CLINICAL ARTICLE

Failed Primary Surgery in Congenital Scoliosis Caused by a Single Hemivertebra: Reasons and Revision Strategies

Ben-long Shi, MD[†], Yang Li, PhD[†], Ze-zhang Zhu, PhD, Wan-you Liu, MM, Zhen Liu, PhD, Xu Sun, PhD, Dun Liu, PhD, Yong Qiu, MD

Spine Surgery, the Affiliated Drum Tower Hospital of Nanjing University Medical School, Nanjing, China

Objective: To analyze the factors causing failure of primary surgery in congenital scoliosis (CS) patients with single hemivertebra (SHV) undergoing posterior spinal fusion, and to elucidate the revision strategies.

Methods: In this retrospective study, a total of 32 CS patients secondary to SHV undergoing revision surgery from April 2010 to December 2017 due to failed primary surgery with more than 2 years follow-up were reviewed. The reasons for failure of primary surgery and revision strategies were analyzed for each patient. The radiographic parameters including coronal Cobb angle, segmental kyphosis (SK), coronal balance (CB), and sagittal vertical axis (SVA) were compared between pre- and post-revision. The complications during revision and follow-up were recorded.

Results: The mean age at revision surgery of the 32 CS patients was 15.8 ± 9.7 years and the average duration between primary and revision surgery was 31.0 ± 35.4 months. The reasons for failed primary surgery were severe post-operative curve progression of focal scoliosis in 14 cases (43.8%), implant failure in 17 (53.1%) and trunk imbalance in 12 (37.5%). The candidate revision strategies included thorough resection of residual hemivertebra and adjacent discs, extending fusion levels, complete pseudarthrosis resection, massive bone graft, replacement of broken rods, satellite rod fixation, horizontalization of upper/lower instrumented vertebrae and rigid fusion of structural compensatory curves were performed individually. After revision surgery, the coronal Cobb angle, SK, CB and SVA showed significant improvement ($P < 0.05$) with no significant correction loss during follow-up ($P > 0.05$). The intra-operative complications included alarming changes of neurologic monitoring in three (9.4%) patients and dual tear in two, while rod fracture re-occurred was detected in one patient at 18 months after revision.

Conclusions: The common reasons for failed primary surgery in CS patients with SHV undergoing posterior spinal fusion were severe post-operative curve progression of focal scoliosis, implant failure and trunk imbalance. The revision strategies including thorough resection of residual hemivertebra and adjacent discs, extended fusion levels to structural curvature, complete pseudarthrosis resection, massive bone graft, replacement of broken internal fixation and horizontalization of upper/lower instrumented vertebrae should be individualized based on the causes of failed primary surgery.

Key words: Congenital scoliosis; Curve progression; Hemivertebra; Implant failure; Reversion surgery

Address for correspondence Ze-zhang Zhu, PhD, Spine Surgery, Drum Tower Hospital, Nanjing University Medical School, Zhongshan Road 321, Nanjing, China 210008 Tel: +86-25-68182022; Fax: +86-25-68182022; Email: zhuzezhang@126.com
Grant Sources: This work was supported by the Natural Science Foundation of Youth Fund Projects of Jiangsu Province (BK20170126); Jiangsu Provincial Key Medical Center (YXZXA2016009); Natural Science Foundation of China (81772304).
Disclosure: The authors have no conflict of interest to declare.
[†]These two authors contributed equally to this work.
This study was approved by the University Institutional Review Board.
Received 3 February 2021; accepted 22 November 2021

Introduction

During the last few decades, the hemivertebra, widely regarded as the most frequent cause of congenital scoliosis (CS), has posed great challenges for treatment[1]. According to previous studies[2–4], the deformity due to single hemivertebra (SHV) is reported to progress at an approximate rate of 2°–3.5° per year for fully segmented and 1° per year for partially segmented hemivertebra in children, which is actually much more severe during rapid growth spurt. The posterior spinal fusion has gradually become a preferred surgical strategy for CS patients secondary to SHV, and satisfactory radiographic and clinical outcomes have been commonly demonstrated in the literature[5–7]. Notably, the *in-situ* fusion, Smith–Peterson osteotomy, pedicle subtraction osteotomy (PSO), SRS-Schwab Grade 4 osteotomy, vertebral column resection (VCR) and even the anterior complementary fusion were individually needed during the correction surgery.

In recent years, the revision surgery have been frequently reported in certain unfortunate patients accompanying with the increasing application of posterior spinal fusion[8–10]. Ruf *et al.* found that the incidence of revision surgery was 14.3% (four of 28) in CS patients aged 1 to 6 years undergoing posterior hemivertebra resection during 3.5 years follow-up[11]. The reasons for revision surgery were reported as infection in one patient (implant removal and another revision surgery for deformity correction) and implant failed in three (two screw/wire combinations and one screw/rod instrumentation). Zhang *et al.* reported six revision surgeries (10.7%) due to the pedicle fractures, instrumentation failures, proximal junctional kyphosis and delayed wound union in 56 CS cases undergoing posterior hemivertebra resection with transpedicular instrumentation[8]. The corresponding revision strategies were summarized as rod change with additional inter-body fusion with a mesh cage, hemivertebra resection with extended fusion levels, bone grafting and debridement. However, to the best of our knowledge, the reasons and revision strategies of failed primary surgery in CS patients undergoing posterior hemivertebra resection needed more in-detailed investigations.

Herein, based on a cohort of CS patients undergoing revision surgery due to failed primary surgery, this retrospective study was designated. The radiographic and clinical data at pre- and post-primary surgery, pre- and post-revision surgery were carefully reviewed for each patient. The objectives of the current study were as follows: (i) to analyze the factors causing the failure of primary surgery in CS patients with SHV undergoing posterior hemivertebra resection; (ii) to elucidate the strategies of revision surgery; and (iii) to evaluate the radiographic and clinical outcomes of revision in this cohort.

Subjects and Methods

Patients

The present study was approved by the ethical committee of our hospital, and written informed consent was obtained from all subjects before enrolling participants into the study. A total of 215 CS patients with SHV undergoing posterior-only spinal fusion and correction surgery from April 2010 to December 2017 in our center were retrospectively reviewed. The inclusion criteria were as follows: (i) patients underwent posterior revision surgery due to failed primary surgery; (ii) with intact radiographic and clinical data at pre- and post-first surgery, pre- and post-revision, and the last follow-up; (iii) with more than 2 years follow-up after revision; and (iv) without any hemivertebra(e) elsewhere. Twenty (9.3%) patients meeting the inclusion/exclusion criteria were included in the study. In addition, 12 patients undergoing revision surgery in our center due to failed primary surgery at other hospital fulfilled the inclusion criteria and were included for analysis as well. Finally, a total of 32 patients (15 males and 17 females) were involved in the study, of whom the reasons for failure of primary surgery and revision strategies were analyzed.

Radiographic and Clinical Evaluations

The radiographic parameters (Fig 1) applied for the assessment of local curve severity and trunk shift were measured on whole standing spinal x-rays of pre-revision, immediate post-revision, and the last follow-up.

Coronal Cobb Angle

The coronal Cobb angle was defined as the angle between the maximally tilted upper and lower end vertebra in coronal plane.

Segmental Kyphosis (SK)

The SK was defined as the angle between the maximally tilted upper and lower end vertebra in sagittal plane.

Coronal Balance (CB)

The CB was defined as the horizontal distance between C_7 plumb line and central sacral vertical line.

Sagittal Vertical Axis (SVA)

The SVA was defined as the distance between C_7 plumb line and the posterior superior corner of S_1. In addition, complications such as infection, neurological deficit, implant failure, curve progression and others during revision and follow-up were recorded for each patient.

Statistical Analysis

The SPSS 22.0 software package (SPSS, Inc., Chicago, IL, USA) was utilized for analysis. Summaries of categorical variables were given as means and standard deviations, The paired Student's *t*-test or Wilcoxon signed ranks test was applied for comparison between pre-revision and post-revision. The chosen level of significance was 0.05.

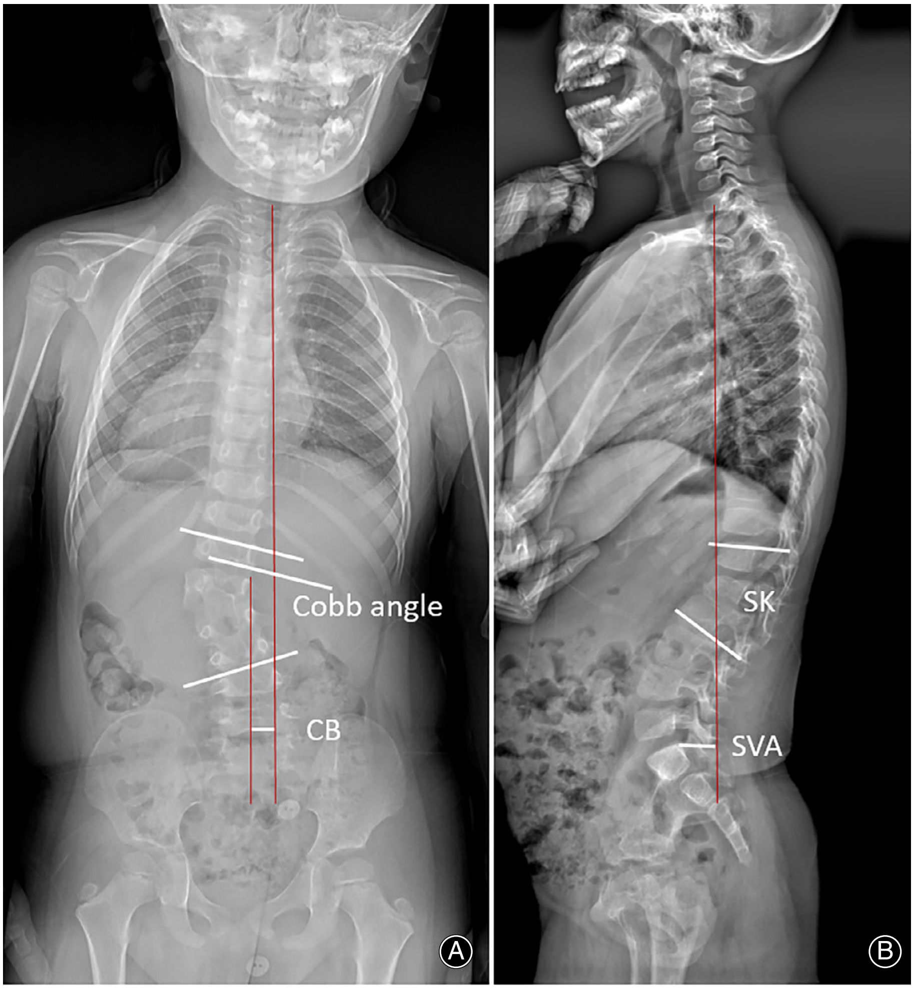

Fig. 1 The patient was diagnosed as CS with T12 hemivertebra at 2 years (A, B). Posterior unilateral and short spinal fusion without hemivertebra resection was performed (C, D). At 4 years post-operation, curve progression of focal scoliosis was found (E, F). Posterior thorough resection of residual hemivertebra with extended fusion levels was performed (G, H). At 2.3 years follow-up, the correction outcomes were well maintained (I, J).

Results

General Data

The locations of SHV were thoracic in three (10.3%) patients, thoracolumbar in 13 (40.6%), lumbar/lumbosacral in 16 (50.0%), respectively. The mean age at revision surgery was 15.8 $\pm$ 9.7 years and the average duration between primary and revision surgery was 31.0 $\pm$ 35.4 months (3 months–10 years), including 12 (37.5%) performed within 1 year after primary surgery, five (15.6%) within 1–2 years, and 15 (46.9%) beyond 2 years. The average operative time of revision was 4.3 $\pm$ 2.7 h and the estimated blood loss of revision was 460.7 $\pm$ 815.3 mL.

Reasons for Failed Primary Surgery

The reasons for failed primary surgery were summarized as follows: (i) severe post-operative curve progression of focal scoliosis in 14 cases (43.8%).The SHV remained or was incompletely resected at primary surgery in 10 patients, leaving sustainably growing plate and body of hemivertebra. Over-short segmental fusion of the main curve was found in four patients. The fusion levels in these four patients were SHV $\pm$ 1 or SHV $\pm$ 2 levels; (ii) implant failure in 17 cases (53.1%). Pseudarthrosis was observed in the patients, including eight cases at osteotomy site and nine cases at thoracolumbar or lumbarsacral region; and (iii) trunk imbalance in 12 cases (37.5%). The causes of trunk imbalance included post-operative insufficient leveling of upper/lower instrumented vertebra in seven patients and progression of compensatory curves (adding-on phenomenon) in five patients, respectively.

Revision Strategy

For the 10 patients with post-operative curve progression due to SHV remaining or incompletely resected, thorough resection of residual hemivertebra and adjacent discs were adopted. The fusion levels were extended for four patients with over-short segmental fusion of main curve. The revision strategies for eight patients who had implant failure at the osteotomy area included complete pseudarthrosis resection, massive bone graft and replacement of the broken internal fixation. In addition to complete pseudarthrosis resection, massive bone graft and replacement of broken rods, extending fusion level was performed in the nine patients with implant failure at the thoracolumbar or lumbosacral junctions. To maximally avoid recurring implant failure, satellite rod fixation around the rod-fracture area was utilized in 10 of the 17 cases. Revision with extended fusion level was performed in the 12 patients with post-operative trunk decompensation. The horizontalized upper/lower instrumented vertebrae and rigid fusion of structural compensatory curves were priorities of revision surgery.

Radiographic Measurements

The mean follow-up after revision was 33.0 ± 14.1 months in the current study. The average coronal Cobb angle was significantly corrected from 40.2° ± 22.4° to 17.3° ± 12.5° at post-revision ($P = 0.000$). The SK was 42.8° ± 27.5° at pre-revision, which decreased to 20.3° ± 10.6° at post-revision ($P = 0.000$). The CB and SVA were 23.6 ± 14.9 mm and 31.0 ± 16.2 mm at pre-revision, 12.1 ± 8.9 mm and 15.7 ± 10.1 mm at post-revision, respectively ($P = 0.000$). At the last follow-up, no significant correction loss was observed in all parameters (all $P > 0.05$, Table 1). Two demo cases are displayed in Figs 2 and 3.

Complications

Alarming changes in neurologic monitoring were reported in three (9.4%) patients during the resection of residual SHV, of whom one (3.1%) patient was found to have crus numbness postoperatively and relief occurred entirely after 2 weeks of conservative treatment. Dual tear was found in two patients during osteotomy, who recovered at post-operation after prompt epidural suture. Rod fracture re-occurred in one patient at 18 months after revision due to implant failure after primary surgery, and the second revision was performed with the utilization of the satellite rods technique.

Discussion

The posterio-only spinal correction and fusion has been recognized as the curative treatment of congenital scoliosis[12], by which the immediate correction outcomes of the deformity are practically visible[13]. However, failed primary surgery is occasionally observed during follow-up and revision surgery seems to be inevitable to a portion of the patients. According to previous studies, it is still controversial as to why these depressing outcomes happen and how to manage afterwards. Therefore, it is critical to summarize the causes of failed primary surgery and the corresponding revision strategies, which we believe could provide more evidence for the individual decision-making of CS patients at both primary and revision surgeries.

Post-Operative Curve Progression

According to the study of Repko *et al.*[12], the correction rate of posterior fusion without resection of hemivertebra was only 22.1% during the initial exploration of surgical intervention for CS. Though hemivertebra resection was thus regarded as essential procedure for satisfactory radiographic outcomes, the residual growth of plate and vertebral body after partial hemivertebra resection might lead to irreversible curve progression during longitudinal follow-up[9]. Therefore, complete resection of both hemivertebra and adjacent discs in CS patients has been emphasized for a well maintained correction outcome, especially in those with SHV[13]. In the current study, curve progression due to hemivertebra remained or incomplete resected was observed in 10 patients, strongly indicating the importance of the key case-removal procedure. Naturally, thorough resection of the residual hemivertebra combined with strong internal fixation should be the critical revision procedure for such patients. In addition, alarming changes in neurologic monitoring were reported in three patients and dual tear in two patients during osteotomy in the current study, implying the osteotomy procedure during revision was more difficult and high-risk than primary surgery. The fusion levels should be extended for patients with significant curve progression of focal deformity and massive bone graft was highly recommended for all patients.

Implant Failure with Pseudarthrosis

Implant failure with pseudarthrosis was a common complication in patients undergoing posterior spinal fusion, of which the average incidence was reported as 4.2% in CS patients undergoing posterior vertebral column osteotomy[14]. The three-column osteotomy as well as long fusion across the thoracolumbar/lumbarsacral region were identified as two independent high-risk factors[15]. In the current study, 17 patients underwent revision surgery due to implant failure, serving as one of the most common reasons for failed primary surgery. Since the bone stock was relatively deficient due to wide osteotomy and stable bone fusion was not yet formed in short-term follow-up, the osteotomy site and adjacent segments suffered from the greatest mechanical

TABLE 1 Comparison of radiographic parameters among pre-, post-revision and last follow-up

Parameters	Pre-revision	Post-revision	Last follow-up	Pre- *vs.* post-revision	Post-revision *vs.* last follow-up
Coronal Cobb angle (°)	39.0 ± 8.8	17.0 ± 7.8	16.8 ± 6.5	$t = 18.929$ $P = 0.000$	$t = 0.219$ $P = 0.828$
Segmental kyphosis (°)	43.7 ± 7.8	22.6 ± 6.6	21.7 ± 4.9	$t = 15.228$ $P = 0.000$	$t = 0.908$ $P = 0.371$
Coronal balance (mm)	22.0 ± 9.9	11.1 ± 6.4	10.3 ± 6.0	$t = 8.668$ $P = 0.000$	$t = 1.074$ $P = 0.291$
Sagittal vertical axis (mm)	30.0 ± 8.0	13.8 ± 6.2	12.8 ± 4.7	$t = 12.070$ $P = 0.000$	$t = 1.118$ $P = 0.272$

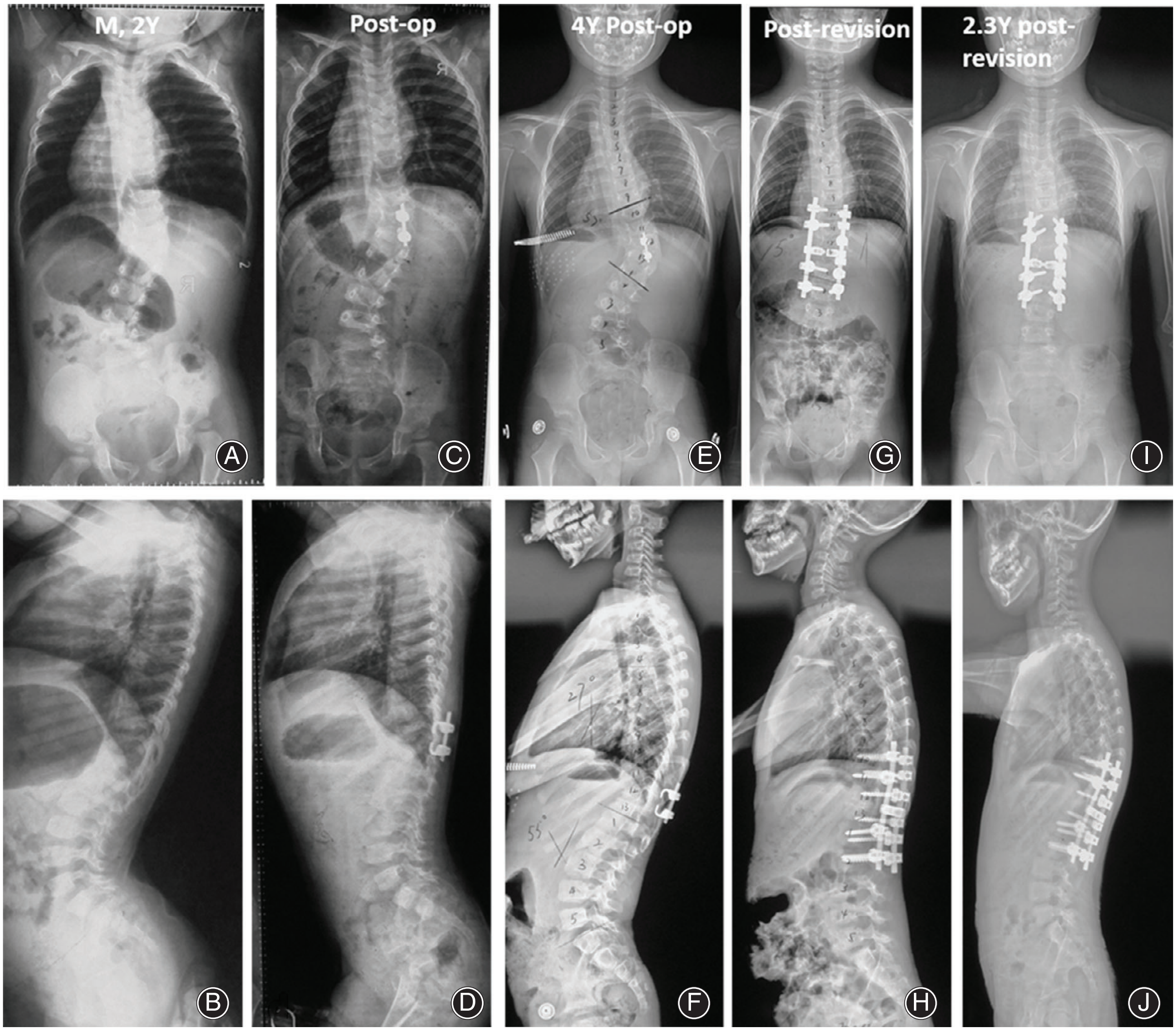

Fig. 2 The patient was diagnosed as CS with L_3 hemivertebra at 23 years (A, B), and posterior hemivertebra resection and fusion was performed (C, D). Significant trunk imbalance and breakage of unilateral S_1 screw were found at 2.7 years post-operation (E, F). Posterior revision with extending lower instrumented level to pelvic was performed. The broken screw was left in the vertebral body and pedicle screws of L_2 were removed in order to better maintain the spontaneous compensation ability (G, H). No deterioration of trunk balance was observed during 2 years follow-up (I, J).

stress[16,17]. A total of eight patients had implant failure at the osteotomy area in our study, which was in accordence with previous studies. Therefore, complete pseudarthrosis resection, massive bone graft and replacement of broken internal fixation were the standard revision strategy for these patients. More importantly, bone-to-bone closure of the osteotomy site, sufficient anterior support and massive bone graft should be enhanced at primary surgery. In addition, the internal fixation at the thoracolumbar and lumbarsacral regions was much easier to fatigue and break than other spinal segments, which was responsible for implant failure of the other nine patients. Rigid internal fixation and solid bone fusion at the thoracolumbar and lumbarsacral regions were thus critical for this cohort. In spite of pseudarthrosis resection, bone graft and replacement of broken rods, extension of the fusion level was often needed and the utilization of satellite rods unilaterally or bilaterally was highly recommended for an integrated and enhanced local fusion structure[18,19]. In our study, no reoccurrence of rod fracture was detected in patients with satellite rod fixation during a minimum of 2-year follow-up.

Post-Operative Trunk Imbalance

According to the study by Xu *et al.*[20], the incidence of post-operative trunk imbalance in CS patients undergoing three-column osteotomy was about 20% depending on the curve type, curve correction rate and lower instrumented vertebra tilt. The authors also demonstrated that the cut-off value of lower instrumented vertebra tilt was 12.3° at immediate post-operation[20]. Bao *et al.*[21] demonstrated the importance of horizontalized

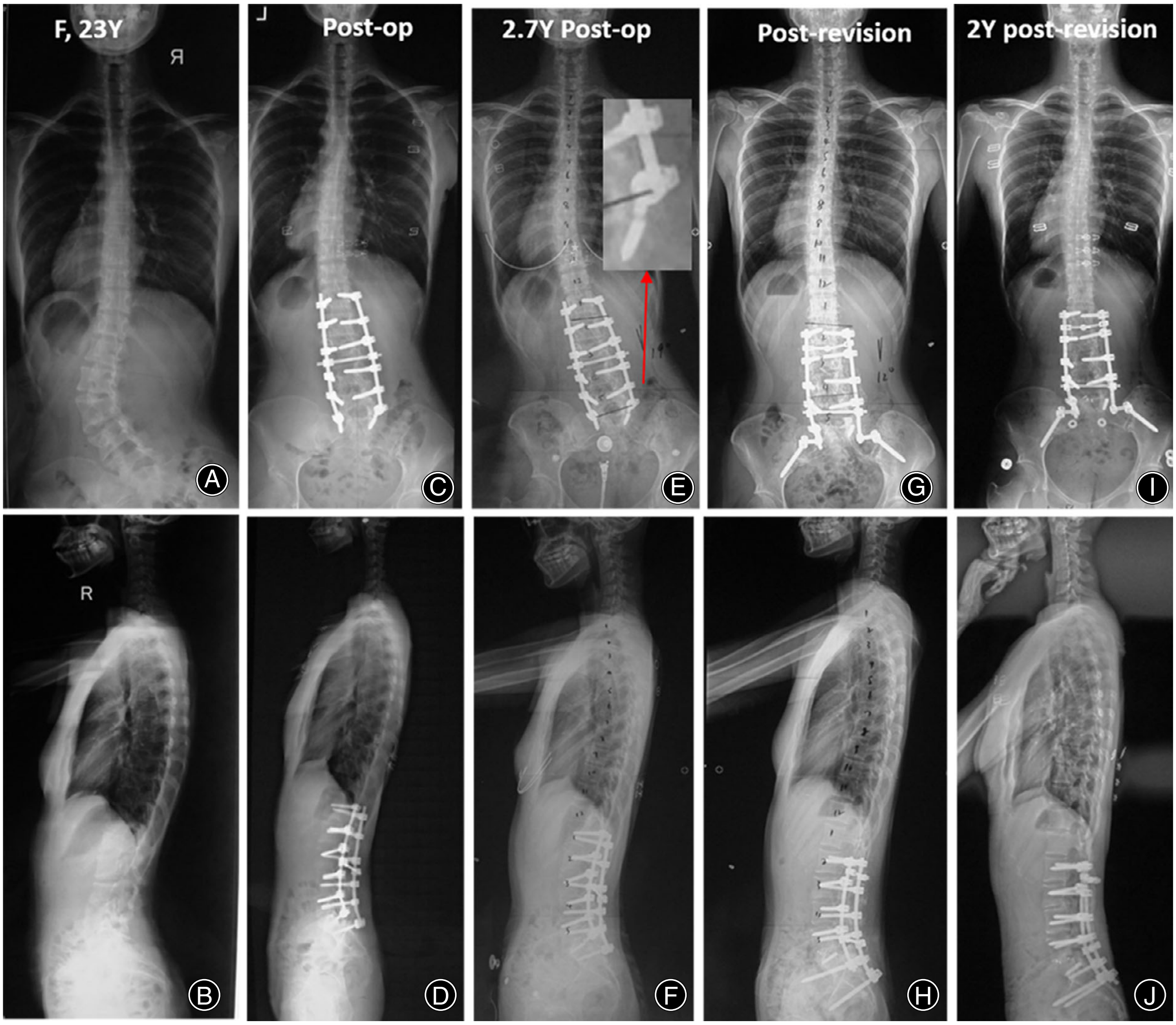

Fig. 3 The patient was diagnosed as CS with L_3 hemivertebra at 23 years (A, B), and posterior hemivertebra resection and fusion was performed (C, D). Significant trunk imbalance and breakage of unilateral S_1 screw were found at 2.7 years post-operation (E, F). Posterior revision with extending lower instrumented level to pelvic was performed. The broken screw was left in the vertebral body and pedicle screws of L_2 were removed in order to better maintain the spontaneous compensation ability (G, H). No deterioration of trunk balance was observed during the 2 years follow-up (I, J).

lumbosacral region in adult spinal deformity in order to avoid the risk of iatrogenic global coronal malalignment. In the present study, insufficient leveling of upper/lower instrumented vertebra was detected in seven patients with trunk imbalance needing revision surgery, serving a critical risk factor for progressive trunk shift during follow-up. Additionally, hemivertebra resection together with short-segment fusion was traditionally recommended for CS patients with relatively young age in order to protect the longitudinal spine growth[22,23]. Zhuang *et al.*[23] reported 14 CS patients secondary to lumbosacral hemivertebra undergoing one-stage posterior lumbosacral hemivertebra resection with short segmental fusion, showing significant improvement in coronal (63%) and sagittal (58%) trunk shift during more than 2 years follow-up. However, our study demonstrated the inappropriate short fusion levels of the main curve and leaving the structural decompensatory curves un-fused were very likely to result in irreversible curve progression during follow-up. Therefore, a longitudinal and regular post-operative follow-up was needed for these patients. Nevertheless, horizontalization of upper and lower instrumented vertebrae, extending fusion to the structural compensatory curves were priorities for both primary and revision surgeries.

Limitations

The potential defects of the study were its retrospective character and relatively small sample size. The various age, location of hemivertebra and fusion levels of the cohort possibly leaded to certain selection bias for the results. Though gratifying outcomes during 2 year follow-ups were demonstrated in the current study, more longitudinal studies with large sample size were needed. In addition, the quality of life before and after revision was not assessed.

Conclusions

The common reasons for failed primary surgery were severe post-operative curve progression of focal scoliosis, implant failure and trunk imbalance for CS patients with SHV undergoing posterior spinal fusion. The revision strategies including thorough resection of residual hemivertebra and adjacent discs, extended fusion levels to structural curvature, complete pseudarthrosis resection, massive bone graft, replacement of broken internal fixation and horizontalization of upper/lower instrumented vertebrae should be applied individually based on the causes of failed primary surgery. The satellite rod fixation at rod the fracture area and the thoracolumbar/lumbarsacral region were highly recommended for an enhanced local fusion structure. Satisfactory radiographic and clinical outcomes during post-revision follow-up could be achieved for those patients with appropriate revision surgery.

References

1. Ruf M, Jensen R, Letko L, Harms J. Hemivertebra resection and osteotomies in congenital spine deformity. Spine (Phila Pa 1976), 2009, 34: 1791–1799.
2. McMaster MJ, Ohtsuka K. The natural history of congenital scoliosis. A study of two hundred and fifty-one patients. J Bone Joint Surg Am, 1982, 64: 1128–1147.
3. McMaster MJ, David CV. Hemivertebra as a cause of scoliosis. A study of 104 patients. J Bone Joint Surg Br, 1986, 68: 588–595.
4. Bollini G, Docquier PL, Viehweger E, Launay F, Jouve JL. Thoracolumbar hemivertebrae resection by double approach in a single procedure: long-term follow-up. Spine (Phila Pa 1976), 2006, 31: 1745–1757.
5. Chen X, Xu L, Qiu Y, *et al*. Incidence, risk factors, and evolution of proximal Junctional kyphosis after posterior hemivertebra resection and short fusion in young children with congenital scoliosis. Spine (Phila Pa 1976), 2018, 43: 1193–1200.
6. Shono Y, Abumi K, Kaneda K. One-stage posterior hemivertebra resection and correction using segmental posterior instrumentation. Spine (Phila Pa 1976), 2001, 26: 752–757.
7. Nakamura H, Matsuda H, Konishi S, Yamano Y. Single-stage excision of hemivertebrae via the posterior approach alone for congenital spine deformity: follow-up period longer than ten years. Spine (Phila Pa 1976), 2002, 27: 110–115.
8. Zhang J, Shengru W, Qiu G, Yu B, Yipeng W, Luk KD. The efficacy and complications of posterior hemivertebra resection. Eur Spine J, 2011, 20: 1692–1702.
9. Shi Z, Li Q, Cai B, *et al*. Causes of the failure and the revision methods for congenital scoliosis due to hemivertebra. Congenit Anom (Kyoto), 2015, 55: 150–154.
10. Yang X, Song Y, Liu L, *et al*. Emerging S-shaped curves in congenital scoliosis after hemivertebra resection and short segmental fusion. Spine J, 2016, 16: 1214–1220.
11. Ruf M, Harms J. Posterior hemivertebra resection with transpedicular instrumentation: early correction in children aged 1 to 6 years. Spine (Phila Pa 1976), 2003, 28: 2132–2138.
12. Xue X, Zhao S. Revision surgery for lumbar hemivertebra in a 7-year-old child with 10-year follow-up-a case report: A CARE-compliant article. Medicine (Baltimore), 2017, 96: e8794.
13. Callahan BC, Georgopoulos G, Eilert RE. Hemivertebral excision for congenital scoliosis. J Pediatr Orthop, 1997, 17: 96–99.
14. Wang S, Aikenmu K, Zhang J, *et al*. The aim of this retrospective study is to evaluate the efficacy and safety of posterior-only vertebral column resection (PVCR) for the treatment of angular and isolated congenital kyphosis. Eur Spine J, 2017, 26: 1817–1825.
15. Kavadi N, Tallarico RA, Lavelle WF. Analysis of instrumentation failures after three column osteotomies of the spine. Scolios Spinal Disord, 2017, 12: 19.
16. La Barbera L, Brayda-Bruno M, Liebsch C, *et al*. Biomechanical advantages of supplemental accessory and satellite rods with and without interbody cages implantation for the stabilization of pedicle subtraction osteotomy. Eur Spine J, 2018, 27: 2357–2366.
17. Yang BP, Ondra SL, Chen LA, Jung HS, Koski TR, Salehi SA. Clinical and radiographic outcomes of thoracic and lumbar pedicle subtraction osteotomy for fixed sagittal imbalance. J Neurosurg Spine, 2006, 5: 9–17.
18. Zhu ZZ, Chen X, Qiu Y, *et al*. Adding satellite rods to standard two-rod construct with the use of duet screws: an effective technique to improve surgical outcomes and preventing proximal junctional kyphosis in posterior-only correction of Scheuermann kyphosis. Spine (Phila Pa 1976), 2018, 43: E758–E765.
19. Wang T, Liu H, Zheng Z, *et al*. Biomechanical effect of 4-rod technique on lumbosacral fixation: an in vitro human cadaveric investigation. Spine (Phila Pa 1976), 2013, 38: E925–E929.
20. Xu L, Chen X, Qiao J, *et al*. Coronal imbalance after three-column osteotomy in thoracolumbar congenital kyphoscoliosis: incidence and risk factors. Spine (Phila Pa 1976), 2019, 44: E99–E106.
21. Bao H, Liu Z, Zhang Y, *et al*. Sequential correction technique to avoid postoperative global coronal decompensation in rigid adult spinal deformity: a technical note and preliminary results. Eur Spine J, 2019, 28: 2179–2186.
22. Polly DW Jr, Rosner MK, Monacci W, Moquin RR. Thoracic hemivertebra excision in adults via a posterior-only approach. Report of two cases. Neurosurg Focus, 2003, 14: e9.
23. Zhuang Q, Zhang J, Li S, Wang S, Guo J, Qiu G. One-stage posterior-only lumbosacral hemivertebra resection with short segmental fusion: a more than 2-year follow-up. Eur Spine J, 2016, 25: 1567–1574.