ratio of the number of fixation elements (hooks/ screws) to the number of vertebrae involved in fusion, depends on instrumentation type. The mean ID is 0.89 and 1.68 for hook and hybrid/pedicle-screw fixation, respectively (Table 4). The surgical outcome also depends on the type of instrumentation used. Hook fixation reduces kyphosis from 79.7° to 44.7°, while hybrid/pedicle-screw fixation decreases the angle from 82.2° to 37.3°. Thus, the use of pedicle screw instrumentation allows for a significant increase in the correction rate (44.9°, 54.7 %) compared to hook fixation (35°, 44 %) and its better maintenance (loss of correction, 3.9° versus 7.1°).

Proximal junctional kyphosis was diagnosed in 21 out of 43 patients (48.8 %). Of 33 patients with the upper end vertebra of the curve included in the fusion, PJK was detected in 15 (45.4 %) individuals, including two cases of severe

kyphosis with subluxation of the vertebra superior to the UIV. These two patients underwent unplanned interventions. All the other individuals with PJK were asymptomatic. PJK developed in six (60 %) out of 10 patients in whom the upper end vertebra was excluded from the fusion. There were no statistically significant differences between the subgroups (p = 0.448). PJK was diagnosed in eight (47.8 %) out of 17 participants with kyphosis correction of $\geq 50 \%$ and in 13 (50 %) out of 26 individuals with deformity correction of < 50 %. Since pedicle and transverse hooks were used for formation of the upper end grip in all cases, the ligamentum flavum was not injured. For this reason, this factor was excluded from the list of possible causes of frequent PIK cases.

The following changes were revealed at the distal end of instrumentation. The angle formed between the cranial endplate of the LIV and the caudal endplate of the distal vertebra averaged 16.9° before surgery, 9.8° immediately after surgery, and 10.1° at the end of the follow-up in the general group (Fig.). The final angle was < 10° in 17 individuals. In addition, two cases of kyphosis (2° and 7°) were noted. The DJK rate was 39.5 % in the general group. LIV was located proximal to the SSV in 16 cases. Of them, 12 patients (75 %) developed DJK. LIV was found either at the SSV level or distal to it in 27 patients; there were five (18.5 %) DJK cases. Thus, a significant difference between these two subgroups (p < 0.001) was noted. There were no indications for unplanned intervention among DJK patients.

Assessing the sagittal balance is hampered by the diversity of pre- and post-operative parameter values. We can only state that negative balance was noted in 54 % of patients before surgery, 36 % of individuals immediately after surgery, and in 81 % of cases at the end of the follow-up. The sagittal balance varied from -89 to +108 mm.

The results of the SRS-24 questionnaire survey demonstrated that the surgical outcome score increases for all domains between the early and late postoperative periods; however, the differences are insignificant (Table 5). Statistically significant changes were noted for the following domains: pain, function after surgery, general function, and function activity. The total score increased proportionally as well. The same applies to answer to the question No. 24 (consent to undergo the same surgical treatment if necessary): the score grew from 82 to 86 %.

Fourteen (32.5 %) out of 43 patients had 16 complications in total. The most frequent complications (11 cases) were the ones associated with implants: rod/ screw fractures and hook displacement (Fig. c). Six individuals required reoperation. Proximal Junctonal Failure (PJF) accompanied by severe kyphosis due to subluxation of the vertebra superior to the UIV was noted in two cases. Both patients required extension of the instrumentation to the cranium. The operation did not provide the desired result immediately. These two patients underwent seven unplanned surgeries in total. Other complications included pyelonephritis and spontaneous pneumothorax, which were successfully treated by conservative treatment. Another patient had impaired sensitivity on the outer thigh surface within four weeks after surgical intervention. There were neither neurological nor purulent complications.

Discussion

We have studied long-term results of surgeries for juvenile kyphosis and consider it necessary to begin the discussion with an overview of the articles that also present treatment outcomes with long-term (over five years) postoperative follow-up. As far as we can judge, there are very few such studies, while both the treatment strategy and the outcomes seem to be ambiguous.

Apparently, the first study of this kind was published by Soo et al. [23] in 2002. The authors evaluated the outcomes of three strategies used for treating 63 patients: observation and exercise, bracing, and surgical correction using Harrington compression rods following anterior release. The results were followed up for 14 (10–28) years. The main exam-

Table 1
Participant characteristics (age, male/female, BMI, Cobb angle) in groups A and B

Parameters	Group A	Group B	P-value
Mean patient age, years	19.2 (14-28)	18.7 (14-33)	0.572
Male to female ratio	6:2	26:9	>0.999
ВМІ	19.6 (16.8-22.6)	20.8 (16.9-31.6)	0.587
Initial Cobb angle of kyphosis, degree	81.7 (64–105)	77.8 (62–87)	0.288

Table 2

Changes in the Cobb angle of kyphosis in the general group depending on the surgical approach

Parameters	General group	Group A	Group B	P-value
Number of patients, n	43	35	8	_
Cobb angle before	80.6 (62-105)	77.8 (62-87)	81.7 (64-105)	0.288
surgery, degree				
Cobb angle on supine	55.5 (27-83)	45.0 (27-48)	57.2 (34-83)	
lateral bending, degree				0.022
Mobility, %	37.6	41.7	30.0	
Cobb angle after surgery,	41.4 (21-72)	40.7 (27-53)	41.6 (21-72)	0.790
degree				
Cobb angle at the end of	47.9 (17-88)	49.8 (28-72)	47.6 (17-88)	0.628
the follow-up, degree				
Correction of the initial	39.2 (48.6)	37.1 (47.7)	40.3 (49.3)	0.492
Cobb angle, degree (%)				
Loss of correction, degree	6.5 (13.7)	9.1 (24.5)	6.0 (12.1)	0.696
(% of the achieved angle)				

Table 7

Changes in the Cobb angle of lumbar lordosis in the general group depending on the surgical approach

General group	Group A	Group B	P-value
43	35	8	_
80.3 (48-112)	78.5 (60–103)	80.7 (48-112)	0.444
52.8 (33–132)	51.6 (34–73)	52.2 (33–132)	0.950
57.1 (36–89)	62.1 (52–79)	53.8 (42-89)	0.229
	43 80.3 (48–112) 52.8 (33–132)	43 35 80.3 (48–112) 78.5 (60–103) 52.8 (33–132) 51.6 (34–73)	43 35 8 80.3 (48–112) 78.5 (60–103) 80.7 (48–112) 52.8 (33–132) 51.6 (34–73) 52.2 (33–132)

 $\label{eq:Table 4} Table \, 4$ Changes in the Cobb angle of kyphosis at different Implant Density values

Parameters	Hooks	Hybrid/pedicle-screw instrumentation	P-value
Number of patients, n	24	19	-
Implant Density	0.89 (0.36-1.20)	1.38 (0.80-1.75)	0.001
Cobb angle before	79.7 (66-100)	82.2 (64-105)	0.440
surgery, degree, degree			
Cobb angle immediately	44.7 (28-72)	37.3 (21-54)	0.048
after surgery, degree			
Cobb angle at the end of the	51.8 (30-88)	41.2 (22-72)	0.009
follow-up, degree			
Correction, degree (%)	35.0 (44.0)	44.9 (54.7)	0.026
Loss of correction, degree	7.1	3.9	0.020

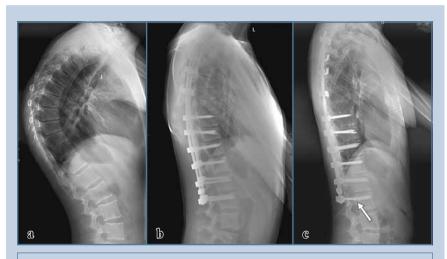


Fig. A 14-year-old patient operated on using the hybrid approach: **a** – before surgery: thoracic kyphosis, 90°; lumbar lordosis, 84°; DJA, 26°; **b** – 10 days after surgery: thoracic kyphosis, 21°; lumbar lordosis, 60°; DJA, 16°; **c** – six years after surgery: thoracic kyphosis, 22°; lumbar lordosis, 69°; DJA, 6°. One of the distal screws was broken without fragment displacement (arrow)

ination methods were radiography and surveys using a questionnaire designed by the authors. The kyphotic curves in patients of the three subgroups were 57°, 64°, and 73° before surgery, and 57°, 51°, and 59°, respectively, at the end of the follow-up. Minor differences between the observation and brace subgroups were expected. About half of the achieved correction (20° out of 37°) was lost in the surgery group. The achieved

kyphotic curvatures were almost identical in the three subgroups by the end of the follow-up period.

The questionnaire results demonstrated no differences in the following parameters between the patents: marital status, general health, educational level, work status, pain degree, and functional capacity. There was no relationship with the treatment mode, as well as type and degree of the kyphotic curve. Patients of

the brace and surgery subgroups noted the greatest improvement in their self-image among other participants, attributing it to the treatment. The lowest functional level was observed in patients with deformities of $>70^{\circ}$ by the end of the follow-up.

The authors conclude that careful selection of the method for treating patients with Scheuermann's disease should be based on age, deformity type, and pain severity. By following this principle, it is possible to achieve similar functional results for fundamentally different therapeutic approaches in the long-term period.

In 2009, Denis et al. [24]. investigated the frequency and risk factors for the development of junctional kyphosis after surgical correction of kyphotic deformities due to Scheuermann's disease. The authors analyzed the results of the treatment of 67 patients who were operated on at least five years ago (mean follow-up, 73 months). A total of 15 individuals underwent one-stage posterior intervention; 52 patients were treated by two-stage surgery. Traditional definitions were used to identify junctional kyphosis. PJK was considered if the proximal junctional angle between the cranial endplates of the UIV and the vertebra two levels cranial to it was >10° or increased by at least 10° after surgery compared to the baseline. Distal junctional kyphosis was determined by the angle between the caudal endplates of the LIV and the vertebra located one level caudal to it. The authors managed to reduce kyphosis from 78° to 45° and almost completely preserve it: the curve angle was 49° at final follow-up. PJK developed in 20 (30 %) patients. The frequency of PJK was 8 % if the proximal end vertebra in the curvature was included in the fusion and 63 % if it was not. The authors mention damage to the ligamentum flavum by a hook or a sublaminar wire as the second most important cause of PJK. PJK development is associated with neither the baseline kyphosis magnitude nor the achieved correction rate. DJK was detected in eight patients (12 %), with seven of them sharing the same feature: the first lordotic disc was

not included in the fusion. Various types of vertebral instrumentation were used in the analyzed group; however, no correlation with the frequency of junctional kyphosis was noted.

In 2016, Graat et al. [25] published the long-term outcomes of surgeries for Scheuermann's kyphosis in 29 patients. The postoperative follow-up was 18 (14– 21) years. Posterior approach was used in 13 cases; combined (anterior–posterior) procedure was carried out in 16 individuals. The initial Cobb angle (82°) was reduced to 69° after surgery by the end of the follow-up. Unfortunately, the authors present radiography data for the general group only and do not differentiate them depending on the surgical approach used. They only mention that the combined and posterior approaches reduced the curvature by 27 and 17 %, respectively, while not considering the difference to be statistically significant. The number of PJK cases increased with the duration of postoperative follow-up: there were nine (31 %) patients during the first year after surgery, 12 (43 %) individuals in the period of eight years after surgery, and 15 (53 %) cases by the end of the follow-up period. The upper end vertebra was fused in eight patients (four PJK cases) and not included in the instrumentation in 19 individuals (11 PIK cases). No revision surgeries for PJK were performed. There were no reported cases of DJK. Implantassociated complications were observed in 20 (69 %) patients and distributed

approximately equally between the two subgroups. Implants were removed in seven individuals; a solid bone fusion was visually confirmed in all of the cases. The correction loss was 5° after implant removal. HRQoL was assessed using the Oswestry Disability Index, Visual Analog Score Pain (SF-36), and EQ-5d. A total of 21 (72 %) out of 29 patients were satisfied with the treatment outcome and would be willing to undergo the same treatment again if they had a similar condition, while the remaining 23 (79 %) participants recommended the procedure to others. The authors consider the radiographic results of surgical treatment as "disappointing". However, they also reasonably correlate them with the data of clinical studies indicating high functional activity of patients for many years after surgery even with a high incidence of postoperative pain. In addition, the patients who underwent the combined surgical treatment demonstrated better functional results than those subjected to posterior fusion only. Despite the ambiguity of the obtained results, the authors note that the outcomes are better than in case of natural disease course, as far as it can be judged from the literature.

In 2019, Hwang et al. [26] published the results of all-pedicle-screw fixation in individuals with kyphosis of various etiologies, including Scheuermann's disease. Juvenile kyphosis was diagnosed in 15 out of 43 patients. The mean post-operative follow-up was 5.8 (5–9.7) years

for these 15 individuals. The average age of the patients with Scheuermann's disease was 19.1 years. Vertebral column resection was conducted in 11 cases in order to increase the mobility of the spinal deformity. The authors chose the length of the instrumented fusion based on the following principles: the number of vertebra involved in fusion should be symmetrical both above and below the kyphosis apex, provided that the disc located cranial to the UIV is lordotic. Kyphosis was 91° at baseline, 48.1° immediately after surgery, and 49.9° at the end of the follow-up, i.e. the average correction loss was only 1.8°. Complications included two cases of PIK, one screw pullout, and one case of signal loss during spinal neuromonitoring, which were followed by complete recovery. Evaluation of the quality of life (ODI and SRS-30) showed significant improvement in all domains.

The paucity of articles on long-term outcomes of surgical correction of kyphosis due to Scheuermann's disease and ambiguity of the results presented in these studies force us to widen the range of considered literature sources (with follow-up periods of two to five years) in order to obtain a more comprehensive picture of the study topic. The key issues are advantages and disadvantages of posterior and anterior-posterior approaches, risk factors, rate of junctional kyphosis (both proximal and distal ones), as well as the quality of life in the postoperative period.

The first fusion surgeries performed by Berg (1948), Stagnara & Perdriolle (1958), and Roaf (1960) are mentioned in the classic book by Sorensen (1964) [3]. The interventions included bone grafting in situ, sometimes with the use of a corrective plaster cast. The first work, which was based on a quite large clinical data (22 patients), was published in 1975 [4]. The study subject was posterior approach (Harrington instrumentation and posterior spinal fusion). The resulting loss of correction exceeded 5° in 16 cases. Moreover, the overall complication rate was so high that the authors recommended surgery only in case of either intense pain or spinal cord compres-

Table 5
The results of HRQoL assessing using the SRS-24 questionnaire

Domain	Early	Late	P-value
Pain	3.74 ± 0.56	3.81 ± 0.52	0.009
General self-image	3.96 ± 0.58	4.12 ± 0.55	0.072
Self-image after surgery	4.39 ± 0.48	4.45 ± 0.43	0.062
Function after surgery	2.03 ± 1.18	2.32 ± 1.43	0.030
General function	3.10 ± 0.75	3.26 ± 0.84	0.042
Function activity	3.50 ± 0.74	3.68 ± 0.71	0.036
Satisfaction with surgery	4.43 ± 0.51	4.49 ± 0.56	0.057
Consent to undergo the same	82 %	86 %	_
surgical treatment if necessary			
Grand total	88.4	91.4	0.058

sion. Taylor et al. [29] obtained similar results. The interest in anterior approach grew [31, 32] thanks to the studies by Hodgson [30]. In 1980, Bradford et al. [5] presented the results of a combined approach to the treatment of juvenile kyphosis. According to the authors, the results were quite acceptable and stable, although the rate of complications associated with thoracotomy was rather high. In 1986, Speck and Chopin [32] proposed a differentiated approach to the choice of the surgical strategy: posterior correction and spinal fusion are sufficient in patients with continued vertebral growth, while the anterior-posterior approach is recommended for adult patients. Comparison of the results of one- and two-stage interventions using Harrington rods showed that two-stage operations allow for a significant reduction in postoperative loss of correction at a similar correction rate achieved after surgery [4, 5, 7, 29, 34–36]. In 1977, Enslin [37] first described the combined anterior and posterior approach, which is currently known as same-day surgery. It also contributed to the widespread use of anterior stage of surgical intervention. The orthopedic community discovered a new implant system, namely CDI, in 1983 [38]. Lowe and Kasten presented the first report on the treatment of juvenile kyphosis using CDI in 1993 [39]. A total of 28 out of 32 operated patients were treated in two stages. A number of articles on the use of segmental instrumentation either with or without anterior stage were published in 2001-2004 [14, 19, 40-42]. The obtained results seemed to be quite satisfactory for both surgeons and patients, while the need to make a decision regarding the choice between one- or two-stage operations was gradually emerging. Johnson et al. [43] was among the first researchers who raised the question and concluded that there are no advantages of one approach over another. This issue was further discussed in numerous publications [9, 11, 15, 21, 22, 44–51]. The main conclusion drawn by almost all of the authors is that the results of posterior and combined approaches are identical. However, the use of isolated posterior fusion results

in less blood loss as well as decreased surgery duration and hospital stay. Moreover, it is less cost-effective and rarely leads to complications. According to some authors [50], the only drawback of the posterior approach is that the fusion area is more extended. This is mainly due to the fact that the widely used pedicle screw (all-pedicle screw/hybrid) fixation provides more reliable placement than hook constructs. Our data confirm that the combined approach has no advantages over the use of one type of instrumentation. Moreover, posterior intervention made it possible to obtain a slightly larger correction and its better preservation.

Proximal junctional kyphosis was noted in 12 works in the period of 1980-2017 [5, 9, 13–15, 39–42, 47, 52, 53]. A total of 268 patients were operated on. PJK was noted in 34 cases, which comprises 12.7 (3-30) %. Only one patient required repeated intervention, while the remaining individuals had asymptomatic kyphosis. The main causes of complications were the following: exclusion of the upper end vertebra from the instrumentation [5], overcorrection of kyphosis [39], damage to the ligamentum flavum at the upper instrumentation level [24], association with the spinal and pelvic parameters [54], and abrupt transition from rigidly fixed to unfused vertebrae [55]. Yanic et al. [56] proposed a specific technique for implanting proximal screws in order to prevent the development of PJK: the screws should not be completely tightened, with two threads left outside the cortical plate. This technique allowed the authors to avoid the development of PJK in all of 60 patients operated on.

Distal junctional kyphosis is mentioned in nine publications [8, 9, 16–18, 39, 54, 56, 57]. In total, the authors operated on 330 patients. DJK developed in 57 (17.3 %) cases. Gong et al. [27] performed a meta-analysis on the topic under discussion (4 articles, 173 patients) and obtained a result similar to the one mentioned above (36 cases, 20.8 %). The authors emphasized that additional intervention was required in 10 (27.8 %) patients to eliminate the complications. Ghasemi et al. [54] reported overcorrec-

tion of kyphosis, extremely negative sagittal balance and (possibly) young age as potential risk factors, while Zho et al. [18] mentioned preoperative kyphosis angle. However, after the study of Cho et al. [16], the main discussion ended in choosing between SSV and the first lordotic vertebra as the LIV. No consensus has been achieved vet. Kim et al. [57], Dikici et al. [17], and Gong et al. [27] state the effectiveness of choosing SSV as the LIV for preventing PJK development. Zhu et al. [18] suggest using SSV and FLV as the LIV in thoracic and thoracolumbar kyphosis, respectively. Yanic et al. [56] do not consider the choice of SSV a necessary stage; instead, they propose limiting the fusion by FLV at the distal end. Gong et al. [27] emphasize the ambiguity of the SSV choice due to the fact that the perpendicular line drew from the posteriorsuperior corner of the sacrum can either divide the first vertebra approximately in half (SSV-B, where B stands for bisected) or pass along its margin (SSV-T, where T means touched). However, the authors do not indicate whether the choice of the SSV subtype affects the risk of DJK.

The rate of junctional kyphosis was significant in the group we studied: 48.8 % for PJK and 39.5 % for DJK. Moreover, the overwhelming majority of the cases were asymptomatic and did not require reoperation. Spinal fusion was successfully extended to the cranium in only two PJK cases with kyphosis reaching 90°, which can be interpreted as proximal junctional failure (PJF). Interestingly, the patients were brothers. The reports on Scheuermann's disease in twins are not rare [58, 59]. However, we have not found any similar observations in the literature. PJK usually develops when UIV is excluded from the fusion, but the differences are insignificant. According to our data, the rate of the major curve correction is not a risk factor for PJK. As for the distal end of the fusion, adverse changes in the disc caudal to the LIV occur immediately after surgery and further deteriorate. As far as we can judge, choosing SSV as the LIV seems reasonable.

We did not find any literature data on the effect of ID on the outcome of juvenile kyphosis correction. According to our data, an increase in ID due to a gradual transition from hook to pedicle crew fixation is accompanied by growth in the achieved deformity correction and yields more stable results.

The assessment of HRQoL after surgical correction of juvenile kyphosis was carried out using numerous measures. In the study by Hosman et al. [14], the Oswestry index reduced postoperatively from 21 to 6.6 in the general group of 33 patients. The Neck Pain Disability index changed insignificantly (from 4.9 to 4.4). The Oswestry index dropped from 40.7 to 11.9 in 14 patients who agreed to surgery because of pain. In other 19 patients, who mainly complained for their physical appearance, the Oswestry index decreased from 6.9 to 2.6. All patients, except for two, expressed satisfaction with their appearance at the end of the follow-up (mean period, 4.5 years). All of them either had a paid job or were homemakers.

Poolman et al. [19] studied the results of surgical treatment of 22 patients using the SRS-24 questionnaire. The findings turned out to be highly controversial. The mean score was 83 points (max, 120). Only 10 patients noted decreased pain, while four individuals experienced deterioration. Furthermore, only 10 participants stated improved self-image, while three patients said that their appearance worsened after the operation. A total of 16 individuals out of the entire group stated that they would undergo the same treatment. Among them, only nine participants were sure of that.

In 2004, Yang et al. [20] presented the results of surgical correction in 16 patients. All participants noted reduced pain, while the ODI score decreased from 37.3 to 6.4 points.

Lee et al. [9] operated on 39 patients and used the SRS-30 questionnaire to assess the quality of life. A total of 29 patients were examined. The mean score was 128 (115–146) points after two-stage operation (anterior release and deformity correction) and 120 (90–142) points after one-stage surgical correction. The same questionnaire was used by Koptan et al. [15] in a study of surgi-

cal outcomes of 33 patients in 2009. The mean score was 134 (98–146) and 120 (90–132) points after one-stage and two-stage surgeries, respectively.

Temponi et al. [21] used the VAS questionnaire to assess pain in 28 patients. Of the 19 patients operated on in two stages, 17 individuals complained of pain (mean score, 6.6 points) before surgery, and only three were bothered by pain (mean score, 0.6 points) by the end of the follow-up. Eight out of nine patients of the one-stage correction group had preoperative pain, and only one participant complained of pain after surgery (5.6 and 0.5 points, respectively).

Koller et al. [22] presented the results of one-stage surgical treatment in 111 patients. The authors assessed the quality of life using several measures: ArM (Approach-Related Morbidity questionnaire), SRS-24, ODI, and SF-36. The following results were obtained: SRS-24 score, 91.4 points; ODI score, 7.7 %; ArM score, 19.2 %, SF-36 PSC (physical component score), 48.6 points; and SF-36 MSC (mental component score), 48.8 points. A clear correlation was found between the following scores: SRS-24 and ODI; SRS-24 and ArM; ODI and ArM. There was a negative correlation between the increase in the fusion length and SRS-24 self-image score. The achieved correction and other radiological parameters did not affect questionnaire results. A total of 87 % of the surveyed patients answered that they would have the same treatment if they had the same conditions.

Etemadifar et al. [46] published the results of surgical treatment of 30 patients using one- and two-stage approaches. ODI and SRS-30 questionnaires were used in the study. Paired t-test showed significant improvement in both groups.

Cobden et al. [47] operated on 20 patients using only the posterior approach. The authors applied the SRS-22 questionnaire and obtained the mean score of 3.9. The best and the worst scores were noted for domains self-image and mental health, respectively.

In 2018, Toombs et al. [60] published a study on the quality of life of patients

operated on for Scheuermann's disease and adolescent idiopathic scoliosis. According to the data of SRS-22 questionnaire, such parameters as preoperative curve angle, correction rate, kyphosis apex level, and BMI do not correlate with baseline scores and changes in healthrelated quality of life (HRQoL) in patients with juvenile kyphosis. Improvement was noted for all domains, with the most pronounced increase in self-image (from 2.8 to 4.4 points). The greatest improvement was observed for self-image, mental health, and total score domains two vears after operation. These data correlate with the VAS questionnaire results: an improvement from 3.69 to 1.51 points.

We used the SRS-24 questionnaire and noted improvement for all the seven domains, although it was not statistically significant in all cases. The consent to have the same surgery if required increased from 82 to 86 %.

The issue of surgical treatment of severe juvenile kyphosis is complicated and diverse. There are no uniform generally accepted indications for surgery. For instance, 2,796 patients were operated on in the United States in the period of 2000–2008, which is 310 cases on average per year [61]. A completely different approach also exists. Jean Dubousset, one of the most famous spinal surgeons, has operated on only nine patients throughout his long professional career and was very strict with selecting the indications for operation [62]. Most surgeons agree that the combined surgery, which is more complicated and traumatic, has no advantages over the posterior approach. Thus, this issue aspect can be considered solved to date. At the same time, the question of choosing the length of the fusion and, hence, the risk of junctional kyphosis is very far from being solved. The accumulation of collective experience is required; and we hope that our colleagues can find our data useful and interesting.

One of the main drawbacks of our work is the relatively small number of patients included in the study. Only 43 out of 152 participants with more than five-year follow-up considered it nec-

essary and possible to undergo examination after the end of the follow-up period. Firstly, it can be explained by the distant clinic location relative to the patient and, hence, the high trip cost. Secondly, this can be also due to the alleged fact that the patients did not consider it necessary to undergo another examination in the absence of complaints. We also did not present any data on changes in the spinal and pelvic parameters. This is because the clinic lacked the opportunity to perform radiography with inclusion of the femoral heads for a significant period. Hence, we are unable to present the pelvic incidence and pelvic tilt parameter values. We considered it wrong to

present the results of changes in the sacral slope only in the absence of other parameters.

Conclusion

The results of long-term (over five years) follow-up of patients operated on for severe kyphosis due to Scheuermann's disease show that the issue is complicated and far from being solved. It is apparent that one-stage surgery including posterior correction and spinal fusion is superior to two-stage procedure (anterior release and posterior correction) because it is less traumatic and does not show worse results. The issue of determining the

length of spinal fusion is still unsettled, as indicated by a large percentage of cases with development of junctional kyphosis, both proximal and distal types. At the same time, there is a reason to believe that surgical treatment improves the quality of patients' life, and this improvement is maintained for a long period.

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