SURGICAL TREATMENT OF CONGENITAL KYPHOSIS IN PATIENTS OLDER THAN 10 YEARS

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Objective. To analyze the outcomes of surgery for congenital kyphotic deformities of the spine in patients over 10 years old.
Material and Methods. A total of 49 patients with congenital kyphosis were operated on. The age of the patients at the time of treatment was at least 10 years (mean 14.8 years), the follow-up period — at least 2 years (mean 3.8 years). All patients were operated on using third generation segmental instrumentation with hook and hybrid fixation.
Results. Congenital kyphotic deformities are of high rigidity. The average magnitude of preoperative kyphosis was 92.9°, mobility — 27.5 %, during the operation it was corrected to 61.7° (33.5 %), postoperative progression was 4°. All anthropometric indicators have improved, the SRS-24 survey also showed positive patient-reported outcomes. At the same time, the complications are quite frequent, as well as initial involvement of the spinal canal content in the pathological process (11 patients).
Conclusion. Congenital kyphotic deformities should be detected as early as possible and timely treated.
Key Words: congenital kyphosis, surgical treatment, patients older than 10 years.

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Congenital kyphosis occurs quite often, and each case is a challenge for the surgeon, which is associated with the choice of treatment. This issue was addressed in a sufficiently large number of studies [4, 5, 11], but we could not find papers reporting outcomes of surgical treatment in patients aged 10 years and older. At the same time, early progression of kyphotic deformities and propensity to involvement of the spinal canal contents in the pathological process are quite common.

The study purpose was to analyze the outcomes of surgical treatment in congenital kyphosis patients older than 10 years of age.

Material and Methods

A total of 355 patients with congenital spinal deformities underwent surgery at the Clinic of Pediatric and Adolescent Spine Surgery of Novosibirsk Research Institute of Traumatology and Orthopedics n.a. Ya.L. Tsivyan (NRITO) in 1996–2015. The information on patients was retrieved from an electronic database in accordance with the following criteria:

– kyphotic spinal deformities (with or without the scoliotic component) due to congenital vertebral malformations;
– not less than 10 years of age at the time of surgery;
– at least 2 years of postoperative follow-up.

General characteristics of patients. A total of 49 (m/f = 17/32) patients met the described criteria. The patients' age ranged from 10 to 39 years, with the mean age of 14.8 years; the mean postoperative follow-up was 3.8 (2.0–10.2) years.

Based on the data of medical history and previous radiography, non-progressive spinal deformity was diagnosed only in 3 out of 49 patients. Seventeen patients of this group had decompensated spinal deformity (the central sacral line was more than 20 mm away from the T1 vertebral centroid), and 24 patients were diagnosed with subcompensated deformity (the line was less than 20 mm away).

Only 5 patients had a mobile kyphotic deformity (correction in an extension position on a roller was more than 30 % of the initial Cobb value), the remaining patients had a rigid deformity.

We could not localize the kyphosis apex in four cases due to the complexity of anatomical changes associated with multiple abnormalities; 34 patients had thoracic kyphosis; 8 patients had thoracolumbar kyphosis; 3 patients had lumbar kyphosis. The scoliotic component of spinal deformity was present in 47 patients. Most primary scoliosis curves (33) were right-sided. Secondary scoliosis curves were diagnosed in 17 patients; in this case, compensatory curves in 9 patients developed both cranial and caudal to the kyphotic deformity: 3 upper thoracic curves, 11 thoracic curves, and 11 thoracolumbar/lumbar curves. A rib

hump on the convex side of the scoliosis curve was present in all patients, being considered as a costovertebral hump in 33 patients.

Spinal malformations. Spinal malformations were verified in 45 out of 49 patients. Mixed abnormalities (24) as well as abnormalities of formation (17) and pure segmentation (4) prevailed. Various rib malformations (fusion, branching, lacking) were diagnosed in 7 patients.

In one case, an MRI study revealed diastematomyelia at the T1–T2 level, with a bony diastematomyelia spur.

Concomitant pathology was detected in 16 patients of this group: pathology of the cardiovascular system (5 cases of mitral valve prolapse and kyphoscoliotic heart disease), gastrointestinal tract (2 cases of gastritis, gastroduodenitis), urinary system, including 1 case of a neurogenic bladder), non-spinal pathology of the musculoskeletal system (5 cases of degenerative spine disease, osteoporosis, isthmic spondylolisthesis of the L5 vertebra, clavicular hypoplasia), Mohr syndrome (1 case), Scheuthauer-Marie-Sainton syndrome (1 case), adhesive disease (1 case), and schizophrenia (1 case). Thus, non-spinal malformations were detected in 3 out of 49 patients.

Eight patients had surgery in medical history: removal of non-spinal tumors (1 case) abdominal surgery (2 cases), ligation of the ductus arteriosus (1 case), anterior spinal fusion using a rigid strut allograft at the Nizhniy Novgorod Institute of Traumatology and Orthopedics (1 case), corrective surgery for spinal deformity at other clinics in Russia and CIS (3 cases).

Local changes. Shoulder obliquity was diagnosed in 37 patients (1–11°) before surgery and in 24 patients (1–12°) at the end of follow-up. Before surgery, the plumb line dropped from the *incisura jugularis* passed through the umbiculus in 22 patients and 3–65 mm away in the others; in the postoperative period, the deviation (1–65 mm) was observed in 19 patients. Before surgery, scapular symmetry was observed in 17 patients, while scapular asymmetry (1 to 23°) occurred in the others; at the end of follow-up, scapular asymmetry (3 to 16°) was observed also in 17 patients. The plumb line dropped from the C7 spinous process passed through the gluteal cleft in 23 cases and 3–40 mm away from the cleft in the remaining patients; at the end of follow-up, the situation slightly changed: the plumb line passed through the gluteal cleft in 20 patients and 1–40 mm away from the cleft in the vast majority of cases.

Anthropometric data. The mean height of patients was 151.1 (129–169) cm before surgery, 155.6 (132–181) cm immediately after surgery, and 157.5 (140–176) cm at the end of follow-up. The mean weight was 46.5 (22–73) kg before surgery, 45.2 (25–68) kg immediately after surgery, and 54.7 (33–93) kg at the end of follow-up. The vital capacity value was 1,973 (1,100–3,600), 1,584 (1,000–2,500), and 2,127 (1,300–3,600) mL, respectively.

Surgical treatment. All patients underwent surgery using third generation segmental instrumentation with hook and hybrid fixation. The location of the upper limit of the instrumented fusion zone ranged from the C7 to T11 vertebra, and that of the lower limit ranged from the T8 to S1 vertebra. Therefore, the length of the posterior fusion zone was 12.1 (6–15) vertebrae, on average. In 27 cases, spinal deformity correction was preceded by surgery on the anterior parts of the vertebral column: mobilizing discectomy on the kyphosis apex and interbody fusion (25 patients) or posterior spinal fusion with a strut graft. Both interventions in all patients, except three cases, were carried out in single anesthesia. In 22 patients, surgery included only posterior correction using vertebral instrumentation and spinal fusion. In 17 cases, corrective surgery was performed under skeletal traction applied to the calvarial bones and supramalleolar area. Three patients underwent gradual (for 3–6 weeks) correction in a halo-pelvic apparatus, after which the achieved result was fixed using vertebral instrumentation.

Patients with symptoms of dural sac compression underwent two decompression surgeries: one through the anterior approach and the other through the posterolateral approach.

Posterior and anterior bone grafting was performed using only an autograft obtained from a rib resected during thoracotomy and from decortication of the posterior vertebral segments in the instrumented fusion area. Cosmetic surgery, resection of the residual rib hump, was performed in three cases.

The development of various complications required 13 operations for rearrangement and 6 operations for removal of an endocorrector.

Results

One-stage surgery (skeletal traction and deformity correction using segmental instrumentation) lasted 153.9 (70–330) min, on average, and was accompanied by a mean blood loss of 503.5 (30–1,700) mL; two-stage surgery (skeletal traction with discectomy and deformity correction using segmental instrumentation) lasted 234.0 (145–440) min, on average, and was accompanied by a mean blood loss of 803.3 (50–3,000) mL. Decompression of the spinal canal contents (in different variants) lasted 245 (55–580) min, on average, and was accompanied by a mean blood loss of 1,266 (250–3,000) mL.

Dynamics of radiographic parameters. The mean kyphosis value was 92.9° (40–159°) in the upright position and 67.3° (29–102°) in an extended position on a roller. The curve mobility before surgery was 25.6° (27.5 % of the initial value). Surgery reduced the deformity to 61.7° (18–115°); correction amounted to 31.2° (33.5 %). At the end of follow-up, the mean kyphotic deformity was 67.7° (17–129°). Therefore, postoperative progression of kyphosis amounted to 6.0° (19.2 % of the initially achieved correction; Fig. 1) in this group of patients.

The scoliotic component of a multiplanar spinal deformity in study group patients demonstrated the following changes. The initial curve value was 56.7° (5–139°). Surgery reduced the curve to 42.8° (1–118°). Correction amounted to 13.9° (24.5 % of the initial deformity value). At the end of follow-up, the com-

pensatory curve value was 45.5° (1–100°). Therefore, postoperative progression of the secondary curve was 2.7° (19.4 % of the achieved correction). The lumbar lordosis was 74° (38–112°) before surgery, 56.9° (35–80°) immediately after surgery, and 61.1° (26–103°) at the end of follow-up.

Trunk balance. The trunk inclination in the frontal plane, which was determined using radiographic data (the distance between the T1 centroid and the central sacral line), was 19.0 (1–65) mm before surgery, 20.0 (1 52) mm immediately after surgery, and 19.3 (1–74) mm at the end of follow-up.

Neurological symptoms. A preoperative examination revealed neurological symptoms of varying severity in 11 (22.4 %) patients.

Before surgery, pyramidal insufficiency symptoms were detected in 4 out of the 11 patients, with one of them being diagnosed only during a traction test (vertical traction under the patient's body weight).

Symptoms in the form of mono- or paraparesis, without pelvic organ dysfunctions, were detected in six cases (two cases in the setting of epiconus abnormalities). Another patient was diagnosed with stenosis of the spinal canal (Verbiest syndrome).

Postoperatively, none of the 11 patients developed worsening; in one case, improvement was observed pyramidal insufficiency in the presence of initial lower paraparesis.

In these 11 patients, the mean kyphosis value was 114.7° (46–159°), and the mean scoliotic component was 80.2° (10–136°).

Complications occurred in 14 patients. The most frequent complications were mechanical ones: rod fracture (4 cases) and failure of caudal (1 case) and cranial (4 cases) fixation. All these cases required repeated surgery: rearrangement or removal of an endocorrector in the long-term (over two years) postoperative period. One patient with the kyphotic apex located in the lumbar spine, who was admitted to the hospital at the age of 9 years, developed pronounced proximal junctional kyphosis; in this patient, displacement of distal hooks was also found. The patient underwent three additional interventions (replacement of the distal hooks with pedicular screws and extension of the fusion zone to the cranial direction, twice), but every time the junctional kyphosis developed at a new level within 1 year, which finally resulted in endocorrector removal.

Wound infection occurred in three cases, twice in the same patient with an interval of 8 years. Two early infections were stopped, and an endocorrector was spared. In one case, a metal construct was removed.

There was one intraoperative complication: bleeding from the intercostal veins at the discectomy level, which

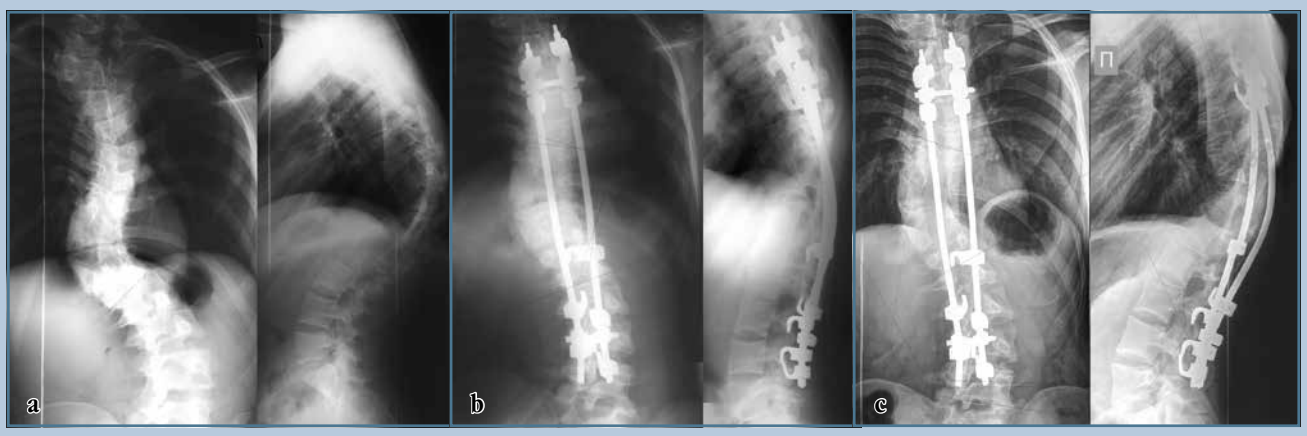

Fig. 1
Radiographs of a 17-year-old female patient T. with congenital uncomplicated progressive subcompensated rigid thoracic kyphosis with a right scoliotic component due to multiple malformations (concrescence of the C_7–T_1 vertebrae, butterfly T_1 vertebra, left-sided wedge-shaped T_3 vertebra, concrescence of the T_2–T_6 and T_{10}–T_{12} vertebral bodies, right-sided wedge-shaped T_{12} and T_{13} vertebrae, multiple rib abnormalities); surgery of 01.03.05 (M.V. Mikhaylovskiy): mobilizing discectomy of the T_{12}–L_1 vertebrae, interbody fusion with an autograft bone, skeletal traction, deformity correction using NRITO-adapted instrumentation, posterior spinal fusion with an autograft bone; time of surgery 265 min, blood loss 750 mL: **a** – before surgery thoracic kyphosis of 77°, a right-sided scoliotic component of 63°, compensatory curves of 38° (L_2–L_5) and 35° (T_5–T_9), lumbar lordosis of 68°; **b** – after surgery thoracic kyphosis of 45°, the right-sided scoliotic component of 50°, compensatory curves of 29° (L_2–L_5) and 48° (T_5–T_9), lumbar lordosis of 50°; **c** – 3 years after surgery thoracic kyphosis of 50°, the right-sided scoliotic component of 43°, compensatory curves of 43° (L_2–L_5) and 50° (T_5–T_9), lumbar lordosis of 61°

required retoracotomy and hemostasis, without any consequences.

Patient survey results. We used the SRS-24 questionnaire containing 24 questions divided into seven groups (domains) to assess the patient condition according to the following indicators: pain, general appearance, appearance after surgery, functions after surgery, overall activity, professional activity, and satisfaction with results of surgery.

Questionnaires were filled out 6, 12, and 24 months after surgery. Each indicator was evaluated on a five-point scale where 1 meant the worst result, and 5 meant the best result.

Six months after surgery, study group patients (Fig. 2) evaluated pain as moderate (3.6 points); later, the pain slightly decreased (3.7 points). Evaluation of the general appearance changed little: 3.6 points postoperatively, 3.7 points one year after surgery, and 3.6 points two years after surgery. The appearance shortly after surgery (6 months) was scored high (4.0), the same score was one year later, but it slightly decreased, up to 3.6 points, after two years. Postoperatively, functions were scored quite low (2.4 points); the score decreased more (2.0 points) one year after surgery and slightly increased (2.2 points) two years after surgery. The overall activity, being relatively low (2.9 points) shortly after surgery, then gradually increased from 3.0 points one year after surgery to 3.4 points two years after surgery. The professional activity was assessed as moderate (3.4 points) shortly after surgery; the assessment was increased to 3.8 points one year after surgery and then slightly decreased to 3.5 points. Satisfaction with the result of surgery was high enough and varied insignificantly: 3.8, 3.9, and 3.7 points 6 months, one year, and two years after surgery, respectively.

In general, the survey results demonstrate that, according to the majority of parameters, patients improved their assessment of the treatment outcomes, sometimes to the maximum score. The assessment of functions was somewhat different, demonstrating lower parameters. Presumably, this results from immobilization of a significant number of vertebral segments, which is less manifested in answers to questions from other questionnaire domains (pain, appearance, etc.).

Discussion

Unfortunately, the modern literature contains a very small number of papers dedicated to the treatment of kyphotic deformities in patients older than 10 years of age. Usually, these patients are part of a group with congenital deformities of different spatial orientations.

Aydogan et al. [2] reported successful treatment of 19 patients, aged 2–22 years, with all deformity types (kyphosis, kyphoscoliosis, scoliosis) of the thoracic and lumbar spine. They used posterior hemivertebrectomy with placement of an anterior cage and transpedicular fixation. The mean follow-up period was 4.6 years. The authors did not observe any significant loss of correction, but noted that surgery was accompanied by massive bleeding.

In 2006, Kawakami and Goto [5] presented the results of surgical correction of kyphotic and kyphoscoliotic deformities using vertebrotomy, followed by posterior instrumented fixation. There were 25 patients with a mean age of 16 years. The kyphotic deformity was reduced from 58 to 24°; the lumbar lordosis was decreased from 73 to 56°. Complications included 4 cases of dura mater injury and 3 cases of transient neurological symptoms.

One of a few studies on surgical correction of congenital spinal deformities combined with intracanal abnormalities was published by Hamsaoglu et al. [4]. They operated on 21 patients (scoliosis, kyphosis) who were diagnosed with tethered spinal cord syndrome and/or diastematomyelia. The mean age of patients was 13 (3 to 19) years. Four patients (all with kyphosis) had a primary neurological deficit. The surgical technique was as follows. Initially, a segmental endocor-

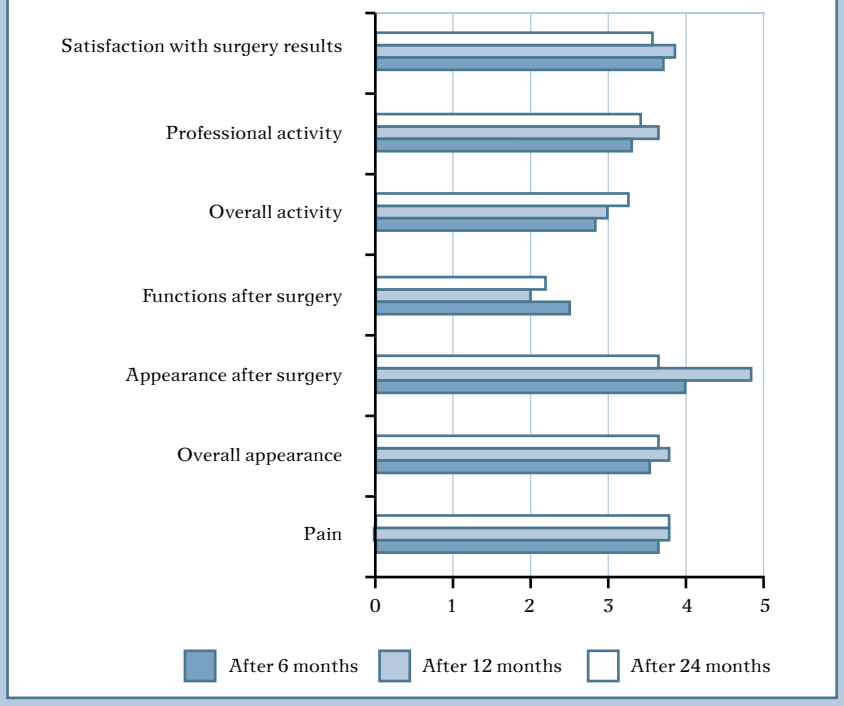

Fig. 2
SRS-24 scores in kyphotic patients

rector was implanted via the posterior approach throughout a targeted fusion area. Next, a team of neurosurgeons performed required surgery on the spinal canal contents. And finally, reasonable correction of the deformity and fixation of the achieved result using an endocorrector were carried out. At the second step, anterior spinal fusion was performed to prevent the crankshaft phenomenon in patients with incomplete growth. The mean duration of the main surgery step was 9.3 (7 to 12) hours, and the mean blood loss was 1,980 (1,500–3,000) mL. The mean follow-up period was 6.8 (2 to 12) years. No worsening or development of a neurological deficit was observed. The mean correction of a spinal deformity, scoliotic or kyphotic, amounted to 23% of the baseline value, and postoperative loss of the correction was less than 10%. There were no cases of infectious complications and pseudarthrosis of the block. The authors suggested that the used technique, which involved the simultaneous use of orthopedic and neurosurgical interventions, was not associated with significant difficulties.

Zhang et al. [10] reported the experience of surgical correction of congenital kyphoscoliosis due to fully segmented hemivertebrae. An abnormal hemivertebra was completely removed through the posterior approach, and instrumented fixation was performed within a short length. The peculiarity of this study was the fact that all 19 patients were adolescents whose mean age was 13 years and 9 months. The postoperative follow-up period was 3 years. The scoliotic component of the deformity was corrected from 40.7 to 10.3° (74.7 %), and the kyphotic component was corrected from 25.2 to 6.5° (74.2 %). Postoperative loss of the correction was insignificant.

Suzuki et al. [9] operated on 31 patients with congenital deformities of the lumbar and thoracolumbar spine. The mean age of patients was 11.8 years; the mean postoperative follow-up period was 13.1 years. Posterior and posterior-anterior surgery using segmental instrumentation was used. Kyphosis was corrected from 38 to 23°, on average. In this case, no loss of the correction occurred during the entire postoperative follow-up period. Sagittal imbalance was reduced from 10 to 5 mm. Repeated surgery was performed in seven patients: in five cases, the cause was decompensation of the trunk; in two cases, the cause was pseudoarthrosis of the block.

Dockendorf and Silva [3] used posterior instrumentation (Kaneda system) in the treatment of congenital kyphosis. There were 4 patients aged 15–17 years. The kyphotic deformity was corrected from 50.3 to 32.0°; the posterior instrumentation was complemented with a fibular strut graft. There were no neurological and infectious complications. One repeated surgery was required during 11-year postoperative follow-up.

Congenital kyphosis, compared to any other spinal deformities, is well known to most often lead to the development of neurological complications. Mushkin et al. [7] who described 61 similar cases emphasized that the rate of neurological symptoms directly depends on the type of abnormality, Cobb angle, and canal stenosis at the deformity apex. The authors indicate that complications occur more frequently in type I and III deformities. This is quite true, but neurological symptoms can develop in type II deformities, although in rare cases [1]. One of similar cases was presented by Khanna et al. [6]. The authors described the development of lower paraparesis in a 17-year-old male patient and emphasized that this was the first case in their practice and in the literature. They performed posterior decompression of the spinal cord and spinal fusion using a rib autograft. After three years, the neurological symptoms completely regressed.

In general, treatment of congenital kyphosis complicated by neurological symptoms is a challenge. This fact was emphasized in a paper by Song et al. [8] who operated on 51 patients with angular (a mean Cobb angle of 72°) kyphosis of varying etiology. Progressive myelopathy was diagnosed in 16 patients. Surgery included anterior decompression and posterior spinal fusion with pedicular fixation, without correction of the spinal deformity. The postoperative follow-up period was 72 months. There were no cases of deformity progression and pseudarthrosis of the block. According to a survey of patients (SRS-24 questionnaire), pain decreased (3.1–4.7), and daily activity increased (2.3–4.2). According to the Frankel scale, 15 patients improved their condition by at least one grade.

Despite the fact that a significant part of the study group patients were admitted to the hospital at the age of over 10 years, the data of medical history and preoperative observations enabled the diagnosis of a non-progressive deformity only in four cases. Most of the patients were diagnosed with multiple malformations occurring in one or more spinal segments. This circumstance should probably be considered as the cause for rigidity of the primary curve, which is objectivized by functional spondylography. In most cases, deformities are decompensated in the frontal plane, probably due to the fact that rigidity reduces the capability for self-correction.

Given the results of surgical treatment, we may state that the appearance normalized, which was confirmed by the data on the changes in local appearance and trunk imbalance. According to the survey data, patients were generally satisfied with the results of treatment. The height and weight of patients also increased.

An analysis of the changes in the primary and compensatory curves provides predictable information. The kyphotic component of the deformity is grossly severe (92.9°) and very rigid (passive correction with a reclination roller amounts only to 27.5 %). This also pertains to the scoliotic component – 56.7° (24.5 %). Surgical correction of both components is small, especially in comparison to idiopathic scoliosis, with postoperative progression of the kyphotic component exceeding, to some extent, that of the scoliotic component 6.0 and 2.7°, respectively. The sagittal contour of the lumbar spine remained within normal parameters at all stages of postoperative follow-up.

It is necessary to note a significant number of mechanical postoperative complications. Probably, rod fractures,

instability of fixations, and the formation of junctional kyphosis are associated with a biomechanical situation produced by a severe kyphotic component of the spinal deformity.

Conclusion

Congenital kyphosis in patients in the second decade of life and older is a serious problem for the spine surgeon. Stiffness, continued progression, and propensity to the development of neurological complications are typical of the discussed deformity. These deformities should be diagnosed and treated as soon as possible. However, these are just velleities because the reality is far from the ideal. Patients "treat" themselves with corsets, shoulder braces, and massage, but apply to specialized clinics lately. Our results are determined by a surgical technique: deformity correction using segmental instrumentation, with or without preliminary discectomy. Comparison with the results of foreign colleagues demonstrates almost complete identity, except radical interventions (PSO, VCR). It is possible to stop progression, achieve reliable but not very significant correction, and improve the appearance, with the positive effect of surgical treatment being objectivized by the results of patient reported outcomes.

Regarding the aforementioned radical surgery, it is indicated relatively rarely due to technical complexity and a high risk of neurological complications. We have performed these interventions, but their results have not been considered in this article due to their relatively small number and an insufficient length of postoperative follow-up.

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