CLINICAL ARTICLE

Failure of Posterior Lower Lumbar/Lumbosacral Hemi-Vertebra Resection: An Analysis of Reasons and Revision Strategies

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Objective: To investigate the causes of failed primary surgery and the revision strategies for congenital scoliosis (CS) patients with lower lumbar/lumbosacral (LL/LS) hemi-vertebra (HV).

Methods: Fifteen CS patients with LL/LS HV (seven females and eight males) with a mean age of 20.4 ± 10.4 years undergoing revision surgery in our center were retrospectively reviewed. The radiographic parameters including Cobb angle, distance between C_7 plumb line and center sacral vertical line (C_7PL-CSVL), thoracic kyphosis (TK), lumbar lordosis (LL) and sagittal vertical axis (SVA) were assessed at pre-revision, post-revision and the last follow-up. The causes of failure in primary operation, and radiographic and clinical outcomes of revision procedures were analyzed.

Results: The revision rate of patients undergoing LL/LS HV resection and correction surgery was 11.4%. The average time interval between primary surgery and revision surgery was 18.2 ± 10.6 months. The operation duration and estimated blood loss of revision surgery were 194 ± 56 min and 326 ± 74 ml, respectively. Reasons for failed primary operations were as follows: internal fixation fracture in 10 cases, curve progression in two cases, implant loose in two cases and post-operative coronal imbalance in one case. The post-revision Cobb angle was significantly improved from 29.9° ± 8.3° to 18.7° ± 6.7° ($P < 0.001$) with a correction rate of 37.5% ± 12.6%. At the final follow-up, the average Cobb angle was 18.9° ± 6.2° and the correction was well maintained ($P = 0.788$). The C_7PL-CSVL at pre-revision, post-revision and at last follow-up were 23.2 ± 9.3 mm, 14.8 ± 4.8 mm and 14.9 ± 5.4 mm, respectively. Significant improvements ($P = 0.004$) were observed after revision surgery and there was no evident loss of correction ($P = 0.703$). There was no significant difference in TK, LL and SVA before and after revision surgery (all $P > 0.05$). At the last follow-up, no significant correction loss of above coronal and sagittal parameters were observed (all $P > 0.05$). The revision methods were individualized according to the primary surgical procedures and the reasons for revision. The recommended revision strategies include incision of pseudarthrosis with sufficient bone graft, fixation of satellite rods, thorough residual HV excision, prolonged fusion to S_2 and transforaminal lumbar interbody fusion at lumbosacral region. Solid bony fusion and no implant-related complication were detected during the follow-up.

Conclusions: The causes of revision surgery for patients with congenital scoliosis (CS) due to lumbosacral HV were verified and implant failure with pseudarthrosis was the main reason for failed primary operation.

Key words: Congenital scoliosis; Hemi-vertebrae resection; Lumbar/lumbosacral hemi-vertebrae; Revision surgery

Introduction

Hemi-vertebra (HV) has been recognized as the most frequent cause of congenital scoliosis (CS), posing a challenge in the prognosis and therapy.[1] The natural history of CS implies that the location of HV is a decisive factor for the curve evolution of the deformity.[2,3] The lower lumbar/

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lumbosacral (LL/LS) HV, defined as HV between L_3 and S_1 vertebrae, often causes early trunk decompensation and a long compensatory curve above since the spine below lacks the ability to compensate.[4,5] As the age increases, the LL/LS HV tends to inevitably result in gross trunk imbalance and pelvic obliquity.[6] As a result, early surgical interventions including posterior spinal fusion and various osteotomy techniques are necessary in the treatment of this particular cohort. Zhuang *et al.*[7] retrospectively reviewed 14 congenital scoliosis due to lumbosacral HV treated by one-stage posterior HV resection with short segmental fusion and the clinical results after at least a 2-year follow-up showed that this strategy can offer excellent scoliosis correction and trunk shift improvement without neurological complications, while saving motion segments as much as possible. Wang *et al.*[8] evaluated the radiological outcomes following posterior-only HV resection and short fusion for the treatment of CS secondary to lumbosacral HV with a minimum of a 5-year follow-up and found that one-stage posterior-only HV resection with short fusion is an effective procedure for lumbosacral HV, and the correction can be well maintained during longitudinal follow-up. Therefore, HV resection at an early age is regarded to be the most direct and efficient strategy in the treatment of CS due to LL/LS HV.

Though satisfactory radiographic and clinical outcomes following HV recession have been revealed in the literature,[9,10] several complications such as pseudarthrosis with implant failure, post-operative trunk imbalance and deformity progression are reported simultaneously with the necessary of revision surgery. Leong *et al.*[9] found that 16.7% of the patients receiving one-stage anteroposterior vertebral column resection (VCR) for LL/LS HV suffered from pseudarthrosis around osteotomy sites within 9-year follow-up. Lyu *et al.*[6] retrospectively reviewed 16 CS patients with lumbosacral HV undergoing VCR, finding one patient with curve progression and coronal imbalance and one patient with pseudarthrosis requiring grafting revision. In addition, in recent years, for young CS patients with LL/LS HV, posterior-only HV resection combined with short segment fusion is often the first choice in order to preserve the growth potential. However, the postoperative compensatory curve progression, as the cost of short segmental fusion, was frequently observed during longitudinal follow-up. Wang *et al.*[11] reviewed 48 CS patients aged 2.5 to 15 years with lumbosacral HV undergoing posterior-only resection and short segmental fusion, and reported that the incidence of postoperative curve progression was as high as 33.3% (16/48) during 48 months follow-up and one of them received revision surgery due to S_1 screw loosening at 1 year follow-up. In summary, the surgical treatment of CS patients with LL/LS HV is challenging and technique demanding with a relatively high revision rate and it is necessary to clarify the reasons for the failure of first operation to adopt the corresponding revision strategy.

To the best of our knowledge, there has been no report specifically focusing on the revision surgery in this particular cohort. In this respect, the causes of failure in primary operation, and radiographic and clinical data at pre-, post-revision surgery and the last follow up were carefully analyzed for each patient. The aims of the retrospective study were as follows: (i) to analyze the reasons for the failure of primary surgery in CS patients with LL/LS HV undergoing posterior HV resection and correction surgery; (ii) to explore the effective surgical strategies; and (iii) to assess the radiographic and clinical outcomes of revision in this cohort.

Methods

Patients

CS patients with single LL/LS HV (HV between L_3 and S_1) undergoing revision surgery due to failed primary surgery at our center from December 2009 to October 2015 were retrospectively reviewed. Patients undergoing revision surgery *via* posterior-only approach and meeting the following inclusion criteria were included: (i) patients with at least 2-year follow-up after revision; and (ii) with intact radiographic and clinical data at pre-, post-revision, and the last follow-up. Patients with less than a 2 yeas follow-up after revision surgery were excluded. At initial surgery, the surgical strategy was determined with reference to pre-operative X-rays, CT, and MRI. The length of fused segments was mainly decided by the property of cranial and caudal end vertebrae of scoliosis and kyphosis. Generally, ideal coronal and sagittal balance was the main goal of the surgery, and short segment fusion was the priority, especially for young patients to preserve more growth potentials. The reasons for failure of primary surgery and the corresponding revision strategies were analyzed. The present study was approved by the ethical committee of our hospital (Approval No.: 2013-079-01).

Radiographic Measurements

Radiographic measurements were performed on standing full spine radiographs at pre-, post-revision and the last follow-up.

Segmental Cobb Angle on Coronal Plane

Segmental Cobb angle on coronal plane was defined as the angle between the superior end plate of the upper end vertebra and the inferior end plate of the lower end vertebra on coronal plane.

Distance between C_7 Plumb Line and Center Sacral Vertical Line (C_7PL-CSVL)

The C_7PL-CSVL was defined as the vertical distance between C_7PL and CSVL.

Thoracic Kyphosis (TK)

The TK was defined as the angle between the superior end plate of T_5 and the inferior end plate at T_{12}.

Lumbar Lordosis (LL)
The LL was defined as the angle between the superior end plate of L_1 and the superior end plate of S_1 on sagittal plane.

Sagittal Vertical Axis (SVA)
The SVA was defined as the distance between C_7PL and the posterosuperior corner of S_1 vertebra on sagittal plane.

Statistical Analysis
All data were analyzed with standardized statistical software (SPSS, version 17.0, Chicago, IL, USA). The paired *t* test was conducted for comparison analysis of radiographic parameters. Statistically significant difference was set at $p < 0.05$.

Results

General Data
A total of 132 patients underwent LL/LS HV resection and correction surgery at our center from December 2009 to October 2015, of whom revision surgery were performed in 15 (11.4%) patients. The age at revision surgery was 20.4 ± 10.4 years and the duration of follow-up was 40.7 ± 16.4 months. The average time interval between primary surgery and revision surgery was 18.2 ± 10.6 months, of which eight (53.3%) revision surgeries were performed within 1 year after the primary surgery. The duration and estimated blood loss of revision surgery were 194 ± 56 min and 326 ± 74 ml, respectively.

Details of Primary Surgery
All patients underwent posterior-only HV resection and instrumentation with pedicle screws at primary surgery. Fusion span averaged 7.3 ± 3.1 levels including two levels in one case, three levels in one, four levels in one, six levels in three, seven levels in four, nine levels in two, 10 levels in one, and 13 levels in two, respectively. Anterior strut grafts were performed in four patients, including one titanium mesh and three cages (Table 1).

Reasons for Failed Primary Surgery
The reasons for failed primary surgery of the 15 patients were summarized as follows: (i) the osteotomy gaps after HV resection were closed incompletely in 10 (66.7%) patients, leading to space dysraphism and consequent implant failure (rod fracture with pseudarthrosis in nine cases and screw brakeage in one); (ii) the HV plate and vertebral body were not fully excised in two (13.3%) patients, of whom curve

TABLE 1 The general data of the 15 patients undergoing revision surgery

Case	Age (years)	Sex	HV location	Type	Fused segments at first surgery	Reasons for revision	Revision strategies	Fused segments at revision	Follow-up (m)
1	10	M	L_3-L_5	FS	L_3-L_5	Curve progression	Complete posterior HV resection, extended fusion	L_1-S_1	72
2	23	M	L_3-L_4	FS	T_{12}-L_5	Rod fracture and pseudarthrosis	Rod replacement, bone graft	T_{12}-L_5	68
3	6	F	L_3-L_4	SS	T_8-L_4	Screw loose and extraction	Screws re-implantment	T_8-L_4	64
4	10	M	L_5-S_1	FS	L_1-S_1	Rod fracture and pseudarthrosis	Revision with satellite rods, extended fusion, bone graft	T_{11}-S_2	54
5	17	M	L_5-S_1	SS	T_{12}-S_1	Rod fracture and pseudarthrosis	Revision with satellite rods, extended fusion, bone graft	T_{12}-S_2	48
6	35	F	L_3-L_4	FS	T_{12}-S_1	Rod fracture and pseudarthrosis	Revision with satellite rods, extended fusion, bone graft	T_{12}-S_2	42
7	29	M	L_3-L_4	FS	T_9-S_1	Rod fracture and pseudarthrosis	Revision with satellite rods, extended fusion, $L_{4/5}$ and L_5/S_1 TLIF, bone graft	T_9-S_2	36
8	26	M	L_4-L_5	SS	T_{10}-S_1	Rod fracture and pseudarthrosis	Revision with satellite rods, extended fusion, bone graft	T_{10}-S_2	32
9	23	F	L_5-S_1	SS	T_6-S_1	Screw fracture and pseudarthrosis	Revision with satellite rods, extended fusion, bone graft	T_6-S_2	28
10	16	F	L_3-L_4	FS	L_2-L_5	Curve progression	Complete posterior HV resection, extended fusion, $L_{4/5}$ and L_5/S_1 TLIF	L_3-S_1	42
11	23	F	L_4-L_5	FS	T_6-S_1	Rod fracture and pseudarthrosis	Revision with satellite rods, extended fusion, bone graft	T_6-S_2	26
12	31	F	L_5-S_1	FS	T_{12}-S_1	Rod fracture and pseudarthrosis	Revision with satellite rods, extended fusion, $L_{3/4}$ TLIF, bone graft	L_2-S_2	26
13	41	F	L_5-S_1	SS	L_1-S_1	Coronal imbalance	Extended fusion, $L_{4/5}$ and L_5/S_1 TLIF	T_6-S_2	24
14	8	M	L_3-L_4	FS	L_3-L_4	Screw loose and extraction	Screws re-implantment	L_3-L_4	24
15	8	M	L_5-S_1	SS	L_1-S_2	Rod fracture and pseudarthrosis	Revision with satellite rods, extended fusion, bone graft	T_{11}-S_2	24

Abbreviations: M, male; F, female; FS, full segmented; SS, semi-segmented.

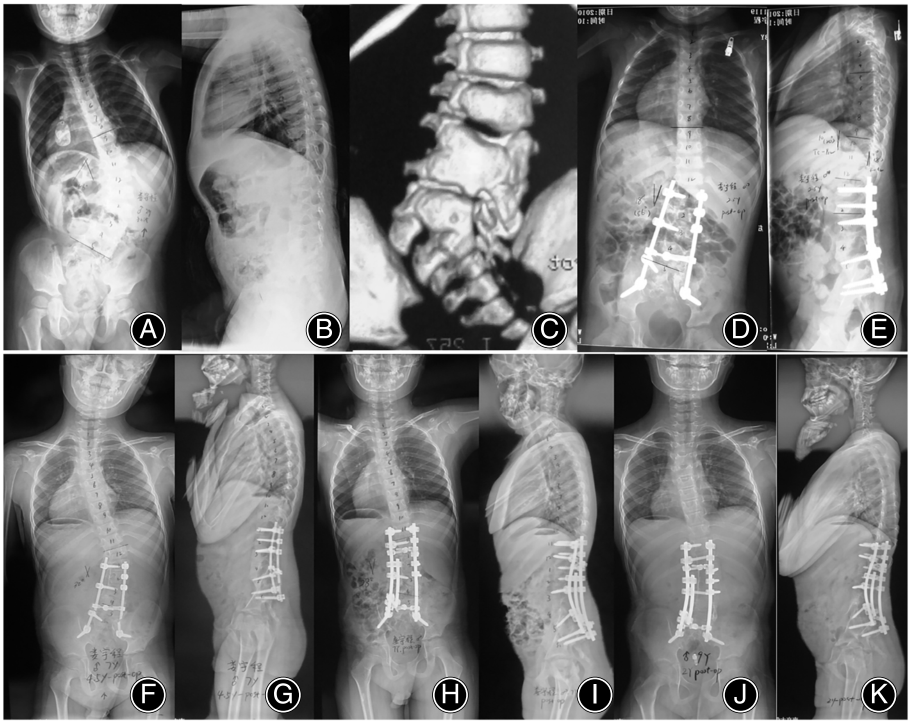

Fig. 1 A 3-year-old boy with lumbosacral HV and sacral dysplasia. The coronal Cobb angle and C_7PL-CSVL were 56° and 41.6 mm, respectively (A, B, C). The patient underwent posterior L_5 HV resection and fusion from L_1 to S_2 (D, E). However, bilateral rod breakage with pseudarthrosis at L_5/S_1 and proximal curve progression were observed at 54 months follow-up (F, G). During revision, the broken rods were replaced with new ones and the upper instrumented vertebra was extended to T_{11}. Fixation of additional satellite rod from L_2 to S_1 was performed *via* dual head connectors simultaneously as a reinforcement (H, I). The coronal and sagittal balance were well maintained during 24 months follow-up (J, K)

progression were detected during follow-up; (iii) screw malposition was observed in two (13.3%) patients, resulting in low pull-out resistance and further internal fixation loose; and (iv) post-operative coronal imbalance due to dissatisfactory reconstruction of the lumbosacral balance was detected in one (6.7%) patient.

Revision Strategies

The revision strategies for 10 patients who had implant failure included complete pseudarthrosis resection, sufficient bone graft and replacement of the broken internal fixation. In order to maximally avoid recurring implant failure, satellite rod fixation around pseudarthrosis area and extended fixation to S_2 with S_2-Alar-Iliac (S_2AI) screws were utilized in eight patients. For the two patients with post-operative curve progression due to incomplete HV resection, thorough HV resection and prolonged fusion were adopted. In two patients with loose implants due to screws malposition, pedicle screws were re-implanted carefully with the assistance of O-arm navigation. Another patient with post-operative global coronal decompensation was revised with extended fusion to S_2 and transforaminal lumbar interbody fusion (TLIF) at L_4/L_5 and L_5/S_1. TLIF was performed at lumbosacral region in four patients during revision to promote lumbosacral fusion and horizontalize the L_4 and L_5 endplates (Table 1).

Typical cases are shown in Fig. 1 and 2.

Radiographic Outcomes

As shown in Table 2, the post-revision segmental coronal Cobb angle evidently improved (29.9° ± 10.7° vs. 18.7° ± 6.7°, $P < 0.001$) with an average correction ratio of 37.5% ± 12.6%. The C_7PL-CSVL decreased significantly from 23.2 ± 9.3 mm to 14.8 ± 4.8 mm ($P = 0.004$). The differences between pre- and post-revision in TK, LL and SVA were not statistically significant. At the last follow-up, there was no significant correction loss in both coronal and sagittal parameters (all Ps > 0.05).

Complications

There was one transient neurological deficit and one dual tear during revision surgery. One patient suffered superficial infection at post-operation, which was cured with antibiotics. During the longitudinal follow-up, no re-occurrence of pseudarthrosis, coronal and sagittal imbalance, or implant failure was detected.

Discussion

Current Status of Treatment

HV located at LL/LS region is a rare but complicated spinal deformity, which may lead to early three-dimensional decompensation and a long compensatory curve.[12,13] As a result, early HV resection is strongly recommended for young patients with LL/LS HV.[10,14] Although satisfactory radiographic and clinical outcomes were reported in the literature, revision surgeries were unfortunately required in certain patients undergoing posterior HV resection. Bollini *et al.*[15] and Michael *et al.*[16] also found that the risk of revision surgery in CS patients with LL/LS HV treated with posterior VCR was evidently higher than those with HV located elsewhere. The unique anatomic characteristics

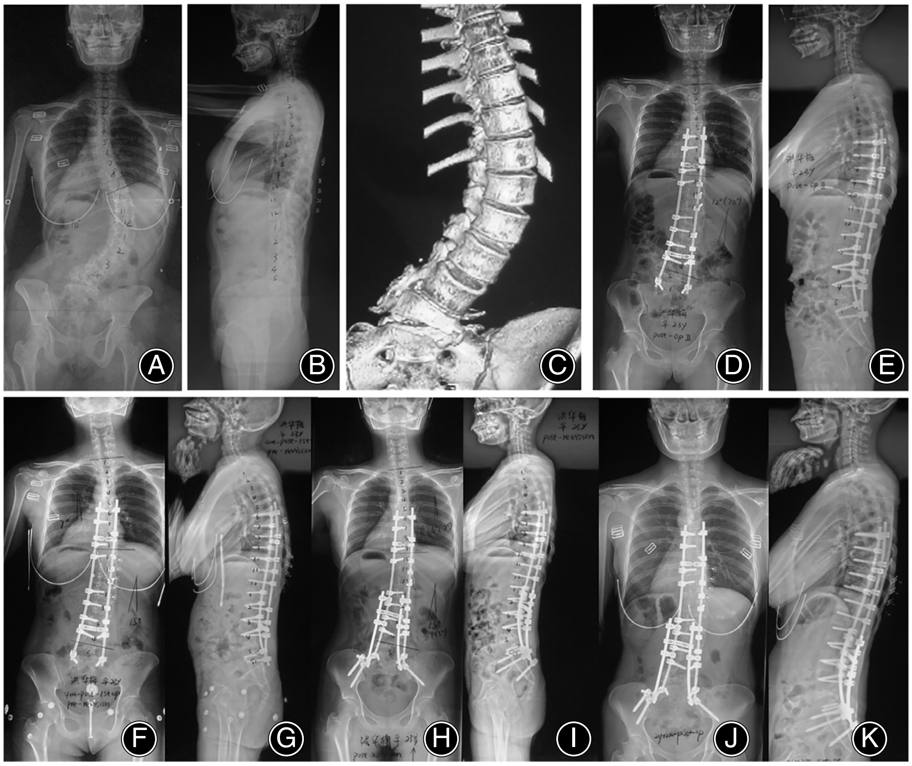

Fig. 2 A 23-year-old female with L_5 HV. The coronal Cobb angle and C_7PL-CSVL were 70° and 50.1 mm, respectively (A, B, C). The patient underwent posterior L_5 HV resection and fusion with traditional pedicle screw instrumentation from T_6 to S_1 (D, E). However, screw fracture was observed at 4 months follow-up (F, G). During revision, the lower instrumented vertebra was extended to S_2 for rigid pelvic fixation. Satellite rods were implanted from L_1 to S_2 for an integrated and enhanced local fusion structure. (H, I). The coronal and sagittal balance were well maintained during 28 months follow-up after revision (J, K)

and mechanical features in the LL/LS region were believed to be responsible for the relatively high risks.[17,18] The mobile lumbar spine was connected to the stable sacrum *via* lumbosacral junction, which results in high mechanical demand in the area.[19] In addition, the high ratio of cancellous bone to cortical bone, thin anterior cortex in sacrum and wide and short S_1 pedicles, together contributed to the difficulty in obtaining solid fusion at the lumbosacral junction.[20–22] Herein, the post-operative complications including implant failure with pseudarthrosis, trunk imbalance and curve progression were frequently detected in this cohort.

Implant Failure with Pseudarthrosis

Implant failure was found in 10 patients in our study, serving as the most common reason for the revision surgery. A rigid internal fixation and sufficient bone graft were the key points for such revision surgeries. The replacement of the broken implants was usually necessary and the use of satellite rods unilaterally or bilaterally was recommended if possible for an integrated and enhanced local fusion structure, which was proved to effectively disperse the stress on internal fixation and consequently lower rate of implant failure.[23,24] In addition, extending fusion to S_2 with S_2AI screws in cases merely using S_1 pedicle screws as distal fixation anchors at

TABLE 2 Comparison of radiographic parameters among pre-, post-revision and the last follow-up

	Pre-revision	Post-revision	Last follow-up	Pre- *VS* post-revision	Post-revision *VS* last follow-up
Segmental Cobb angle (°)	29.9 ± 8.3 (22–46)	18.7 ± 6.7 (8–28)	18.9 ± 6.2 (10–27)	$t = 9.155$ $P < 0.001$*	$t = -0.281$ $P = 0.788$
C_7PL-CSVL (mm)	23.2 ± 9.3 (18.7–40.3)	14.8 ± 4.8 (8.7–21.4)	14.9 ± 5.4 (7.1–22.5)	$t = 4.441$ $P = 0.004$*	$t = -0.400$ $P = 0.703$
TK (°)	24.3 ± 9.8 (10–38)	23.4 ± 9.9 (12–40)	23.3 ± 10.0 (12–41)	$t = 0.795$ $P = 0.457$	$t = 0.420$ $P = 0.689$
LL (°)	31.0 ± 11.7 (15–51)	30.6 ± 8.7 (22–47)	30.9 ± 9.4 (21–48)	$t = 0.208$ $P = 0.842$	$t = -0.679$ $P = 0.522$
SVA (mm)	15.1 ± 9.2 (7.3–31.5)	13.7 ± 5.9 (5.5–20.1)	14.1 ± 6.7 (4.9–23.4)	$t = 0.543$ $P = 0.607$	$t = -0.782$ $P = 0.464$

Abbreviations: C_7PL-CSVL, distance between C_7 plumb line and center sacral vertical line; TK, thoracic kyphosis; LL, lumbar lordosis; SVA, sagittal vertical axis.
* Statistically significant if $p < 0.05$.

primary surgery could effectively decrease the incidence of implant-related complications in patients undergoing revision.[6] In the present study, no reoccurrence of rod or screw fracture was detected during a minimum 2-year follow-up, demonstrating the feasibility of the preferred revision procedures.

Post-Operative Curve Progression

According to previous studies, the full resection of malformed HV could directly remove the deformity factors and control the scoliosis development immediately,[25] emphasizing the importance of the cause-removing procedure during the correction surgery. A total of two patients suffered from post-operative curve progression due to the incomplete resection of HV in the current study. As a result, the residual malformation led to the deformity progression. Therefore, the critical revision procedures for such patients should be the thorough resection of residual HV with strong internal fixation.

Internal Loose Fixation

As a general consensus, the width of vertebral pedicle in young children was often too small for the accurate implant of pedicle screws.[26] Moreover, pedicles on the concavity were significantly narrow, which can be aggravated in cases with severe axial rotation.[27] In our research, internal fixation loose occurred in two cases after the primary surgery duo to the screw malposition and the consequent low extraction torques. Pre-operative CT scan parallel to pedicle and 3D reconstruction were critical references for surgical evaluation helping to choose appropriate implants.[28] The O-arm navigation system was also conducive to complex cases providing real-time multidimensional images optimized for spine surgeries.[29]

Post-Operative Coronal Malalignment

In addition, the post-operative coronal malalignment was observed in one patient after posterior L_5 HV resection due to the unsatisfactory horizontalization of L_4 and L_5 endplates, which was regarded as the foundation of the upper spine. During revision, the patients underwent a prolonged fixation to S_2 and TLIF at L_4-S_1, and the trunk balance was well restored. According to a study by Bao *et al.*,[30] TLIF at lumbosacral region helped to horizontalize the foundation of the spine, and promote the fusion of lumbosacral region. S_2AI screws were also strongly recommended to obtain a rigid pelvic fixation in this cohort since it was of great importance to achieve a both flat and stable base to avoid reoccurring coronal imbalance.

Limitations

Our study has several limitations. First, a small series of cases were included, and selection bias might be caused consequently. Since HV located at LL/LS region is a rare spinal deformity, we have tried our best to include more patients in the analysis. Second, the average follow-up was 40.7 months, loss of correction, pseudarthrosis, implant failure and coronal imbalance would be still possibly encountered in the future. Even though the details of the 15 patients such as age, gender, HV location, type of HV, fused segments at first surgery, reasons for revision and revision strategies were summarized in Table 1, the bone graft figure during revision was not available for this cohort. In addition, the high resolution CT was not routinely performed at follow-up due to the ethic consideration. Hence, further prospective studies with a large sample and long follow-up are urgently required.

Conclusions

For CS patients with LL/LS HV undergoing posterior HV resection, implant failure with pseudarthrosis was the main reason for revision, followed by internal fixation loose, curve progression and post-operative coronal imbalance. Revision strategies should be decided individually according to the primary surgical procedures and reasons for revision, including the incision of pseudarthrosis with sufficient bone graft, fixation of satellite rods, thorough residual HV excision and TLIF at lumbosacral region. Moreover, prolonged fusion to S_2 with S_2AI screws was usually needed during revision surgery.

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Conflict of Interest

The authors have no conflict of interest to declare.

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