

Clinical and radiological outcomes of hemivertebra resection for congenital scoliosis in children under age 10 years

More than 5-year follow-up

Jae Hyuk Yang, MD, PhD^a, Dong-Gune Chang, MD, PhD^{b,*}, Seung Woo Suh, MD, PhD^a, Woon Kim, MD^a, Jewel Park^c

Abstract

The surgical treatment of congenital scoliosis under 10 years is difficult as it involves resecting vertebrae. Moreover, patient follow-up after surgery is extremely important because the patient is a child whose growth has not been completed. However, there are very few long-term follow-up studies after surgical treatment of congenital scoliosis. Therefore, this study is designed to evaluate surgical outcomes after posterior hemivertebra resection in children under 10 years and its long-term effects.

Twenty-one patients with congenital scoliosis who were younger than 10 years at the time of the surgery and received posterior hemivertebra resection and fusion using pedicle screw fixation were included in this study.

There were significant improvements in the main curve, coronal balance, sagittal vertical axis, thoracic kyphosis, and lumbar lordosis after surgery ($P < .001$, .021, .047, .043, .006, respectively). Coronal balance, sagittal vertical axis, thoracic kyphosis, and lumbar lordosis remained within the normal range during the follow-up period; however, main curve deteriorated significantly ($P = .005$). Trunk appearance of perception scale improved significantly after surgery ($P = .031$) and was maintained during the follow-up period ($P = .078$).

In conclusion, posterior hemivertebra resection and fusion using pedicle screw fixation in patients under 10 years old with congenital scoliosis is a safe and effective procedure that can achieve rigid fixation and deformity correction. Complete resection of the hemivertebra is important for deformity correction and prevention of curve progression, and careful long-term follow-up is necessary.

Abbreviations: CB = coronal balance, EBL = estimated blood loss, LL = lumbar lordosis, TAPS = trunk appearance perception scale, TK = thoracic kyphosis, SVA = sagittal vertical axis.

Keywords: spine, congenital scoliosis, hemivertebra, surgical treatment, long-term outcomes

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^aDepartment of Orthopaedic Surgery, Korea University Guro Hospital, College of Medicine, Korea University, ^bDepartment of Orthopaedic Surgery, Sanggye Paik Hospital, College of Medicine, Inje University, ^cCollege of Medicine, Korea University, Seoul, Korea.

*Correspondence: Dong-Gune Chang, Department of Orthopaedic Surgery, Sanggye Paik Hospital, 1342 Dongil-ro, Nowon-gu, Seoul, 01757, Korea (e-mail: dgchangmd@gmail.com).

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1. Introduction

Hemivertebra is the most common cause of congenital scoliosis. Since a segmented hemivertebra has the same growth potential as a normal vertebrae, it can lead to substantial spinal deformity due to unbalanced spinal growth if left untreated.^[1,2] The progression of congenital scoliosis depends on the age, type, shape, the number of hemivertebrae, and the location at which it occurs.^[1,2] If hemivertebra is not treated properly at the appropriate time, spinal deformity could become severe; also, internal organ function may deteriorate due to anatomically unbalanced growth of hemivertebra and structural differentiation of surrounding spine.^[3,4] Thus, hemivertebra should be treated before the deformity progresses.

For treatment of congenital scoliosis with hemivertebra, early surgical intervention in young children is increasingly being preferred since it prevents the development of local deformities and secondary structural curves, allowing unaffected parts of the spine to achieve normal growth.^[3,5–8] Many authors have described the surgical treatment of hemivertebra, including the first ever report by Royle in 1928, 2-stage anterior-posterior procedure for hemivertebra resection by Leatherman et al,^[9] combined anterior and posterior approach performed in a single stage procedure by Bollini et al,^[5] and resection of hemivertebra using a posterior-only approach with transpedicular instrumentation in very young patients by Ruf and Harms.^[3,4] Recently, this surgical technique of Ruf and Harms has become more popular.

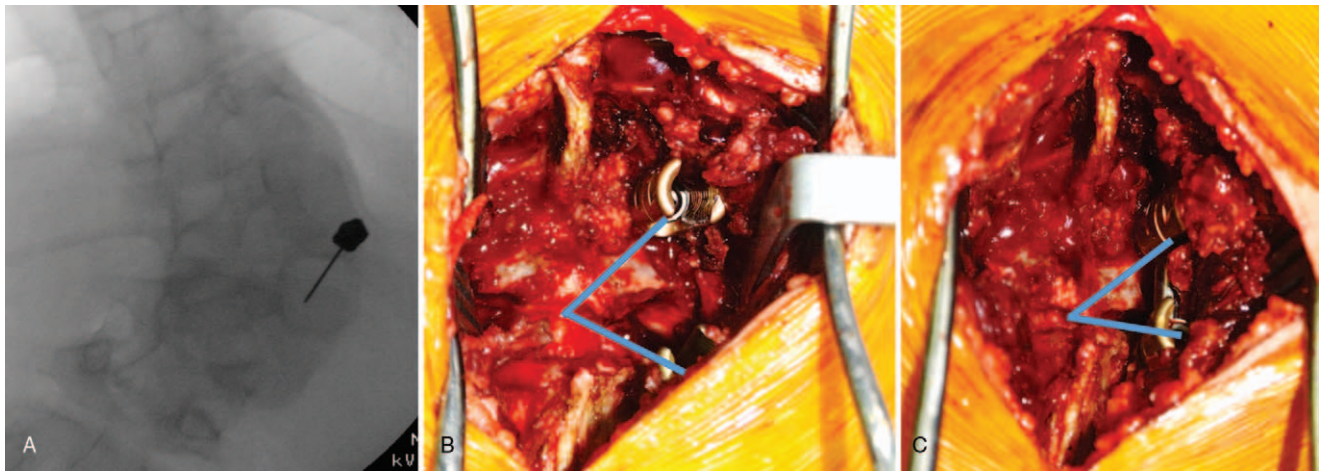


Figure 1. Surgical correction procedure of hemivertebra. A skin incision is performed and the hemi-vertebrae requiring resection is identified under the image intensifier (A). Pedicle screws are inserted into the upper and lower vertebrae of the hemivertebra, and the lamina is incised using a motorized burr. After exposure and protection of the dura, the hemivertebra is resected using a motorized burr and bone curette (yellow dotted arrow) (B). After making a construct between the pre-contoured rod and the pedicle screw, correction of the spinal deformation was done by compress pedicle screws on the side where the hemivertebra was resected while distraction of the opposite pedicle screw (compression amount was identified with blue line) (C).

However, only few reports have described hemivertebra resection through a posterior approach using pedicle screw fixation for early surgical intervention.^[6,7] Moreover, considering the fact that surgery is performed on a young child, it is important to verify the long-term surgical outcome; almost no previous studies have explored this aspect.

For these reasons, this study was designed to evaluate the clinical and radiological outcomes after posterior hemivertebra resection and its long-term effects on deformity correction in children under 10 years old with congenital scoliosis.

2. Materials and Methods

2.1. Study Patients

From 2006 to 2012, 67 patients underwent surgical correction of congenital scoliosis with hemivertebra, and 21 of these patients were under 10 years of age (12 boys and 9 girls) and had been followed up for a minimum of 5 years. We retrospectively reviewed their records to collect data pertaining to deformity correction and its long-term effects on the growing spine. Institutional Review Board approval was obtained before collecting and analyzing data. All patients underwent posterior hemivertebra resection and fusion using pedicle screw fixation. The inclusion criteria were:

- (1) diagnosis of congenital scoliosis due to hemivertebra requiring surgical treatment (curve magnitude greater than 25 degrees with rapid progression, and documented curve progression greater than 5 degrees in 6 months and/or failure of conservative treatment);
- (2) age at surgery below 10;
- (3) a minimum of 5-year follow-up.^[7,8]

Patients who underwent surgery through the anterior approach, had revision surgery, or had scoliosis of other etiologies were not included in this study.

2.2. Surgical Procedures

All patients were surgically corrected with posterior hemivertebra resection and fusion using pedicle screw fixation.^[8] Intraoperative neurophysiological monitoring, including somatosensory and motor evoked potentials, was used to identify and prevent injury to neurovascular structures during surgery. Under general anesthesia, patients were positioned prone on a radiolucent operation table. Pedicles of the hemivertebrae and adjacent vertebrae were identified using needles and intraoperative fluoroscopy (Fig. 1A). Pedicle screws were inserted into the vertebral bodies above and below the level of the hemivertebra. The posterior elements of the hemivertebra, including the upper and lower facets, the lamina, and the transverse process, were removed so that the pedicle and the nerve roots above and below the hemivertebra were exposed. The pedicles, the vertebral body, and the upper and lower disks were exposed and removed until a bleeding bone was visualized in the osteotomies. Additionally, in the thoracic spine, the rib heads and the proximal part of the surplus rib on the convex side were exposed and resected. After applying compression to close the osteotomy gap, caution was taken to ensure that the exiting nerve roots and the dura were not impinged (Fig. 1B and 1C). Autogenous bone from the hemivertebra was used for posterolateral fusion, and negative pressure drainage was retained.^[10,11]

2.3. Postoperative Management

Patients were mobilized at 1 week with a thoraco-lumbo-sacral orthosis for 3 to 6 months.

2.4. Radiographic Measurements

Whole-spine standing anteroposterior and lateral radiographs were reviewed to assess deformity correction and spinal balance. The Trunk Appearance Perception Scale (TAPS) was used to measure clinical satisfaction.^[12]

The magnitude of the curve was measured for curve parameters. Coronal balance (CB), sagittal vertical axis (SVA),

Table 1
Demographic and operative data†.

Demographic/Operative parameter	Mean (range)
Age (yr) at the time of surgery	4.7±2.0
Follow-up (yr)	8.3±2.2
Fused segments (n)	2.5
Operative time (min)	200±60
Estimated blood loss (mL)	686±419

†Data represents the mean values for each group.

thoracic kyphosis (TK), and lumbar lordosis (LL) were measured as balance parameters.^[6] The magnitude of the main curve was measured by the Cobb method using the end vertebra based on standing radiographs taken before surgery. CB was measured as the deviation of the C7 plumb line from the center sacral vertical line. Deviation was considered significant when it exceeded 20 mm. SVA was measured as the distance from the C7 plumb line to a perpendicular line drawn from the posterosuperior corner of the sacrum and designated as positive (+) when the C7 plumb line was anterior to the posterosuperior corner of the sacrum and negative (-) when the C7 plumb line was posterior to the posterosuperior corner of the sacrum. A deviation greater than 20 mm was considered as a decompensation. LL was measured from the upper plate of T12 to the upper end plate of the sacrum. TK was measured from the upper end plate of T5 to the lower end plate of T12.

Perceived body image is important in assessing health-related quality of life in scoliosis patients^[13,14]; thus, we used TAPS, which includes self-image scales, to evaluate these patients. This includes three sets of figures that depict the trunk from 3 viewpoints: looking toward the back, looking toward the head with the patient bending over (Adam’s forward bending test), and looking toward the front. For this last viewpoint, men and women receive different drawings. Each drawing receives a score from 1 (greatest deformity) to 5 (smallest deformity) and a mean score is calculated by adding the scores for the 3 drawings and dividing that number by three.^[12] Among the drawings presented for each item, patients choose the drawing that most closely matches their perception of their body appearance.

All radiological and clinical assessment data were reviewed before the operation, immediately after the operation, and at 1, 3, 5 year and final follow-up, and are presented as means ± standard deviations. In order to minimize measurement error due to variation between observers, all radiographs were measured by

Table 2
Location of hemivertebra.

Location	Patients (n)
Thoracic	4
Thoracolumbar	10
Lumbar	7

2 authors who did not participate in the operation. The means of the measurements were used for analysis.

2.5. Statistical Analysis

For statistical analysis, SPSS 20.0 (IBM, Armonk, NY) was used. All factors for coronal and sagittal balance of spine were statistically analyzed using the paired Student *t*-test. *P*-values under .05 were considered as statistically significant.

3. Results

3.1. Deformity Correction

All patients were under 10 years of age at the time of surgery with a mean age of 4.7±2 years, and the mean follow-up period was 8.3±2.2 years (Table 1). Resected hemivertebrae were thoracic in 4 patients, thoracolumbar in 10 patients, and lumbar in 7 patients (Table 2).

The preoperative main curve Cobb angle of 32.7° improved to 10.3° postoperatively and was 15.4° at the last follow-up (Table 3) (Fig. 2) (Fig. 3A, 3B, and 3C). The correction rate was 68.5% postoperatively and 52.9% at the last follow-up compared with the preoperative value. There was a loss of correction of 15.6% during the follow-up period, indicating significant deterioration (*P* = .005) (Fig. 2) (Fig. 4A, 4B, and 4C).

3.2. Coronal and Sagittal Balance (SVA)

The preoperative CB of 10.4 mm improved to 2.2 mm postoperatively and was 9.7 mm at the last follow-up. The preoperative SVA of 6.7 mm increased to 24.9 mm postoperatively and was 7.8 mm at the last follow-up (Table 3). CB and SVA changed significantly after surgery. Compared to SVA, CB was less influenced by surgery. SVA was maintained during the follow-up period (*P* = .070), but CB deteriorated during the follow-up period (*P* = .028). However, both CB and SVA remained within normal ranges.

Table 3
Radiological and clinical assessment.

Radiological and clinical assessment	Preoperative	Postoperative				
		Immediate (<i>P</i> value)	1 yr	3 yr	5 yr	Final follow up (<i>P</i> value)
Main curve (Cobbs angle) (°)	32.7±6.7	10.3±7.2* (.001)	11.7±7.0	12.9±7.4	13.5±6.8	15.4±8.1** (.005)
Coronal balance (mm)	10.4±28.5	2.2±11.6* (.021)	2.6±17.6	8.0±10.0	8.6±13.4	9.7±18.2** (.028)
Sagittal vertical axis (mm)	6.7±27.6	24.9±26.2* (.047)	16.7±32.8	12.4±38.3	11.4±1.9	7.8±49.0 (.070)
Thoracic kyphosis (°)	33.0±13.0	24.3±13.9* (.043)	29.7±15.6	30.5±22.9	28.8±9.9	29.3±22.8 (.180)
Lumbar lordosis (°)	33.9±17.2	27.5±12.6* (.006)	37.2±15.6	38.2±19.6	39.2±7.0	41.8±19.9** (.002)
Trunk appearance perception scale	3.4±0.5	4.4±0.6* (.031)	4.5±0.8	4.2±0.7	4.1±0.7	4.0±0.5 (.078)

* Paired *t*-test between preoperative and immediate postoperative.
 ** Paired *t*-test between immediate postoperative and final follow-up.
 Significant differences were considered for *P* value less than .05

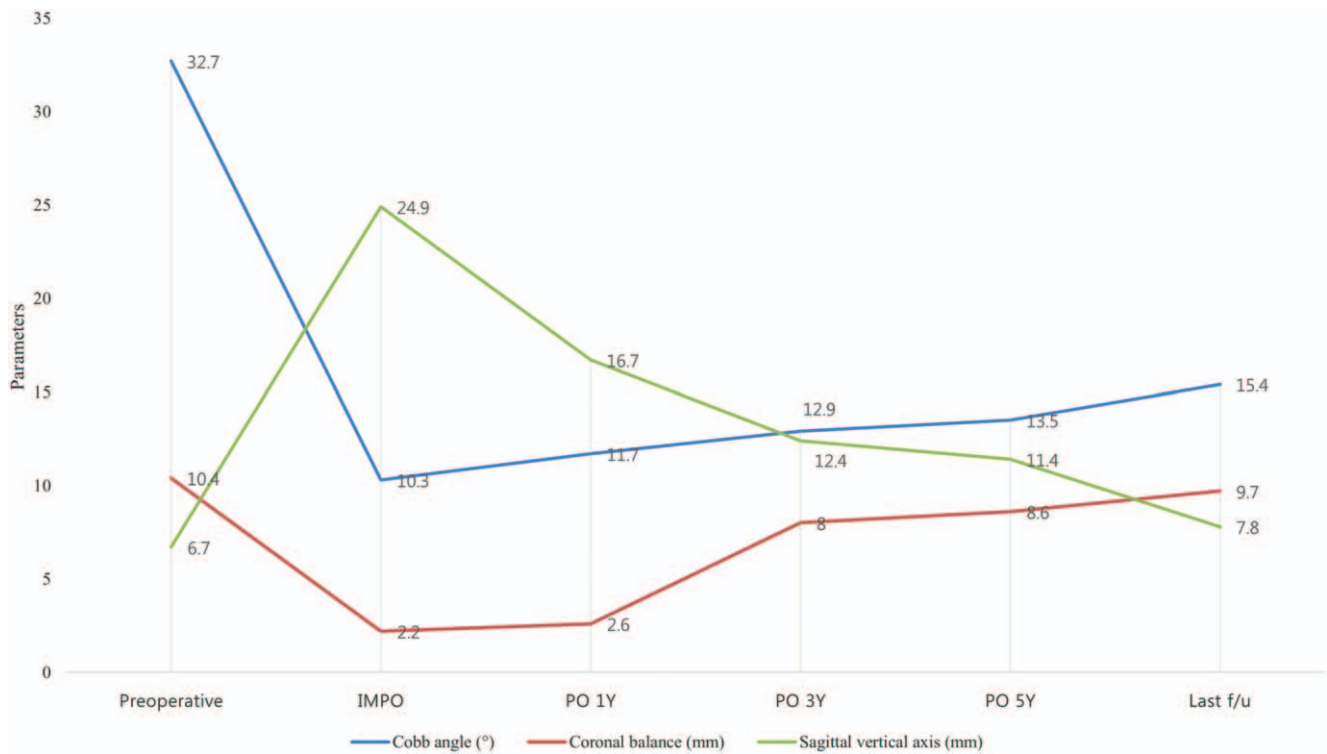


Figure 2. Pattern of changes in Cobb angle, coronal balance and sagittal vertical axis (SVA) during follow-up of patients. The patient's preoperative, immediate postoperative, postoperative 1, 3, 5 years and final follow-up results were analyzed graphically. Cobb angle was found to be statistically corrected immediately after surgery. The Cobb's angle deteriorates continuously depending on the observation period ($P = .005$). Coronal balance and SVA are in the normal or preoperative category, gradually changing to preoperatively as compared to immediately after surgery depending on the duration of follow-up observation.

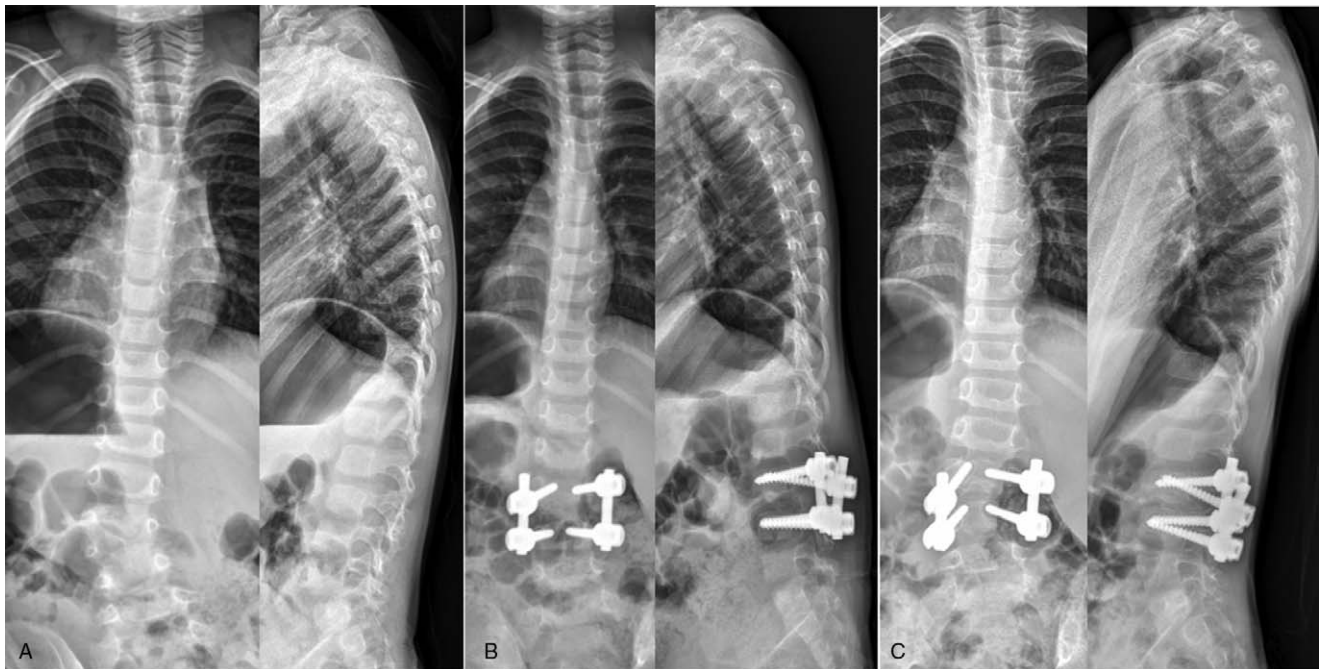


Figure 3. Case of congenital scoliosis with L3 hemivertebra. A 4-year-old male who had congenital L3 hemivertebra with a 30° left lumbar main curve. The L3 hemivertebra was resected using the posterior-only approach and pedicle screws were inserted at L3 and L4 (A). Immediate postoperative radiographs showed that the main curve improved to 12° with satisfactory deformity correction (B). After 7 years of follow-up, radiographs showed that the main curve was maintained at 12° without curve progression (C).

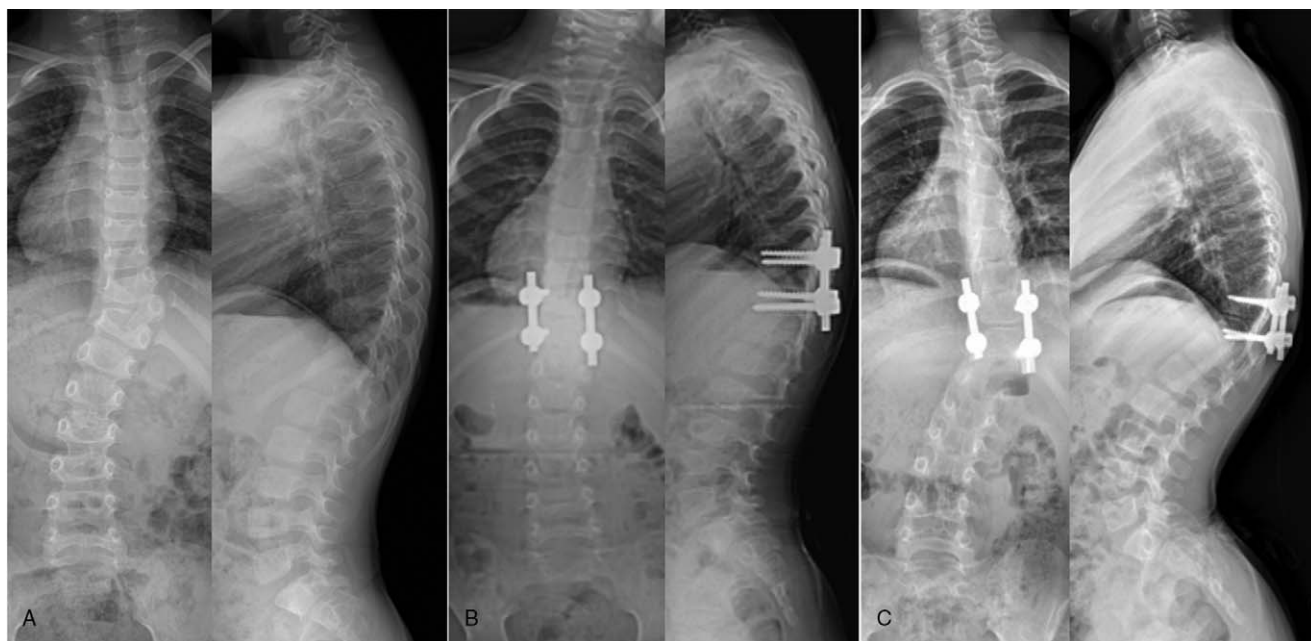


Figure 4. Case of congenital scoliosis with T10 hemivertebra. A 4-year-old female who had congenital T10 hemivertebra with a 27° right thoracic main curve. The T10 hemivertebra was resected using the posterior-only approach and pedicle screws were inserted at T9 and T11 (A). Immediate postoperative radiographs showed that the main curve improved to 4° (B). After three years of follow-up, radiographs showed that the corrected coronal balance deteriorated with a loss of correction (change in Cobb's angle from 4° to 17°) (C).

3.3. Sagittal Plane (TK and LL)

The mean value of TK was 33° before surgery, 24.3° after surgery, and 29.3° at the last follow-up, and mean value of LL was 33.9° before surgery, 27.5° after surgery and 41.8° at the last follow-up (Table 3). TK improved after surgery ($P=.043$) and was maintained postoperatively with no significant changes during the follow-up period ($P=.180$). LL improved after surgery ($P=.006$) and changed during the follow-up period ($P=.002$), but remained within the normal range.

3.4. Operative Data

Operative data is reported in Table 1. The mean operative time was 200 ± 60 min and the estimated blood loss was 686 ± 419 mL.

3.5. Complications

There were 5 cases of complications in 5 patients (Table 4). The overall incidence of complications was 23.8%. There was 1 case of postoperative transient neurologic deficit that resolved within 3 months and 1 case of superficial wound infection that was managed by incision and drainage. There were 2 cases of late postoperative complications, which were both add-on deformities with progression of the curve. Of these 2 patients, 1 was treated with a brace and the other was treated with revision surgery. There were no cases of crankshaft phenomenon at the most recent follow-up.

4. Discussion

Congenital scoliosis, which is seen in about 1 in 1000 births, is the most common congenital spinal disorder, followed by congenital kyphosis and lordosis.^[15-17] Congenital scoliosis occurs due to

the damage caused to the fetus between the fifth and eighth week of gestation. Thus, it is often associated with congenital heart disease (10%-15%), spinal cord dysraphism, and congenital kidney disorders.^[17,18] The severe restriction of pulmonary function in patients with large curves suggests that congenital scoliosis may coexist with hypoplastic lung development.^[18]

Whether a case of congenital scoliosis is indicated for surgery depends on the degree of scoliosis at the time of diagnosis and the expected progression. Segmented hemivertebrae have normal growth plates, which worsens the deformity with further growth.^[3,4] Secondary curves will develop to equilibrate the trunk. The aim of surgery in congenital scoliosis is to achieve a balanced spine with a physiologic sagittal profile using the shortest possible fusion segment.^[19] Delayed treatment requires

Table 4
Complications.

	Patients (n)
Early complications	
Transient neurologic deficit	1
Dura tear	1
Hematoma	0
Wound infection, superficial	1
Wound infection, deep	0
Screw malposition	0
Major vascular injury	0
Late complications	
Permanent neurologic deficit	0
Metal failure or screw pull-out	0
Curve progression or adding-on	2
Wound infection	0
Crankshaft phenomenon	0

long fusion segments, as secondary structural curves would have to be included, and correcting these rigid curves is not only more challenging but also accompanied by a higher risk of neurologic impairment.^[19] As the spinal canal is almost completely formed in the first 2 years of life, inserting screws into the growing pedicles does not cause the posterior part of the vertebral canal to narrow and only causes partial impairment in the vertebral somatic area.^[20–22] Regardless, some authors have expressed concerns over spinal canal growth impairment in young children whose pedicles are inserted with screws.^[23]

Posterior hemivertebra resection with transpedicular instrumentation was first reported in 1991 by Jürgen Harms. In comparison to 1-stage or 2-stage AP hemivertebra resection, posterior hemivertebra resection has many advantages, including good correction results on the coronal and sagittal planes, reduced invasiveness, lower complication rate, and shorter postoperative recovery period. These advantages make it more favorable for young children.^[24]

Main curve correction rates in our study were similar to those in previous studies. Our main curve correction rate was 68.5% after surgery and 52.9% at the last follow-up visit; Ruf and Harms^[3,4] reported short-term corrections of 66% and 72%, Chang et al^[25] reported 75% after surgery and 62.5% at the last follow-up, and Aydogan et al^[26] reported 75% with shorter instrumentation and fusions. However, our study shows a 15.6% loss of main curve correction during the follow-up period, indicating significant deterioration. The cause of curve progression may be attributed to incomplete hemivertebra resection with insufficient reduction of the deformity.^[7,8,10,25] In treating congenital scoliosis due to hemivertebra, the hemivertebra should be completely removed along with the adjacent disc. If this procedure is overlooked, the remnant hemivertebra may grow and cause curve progression.^[25]

Regarding SVA and CB, SVA was maintained at the last follow-up visit, but CB deteriorated during the follow-up period. This suggests that while CB may deteriorate during growth, SVA could remain within normal limits for a child or adolescent. Nevertheless, the spine was balanced in most of the patients at the last follow-up. As for sagittal parameters, both TK and LL were improved after surgery. TK was maintained during the follow-up period; however, LL improved to the normal range during the follow-up period. The values of TK and LL were less influenced by hemivertebra resection because the adjacent segments were compensated for.^[3,4,25,27]

Cosmesis in congenital scoliosis is an important factor in patients' satisfaction with surgical outcomes. Perceived body image plays an important role in assessing health-related quality of life in scoliosis patients^[14]; for this reason, self-image scales were included in the specific instruments used to evaluate these patients.^[13] We used TAPS, described by Bago et al^[12], for our clinical assessment. The criteria used in TAPS have several advantages over the Walter-Reed Visual Assessment Scale, which was described by Sanders et al.^[28] The former includes a more realistic illustration of body image, has a shorter format, and uses three images instead of 2 figures in Walter-Reed Visual Assessment Scale that correspond to the views of the trunk from the back and in the axial plane. Incorporation of the frontal view in TAPS is important because this corresponds to what patients see in the mirror, and is thus a more realistic perception of one's body.^[12]

As such, TAPS tries to measure trunk deformity from the patient's perspective. Objective methods, such as the Quantec system, are based on optoelectronic technology and, and are

hampered by the high cost and the requirement of both the skill of the examiner and accurate positioning of patients for reliable measurements.^[29] Bago et al^[12] also noticed a correlation between TAPS and the Cobb angle ($r = -0.55$), and their results are similar to the findings of Theologis et al.^[30] They also studied clinical photographs (posterior, lateral, and forward bending) on a scale of 1 to 10 and concluded that the correlation coefficient of 0.46 between their cosmetic score and the Cobb angle. Donaldson et al^[31] reported a clinical correlation coefficient of 0.53 with the Cobb angle. In our study, TAPS scores at 1, 3, and 5 years and at final the follow-up post-operatively were 4.5, 4.1, 4.1, and 4.0 respectively; it gradually deteriorated at the final follow-up visit ($P < .005$).

When the posterior column is fused, the anterior column grows continuously, which results in the crankshaft phenomenon due to the rotation of the fused segments; this phenomenon mainly occurs in young patients who have open triradiate cartilage.^[26] There was no crankshaft phenomenon observed in our study because the biomechanical characteristics of the pedicle screws allowed them to serve as a structural tie and provide resistance to the longitudinally directed force through all three columns of the vertebra, achieving near complete correction and a short fusion.

The limitations of this study were its retrospective design and the small number of patients included. Moreover, cervical lordosis could have been included in order to measure the global sagittal balance more accurately. However, change of cervical lordosis after the resection of hemivertebra on the thoracic, thoraco-lumbar and lumbar vertebrae was considered to be an indirect 1 to compensate for the sagittal balance. Thus, we only measured the balance of the trunk vertebrae using SVA. Larger prospective studies designed to measure global sagittal balance are needed to confirm our results.

In conclusion, posterior hemivertebra resection and fusion using pedicle screw fixation in patients under 10 years of age with congenital scoliosis is a safe and effective procedure that can achieve rigid fixation and deformity correction. Complete resection of hemivertebra is important for deformity correction and prevention of curve progression, for which careful long-term follow-up is necessary.

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Author contributions

Conceptualization: Dong-Gune Chang, Seung Woo Suh.

Data curation: Woon Kim, Jewel Park.

Formal analysis: Jae Hyuk Yang.

Investigation: Dong-Gune Chang.

Methodology: Jae Hyuk Yang, Woon Kim, Jewel Park.

Project administration: Dong-Gune Chang, Seung Woo Suh.

Supervision: Dong-Gune Chang.

Validation: Jae Hyuk Yang, Dong-Gune Chang, Seung Woo Suh.

Writing – original draft: Jae Hyuk Yang.

Writing – review & editing: Dong-Gune Chang.

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