TREATMENT OF CONGENITAL SPINAL DEFORMITIES IN CHILDREN: YESTERDAY, TODAY, TOMORROW

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The paper presents an unsystematized review of technologies, techniques and options for surgical treatment of congenital spinal deformities in children over the past 40 years. The main trends in the surgery of spinal deformities are highlighted: evolution of methods of visual diagnostic, treatment planning, and surgeon action control, introduction of adapted functional status scales and questionnaires for quality of life, hybridization of surgical techniques, evolution of spinal implants and instruments, and progress of anesthetic management. At the same time, new clinical and scientific problems are also discussed in the paper: questions of unifying terminology, planning the volume of treatment, the difficulty of comparing treatment methods and technologies, education, and integration.

Key Words: spinal malformations, congenital scoliosis, deformities, spine, children, technologies, trends, treatment.

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This publication was inspired by the proposal made by the organizers of the 9th Meeting of the Association of Spinal Surgeons of Russia «Spine Surgery: the Achievements and Unsolved Questions» to one of the authors to deliver a speech on the current trends in surgery of congenital spinal deformities. This analysis has gone beyond the scope of the conventional reviews and was actually shaped as a public dialogue.

The questions related to surgical treatment of spinal anomalies in Russia started to be studied in the late 1970s–early 1980s [1–3]. However, their development in the USSR and during the first several decades of existence of the Russian Federation was characterized by a number of political (the lack of scientific or practical networking), economic («backyard» production of implants and the insufficiently developed technical equipment), and psychological features (many top orthopedists believed that reconstructive stabilization surgeries were justified only for preadolescent and adolescent patients). These limitations have to a certain extent hindered the evolution of surgery of the axial skeleton in children of early age. At that time, systematic development of this field was started only at the Leningrad Pediatric Medical Institute (its name being later changed to St. Petersburg Medical Academy). Although the ideology of the institute back then was virtually not different from those of the leading clinics abroad, very few Russian studies have been published in foreign journals and have mostly focused on simultaneous two-stage (later single-stage) hemivertebral excision involving instrumented correction/fixation or presented justification of the benefits of compression as the main correction maneuver characterized by greater safety [4–10].

Over the past 10–15 years, there has been sufficient progress in technical facilities and treatment outcomes in pediatric spine surgery in Russia, bringing it to the level comparable to that in top foreign clinics. The geography of Russian publications has been broadened, although the number of articles published in foreign journals still remains limited [11–16]. Comparison with the modern global trends reveals the following patterns in surgery of spinal deformities:

– the evolution of visual diagnosis methods (the EOS system, side-bending X-ray, the emergence of gravity computed tomography or magnetic resonance imaging, and 3D printing), which has modified treatment planning and made it possible to elaborate tailored surgical approaches with allowance for the type of vertebral malformations;

– modification of the criteria used to evaluate the intervention efficiency: they started to involve not only the assessment of deformity correction but also the traumaticity degree of an intervention (blood loss volume and duration), which has led to implementing blood-saving techniques and methods for controlling surgeon's actions (3D navigation, intraoperative CT/MRI, robot-assisted technologies, and neuromonitoring);

– implementation of the adapted functional status scales and quality of life questionnaires (e.g., SRS);

– application of different types of instrumentation in the same patient, as well as performing simultaneous interventions for the spine, spinal cord, and the thorax, has made it necessary to use hybrid surgical methods; more accurate patient selection has led to the revival of external fixation methods (ExFix, Halo-pelvic, and Halo-gravity);

– the evolution of spinal implants (third- and fourth-generation ones, Lenke frame, reduction and uniaxial screws) and equipment (tables, osteoplasty kits, power-driven equipment, and ultrasonic bone cutters);

– the progress in anesthesia management has made it possible to monitor the depth of anesthesia, perform video-assisted intubation, and use drugs for hemostasis management [7, 8, 17–19].

However, new clinical and fundamental problems have also emerged:

– terminology unification was required for refining developmental malformations;

– treatment planning became more challenging: first of all, there appeared some new factors that can hardly be analyzed in children of young age (e.g., balance of the body or certain spinal segments);

– the advances in medicine and economics have made it necessary to compare the treatment methods and techniques using the criteria that can hardly be compared between different countries (e.g., the cost/benefit analysis);

– interdisciplinary integration started to play an exclusively administrative and organizational role: so it was necessary to form a complimentary team consisting of surgeons, orthopedists, neurosurgeons, anesthetists, pediatricians, general practitioners, neurologists, neurophysiologists, rehabilitation therapists, and physical therapists; all the team members need to be trained, and the learning curves will inevitably have an effect when new tools start to be used;

– there has been controversy between the new knowledge (systematization of nonconventional rare malformations and justification of their management) and the limitations related to the regulatory statistical tools (first of all, the ICD), which does not involve this detalization.

The following aspects are typical of our country: the strict regulations of surgical activity limiting any innovations related to the choice of treatment modality; the need to prove that a certain procedure is superior to another one, while there are no criteria for this assessment or they are rather inconsistent; blatant disregard (or, vice versa, hypertrophy) of certain expert's personal opinion. This is especially true for rare spinal malformations when it is very difficult to find enough patients to form study and control groups that would be sufficiently large for statistical analysis.

Taking these aspects into account, we will try to focus on the questions outlined above.

Terminology and Classification

Strange as it may seem, these questions are of the utmost importance in Russia. Codes for almost all congenital spinal malformations are listed in the ICD-10 not only in the Q group (Q00–Q99 «Congenital malformations/developmental anomalies») but also correspond to «deforming dorsopathies» (M40–M43), «spondylopathies» (M45–M49), «other dorsopathies» (M50–M54), and «chromosomal abnormalities.» It is worth mentioning that the International Classification of Diseases (ICD) is a statistical classifier rather than the clinical one and is used as a tool for administrative control in maintenance of clinical records (patient's records) and, unfortunately, for financial reasons. Since a pathology needs to fall into a certain ICD class, the clinicians tweak the diagnosis to a more convenient (sometimes the «more expensive») ICD code, which has nothing to do with standardization of surgical approaches, the strategy of dynamic patient management, and prediction treatment outcomes, but makes clinical evaluation of the pathology more difficult.

One should bear in mind that the classifications of vertebral developmental anomalies proposed by MacEwen et al. [20] and complemented by Winter et al. [21], McMaster and Ohtsuka et al. [22], E.V. Ulrikh [4], and Kawakami et al. [23] do not take into account the modern potential of using CT (including 3D CT) to evaluate the pathology (Fig. 1), so many complex deformities are classified as combined malformations (i.e., those including abnormal vertebral segmentation, formation, and coalescence). Meanwhile, to choose a proper treatment modality, it is fundamentally important to identify the key anomaly that is responsible for certain spinal deformity and malformation progression or a combination of anomalies, which allows one to differentiate the approach used for treating its harmful effect on spinal growth and development [4]. This idea has also been presented in other studies [5, 24].

Figure 1 shows an example of combined malformation with the noncompensated triad of key anomalies: asymmetric L2 butterfly vertebra (in combined malformations, the number of vertebrae is regarded as conditional), the semisegmented T7 hemivertebra with contralateral block and sacralization of the L6. The risk of progression of any of these components makes it necessary to perform differentiated planning of the strategy for surgical treatment of the deformity; neither the supernumerary anomalous vertebrae nor the complex of fused vertebrae in the upper thoracic spine affects the deformity progression.

Modern Diagnosis Techniques

The emergence of Slot radiography, which allows one to obtain full-body radiographs followed by 3D processing and analysis of the deformities of the axial skeleton and body balance (the EOS system), has been the key trend over the past decade [25, 26]. Functional radiographs still remain important in evaluation of the mobility of structural and nonstructural curves; however, their analysis in the 3D model is fundamentally different. The digital platforms for processing the radiography data (Surgimap, MediCAD, and SagittalMeter) allow one to take into account an extended set of parameters: the length and Cobb angle of the structural or nonstructural curves, torsion of the apical vertebrae, deviation from the central sacral vertical line (CSVL), curve mobility parameters, rotation of the cranial and caudal vertebrae, the horizontal position of the distal neutral vertebra, the stability zone, shoulder balance, T1 slope, deviation of the C7 vertebra from the posterior sacral

vertical line (PSVL), etc. Today, routine examination in the leading clinics already involves radiographic telemetry of the spine, CT in the 3D VRT and MPR modes, and MRI. The first reports on evaluation of both static and dynamic body balance have been published [27]. It is fair to expect that the options of vertical (gravity) CT and MRI, as well as evaluation of the dynamic body balance, will become available in the near future.

The emergence of virtual (computer-assisted) and real-world 3D modeling (3D printing) of the anomalous spine gives grounds to expect that a checklist of surgeon's actions will be elaborated and will be based on 3D classification of the malformation, identification of its key components, allowance for the metric and spatial parameters of the instrumentation zone, the geometry and trajectory of screw placement, as well as the type and level of osteotomy. However, it still remains unclear how decision-making regarding the treatment tactics depends on the geometrically increasing number of evaluated characteristics and whether they actually need to be evaluated for this purpose.

Evolution of Implants

The emergence of Harrington distraction rod in the 1960s, as well as the Luque fixation procedure and ventral fixation systems in the 1970s (Dwyer and Zielke), to treat patients with congenital vertebral malformations ensured only moderate deformity correction [28, 29]. An absolutely new stage became possible in the 1980s as the CD instrumentation [30] started to be used and transpedicular fixation was popularized [31, 32]; the assortment of anchoring elements (hooks, transpedicular and pelvic screws) was broadened in the 1990s [3–5, 33–35]. Meanwhile, personalized correction of congenital scoliosis of children of early age (primarily having hemivertebrae) has become feasible as surgeons started to use the compression maneuver instead of distraction already when Harrington rods threaded in opposite direction with respect to the distractor were launched into practice and subsequently, due to designing low-profile instrumentation having small diameter (baby CD) especially for this age cohort. The instrumentation further evolved as an apical vertebral derotation frame, percutaneous fixation, and various systems for monitoring the axial growth in children started to be used in the 2000s. The latter systems are generally classified to distraction-type (growing rods, VEPTR, and magnetic rods – Phenix/MAGEC), compression-type (stapling and using cable tension systems), and guided growth systems (Luque–Trolley and Shilla systems) [36–46].

If one views the instrumented fixation systems as a «technology – method – option» algorithm, the first two decades of the 21st century can be characterized as the evolution of instrumentation options.

Choosing the Fixation Zone

According to the Dubousset cone of economy concept [47], the evolution in techniques used for analyzing the radiographic and functional parameters characterizing the static and dynamic frontal and sagittal spinal profile (the so-called spinal alignment and balance) has made it topical to choose the fixation zone. The capabilities of the main four correction maneuvers (translation, apical vertebral derotation, distraction, and compression) have reached the maximum possible capabilities of segment-wise correction of all the deformity components [48].

As mentioned earlier, in order to process the data using progressive digital platforms, one needs to understand the biomechanics of axial skeleton deformation whose key components can be presented as follows:

(1) segmental and global gravitational balance [47];

(2) mobility of deformity curves and imbalance compensation; and

(3) optimization of correction technologies, methods, and procedures that involve the potential of operative approach, the levels and types of osteotomy and variants of spinal fixation.

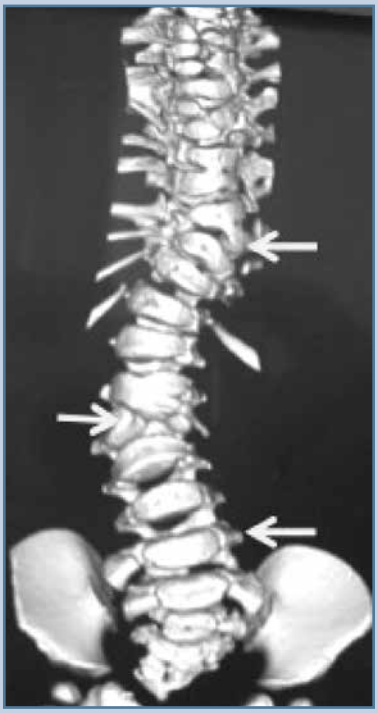

Fig. 1
A CT scan (3D multiplanar reconstruction, anterior view) of an 18-month-old child having congenital scoliosis and multiple developmental anomalies of the vertebrae: the key components of the deformity are shown with arrows (see explanation in the text)

Growth Control

The broad range of technologies and implants used for spinal growth control makes it difficult to choose the best option. The key trends in spinal growth control are as follows: the predominant use of bilateral systems [36, 49] or controlling growth and volume of the thorax [38, 39–41, 50, 51]. The efficiency of correction is compared to the rate of complications, the functional outcomes, and patients' quality of life. It has been reported that minimally invasive approaches within the zone of instrumentation placement and insertion of thoracic rods via the axillary approach are beneficial [52]. The risk of complications (mainly,

implant instability and surgical site infection) decreases with age at which a patient has undergone primary surgery, while simultaneously the outcomes of correction are worsened [43] and parameters of the body balance and thoracic volume (the SAL index and the thorax asymmetry index) are improved [53]. The advantages of using distractors ensuring single- or double-tip rib fixation in patients with thoracic insufficiency syndrome have also been demonstrated [42, 45, 54].

Release Procedures and Control over Surgeon's Actions

The wide implementation of corrective spinal osteotomies and their systematization are especially relevant for congenital deformities. Three-column osteotomies (Schwab grades 3–6) can be regarded as the main tool for correcting severe and decompensated deformities; the posterior approach is associated with fewer complications [55]. The trend to limit the fixation zone (including via achieving local mobility) observed for three-column osteotomies fully agrees with the viewpoint of Dubousset [47], who has repeatedly suggested that extensive fixation should be avoided if it is possible to perform local correction (Fig. 2) [56–59].

The technical features of limited interventions include the use of navigation or endoscopy to control the osteotomy volume [60], as well as the application of ultrasonic bone cutter to resect osseous structures of the spine [59, 61, 62].

An individual trend in evolution of spinal surgery involves controlling surgeon's actions to reduce the number of most severe complications (the neurological and infectious ones) after deformity correction surgeries [63]. Briefly, the main intraoperative measures involve controlling the instrumentation insertion (radiography, CT, MRI, or 3D template-assisted navigation), controlling the status of spinal cord conduction (intraoperative neuromonitoring), and controlling the osteotomy volume (bone tool navigation and endo video assistance). However, the problems related to interpretation of the results of intraoperative neuromonitoring with allowance for false negative and, more importantly, false positive data, still persist. The key studies focus on elaboration of protocols of neurophysiological control [64–67] and measures to be taken if signal is lost (the NASCIS II, NASCIS III protocols, etc.) [68–71]. To reduce the risk of complications, it is justified to use checklists for preoperative examination and preparation of patients by a multidisciplinary medical team and checklists for the team's actions during a surgery to correct spinal deformities (including the congenital ones) [72].

Hybridization of Techniques

One of the current trends in surgery of congenital spinal malformations is to simultaneously use several techniques for treating a combined pathology. This largely has been done due to the establishment of spinal surgery, a subdiscipline that simultaneously uses the neurosurgical and orthopedic methods. Combination of meningolysis and meningoradiculolysis involving spinal fixation (Fig. 3) [14, 73], as well as spinal osteotomy with dynamic or total interstitial instrumentation [63] and/or temporary external extrafocal transpedicular fixation, halo-gravity, halo-pelvic, and halo-cast apparatuses [14, 16, 73], are the most in-demand ones.

Conclusions

Today, prospective, multicenter, inter-rater concordance trials should be regarded as the most efficient studies for elaborating classifications and protocols of managing patients with different pathologies. However, for the group of congenital developmental anomalies of the spine, this resource can be limited because of the small number of cohorts of patients having homotypic anomalies or their combinations [57]. For this very reason, it still is extremely important to perform multicenter retrospective evaluation and systematization of reviews to develop a unified terminology to be used in evaluation and management protocols for these patients.

In our opinion, the key current trends of surgery of congenital spinal deformities are as follows:

– the diagnostic one, which is related to 3D X-ray imaging, detalization of the anomaly structure, the use of digital platforms for parameter assessment, and refinement of the body balance;

– the multidisciplinarity of syndromic evaluation of the patient's status and the related treatment risks;

– comprehensive diagnosis of the vertebral and concomitant anomalies inevitably leads to refinement of planned surgery volume, which striving for correction of the deformity and body imbalance;

– the low-trauma nature of the approach (including that used for spinal osteotomy) and justified minimization of the length of instrumented spinal fixation zone at an early age are preferred; however, the role of the instrumentation controlling growth of children with extensive spinal deformities will be refined as more data become available;

– if there are no large-size homogeneous clinical cohorts, the nonconventional (including novel) approaches to treatment of rare spinal malformations cannot be regarded as evidence-based; pooling the data obtained at different study sites, elaborating the checklists for surgeon's actions, and unifying the analysis results using the principle of inter-rater concordance at the initial stage can rather quickly give rise to conclusions whose reliability will also be proved by statistical analysis.

We will appreciate any comments, additional information, and proposals on designing perspective multicenter studies.

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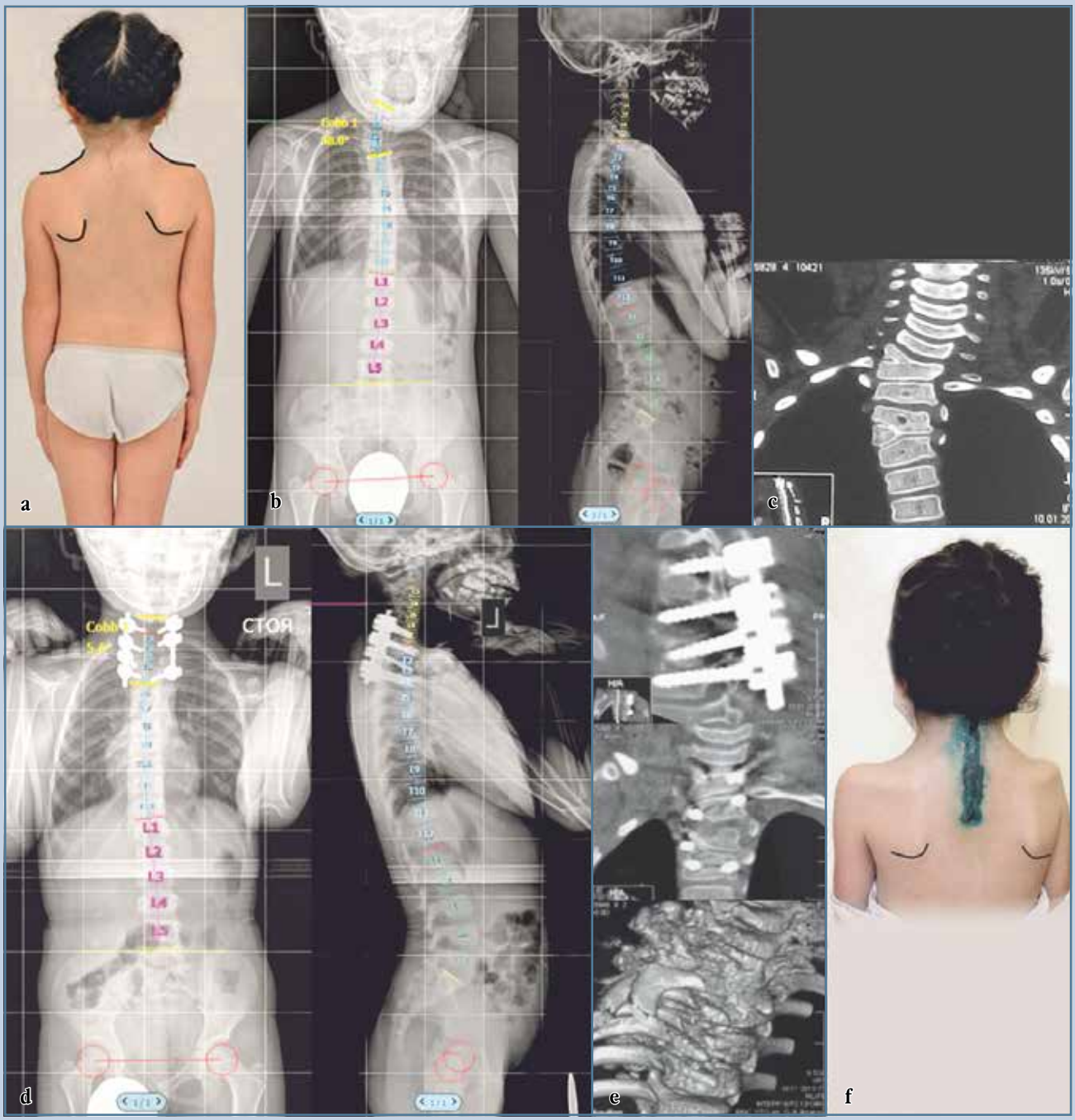

Fig. 2
The data for a 7-year-old girl with congenital scoliosis; the main malformation is developmental and segmentation anomaly of the C7–T5 vertebrae: **a** – a photo taken prior to treatment: severe shoulder girdle imbalance; **b** – preoperative frontal and lateral radiographs of the spine (the Surgimap software was used for digital analysis); **c** – CT scan in the MPR mode; frontal view: the asymmetric form of T1–T6 vertebral segmentation anomaly, unsegmented lateral T1–T5 hemivertebrae, Cobb angle, 38°; **d, e** – postoperative frontal and lateral radiographs (the Surgimap software was used for digital analysis): asymmetric bone–disc–bone osteotomy (BDBO, Schwab grade 4) at the T4 level, local posterior instrumented fixation of T1–T5 anomaly zone using pediatric instrumentation, deformity correction, local fusion, Cobb angle, 5.6°; **f** – a photo taken after the treatment: the shoulder girdle balance has been restored

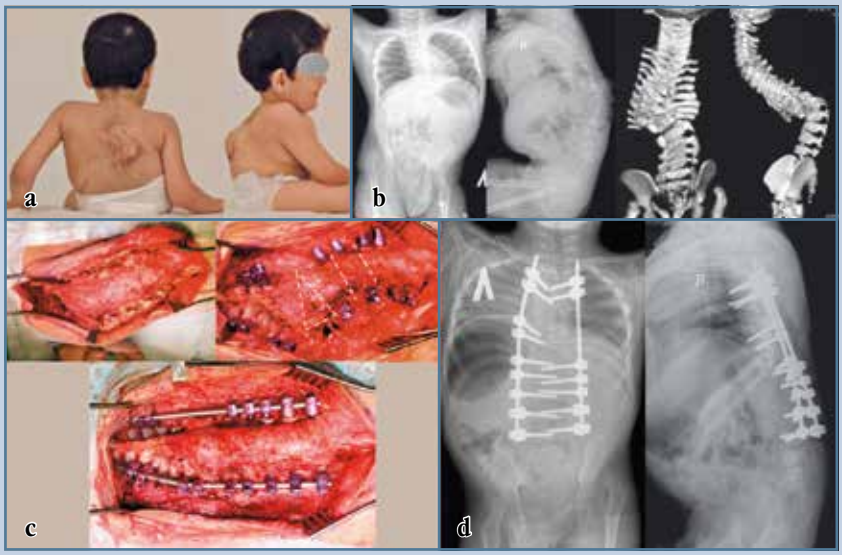

Fig. 3
The data of a 6.5-year-old girl with congenital kyphoscoliosis associated with congenital spinal dysraphism (myelomeningocele) starting at the T6 level: **a** – a photo before treatment: severe frontal imbalance; **b** – preoperative frontal and lateral radiographs of the spine, CT in the VRT mode (posterior and lateral views): the asymmetric form of the T6–T12 vertebral segmentation anomaly, conditionally unsegmented lateral T9 and T11 hemivertebrae; **c** – intraoperative photos: asymmetric PSO osteotomy (Schwab grade 3) at the T10–T11 level, bone–disc–bone osteotomy (BDBO, Schwab grade 4) at the L1–L2 and L2–L3 levels, posterior instrumented fixation with pediatric instrumentation at the T3–L5 level, deformity correction, local fusion, correction of the kyphotic and scoliotic components of the deformity (96 and 78%, respectively); **d** – postoperative frontal and lateral radiographs of the spine: the sagittal and frontal balance has been restored

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