



SURGICAL TREATMENT OF CHILDREN WITH CONGENITAL SPINAL DEFORMITIES ASSOCIATED WITH MULTIPLE MALFORMATIONS: A LITERATURE REVIEW

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The tactics of surgical treatment of patients with spinal deformities associated with multiple malformations is of great importance because of aggressive course of disease, limited opportunities for radical correction of deformity, and a high risk of complications. The review analyzes the literature devoted to this problem. The theoretical basis behind the need for surgical treatment is considered. The current methods of surgical intervention are presented, and their principal ideological and practical differences are demonstrated. Surgical treatment of patients with spinal deformity associated with multiple malformations is of particular interest in the practice of a spine specialist. Most patients at an early age require multi-stage surgical treatment which threatens to entail serious complications. In order to minimize risks and improve the final result of treatment, it is necessary to apply existing surgical techniques, as well as correctly arrange a sequence and frequency of their implementation.

Key Words: spinal malformation, infantile scoliosis, children, surgical treatment.

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The choice of treatment tactics for pediatric patients with congenital spinal deformities associated with multiple vertebral malformations remains one of the most important issues in pediatric spinal surgery. With the term “multiple malformations” we mean the presence of two or more vertebrae with similar type of malformations or a combination of various congenital vertebral malformations, including both formation and segmentation failure, which are often accompanied by rib synostosis. The malformation pattern determines the natural course of congenital spinal deformities (scoliotic, kyphotic, kyphoscoliotic) and the rates of curve progression during growth and development in children.

The importance of this issue is due to the peculiarities of the disease: severity and rigidity of the deformity, persistency and rapidness of curve progression, development of the compensatory

curves as a measure of counterbalance for the main congenital curve, as well as a significant decline in quality of life and lifespan of these patients [7, 12, 13, 18, 19, 31]. In the vast majority, these patients require surgical correction of the present deformity, except for children with alternating types of malformations. There are a number of challenges in the treatment of pediatric patients. First, it is necessary to perform surgical procedures as early as possible because of rapid progression of congenital curvature. At the same time, not in every case it is possible to perform a radical correction of the deformity in skeletal immature children. Another matter is the high rate of complications, both intraoperative and postoperative, as well as the necessity of multi-staged surgical treatment [3, 39, 51].

Russian literature highlights quite in detail the tactics of surgical treatment of infants with isolated congenital defor-

mities of the thoracic and lumbar spine [1, 7]. Studies address the methods for surgical stabilization of spinal deformity in congenital vertebral malformations in older children [2]. However, the Russian literature hardly contains any research devoted to the surgical correction of congenital spinal deformities associated with multiple vertebral malformations.

The real rate of congenital spinal deformities is unknown: part of the patients are asymptomatic due to the compensated types of malformations. According to population-based screening research using fluorographic findings, the rate of malformations in the thoracic spine was reported to be 0.5–1 per 1000 [52]. The findings of ultrasonographic screening of the fetus show the rate of 0.1–0.3 per 1000 newborns [27, 32]. Additionally, severe and very severe congenital spine and chest wall deformities constitute 32.5 to 65.4 % in children before the age of 7 years [5, 8]. In gen-

eral, congenital spinal deformities are quite rare.

Pediatric patients with multiple congenital spine deformities have been reported to suffer from a high mortality rate due to cardiac and pulmonary diseases [55], which are associated with extrinsic restrictive lung disease followed by pulmonary hypertension (*cor pulmonale*). This condition was first described by Campbell [18] as the thoracic insufficiency syndrome. Research by Dimeglio and other authors demonstrates that alveologenesis is most active during the first five years of a child's life and the number of alveoli after 7–8 years does not change [15, 24, 29, 47, 53]. The reduced elasticity of the thoracic cage and its severe deformity impair lung development in children and cause secondary changes [5, 6, 18, 20, 36, 59]. Therefore, a surgeon should avoid performing long thoracic spine fusions in children before the age of 5 years [21, 36].

The surgical treatment of patients with congenital spinal deformities associated with multiple vertebral malformations is aimed to restore or improve the trunk balance, prevent the thoracic insufficiency syndrome and neurological disorders, as well as to maintain the length of the spine.

The ideology of surgical treatment is based on the principle of correction and stabilization at as early age as possible before the development of profound deformities. Commonly shared opinion states that the correction of already developed deformities in patients of older age is accompanied by a greater rate of complications and carries a poorer functional outcome [1, 4, 6, 33, 48]. This principle proved itself in the surgical treatment of children with isolated congenital vertebral malformations of the thoracic and lumbar spine [1].

The stabilization of deformity in situ or implementation of hemiepiphysiodesis/hemiarthrodesis at the apex of deformity in multiple congenital malformations of the spine carry a lower number of intraoperative complications. However, these procedures do not provide effective control over the condition of a developing spinal column in children,

leads to the development of a crankshaft phenomenon or a significant growth restriction of the spine and impaired development of the organs within the thoracic cage [18, 36, 37, 57]. This option uses implantation of a metal construct to restrict growth along the convex side of the spine and prevent further curve progression. A certain percent of gradual correction at the concave side can be expected over time with a normal growth plate on the concave side. This technique is indicated and has been most justified to treat early age patients with nonprofound congenital deformities, which are expected to progress along with a child's growth [37].

Different options of staged surgical procedures became most commonly used in treatment of pediatric patients.

In some cases, correction is performed via the resection of a malformed vertebra and stabilization using a metal spinal implant. The ideal case for this intervention is the deformity associated with isolated vertebral malformation [1], but the option can also be used in patients with multiple vertebral malformations contributing to the formation and development of the most significant deformity (Fig. 1).

The advantage of this technique is that it enables radical curve correction [1, 34]. Staged surgical procedures are conducted subsequently, generally when the course of the disease requires an intervention: in the presence of malformed vertebrae in the above- and lower-lying regions of the spine, the formation of compensatory curves, and the nature of the previous surgery. The inevitable consequence is the formation of bone block, which can halt spinal growth in length or cause a secondary deformity. Often, the only option reserved for staged surgery is the vertebral column resection (VCR), which is associated with a quite high rate of complications, particularly, neurological [39, 51, 54].

Many authors prefer to use hook fixators as anchoring points for the rods. Placement of these fixators is technically less-challenging especially in the presence of abnormal bony anatomy in children. In addition, an experimental

study showed that transpedicular screw fixation resulted in early closure of neurocentral synchondrosis area leading to narrowing of the spinal canal [23].

The use of growing metal rods is another option for surgery. Moe et al. [43] was first to describe this technique using Harrington instrumentation. The indications for using growing metal rods include cases of rather mobile deformities in the malformed region or implementation for the correction of compensatory curves. An advantage is control over spinal deformities preserving spinal growth due to rod insertion along the curve without opening the bone structure with fusionless surgery. Originally, a single distractor was installed on pre-augmented vertebral arches. However, dual growing rods with tandem connectors have become a popular treatment option. The anchoring constructs include transpedicular screws, laminar hooks, rib fixators and hook for transverse processes, and support on the iliac crest. The outcomes of the long-term follow-ups have shown that the length of the spine increases on average 1.8 cm per year with a dual growing rod technique [9]. In some cases, metal construct placement is performed in conjunction with the resection of malformed bone structures or an arthrodesis is performed at the apex of the deformity. However, based on the results of comparative studies, the group of patients who underwent only dual growing rod procedure had a larger increase in the spine length (1.5 cm per year), better curve correction and there were no cases of local kyphosis formation compared to the group of patients in whom implantation was supplemented with apical fusion [58]. Experimental studies showed that growing rods technique has no kyphogenic effect on anterior spinal column: there was no increase in the intradiscal pressure with using the growing rod technique [41]. The disadvantages of this method include the necessity for staged distraction to be performed approximately each 6 months, the high rate of metal construct destabilization, and a high rate of infectious and neurological complications. The total rate of compli-

cations was shown to be 72 % and each subsequent surgical lengthening raised the risk of complications by about 24 % [9, 11, 14, 49]. A noninvasive magnetically controlled growing rod technique has been devised to reduce the frequency of invasive distractions [10, 22]. At present, the greater efficacy of magnetically controlled growing rods compared to conventional implants has not been proved. This procedure exerts an even higher rate of metal-related problems, no effect of lengthening, and the occurrence of flat spine syndrome because of no opportunity to perform sagittal spinal profile correction [56].

The stability of any of the aforementioned metal constructs determines the choice of certain anchor, the number and combination pair of anchors for proximal and distal fixation. A significant stability of transpedicular screws has been shown in experimental studies; hook metal constructs are stronger in the lumbar versus thoracic spine [40]. A screw-screw pair was most stable in proximal and distal fixation. When it is impossible to use transpedicular anchors, the use of at least four rib-anchored hook fixators appears reasonable for proximal fixation [9, 60]. The outcomes of using various spinal rod connectors have been studied – with circular slots and V-groove slots: connectors with V-groove slots that mechanically lock the rod had 10 times fewer failures due to three-point fixation [38].

The issues that have not been fully resolved relate to the selection of the optimal age or the curve magnitude as when to start the treatment, necessity to perform definitive fusion, and stabilization of the deformity using conventional metal constructs as a separate stage of surgery. Most authors believe that curves with a Cobb angle greater than 60° in children at the age of 4–9 years is an indication for using the growing rod technique [45]. Comparison of the outcomes of treatment of patients has shown that termination of staged surgery without metal construct replacement is also associated with spontaneous fusion and retention of the achieved result of treatment [16, 26]. This theory is



Fig. 1

Resection of malformed vertebrae, correction and stabilization of deformity using spine implanted metal construct

indirectly supported by monitoring over the extent of performed staged surgeries over time: the first surgery resulted in the greatest correction effect and the amount of correction decreased gradually with each subsequent procedure and over time due to an increase in rigidity of the primary curve [50].

In case of rib malformations with unilateral rib synostosis, the placement of distraction metal constructs on the ribs of a patient is indicated to increase the volume of the hemithorax (Fig. 2).

Fused rib osteotomy is performed in most of these cases. The VEPTR instrumentation was the first proposed system and represents one of the frequently used constructs. VEPTR was first used by Campbell and Smith in 1989 for the treatment of a patient with thoracic insufficiency syndrome [17]. The treatment outcomes show that the correction of deformity, an increase in the spinal length at the deformed side by about 8.0 mm per year, improved lung func-

tion, an increase in the pulmonary perfusion volume in radioisotope studies, and lung tissue growth on CT findings can be achieved [4, 6, 25, 28, 35]. However, there are reports on a negative effect of the VEPTR technique on lung function due to a presence of metal implant and post-operative scarring restricting chest wall excursion [44]. These metal constructs have recently found a rising application in the treatment of children with infantile scoliosis of any etiology.

The choice of the age to perform a surgical procedure is very important, because the increased hemithorax volume in children who were older than 6 years of age resulted in only emphysematous lung tissue enlargement without significantly improved lung function versus younger children [44]. A limitation of this surgical procedure is the high rate of complications (up to 72 %), which are mainly related to impaired operative wound healing, infection, injuries to the pleura, anchoring construct migra-

tion, and injury to the brachial plexus [30, 49]. The rate and frequency of staged distractions have not been established: similar to growing rods, stage distractions are performed approximately each 6–9 months, before bone maturity is reached allowing definitive surgery.

Surgical procedures using Shilla instrumentation [42] and the Luque trolley technique [46] have not become popular. The procedure is based on insertion of spinal anchors that glide free to travel along the rods during spinal growth. The advantage is a lower number of repetitive surgery procedures and a lower rate of complications as shown in studies comparing the treatment outcomes using gliding instrumentation and the dual rod technique. However, although this procedure allows children stabilization of the deformity, there were an about 50 % lower rate of spinal growth in length and lesser opportunity for correction [42]. At present we have no sufficient number of patients and adequate monitoring periods to make a conclusion on the efficacy and safety of this procedure.

Spinal deformities associated with multiple malformations are characterized by severity and aggressiveness of natural course of the disease. The major cause of a poorer quality of life and a lower lifespan of these patients is the impaired development of the lung tissue necessitating surgical intervention already at an early age. Surgical treatment is associated with the high rate of complications, which can be reduced through strict compliance to the surgical procedure. The wide variety of available methods and metal constructs as well as the lack of studies comparing the outcomes when using different techniques leave open an issue related to the choice of tactics of treatment of these patients.

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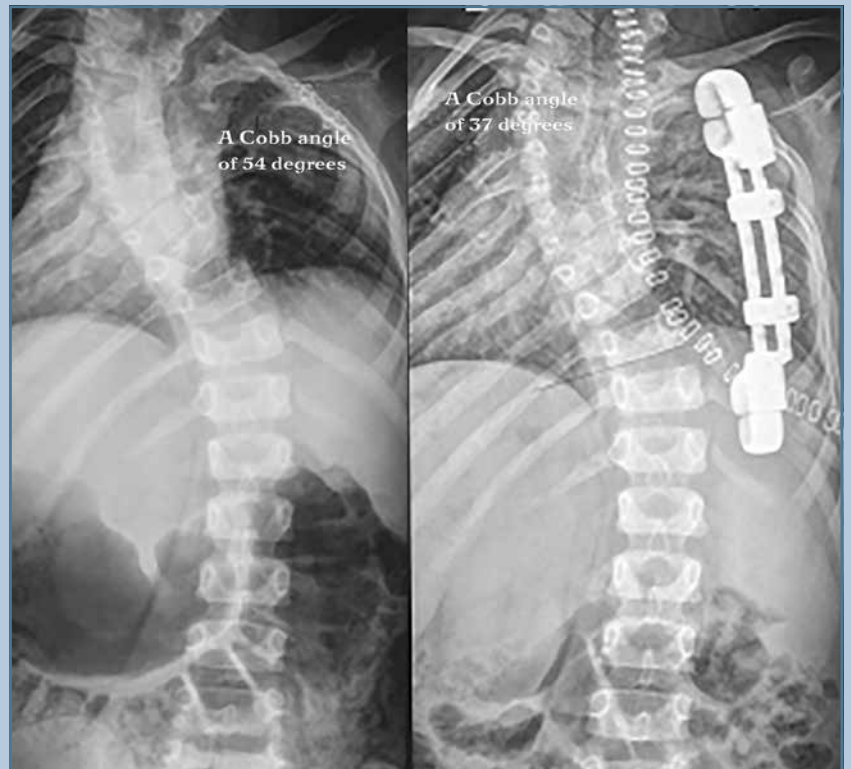


Fig. 2

Correction of spinal deformity associated with rib synostosis using rib-rib corrective metal structure

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