Clinical Case Report

Medicine®

OPEN

Posterior-only vertebral column resection for revision surgery in post-laminectomy rotokyphoscoliosis associated with late-onset paraplegia

A case report and literature review

Youping Tao, MD, Jigong Wu, MD, PhD*, Huasong Ma, MD

Abstract

Rationale: Severe post-laminectomy spinal deformity associated with late-onset paraplegia is a complex and rare disorder. Little is known about revision surgery in post-laminectomy rotokyphoscoliosis associated with late-onset paraplegia treated by the single stage posterior-only vertebral column resection (VCR) procedure.

Patient concerns and diagnoses: The patient was a 14-year-old male diagnosed as post-laminectomy rotokyphoscoliosis associated with late-onset paraplegia. He underwent posterior total laminectomy through the thoracic spine for intramedullary spinal cord tumors at the age of 3 years in another hospital. He then developed kyphosis deformity 1 year after laminectomy, and underwent posterior spinal fusion without instrumentation at 9 years of age. However, the deformity gradually progressed over the years. Seven months before admission to our hospital, he developed a significant progression of neurological deficits, including weakness of strength and sensation in lower extremities bilaterally, with no bladder or bowel dysfunction. There was no improvement of spinal cord function with conservative measures, and he required a wheelchair for movement.

Interventions: The patient underwent posterior-only VCR by single stage with the purposes of spinal cord decompression and spinal deformity correction.

Outcomes: Postoperatively, he was transferred to the intensive care unit (ICU) and required positive pressure ventilation support to improve his respiratory condition. The child experienced cerebrospinal fluid leak (CSF) which resulted in an unplanned return to the operating room. The neurological function improved from preoperative Frankel C to Frankel D within 12 months of surgery, and recovered completely to Frankel E by 18 months. At the 24 month follow-up, the good neurological function was maintained; pulmonary function tests (PFTs) revealed improved forced vital capacity (FVC) and forced expiratory volume for 1 second (FEV1). The patient's coronal major curve and sagittal kyphosis were corrected from 70° to 21°, and 170° to 75°, respectively.

Lessons: These findings demonstrated that single-stage posterior-only VCR is efficacious but challenging for revision surgery in post-laminectomy rotokyphoscoliosis associated with late-onset paraplegia.

Abbreviations: 3D CT = three-dimensional-computed tomography, FEV 1 = forced expiratory volume in 1 second, FVC = forced vital capacity, MEP = motor evoked potential, MRI = magnetic resonance imaging, PFTs = pulmonary function tests, SEP = somatosensory evoked potential, VCR = vertebral column resection.

Keywords: paraplegia, posterior vertebral column resection, post-laminectomy rotokyphoscoliosis, revision

1. Introduction

Spinal cord tumors are not common and account for 1 to 10% of all pediatric central nervous system tumors.[1–3] Most intramedullary spinal cord tumors require surgical treatment as early as possible.[4] Spinal deformity following posterior laminectomy for spinal cord tumors in children has been reported by a number of studies.[5–14] When the spinal deformity is corrected with a plaster cast or only posterior fusion without instrumentation, the deformity would exhibit progression and require revision operation.[7] In 2012, Duman et al[10] first reported a case of post-laminectomy rotokyphoscoliosis causing late-onset paraplegia. However, the patient declined surgical intervention, and no neurological status improvement was obtained.

It is widely accepted that limited correction may be provided without osteotomy in severe spinal fusion deformity.[15] Posterior spinal osteotomy procedures, such as Smith Peterson, Ponte, and pedicle subtraction osteotomy (PSO) may not yield a great amount of deformity correction in very severe spinal deformity cases.[15,16] The vertebral column resection (VCR) technique may provide an effective option to correct these severe and rigid spinal deformities with limited flexibility. The conventional VCR procedure can be performed by 2-stage anterior and posterior

Editor: Johannes Mayr.

The authors have no funding and conflicts of interest to disclose.

Department of Orthopedic Surgery, the 306th Hospital of People's Liberation Army (PLA), Beijing, China.

** Correspondence: Jigong Wu, igong Wu, Department of Orthopedic Surgery, the 306th Hospital of People's Liberation Army (PLA), Beijing 100101, China (e-mail: docwjg@126.com).*

Medicine (2017) 96:1(e5690)

Received: 27 July 2016 / Received in final form: 28 November 2016 / Accepted: 29 November 2016

http://dx.doi.org/10.1097/MD.0000000000005690

vertebral column resection, fusion, and segmental spinal instrumentation.[17,18] In 2002, Suk et al[19] first introduced posterior-only VCR, which was popularized by Lenke's team.[20] Compared with combined anterior and posterior VCR, the posterior-only VCR technique is a single procedure and obviates the need to open the thoracic cavity, with additional negative effects on pulmonary function. More recently, several investigators have reported experiences with posterior-only VCR for severe and rigid spinal deformity, in both primary and revision patients.[15,21–27]

However, to the best of our knowledge, studies applying posterior-only VCR for revision surgery in postlaminectomy rotokyphoscoliosis associated with late-onset paraplegia are scarce. We here present a rare case of postlaminectomy rotokyphoscoliosis associated with late-onset paraplegia, who successfully underwent revision surgery by single-stage posterior-only VCR.

2. Consent

Written informed consent was obtained from the patient's parents on behalf of the child, for the publication of this case report and any accompanying images. A copy of the written consent is available for review by the editor of this journal.

3. Case report

A 14-year-old male was admitted to our clinic for treatment of extremely severe spinal deformity associated with progressive late-onset paraplegia. His medical history included the diagnosis of intramedullary astrocytoma at the age of 3 years in another hospital, where he underwent posterior total laminectomy through the thoracic spine (from T4 to T8), with tumor excision performed without fusion. Intramedullary astrocytoma represents 29% of pediatric spinal cord tumors in the literature.[28]

Figure 1. The patient could not walk independently and required a wheelchair for movement.

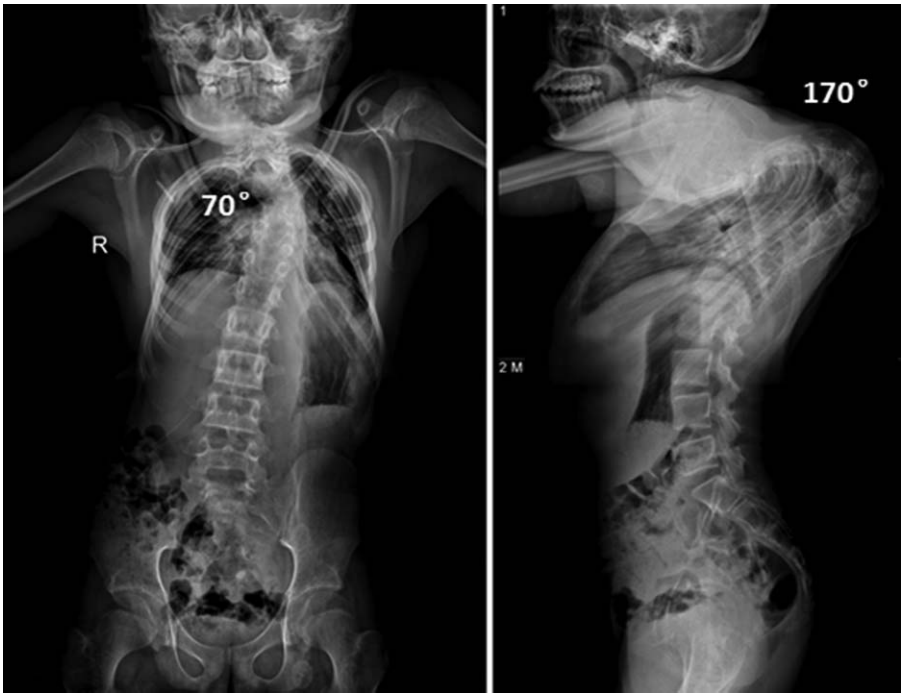

Figure 2. Preoperative scoliosis radiography showed coronal and thoracic kyphosis Cobb's angles of 70° and 170°, respectively.

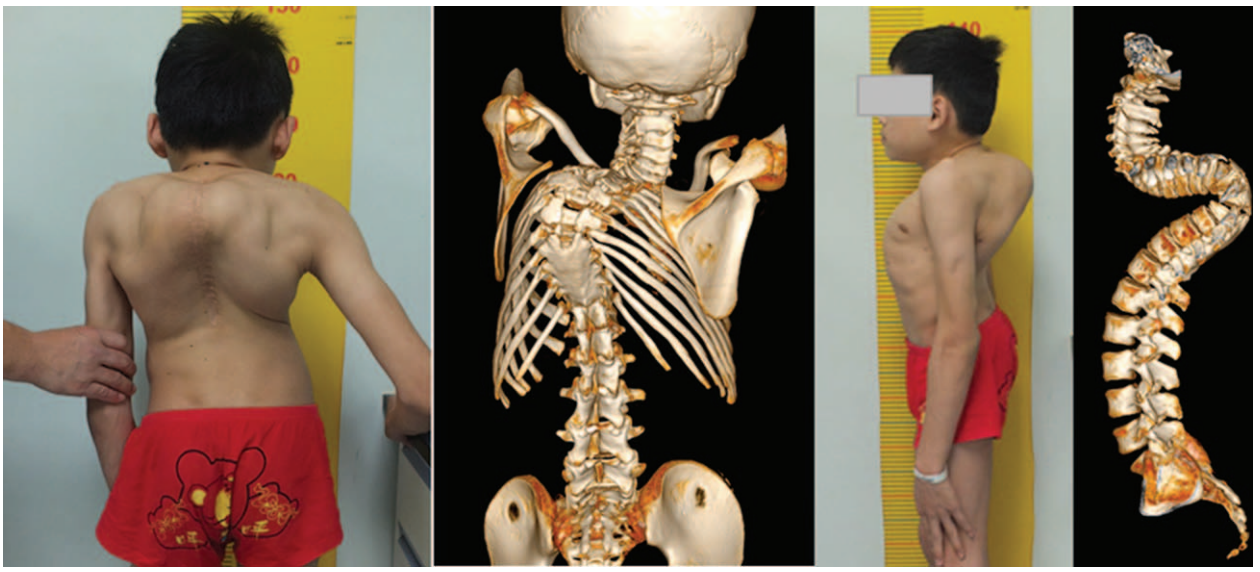

Figure 3. Preoperative 3D CT scans showing extremely severe postlaminectomy thoracic kyphotic deformity and posterior spinal fusion. 3D CT = three-dimensional-computed tomography.

He reported that kyphosis deformity began to appear 1 year after posterior laminectomy. Later, he underwent posterior spinal fusion without any instrumentation at 9 years of age. However, the deformity gradually progressed over the years. Seven months before admission to our hospital, the patient began to feel lack of strength and sensation in lower extremities bilaterally, with no complains regarding bladder or bowel dysfunction. No improvement in spinal cord function was achieved with conservative measures, and the patient gradually lost the ability to walk and required a wheelchair for movement (Fig. 1). The patient and his family denied any recent or remote history of trauma, tuberculosis, or any other infections. The family history was unremarkable; meanwhile, the patient exhibited signs of serious psychosocial problems since he had to use a wheelchair in daily life.

The patient's preoperative coronal and sagittal thoracic kyphosis Cobb's angles were 70° and 170°, respectively (Fig. 2). Preoperative whole spine three-dimensional-computed tomography (3-D CT) scans showed extremely severe post-laminectomy thoracic kyphotic deformity; with the apex at T6 vertebra and previous spinal fusion (Fig. 3); magnetic resonance imaging (MRI) of the spinal canal revealed spinal cord compression at the apex of the kyphosis (Fig. 4). There was no neurological abnormality such as split cord malformation or tethered cord. The electrodiagnostic investigation demonstrated neurological deficit. Pulmonary function tests (PFTs) revealed forced vital capacity (FVC) and forced expiratory volume for 1 second (FEV1) of 31.8% and 33.8%, respectively. Cardiac Doppler ultrasonographic investigation demonstrated mitral and tricuspid regurgitation, with no anomalies of other visceral organs.

Physical examination showed skin hypoesthesia below the T6 level and lower limb spasticity. The patient had weakness of the intrinsic musculature of lower limbs, with left and right lower extremity Grade 3/5 and Grade 1/5 motor strength in all muscle groups, respectively. His Achilles and knee tendons showed hyperreflexia, and the Babinski sign was positive. The overall spinal cord function was classified as Frankel C.

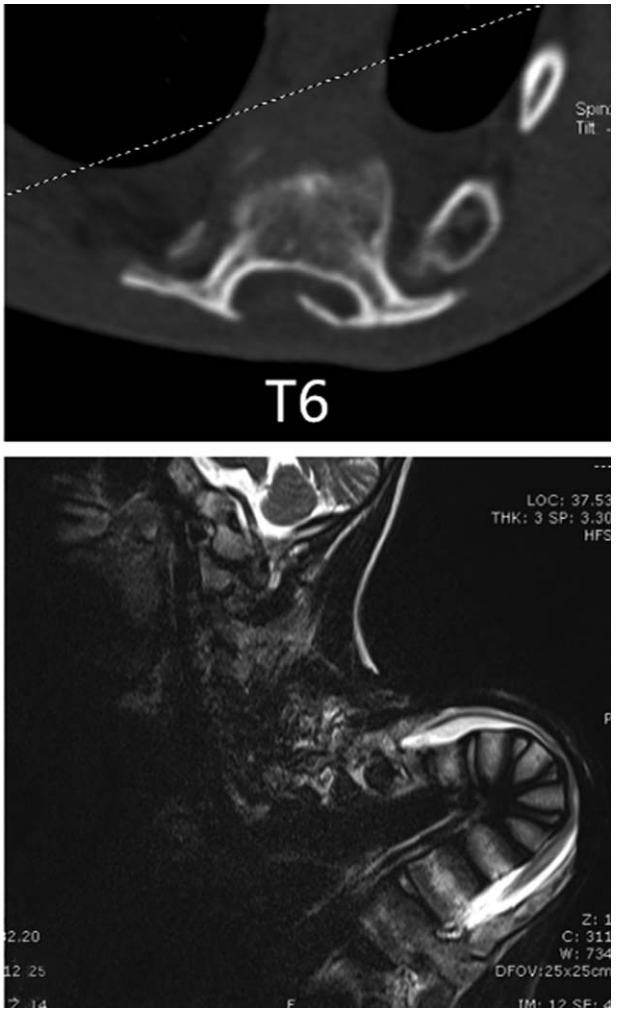

Figure 4. Magnetic resonance imaging (T2-weighted) and CT scans of the spinal canal revealing spinal cord compression at the apex of the kyphosis in the thoracic spine (T6). CT = computed tomography.

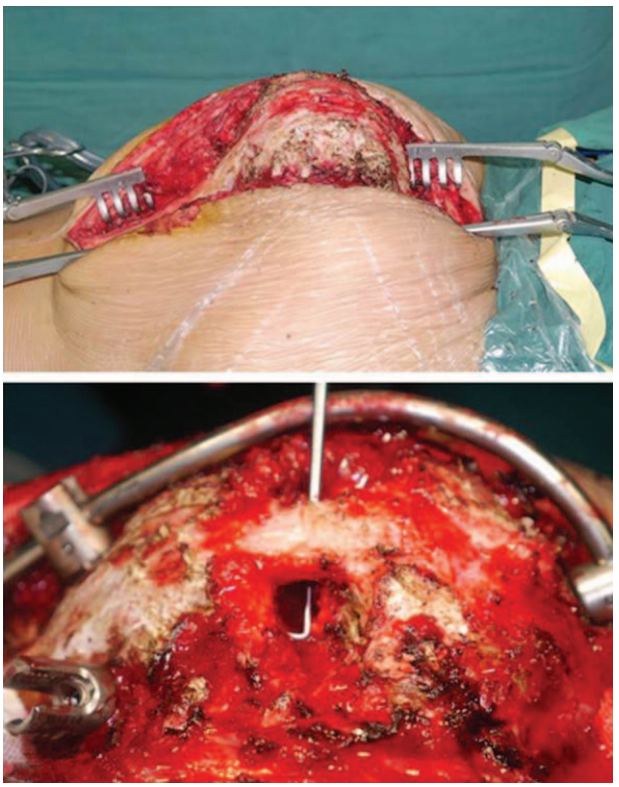

Figure 5. Intra-operative photograph showing posterior vertebral column resection (T6) performed and posterior spinal fusion with pedicle screw fixation from T2 to L3.

In 2014, the patient underwent preoperative assessment by a team of pediatrics, neurosurgery, anesthesiology, and respiratory specialists, and diagnosis of postlaminectomy rotokyphoscoliosis associated with late-onset paraplegia and severe pulmonary ventilation disorder were confirmed based on the findings. The patient was suggested a revision surgical treatment, and the single stage posterior-only VCR procedure was scheduled. In brief, pedicle screws were inserted using the free-hand technique based on previous studies.[29,30] The patient underwent a VCR at T6 for spinal cord decompression, deformity correction, and posterior spinal fusion from T2 to L3 (Fig. 5). The autograft and allograft were used for posterior final spine fusion. During the operation, motor evoked potential (MEP), somatosensory evoked potential (SEP), and wake up test were performed to assess intraoperative complications of spinal cord injury.

The operation time was 450 minutes, and blood loss was 1500 mL. Intraoperatively, a dural tear was noted and repaired; MEP and SEP signals were lost during the entire operation, and the wake-up test was positive. Postoperatively, the patient was transferred to the intensive care unit (ICU) and required positive pressure ventilation support to improve his respiratory condition. On postoperative day 2, he was extubated and transferred to the orthopedic ward, where he received neurotrophic medicines and rehabilitation exercises. The patient required a chest tube for postoperative pleural effusion. The child experienced unplanned return to the operating room for cerebrospinal fluid leak (CSF) on postoperative day 18. The plastic thoracolumbosacral orthosis braces were preserved for 6 months after revision operation.

The spinal cord function improved gradually after the posterior-only VCR procedure. At 3 months, left and right intrinsic musculature of lower limbs both exhibited Grade 3/5 motor strength in all muscle groups. Left and right intrinsic musculature of lower limbs recovered to Grade 4/5 motor

Figure 6. At the final follow-up, long-standing scoliosis radiographs revealed the coronal main curve was corrected to 21° and sagittal kyphosis curve to 75°.

strength at 12 months postoperatively. At 18 months postsurgery (follow-up visit), intrinsic musculature of lower limbs displayed Grade 5/5 motor strength, and sensation in lower extremities bilaterally had returned to normal, with the neurological status improving from preoperative Frankel C to Frankel E. At the 24-month follow-up, the good spinal cord function was maintained. Long-standing scoliosis radiographs demonstrated that the major curve at coronal plane was improved to 20° immediately after surgery and 21° (correction rate, 70%) at final follow-up; the thoracic sagittal kyphosis curve was improved to 76° immediately after surgery and 75° (correction rate, 55.9%) at the final follow-up (Fig. 6). PFTs revealed FVC and FEV_1 of 40.5% and 43.5%, respectively. Postoperative clinical photographs showed overt improvement (Fig. 7).

4. Discussion

Several reports have described spinal deformity as a common complication after laminectomy to remove spinal tumors. In 1965, Tachdjian and Matson[13] reported spinal deformities in 26% of infants and children treated with laminectomy for intraspinal tumors. In 2005, de Jonge et al[7] reported that of 76 children with malignant tumors treated with laminectomy or laminoplasty and/or radiation therapy, 67 developed late-onset spinal deformity. Spinal deformities following laminectomy can be prevented by surgical and nonsurgical treatments; however, when corrected with only posterior fusion without instrumentation, post-laminectomy spinal deformities may progress during the period of rapid growth in children.[3] The present patient had undergone posterior spinal fusion without instrumentation, with spinal deformity gradually progressing over the years; the boy developed paraplegia due to severe postlaminectomy spinal deformity.

Figure 7. Clinical photographs showing overt improvement at the final follow-up.

In 2005, Suk et al[22] reported the use of posterior-only VCR in 16 patients with severe and rigid spinal deformity (scoliosis more than 80°); mean preoperative scoliosis of 109.0° was corrected to 45.6°, with a correction rate of 59%. In 2009, Lenke et al[20] reported that posterior-only VCR is a safe but challenging technique for severe primary or revision pediatric spinal deformities (58% coronal correction and 53% sagittal correction). In the present case, coronal and sagittal correction rates were 70% and 55.9%, respectively.

Revision surgical treatment for severe and rigid spinal deformity poses challenges to spine surgeons, due to the absence of bony landmarks, high risks of neurological deficit, and high blood loss or long surgery time.[30] Glassman et al[31] showed that patients are more likely to have complications if they had undergone previous spine surgery. Previous studies described revision surgical treatments for severe pediatric or adult deformity that were performed with staged posterior procedures or combined anterior/posterior surgeries.[23,32,33] Although staged or combined anterior/posterior procedures for revision surgeries are effective, staged procedures expose patients to anesthetic risks and operation can be performed more than once; in addition, the anterior/posterior spinal fusion could open the thoracic cavity, with further negative effects on pulmonary function. The current findings revealed that single stage posterior-only VCR can achieve a similar correction rate for post-laminectomy rotokyphoscoliosis associated with late-onset paraplegia.

Suk et al[22] reported that posterior-only VCR provides adequate chance for spinal canal decompression. The present patient underwent posterior-only VCR for spinal deformity correction and spinal cord decompression at the apex of kyphosis. Postoperatively, although he experienced major surgical complications, including pleural effusion, CSF, sensation and intrinsic musculature in lower extremities bilaterally gradually improved to normal. He has been able to walk independently and participate in normal daily activities with a high degree of satisfaction.

In 2004, Bumpass et al[34] reported small but statistically significant increases in absolute pulmonary measures in pediatric patients after posterior-only VCR, especially those with angular kyphosis most strongly associated with improved postoperative pulmonary function. In the present case, postoperative PFTs revealed that FVC and FEV1 were overtly improved.

The strength of this article is that after single stage posterior-only VCR with no anterior approach for revision surgical treatment of postlaminectomy rotokyphoscoliosis associated with late-onset paraplegia, the neurological status improved from preoperative Frankel C to Frankel E. Postoperatively, the child experienced improvement of pulmonary function, and overt coronal and sagittal correction rates. As limitations, the retrospective nature of this study and the relatively short follow-up should be mentioned. Longer follow-up is warranted to assess postoperative clinical and radiographic results.

5. Conclusion

Overall, our results revealed that posterior laminectomy for treatment of spinal cord tumors in childhood can lead to spinal deformity. When the patient undergoes posterior spinal fusion without instrumentation, spinal deformity may gradually progress during the periods of fast growth. Extremely severe postlaminectomy thoracic kyphotic deformity is relatively complex and rare, and could lead to serious neurological complications, including paraplegia. Single-stage posterior-only VCR might be an efficacious but challenging option for revision surgery in postlaminectomy rotokyphoscoliosis associated with late-onset paraplegia.

References

[1] Stiller CA, Nectoux J. International incidence of childhood brain and spinal tumours. Int J Epidemiol 1994;23:458–64.
[2] Hardison HH, Packer RJ, Rorke LB, et al. Outcome of children with primary intramedullary spinal cord tumors. Childs Nerv Syst 1987;3: 89–92.
[3] Nadkarni TD, Rekate HL. Pediatric intramedullary spinal cord tumors. Critical review of the literature. Childs Nerv Syst 1999;15:17–28.
[4] Yang S, Yang X, Hong G. Surgical treatment of one hundred seventy-four intramedullary spinal cord tumors. Spine 2009;34:2705–10.
[5] Raab P, Juergen K, Gloger H, et al. Spinal deformity after multilevel osteoplastic laminotomy. Int Orthop 2008;32:355–9.
[6] Lonstein JE. Post-laminectomy kyphosis. Clin Orthop Relat Res 1977; 128:93–100.
[7] de Jonge T, Slullitel H, Dubousset J, et al. Late-onset spinal deformities in children treated by laminectomy and radiation therapy for malignant tumours. Eur Spine J 2005;14:765–71.
[8] Papagelopoulos PJ, Peterson HA, Ebersold MJ, et al. Spinal column deformity and instability after lumbar or thoracolumbar laminectomy for intraspinal tumors in children and young adults. Spine 1997;22: 442–51.
[9] Peter JC, Hoffman EB, Arens LJ, et al. Incidence of spinal deformity in children after multiple level laminectomy for selective posterior rhizotomy. Childs Nerv Syst 1990;6:30–2.
[10] Duman I, Guzelkucuk U, Yilmaz B, et al. Post-laminectomy rotokyphoscoliosis causing paraplegia in long term: case report. J Spinal Cord Med 2012;35:175–7.
[11] McGirt MJ, Chaichana KL, Atiba A, et al. Incidence of spinal deformity after resection of intramedullary spinal cord tumors in children who underwent laminectomy compared with laminoplasty. J Neurosurg Pediatr 2008;1:57–62.
[12] Arkin AM, Simon N. Radiation scoliosis. J Bone Joint Surg Am 1950; 32:396–404.
[13] Tachdjian MO, Matson DD. Orthopaedic aspects of intraspinal tumors in infants and children. J Bone Joint Surg Am 1965;47:223–48.
[14] O'Sullivan C, Jenkin RD, Doherty MA, et al. Spinal cord tumors in children: long-term results of combined surgical and radiation treatment. J Neurosurg 1994;81:507–12.
[15] Ozturk C, Alanay A, Ganiyusufoglu K, et al. Short-term X-ray results of posterior vertebral column resection in severe congenital kyphosis, scoliosis, and kyphoscoliosis. Spine 2012;37:1054–7.
[16] Cho KJ, Bridwell KH, Lenke LG, et al. Comparison of Smith-Petersen versus pedicle subtraction osteotomy for the correction of fixed sagittal imbalance. Spine 2005;30:2030–7.
[17] Bradford DS, Tribus CB. Vertebral column resection for the treatment of rigid coronal decompensation. Spine 1997;22:1590–9.
[18] Boachie-Adjei O, Bradford DS. Vertebral column resection and arthrodesis for complex spinal deformities. J Spinal Disord 1991;4: 193–202.
[19] Suk SI, Kim JH, Kim WJ, et al. Posterior vertebral column resection for severe spinal deformity. Spine 2002;27:2374–82.
[20] Lenke LG, O'Leary PT, Bridwell KH, et al. Posterior vertebral column resection (VCR) for severe pediatric deformity: minimum 2-year follow-up of 35 consecutive patients. Spine 2009;34:2213–21.
[21] Lenke LG, Sides BA, Koester LA, et al. Vertebral column resection for the treatment of severe spinal deformity. Clin Orthop Relat Res 2010;468: 687–99.
[22] Suk SI, Chung ER, Kim JH, et al. Posterior vertebral column resection for severe rigid scoliosis. Spine 2005;30:1682–7.
[23] Lenke LG, Newton PO, Sucato DJ, et al. Complications after 147 consecutive vertebral column resections for severe pediatric spinal deformity: a multicenter analysis. Spine 2013;38:119–32.
[24] Tao Y, Wu J, Ma H, et al. Posterior vertebral column resection for severe and rigid spinal deformity associated with neurological deficit after implant removal following posterior instrumented fusion: a case report and literature review. Spine 2015;40:E794–8.
[25] Xie J, Zhang Y, Wang Y, et al. The risk factors of neurologic deficits of one-stage posterior vertebral column resection for patients with severe and rigid spinal deformities. Eur Spine J 2014;23:149–56.
[26] Xie J, Wang Y, Zhao Z. Posterior vertebral column resection for correction of rigid spinal deformity curves greater than 100°. J Neurosurg 2012;17:540–51.
[27] Stoker GE, Lenke LG, Dorward IG. Posterior vertebral column resection for the treatment of dystrophic kyphosis associated with type-1 neurofibromatosis: a case report and review of the literature. Spine 2012;37:E1659–64.
[28] Wilson PE, Oleszek JL, Clayton GH. Pediatric spinal cord tumors and masses. J Spinal Cord Med 2007;30(suppl 1):S15–20.
[29] Kim YJ, Lenke LG, Bridwell KH, et al. Free hand pedicle screw placement in the thoracic spine: is it safe? Spine 2004;29:333–42.
[30] Yang JH, Suh SW, CW, et al. Effect of posterior multilevel vertebral osteotomies on coronal and sagittal balance in fused scoliosis deformity caused by previous surgery: preliminary results. Spine 2014;39:1840–9.
[31] Glassman SD, Hamill CL, Bridwell KH. The impact of perioperative complications on clinical outcome in adult deformity surgery. Spine 2007;32:2764–70.
[32] Rhee JM, Bridwell KH, Lenke LG, et al. Staged posterior surgery for severe adult spinal deformity. Spine 2003;28:2116–21.
[33] Bridwell KH, Lewis SJ, Lenke LG, et al. Pedicle subtraction osteotomies for fixed sagittal imbalance. J Bone Joint Surg Am 2003;85:454–63.
[34] Bumpass DB, Lenke LG, Bridwell KH, et al. Pulmonary function improvement after vertebral column resection for severe spinal deformity Spine 2014;39:587–95.