RESULTS OF SIMULTANEOUS AND STAGED SURGICAL TECHNIQUES FOR THE CORRECTION OF CONGENITAL SPINE DEFORMITIES ASSOCIATED WITH INTRACANAL ANOMALIES IN CHILDREN

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Objective. To compare the results of simultaneous and staged corrective surgical interventions for congenital spine deformities associated with intracanal anomalies.

Material and Methods. Design: retrospective mono-center observational case-control study. The study included the results of 127 surgeries performed in 49 patients aged from 4 months to 17 years at the time of surgery (mean age 9.2 years). The average follow-up period was 49 months. The study group included 33 patients who underwent simultaneous (in one surgical session) removal of the intracanal component and correction of spine deformity. In the control group (16 children), these interventions were performed in stages. The analysis criteria were: the comparability of groups: the structure of anomalies of the spine, spinal canal and spinal cord, concomitant maldevelopment of organs and systems, and the presence of skin markers; and the comparison of groups: the total volume of blood loss, the duration of surgery, the magnitude of the spine deformity and the neurological status of patients assessed before and after corrective surgery, and complications.

Results. Spine pathology was predominantly represented by variants of segmentation failure in 41 (59.0 %) patients and multiple vertebral malformations with a leading component of segmentation failure in 22 (33.0 %) patients. Among pathologies of the spinal canal, Arnold-Chiari malformation (17 patients, 25.0 %) and type 1 diastematomyelia (22 patients, 32.0 %) dominated. In the neurological status, Frankel types D and E prevailed at baseline, accounting for 27.3 % and 42.4 % in the study group, and 25.0 % and 37.5 % in the control group, respectively. Skin markers were detected in 34 (69.0 %) patients. The scoliotic component of the deformity prevailed; the kyphotic component was detected in 17 (35.0 %) cases. The mean magnitude of scoliotic deformity before surgery was 28° (min 20°; max 105°), after surgery 10° (min 0°; max 70°). Correction in the study group was 68 %, in the control one – 57 %. The average duration of surgery in the study group was 227 min, in the control group – 198 min for the first operation and 204 min for subsequent ones. The average volume of blood loss in the study group was 286.6 ml, in the control group – 247.5 ml during the first operation and 266.6 ml during the subsequent ones. There were no significant changes in neurological status after surgery in both groups. Transient neurological complications were observed in 2 (4.5 %) patients of the study group and in 1 (4.0 %) in the control group. It was statistically proven that simultaneous intervention provides significantly lower blood loss with a comparable orthopedic result of surgery, which indicates the preferred use of a combined surgical technique.

Conclusion. Performing neurosurgical and orthopedic (correction) stages in children with congenital spine deformities and spinal canal malformations in one surgical session is not statistically associated with a greater surgical aggression and complication rate, but is accompanied by a significantly lower total blood loss. Simultaneous intervention provides somewhat better initial correction of the deformity, eliminates the need for staged interventions, and also reduces the likelihood of complications potentially associated with each surgery.

Key Words: spine malformations, intracanal abnormalities, congenital scoliosis, deformities, children, dysraphia, diastematomyelia.

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The incidence of congenital scoliosis in the population is 1–3 per 1,000 newborns [1, 2], while the combination of intraspinal anomalies and congenital spinal deformities is found at a rate of 15 to 38 % [3]. Multiple vertebral malformations with segmentation failure as the leading component predominate among combined anomalies; malformations are usually associated with diastematomyelia, lipomas, lipomyelocele, teratoma, and syringomyelia in the presence of Arnold–Chiari malformation [1, 4–6]. These combinations are characterized by rapid deformity progression, neurological deficit, cardiopulmonary dysfunction [7, 8], and a high risk of neurological complications during surgery [9, 10]. Perioperative risk is aggravated by high incidence of concomitant malformations

of other organs and systems such as nervous, genitourinary, cardiovascular, etc. [2], which is often segmental [4, 11].

There are different opinions on the strategy for treating the pathology. Some authors prefer the conventional approach, which includes primary neurosurgical intervention followed by staged orthopedic correction of scoliosis using dynamic instrumentation after 3–6 months [1–5]. Their opponents favor one-stage surgery [6–9]. A number of authors argue the need to remove the intracanal component before surgical correction of the spinal deformity [13–16]. Most of the studies are retrospective analysis of case series or small cohorts (evidence level 4 according to UK Oxford CEBM, version 2009) lacking comparison of the results using common assessment criteria. This motivated the authors of the current article to conduct an observational case-control study in order to respond to the key question: which technique for correcting congenital spinal deformities associated with intracanal anomalies correlates with higher surgical invasiveness and complication rate in children?

The aim of the study is to compare the results of simultaneous and staged corrective surgeries for congenital spinal deformities associated with intracanal anomalies in children.

Material and Methods

The study design is a monocentric observational retrospective case-control study.

The study has evidence level 3a (UK Oxford CEBM, version 2009).

Patient recruitment period is 2007–2018. The strategy for forming inclusion groups is presented in Fig. 1.

Patients were divided into groups based on the following inclusion criteria:

– the principle of cohort: unity of place (Clinic of Spinal Pathology and Rare Diseases of National Ilizarov Medical Research Center for Traumatology and Orthopedics, Kurgan), of surgical team (unified planning of the surgical protocol, all patients were operated on by the first two authors), and of time (recruitment period, 2007–2018);

– nosological principle: congenital spinal deformities combined with intracanal anomaly requiring correction of both orthopedic and neurosurgical components;

– the follow-up is ⩾12 months after completion of both treatment stages.

The indications for surgery were the following: spinal deformity of ⩾20°, its rapid progression (>5° per year), occurrence or increase of neurological deficit by at least one Frankel grade (pediatric version) [17].

The exclusion criteria were the following:

– different types of myelocele;

– destructive pathology of the spine and the spinal canal;

– absence of registered data on the long-term outcome.

The results of 127 surgeries performed in 49 patients (35 females and 14 males) were included in the study based on the above-mentioned criteria. The mean age at the time of surgery was 9.25 years (min age, 1 year 4 moths; max age, 17 years). The mean follow-up period was 49.06 ± 8.60 months (range, 6–144 months). Patients were divided into groups according to N.P. Gundobin [18]: early childhood (⩽ 3 years 11 months and 30 days), preschool (⩾ 4 years ⩽ 6 years 11 months and 30 days), junior school (⩾ 7 years ⩽ 11 years 11 months and 30 days), and senior school (⩾ 12 years ⩽ 17 years 11 months and 30 days) age groups.

Two groups were formed during a retrospective analysis:

– the study group, which included 33 patients who underwent simultaneous one-stage resection of the intracanal component and spinal deformity correction;

– the control group consisting of 16 patients who were first subjected to primary neurosurgery and then spinal deformity correction. This group also included individuals who have had dual growing rods implanted at the second stage followed by staged distraction.

Age distribution of patients is presented in Table 1.

In order to assess identity of the groups, the following criteria were used:

a) uniform parameters of group comparability:

– nature of congenital vertebral malformations;

– types of intracanal anomalies: spinal dysraphism in the form of diastematomyelia, lipoma, terminal tethering either alone or in the presence of Arnold – Chiari type II malformation and/or syringomyelia;

– age (range, 1–18 years) at the moment of surgery with taking into account age subgroups;

– >20° Cobb angle of the apical curve in the frontal and/or sagittal planes before surgery; surgical deformity correction magnitude according to Cobb;

– long-term outcomes are assessed ⩾ 12 months after corrective surgery;

b) uniform comparison criteria characterizing surgery invasiveness, as well as the presence and nature of complications:

– blood loss volume measured with a gravimeter (mL);

– surgery duration (min);

– neurological status before and after surgery according to Frankel (pediatric version) [17];

– complications.

The result of the first corrective surgery performed either simultaneously or after neurosurgery was assessed as part of analysis for comparing orthopedic stage effectiveness. The issues of deformity changes in staged distraction unrelated to the study purpose were not considered.

Long-term results were followed up to five years (mean period, 29.06 ± 8.60 months) with assessing changes in neurological status, loss of correction (%), and implant stability.

The data were statistically analyzed using Statistical Package for the Social Sciences (SPSS) software, version 22.0. Since the statistical distribution of the analyzed parameters (blood loss volume, correction rate, percentage of correction) differed from baseline values (a histogram with normal distribution curve, Kolmogorov–Smirnov test, values of asymmetry and kurtosis), the significance

of the differences between the study and the control groups (at two-tailed p-value < 0.05) was assessed using the nonparametric Mann – Whitney test. Differences in the frequencies of qualitative characteristics were assessed using the χ^2 test.

Surgical technique. Simultaneous surgery (study group) included skeletonization, marking the levels of anchoring element implantation, and placement of transpedicular screws. The neurosurgical stage, which included removal of the pathological intracanal formation and, if necessary, revision and duraplasty, was performed after the positions of the reference points were confirmed radiologically. Next, the rods were placed, and correction procedures (translation, apical vertebral derotation, and compression), with the exception of distraction, were conducted cranial to the region of intracanal malformation (Fig. 2). All stages of surgery were performed under neurophysiological monitoring of motor/somatosensory evoked potentials and spontaneous EMG activity.

All stages of surgery were carried out under neurophysiological monitoring. Electrophysiological parameter values obtained after the patient was anesthetized immediately before the start of surgery were used as baseline, with taking into account the specific effect of anesthesia components on the studied parameters [20].

Staged treatment (control group) was performed according to the standard protocol: neurosurgery was conducted at the first stage, which included intracanal pathology elimination and placement of spinal instrumentation (the so-called provisional, or phantom points). Positioning of provisional reference points allows one to reduce surgery duration and risk of damage to the spinal structures and membranes during approach at the second stage, since it eliminates the need for precise skeletonization of the spine. Transpedicular fixation and spinal deformity correction were carried out at the second stage after 3–6 months (Fig. 3). Both surgical stages were conducted under neurophysiological monitoring. Staged distraction was performed every 6–8 months according to Akbarnia et al. [19] in cases when dynamic instrumentation such as VEPTR and dual growing rods were used. The results of staged correction were not analyzed in the study.

Results

Spinal pathologies (Table 2) in the groups include different types of segmentation failure, multiple vertebral malformations with either segmentation failure, hemivertebrae or their combination as the leading component. The groups are comparable in the distribution of malformation types: segmentation failure either alone (51.0 %) or as the leading component in multiple vertebral malformations (36.8 %) generally prevailed.

Types of intracanal anomalies are presented in Table 3. Severe tethering of the spinal cord such as either type I diastematomyelia (SCM I) according to Pang [20] or Arnold – Chiari type II malformation prevailed in both groups. The latter one was excluded from the list of considered neurosurgical pathologies, since no patient had indications for decompression surgery at the craniocervical junction during the mentioned observation period. Cases of caudal fixation accompanied by Arnold – Chiari malformation were accounted for as filum terminale and secondary tethering of the spinal cord.

Intracanal abnormality was localized in the cervical spine in five (10.2 %) cases, in the thoracic region in nine (18.4 %) patients, and in the lumbar spine in 35 (71.4 %) individuals.

Concomitant malformations of organs and systems are most often represented by either a single or horseshoe-shaped kidney, paralytic foot deformity, hypotrophy of (typically) one lower limb, and combined contractures of the joints of the lower limbs. Skin markers were noted in 34 (69.0 %) patients and were represented by hypertrichosis, age spots, skin dimpling, and congenital cicatricial defects of the skin.

Scoliotic deformities prevailed in all children. Kyphosis was diagnosed in only 17 cases. The mean values of the scoliotic and kyphotic curves, their changes after corrective surgery, and correction rates are presented in Table 4. There are no significant differences in these parameters between the groups.

The presented data indicate that the study and the control groups are completely comparable by all the criteria considered above. This makes it possible to

Table 1
Patients' age at the time of surgery

Age group	Group criterion (complete years + months + days)	Patients, n (%)			p
		total	study group	control group	
Early childhood	≤ 3 +11+ 30	13	8 (24.0)	5 (31.0)	0.140
Preschool age	4 ≤ ... ≤ 6 +11+ 30	12	10 (31.0)	2 (12.5)	0.315
Junior school age	7 ≤ ... ≤ 11+11+ 30	10	7 (21.0)	3 (19.0)	0.141
Senior school age	12 ≤ ... ≤ 17 +11+ 30	14	8 (24.0)	6 (37.5)	0.469
Total	–	49	33 (100.0)	16 (100.0)	–

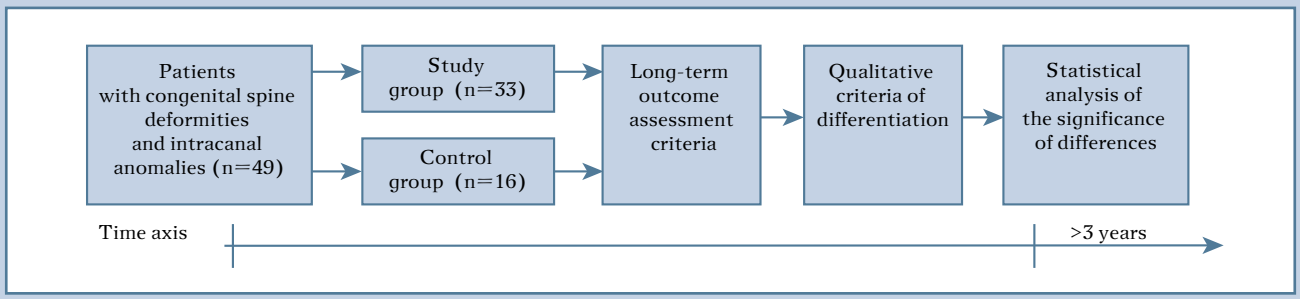

Fig. 1
Strategy for forming inclusion groups

compare the parameters of surgery invasiveness between the groups.

The mean volume of intraoperative blood loss in the study group was 286.6 (min 5.0; max 1,600.0) mL. The mean blood loss in the control group was 247.5 (min 10.0; max 1,100.0) and 266.6 (min 10.0; max 650.0) mL during the first and the second surgeries, respectively.

The mean surgery duration was 227 (min 15; max 600) min in the study group, 198 (min 15; max 530) and 204 (min 45; max 530) min in the control group during the first and the second surgeries, respectively.

Thus, the total blood loss during staged surgical treatment was 1.8 times higher than that of simultaneous surgery ($p = 0.011$), while the total duration of staged surgery was 43.5 % higher than that of single-stage operation ($p = 0.001$).

The severity of neurological deficit and its dynamics immediately after surgery are presented in Table 5. Frankel types D and E prevailed in both groups at baseline without a significant difference between the groups. No significant changes in the neurological status after surgery were observed in both groups.

Transient neurological complications were found in two patients in each group (6.0 and 12.5 %, respectively). Neurological deficit regressed to baseline in the study group after eight-month therapy in both cases. In one patient of the control group, neurological deficit deteriorated to Frankel type A and did not change during the entire observation period.

Long-term results were followed up from one to two years in all patients. Destabilization of the implant was detected radiographically in three (9.0 %) patients of the study group and two (12.5 %) patients of the control group during this period. Elements of spinal instrumentation were re-implanted; no cases of loss of correction were revealed.

Discussion

There are three main opinions on the strategy for treating patients with congenital spinal deformities associated with intracanal anomalies in the literature.

Most authors [1–5] favor staged treatment, which includes neurosurgery with elimination of intracanal pathology as the first stage and spinal deformity correction as the second stage performed after 3–6 months. According to Ulrikh [4], bone spicule position is of fundamental importance: "...apparently, the septum above the L1–L2 vertebrae cannot cause tension in the spinal cord during its growth. Its localization at a lower level in combination with a low position of conus medullaris, intracanal adhesions, and abnormalities of the filum terminale, on the contrary, can lead to tension in the spinal cord during its growth".

Other authors [6–9] prefer simultaneous interventions because one-stage surgery makes it possible to perform single approach and also reduce the number of complications associated with staged procedures.

A number of authors [13–16] argue the need to remove the intracanal component and provide encouraging data on the correction of these deformities. In their opinion, this strategy does not result in neurological deficit, while the surgery for congenital intracanal anomalies has a high risk of these complications. The authors take into account neither level of spinal cord fixation nor conus medullaris position in their study. They also do not analyze the relationship between deformity correction rate and the number of neurological complications.

We would like to emphasize that most studies present a retrospective analysis of case series or small cohorts without comparing the results using common assessment criteria.

We considered it possible to narrow comparison analysis of the two main techniques involving resection of the intracanal component to finding an answer to the following question: which technique for correcting congenital spinal deformities associated with intracanal anomalies correlates with higher surgical invasiveness and complication rate in children? An observational retrospective case-control study of the groups absolutely comparable in age distribution, deformity rate, and nature of vertebral and neural anomalies revealed comparable results of deformity correction and neurological complication risks. The total blood loss volume was 1.8 times higher in case of staged treatment, while the total duration of surgery was 43.5 % lon-

ger than that of simultaneous intervention ($p < 0.05$ for both parameters).

The level of spinal fixation and the level/length of deformity curve can be important aspects when choosing between simultaneous and staged correction. Intracanal anomalies were localized caudal to L3 in all patients of the study (with the exception of Arnold–Chiari malformation in the absence of caudal tethering: its treatment aspects were not analyzed in the current work). In our opinion, simultaneous interventions including deformity correction are advantageous in case of a short deformity curve when fixation of either thoracic or thoracolumbar spine is required. Distraction performed as part of staged correction should be considered as the main corrective procedure in distal fixation of either lumbar or sacral spine with a long structural curve located above the fixation region. However, this narrative is the subject of a separate study, which can be carried out when enough data is accumulated in order to conduct a statistical analysis.

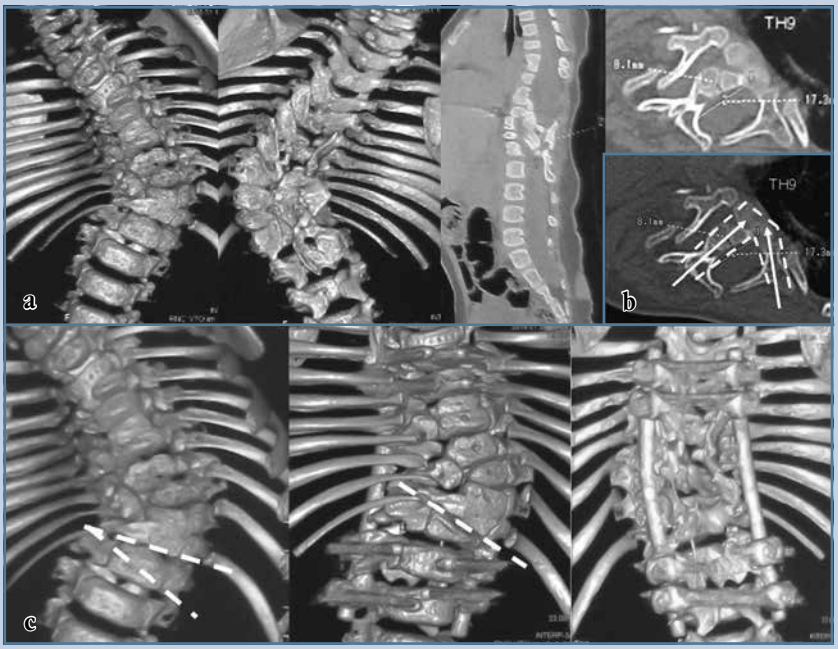

Fig. 2
CT scan of a patient aged 1.5 years with congenital scoliosis in the presence of segmentation failure of the lower thoracic spine, diastematomyelia (SCM I) at T10–T11: **a** – 3D VRT reconstruction and MPR, sagittal and axial scans of the spine at the level of diastematomyelia; **b** – MPR, sagittal and axial scans of the spine at the level of diastematomyelia; a schematic representation of penetration direction during vertebrotomy through the pedicle base and the bone spicule base; **c** – 3D VRT reconstruction of the spine before and after surgery; dotted lines represent the extent of surgical resection and vertebrotomy level

Conclusion

One-stage neurosurgical and orthopedic correction in children with congenital spinal deformity in the presence of intracanal anomaly is not statistically associated with higher surgical invasiveness and complication rate than staged intervention. The main statistically confirmed advantage of simultaneous surgery is reduction in blood loss volume and total duration of surgery. We did not manage to confirm other possible advantages of simultaneous interventions such as better primary correction and a

Table 2
Types of vertebral malformations

Type of vertebral anomaly, the leading component	Patients, n (%)			p
	study group (n = 33)	control group (n = 16)	total (n = 49)	
Failure of segmentation	17 (51.50)	8 (50.00)	25 (51.00)	0.163
Multiple vertebral malformations with failure of segmentation	13 (39.40)	5 (31.25)	18 (36.80)	0.189
Multiple vertebral malformations with hemivertebrae	1 (3.00)	2 (12.50)	3 (6.10)	0.492
Multiple vertebral malformations with segmentation failure and hemivertebrae	2 (6.00)	1 (6.25)	3 (6.10)	0.458

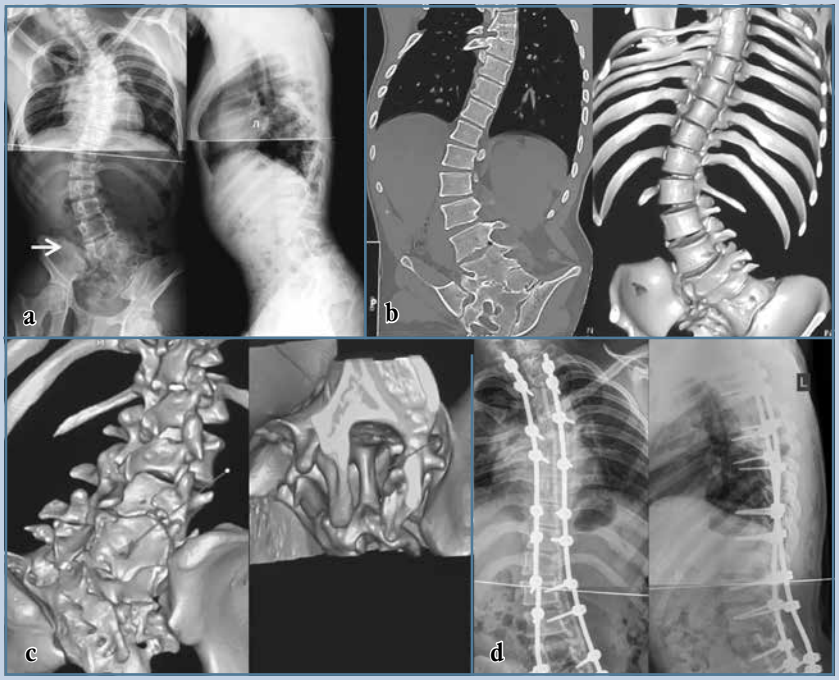

Fig. 3
An example of staged treatment of a 15-year-old patient with congenital spinal and intracanal anomalies, kyphoscoliosis, type II frontal and sagittal imbalance, diastematomyelia (SCM I) at L3–L4: **a** – spine radiographs in the frontal and lateral projections, the arrow indicates the shadow of the bony septum of the spinal canal; **b** – CT scan of the thoracic and lumbar spine, MPR, frontal scan and a 3D VRT reconstruction: failure of segmentation, diastematomyelia (SCM I) at L3–L4 (conditionally), asymmetrically impaired formation, fusion, and segmentation of vertebrae in the thoracic and lumbosacral regions; **c** – a 3D CT reconstruction at the level of the bony septum; **d** – spine radiographs after stage II surgical treatment: correction and posterior instrumented fixation using transpedicular instrumentation were performed

decreased risk of complications at each stage of surgery.

Limitations on the reliability of the results. The study is monocentric with limited comparison groups and long-term follow-up period. As already mentioned, assessing staged corrections with the use of dynamic instrumentation was beyond the study scope. For this reason, we consider it impossible to make any categorical conclusions regarding other advantages and disadvantages of the studied approaches. In our opinion, increasing the duration of long-term follow-up and collecting larger datasets in multicenter studies will allow one not only to assess various combinations of spinal and intracanal malformations in more detail but also find the optimal treatment strategy.

The study had not sponsorship. The authors declare that there is no conflict of interests.

Table 3
Types of intracanal anomalies

Intracanal anomaly	Patients, n (%)			p
	study group (n = 33)	control group (n = 16)	total (n = 49)	
Diastematomyelia (SCM I)	14 (42.50)	6 (37.50)	20 (40.80)	0.077
Diastematomyelia (SCM II)	7 (21.20)	2 (12.50)	9 (18.30)	0.299
Spinal lipoma	4 (12.10)	3 (18.75)	7 (14.30)	0.113
Filum terminale	4 (12.10)	1 (6.25)	5 (10.20)	0.131
Secondary tethering of the spinal cord structures	4 (12.10)	4 (25.00)	8 (16.40)	0.488
Arnold–Chiari type II malformation*	7 (21.20)	4 (25.00)	11 (22.40)	0.094
Number of cases with anomalies	40	20	60	–

*a separate disease group, which did not require neurosurgical intervention.

Table 4

Spinal deformity at baseline and its changes after surgical correction, Cobb angle (deg.)

Deformity		Scoliosis			Kyphosis		
		study group	control group	p	study group	control group	p
Before surgery	mean	28.7	32.1	0.282	35.1	31.0	0.907
	min; max	20.0; 105.0	26.0; 101.0		5.0; 45.0	8.0; 35.6	
After surgery	mean	9.2	12.0	0.100	7.6	8.4	0.804
	min; max	0; 40	0; 65		0; 13	0; 9	
Correction rate, %		66	57	0.175	78	76	0.649

Table 5

Baseline neurological deficit in lower limbs according to the Frankel grading system [17] and its changes after surgery

Frankel grade	Patients, n (%)					
	before surgery			after surgery		
	study group (n = 33)	control group (n = 16)	p	study group (n = 33)	control group (n = 16)	p
A	0 (0.00)	0 (0.00)	0.500	0 (0.00)	1 (6.25)	0.291
B	3 (9.10)	3 (18.75)	0.384	5 (15.10)	3 (18.75)	0.073
C	7 (21.20)	3 (18.75)	0.141	7 (21.20)	4 (25.00)	0.053
D	9 (27.30)	4 (25.00)	0.140	9 (27.20)	3 (18.75)	0.233
E	14(42.40)	6 (37.50)	0.015*	12 (36.30)	5 (31.25)	0.026
Total	33 (100.00)	16 (100.00)	–	33 (100.00)	16 (100.00)	–

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