Analysis of the length of fixation during hemivertebra resection. A monocentric retrospective cohort

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Abstract

Background: We aimed at reviewing results of surgical correction of spinal deformity due to hemivertebra with regard to the age, severity of deformity and length of instrumentation.

Methods. Study comprised 117 patients with congenital spinal deformity treated between 2010 and 2018. Patients’ aged 1 to 18 years. Mean follow-up was 3 years (1 to 8). Posterior approach was used in all cases. Patients were subdivided into 4 groups.

Result:

Unilateral mono-segmental fixation (Group I) for 15 children with mean age of 48.3 months. Cobb angle for scoliosis and kyphosis were 31.1° ± 6.4° and 29.3° ± 11.9° corrected to 7.8° ± 6.8° and 4.7° ± 4.0° respectively. Blood loss was 213.6 mL. Operating time was 165 minutes.

Bilateral mono-segmental fixation (Group II) for 24 patients with mean age of 53.1 months. Cobb angle for scoliosis and kyphosis were 32.5° ± 8.6° and 30.9° ± 4.3° corrected to 5.3° ± 2.8° and 0.2° ± 11.6° respectively. Blood loss was 215.4 mL. Operating time was 160.5 minutes.

Bilateral three-segmental fixation (Group III) for 29 patients with mean age of 78.2 months. Cobb angle for scoliosis and kyphosis were 36.6° ± 10.6° and 37.6° ± 14.6° corrected to 6.2° ± 6.1° and 5.0° ± 5.2° respectively. Blood loss was 342.7 mL. Operating time was 197.0 minutes.

Bilateral poly-segmental fixation (Group IV) for 49 children with mean age of 112.7 months. Cobb angle for scoliosis and kyphosis were 40.2° ± 14.8° and 58.2° ± 35.6° corrected to 10.7° ± 9.6° and 10.7° ± 10.3° respectively. Blood loss was 549.3 mL. Operating time was 288.8 minutes.

Conclusion: Choice of spinal fixation technique and the length of fixation were determined on the basis of patient’s age, magnitude of the deformity and concomitant vertebral abnormalities.

Background

The first report of the removal of a portion of the spine for a fixed deformity was by N. Royle (1928). Initially hemivertebra excision was done via an anterior-only approach without metal construct.[2] Thereafter, the procedure was performed from two approaches without usage of implants and provided a good correction of scoliosis but later presented with local kyphotic deformity and progressive neurological deficit leading to paraplegia.[3,4] The complications could be avoided with application of metal constructs. M. Bergoni (1981) offered a one-stage hemivertebra excision via a combined anterior-posterior approach with the use of metal fixators.[5,6,7] Over time, with steady advances in instrumentation and metal constructs, the procedure could be done through a posterior approach.[8,9,10,11] and screw fixation systems could be used for younger children. Follow-up dispelled worries of several researchers and proved to be of value demonstrating pedicle growth potential and no spinal stenosis.[12,13,14,15] Choice of techniques and options of surgical treatment of congenital scoliosis resulting from hemivertebra remains a challenging issue. This is confirmed by stability in incidence rate and evolution of surgical correction techniques. There is paucity of data regarding optimal length of fixation in the literature and this moved the authors to retrospectively review own results.

Objective

We aimed at reviewing results of surgical correction of spinal deformity due to hemivertebra with regard to the type and length of instrumentation and show the correlation of the type and length of fixation with the age of children.

Design of the study

Retrospective cohort study with intracohort intergroup comparison; level of evidence: 4 done at Russian Ilizarov Scientific Center, Kurgan, Russian Federation. (Case series and poor quality cohort and case-control studies, UK Oxford, v.2009).[16]

Recruitment period: 2010-2018.

Inclusion criteria: Patients with congenital spinal deformity with a leading hemivertebra, not previously operated on up to the age of 18.

Exclusion criteria: Age over 18 years old, previously performed surgery to correct spinal deformity. Multiple congenital spinal deformities for more than 6 segments.

Methods

The study was based on the results of examination and treatment of 117 patients aged from 14 months to 18 years (mean age 83.7 ± 57.5 months) having different variants of hemivertebra and asymmetric types of vertebral fusion.

Surgical intervention was done via posterior approach in all cases and comprised hemivertebra resection from pedicular approach and instrumented fixation. Patients were subdivided into 4 groups depending on type and length of fixation (Fig. 1):

- Group I – unilateral mono-segmental fixation;
- Group II – bilateral mono-segmental fixation;
Types of vertebral malformations, defects of vertebra formation, and variants of segmentation are presented for each of the groups (Table 1)

Long-term follow-ups were available in all the cases from 12 months to 8 years.

Statistical analysis

Statistical analysis was performed using Statistical Package for Social Sciences (SPSS), version 22.0 (SPSS Inc., Chicago, IL, USA). All evaluation parameters had normal distribution. Independent t test was used for contrasting means in two study groups: scoliotic and kyphotic deformity before and after surgery, operation time, and blood loss. A p value < 0.05 was considered statistically significant.

Results

Unilateral monosegmental fixation (Group I, Fig. 2) was done for 15 children aged from 14 months to 10.5 years (Table 2) with the median age of 28 months. There were 11 females and 4 males. Two eggshell procedures of hemivertebra resection and 13 pedicular excisions were performed. Cobb angle for local scoliosis measured 21.1° to 45.4° preoperatively, 0° to 20.9° postoperatively, was corrected to 74.9% (p=0.0001) and ranged from 4.1° to 28.7° at a long-term follow-up. Cobb angle for local kyphosis measured 15.1° to 44.1° preoperatively, 0.4° to 10° postoperatively, was corrected to 84.0% (p=0.042) and ranged from 3.2° to 19.0° at a long-term follow-up. Intra-operative blood loss was 50 mL to 600 mL (p=0.002). Operating time ranged from 120 minutes to 250 minutes (p=0.0001).

Neurologic status was preoperatively classified as Frankel E level in all the patients. No intra- and postoperative neurologic complications were reported. Complications detected in 5 patients of the group (30%) included progression of the curve that required re-operation in three cases at 3-year follow-up with removal of residual hemivertebra (n=1), the crankshaft phenomenon (n=1), adding-on in lumbar spine (n=1) and progression of deformation outside the zone of instrumental fixation in the remaining patients (n=2).

Specific feature with the group included progressive curve extending outside instrumentation segments due to the second hemivertebra in case of multiple vertebral malformations (n=2).

The second surgical intervention comprised either hemivertebra excision or increased length of fixation in these patients.

Bilateral monosegmental fixation (Group II, Fig.3) was performed for 24 patients aged from 14 months to 13.8 years (Table 2) with the median of 35 months. There were 16 female and 8 male patients. An eggshell procedure of hemivertebra resection and 23 pedicular excisions were performed. Cobb angle for local scoliosis measured 14.8° to 51.9° preoperatively, 0° to 25.5° postoperatively, was corrected to 83.7% (p=0.0001) and ranged from 0° to 27.5° at a long-term follow-up (Table 2). Cobb angle for local kyphosis measured 27.8° to 35.8° preoperatively, -12.8° to 10° postoperatively, was corrected to 100.0% (p=0.077) and ranged from 0.9° to 3.2° at a long-term follow-up (Table 2). Intra-operative blood loss was 48 mL to 750 mL (p=0.0001). Operating time ranged from 85 minutes to 300 minutes (p=0.0001).

Neurologic status was preoperatively classified as Frankel E level in all the patients. No intra- and postoperative neurologic complications were reported. Complications were detected in 7 patients of the group (29%) and included necrosis of the wound edges (n=1), unstable screws (n=2, one with pedicle fracture) followed by revision surgery; broken screw at 2-year follow-up (n=1 with radicular syndrome) that entailed reassembly of metal construct; progressive curve at the site of instrumentation fixation that resulted in PJK (n=3) ; they had intervention which included bracing in one and re-operation in two.

Specific features with the group included progressive curve above instrumented fixation level due to the second hemivertebra in cases of multiple vertebral malformations (n=3) that was treated at the second stage by bracing, hemivertebra excision or increased length of fixation at one-to-three-year follow-up.

A screw appeared to cut through the bone intra-operatively being unstable (n=1) due to evidently dysplastic pedicle with fixation increased to one level and was thereafter stable at the fixation site (n=1).

Bilateral three-segmental fixation (Group III) (Fig.4) was performed for 29 patients aged 18 months to 17 years (Table 2) with the median age of 52 months. There were 15 female and 14 male patients. One eggshell procedure of hemivertebra resection, 19 pedicular excisions, 3 asymmetric PSO and 6 VCR type vertebrectomies were done. Cobb angle for local scoliosis measured 16.7° to 55.1° preoperatively, 0° to 23.6° postoperatively, was corrected to 83.1% (p=0.0001) and ranged from 0.2° to 28.3° at a long-term follow-up (Table 2). Cobb angle for local kyphosis measured 19.7° to 65.6° preoperatively, 0° to 20.9° postoperatively, was corrected to 86.7% (p=0.0001) and ranged from 1.1° to 11.4° at a long-term follow-up (Table 2). Intra-operative blood loss was 100 mL to 740 mL (p=0.0001). Operating time ranged from 125 minutes to 445 minutes (p=0.0001).

Two patients developed neurological deficit postoperatively:

1. , 4-year-and-5-month-old. Distal paraparesis without progression in postoperative period, Frankel type D.
2. , 4-year-and-4-month-old. Distal paraparesis, hypoesthesia of the skin of the legs. No progression was observed postoperatively, Frankel type D.
Complications observed in 5 cases of the group (17.2%) included mal-position of screws with radicular syndrome (n=1), the patient had re-operation and the mal-positioned screws were reinserted; damage of the dural sac with formation of cerebrospinal fluid cyst and distal paresis on the right leg (n=1) that improved at 6 months after surgery. Progressive deformity that required extension of the construct to one segment below the level of the initial construct at 4 years after previous surgery (n=1), progression of deformity (n=1) due to weight gain of more than 20 kg and non-compliance with recommendations leading to the migration of metal constructs, this required an installation of Dual Growing Rod systems, progressive curve was treated with bracing in one patient (n=1).

Specific features with the group included intra-operative instability with a screw cutting through C6 vertebra and being replaced with a hook.

Progressive curve resulted from a poor quality brace in another patient.

*Bilateral polysegmental fixation* (Group IV, Fig. 5, 6) was performed for 49 children aged 20 months to 18 years (Table 2) with the median of 108 months. There were 24 female and 25 male patients. Seven eggshell procedure of hemivertebra resections, 22 pedicular excisions, 12 asymmetric PSO and 10 VCR type vertebrectomies were done. Cobb angle for local scoliosis measured 19.2° to 79.0° preoperatively, 0.1° to 34.8° postoperatively, was corrected to 72.5% (p=0.0001) and ranged from 0.1° to 18.6° at a long-term follow-up. Loss of correction was 12.1%, P=0.21 (Table 2). Cobb angle for local kyphosis measured 25.0° to 161.1° preoperatively, 0° to 43.8° postoperatively, was corrected to 81.6% (p=0.0001) and ranged from 0° to 17.6° at a long-term follow-up (Table 2). Intra-operative blood loss was 50 mL to 1800 mL (p=0.0001). Operating time ranged from 115 minutes to 605 minutes (p=0.0001).

Several patients had pre-operative neurological deficiencies:

1. 6-year-and-3-month-old. Distal paraparesis without progression observed postoperatively (Frankel type C).
2. 8-year-old. Distal flaccid paraplegia. No progression seen postoperatively (Frankel type B).
3. 14-year-old. Distal paraparesis without progression observed postoperatively (Frankel type C).
4. 3-year-and-9-month-old. Distal flaccid paraparesis with underlying myelopathy and paresis aggravated postoperatively. A course of test electric stimulation and neurotropic therapy resulted in some neurological improvement with improved tolerance to physical exertion (Frankel type C).
5. 9-year-old. Distal paraplegia and pelvic organs dysfunction without postoperative improvement (Frankel type A).

Complications revealed in 8 patients of the group (16.3%) comprised progressive curve at the site of instrumentation (n=2) and PJK (n=1) that required re-operation at 2-3 years post-operatively and bracing (n=1). Five patients developed neurologic deficiency including postoperative monoparesis on the left hand without progression (n=1), persistent distal paraplegia (the patient died 5 years post-surgery) (n=1), distal paraparesis after surgery that completely resolved (n=1), left-sided distal paraparesis improved at 3 months (n=1), postoperative pleural effusion and distal paraparesis that resolved (n=1).

Metal construct reassembled at 2-year follow-up due to the fracture of both rods (n=1).

Specific features. One patient died 5 years after surgery, and the cause of death was unknown.

Design of the table is intended as an illustrative tool for comparison between the groups showing the difference in absolute values and statistical significance of the variables.

**Discussion**

Surgical correction is indicated in patients with congenital spinal deformities early before deformity progression and development of compensatory curves prior to the first growth spurt at the age of 1 to 5 years.[17,18,19] (The curve in Group IV was significantly greater than that in Groups I and II with P=0.003 and P=0.010). Volume of operative intervention is considered with regard to patient's age, pattern of underlying disorder, 'maturation' of bone structures, anatomy of malformation, abnormality of adjacent vertebrae, spread of major arch and structural properties of compensatory curves.

Unilateral monosegmental fixation is no longer used for children because it has not demonstrated significant advantages in the volume of deformity correction (no possibility of directional maneuver on the contralateral side of the curve), operating time and blood loss compared to those provided by other fixation techniques. The method has shown worse outcome at long-term follow-up as compared to those secured with other techniques (loss of correction was 4.6° on average).[20,21]

Radical pedicular hemivertebra excision within the range of endplates of adjacent vertebrae (PVCR) and bilateral monosegmental transpedicular fixation with acute deformity correction was shown to be effective among correction techniques employed for preschool children aged from 1 to 3 years with single, two or more hemivertebrae located between two segments in addition to accompanying spine pathology that requires no surgical correction, with local major curve without structural compensatory curves.[22] The practice provides comparable results of deformity correction of 27.2° of baseline curve, significant reduction in blood loss (215.4 mL) in comparison to group III (342.7 mL P=0.018) and group IV (549.3 mL P=0.000), minimal operating time of 160.5 minutes in comparison to group III (200.6 min. P=0.077) and group IV (288.8 mL P=0.000) with good long-term follow-up (loss of correction 2.2°). A staged surgical procedure can be performed in two or more hemivertebrae with an interval of 3 to 6 months between the stages.

Congenital kyphosis or kyphoscoliosis in early and middle childhood (6-7 to 10-12 years old) is characterized by structural changes in the apical vertebrae with decreased anterior-posterior size, increased wedging at the apex of the curve and agenesis or hypokinetic vertebral arches.[23,24] Surgical treatment is indicated
for the patients including radical hemivertebra excision within the range of endplates of adjacent vertebral arches combined with posterior bilateral pedicular fixation two segments cranial and caudal to the apex of the curve and acute correction of the physiological curves (three segmental fixation). The method provided excellent results of deformity correction with comparable blood loss and operating time, good long-term follow-up (loss of correction for scoliosis 0.6° and for kyphosis 2.1°). More aggressive technique is indicated for severe deformity of adjacent segments employing vertebrectomy and resection of adjacent segments PSO or VCR types (Schwab grade 4 and 5) and repair of defect of anterior column with mesh implant that constitutes the supporting point for reclination maneuver during deformity correction. Radical excision of the leading anomaly or multiple anomalies and polysegmental transpedicular screw fixation is practical for middle childhood (7 to 11 years old) and adolescence (12 to 17-18 years old) with extensive deformity and structural compensatory curves but the procedure is associated with greater blood loss and operating time.

The strength of this article is that it is a large case series and allow for comparison of the four different groups whereas the weakness is that it is a retrospective study, the different types of surgeries done were not well stratified and the surgeries were done by different surgeons with varied experience.

**Conclusion**

The choice of the fixation type and length is determined by the patient's age, the magnitude of the deformity, and accompanying anomalies in the development of the vertebrae.

Monosegmental bilateral fixation with extirpation of an abnormal vertebra can be used in children under 3 years of age. Monosegmental unilateral fixation is not justified because of the comparable blood loss and the timing of the operation; the scoliotic arch clearly progresses in the long-term period.

At the age of 3-7 years, with a kyphotic component and an aggressive course of scoliotic deformity, the use of volumetric vertebrectomy and bilateral three-segment fixation for deformity correction is justified, but it increases blood loss and the duration of the operation.

The use of bilateral polysegmental fixation is effective in children over 7 years of age with extensive deformities. However, this increases blood loss and surgery time. The data are not final and require further monitoring until the end of the growth period.

**Abbreviations**

UK - United Kingdom

SPSS - Statistical Package for Social Sciences

USA - United States of America

IL - Illinois

p value - calculated probability

ml - Millilitre

% - Percentage

n - Number

PJK - Proximal junctional kyphosis

Kg - Kilogram

C6 - Cervical vertebra 6

VCR - Vertebra Column Resection

PSO - Pedicle Subtraction Osteotomy

♀ - Female

♂ - Male

PVCR - Posterior Vertebral Column Resection.

**Declarations**

**Compliance with ethical standards/Conflict of interest:** The authors declare that they have no conflict of interest.

**Ethics approval and Consent to participate** - The study was approved by the local ethics committee of the Ilizarov Scientific Center, Kurgan, Russian Federation and consent to participate obtained from the parent/guardian of patients less than 16 years. The ethics committee approval attached as file in the submission.
Consent for publication - A copy of the consent is attached as file.

Availability of data and materials - The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing interest - The authors declare that they have no competing interests.

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Authors' contribution -

RSO: conception and design; data acquisition; analysis of data; drafting of manuscript; administrative support; supervision.

FEY: data acquisition, analysis of data, drafting of manuscript

MJO: drafting of manuscript; critical revision

SDM: data acquisition; administrative support

GAB: data acquisition; administrative support; supervision.

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**Tables**

**Table 1**: Distribution of vertebral malformations by types of disorders

| Group | Type of vertebral malformation | Lateral hemivertebra | Posterolateral hemivertebra | Posterior hemivertebra | Body agenesis - segmented spinal dysgenesis | Butterfly vertebra | Fully segmented hemivertebra | Partially segment hemivertebra |
|-------|--------------------------------|----------------------|-----------------------------|------------------------|---------------------------------------------|-------------------|----------------------------|--------------------------------|
| I     | 9                              | 6                    |                             |                        |                                             | 4                 | 3                          |                                 |
| II    | 18                             | 5                    |                             |                        |                                             | 1                 | 16                         | 6                              |
| III   | 16                             | 5                    | 3                           | 4                      | 1                                           | 16                | 8                          |                                 |
| IV    | 31                             | 10                   | 6                           | 4                      | 5                                           | 22                | 12                         |                                 |
| Total | 74                             | 24                   | 9                           | 8                      | 7                                           | 58                | 29                         |                                 |

**Table 2**: Evaluation Criteria and Intergroup comparison data
|                      | 1                  | group 1 vs 2 | group 1 vs 3 | group 1 vs 4 | 2                  | group 2 vs 3 | group 2 vs 4 | 3                  | group 3 vs 4 | 4 Total |
|----------------------|--------------------|--------------|--------------|--------------|--------------------|--------------|--------------|--------------------|--------------|---------|
| Age M ± m (months)   | 48.3 ± 38.7        | 0.708 ± 0.055 | 0.000 ± 0.000 | 0.076 ± 0.048 | 78.2 ± 61.1        | 0.012 ± 0.054 | 0.000 ± 0.000 | 0.837 ± 54.4       | 112.7 ± 57.6  |
| Pre-op Cobb angle for scoliosis M ± m (degrees) | 31.1 ± 6.4        | 0.604 ± 0.057 | 0.003 ± 0.015 | 0.140 ± 0.010 | 36.6 ± 10.6        | 0.307 ± 0.148 | 0.010 ± 0.010 | 0.402 ± 11.9       | 36.1 ± 11.9  |
| Cobb angle for scoliosis correction M ± m (degrees) | 23.3 ± 6.1        | 0.154 ± 0.014 | 0.015 ± 0.026 | 0.266 ± 0.397 | 30.4 ± 10.0        | 0.759 ± 11.5  | 0.010 ± 0.010 | 0.475 ± 10.3       | 28.2 ± 10.3  |
| Cobb angle for post-op scoliosis M ± m (degrees) | 7.8 ± 6.8         | 0.235 ± 0.446 | 0.291 ± 0.613 | 0.008 ± 0.028 | 6.2 ± 6.1          | 0.028 ± 10.7  | 0.008 ± 0.028 | 0.107 ± 9.6        | 7.9 ± 8.6    |
| Cobb angle for loss of scoliosis correction M ± m (degrees) | 4.6 ± 8.7         | 0.035 ± 0.009 | 0.075 ± 0.600 | 0.612 ± 5.7   | 0.291 ± 8.8        | 0.018 ± 35.6  | 0.010 ± 0.010 | 0.582 ± 29.7       | 47.4 ± 29.7  |
| Cobb angle for residual scoliosis M ± m (degrees) | 14 ± 7.8          | 0.035 ± 0.009 | 0.075 ± 0.600 | 0.612 ± 5.7   | 0.291 ± 8.8        | 0.018 ± 35.6  | 0.010 ± 0.010 | 0.582 ± 29.7       | 47.4 ± 29.7  |
| Cobb angle for pre-op kyphosis M ± m (degrees) | 29.3 ± 11.9       | 0.835 ± 0.311 | 0.124 ± 0.449 | 0.204 ± 37.6  | 0.600 ± 31.5       | 0.060 ± 31.5  | 0.014 ± 0.028 | 0.592 ± 26.2       | 47.9 ± 26.2  |
| Cobb angle for kyphosis correction M ± m (degrees) | 24.6 ± 14.4       | 0.594 ± 0.367 | 0.169 ± 0.877 | 0.388 ± 32.7  | 0.600 ± 47.5       | 0.060 ± 10.3  | 0.010 ± 0.010 | 0.77 ± 9.1         | 39.8 ± 9.1   |
| Cobb angle for post-op kyphosis M ± m (degrees) | 4.7 ± 4           | 0.457 ± 0.913 | 0.262 ± 0.223 | 0.098 ± 5.2   | 0.223 ± 10.7       | 0.060 ± 0.029 | 0.010 ± 0.010 | 0.107 ± 9.1        | 7.7 ± 9.1    |
| Cobb angle for residual kyphosis M ± m (degrees) | 8.4 ± 9.1         | 0.418 ± 0.739 | 0.933 ± 0.562 | 0.439 ± 6.4   | 0.518 ± 8.1        | 0.058 ± 14.8  | 0.010 ± 0.010 | 0.74 ± 5.6         | 1.4 ± 5.6    |
| Cobb angle for loss of kyphosis correction M ± m (degrees) | 3.7 ± 4.9         | 0.16 ± 0.6    | 0.26 ± 1.6    | 0.36 ± 1.7    | 0.58 ± 1.4         | 0.1 ± 3.2     | 0.010 ± 0.010 | 1.4 ± 3.8          |