



Review Article

Surgical correction of pediatric spinal deformities with coexisting intraspinal pathology: A case report and literature review

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ABSTRACT

Background: Surgical correction of spinal deformities with coexisting intraspinal pathology (SDCIP) requires special consideration to minimize risks of further injury to an already abnormal spinal cord. However, there is a paucity of literature on this topic. Here, the authors present a pediatric patient with a residual pilocytic astrocytoma and syringomyelia who underwent surgical correction of progressive postlaminectomy kyphoscoliosis. Techniques employed are compared to those in the literature to compile a set of guidelines for surgical correction of SDCIP.

Methods: A systematic MEDLINE search was conducted using the following keywords; “pediatric,” “spinal tumor resection,” “deformity correction,” “postlaminectomy,” “scoliosis correction,” “intraspinal pathology,” “tethered cord,” “syringomyelia,” or “diastematomyelia.” Recommendations for surgical technique for pediatric SDCIP correction were reviewed.

Results: The presented case demonstrates recommendations that primarily compressive forces on the convexity of the coronal curve should be used when performing *in situ* correction of SDCIP. Undercorrection is favored to minimize risks of traction on the abnormal spinal cord. The literature yielded 13 articles describing various intraoperative techniques. Notably, seven articles described use of compressive forces on the convex side of the deformity as the primary mode of correction, while only five articles provided recommendations on how to safely and effectively surgically correct SDCIP.

Conclusion: The authors demonstrated with their case analysis and literature review that there are no clear current guidelines regarding the safe and effective techniques for *in situ* correction and fusion for the management of pediatric SDCIP.

Keywords: Intramedullary spinal tumor, Intraspinal pathology, Kyphoscoliosis, Pediatric spinal deformity correction

INTRODUCTION

Surgical techniques for the correction of idiopathic pediatric scoliotic deformities are well documented. However, various intraspinal pathologies, such as diastematomyelia, tethered cord, syringomyelia, and intramedullary spinal cord tumors (IMSCTs), create a unique set of

considerations to be addressed during surgical planning and management of these three-dimensional spinal deformities. Unfortunately, there is little documentation of the appropriate surgical technique and guidelines for safely performing correction of spinal deformities with coexisting intraspinal pathology (SDCIP).

Here, the authors reviewed the literature on surgical techniques/recommendations for managing pediatric SDCIP, while also presenting an illustrative case of progressive postlaminectomy kyphoscoliosis in a pediatric patient 9 years after subtotal resection of an intramedullary spinal cord tumor (IMSCT).

MATERIALS AND METHODS

A pediatric case of SDCIP with IMSCT was reviewed and accompanied by a literature search of MEDLINE using of the keywords; “pediatric,” “intradural spinal tumor,” “spinal tumor resection,” “deformity correction,” “postlaminectomy,” “intraspinal pathology,” “tethered cord,” “syringomyelia,”

or “diastematomyelia” [Figure 1]. Exclusion and inclusion criteria for the review are described in Table 1.

CASE REPORT

History

In 2007, at the age of 2, the patient underwent thoracic six (T6) to lumbar two (L2) laminectomies, for subtotal resection (STR) of a pilocytic astrocytoma with expansion duraplasty [Figure 2] followed by adjuvant chemotherapy. Postoperatively, her only neurologic deficit was a neurogenic bladder, which required intermittent self-catheterization.

At 5 years of age, the patient had shoulder asymmetry in the horizontal plane and an increased thoracic kyphosis that was initially managed conservatively with external bracing. Over the next few years, the patient’s kyphoscoliosis continued to progress [Figure 3], while interval MRIs demonstrated gross stability of her IMSCT [Figure 4]. By 2016, despite stability of her intraspinal tumor pathology, serial standing scoliosis

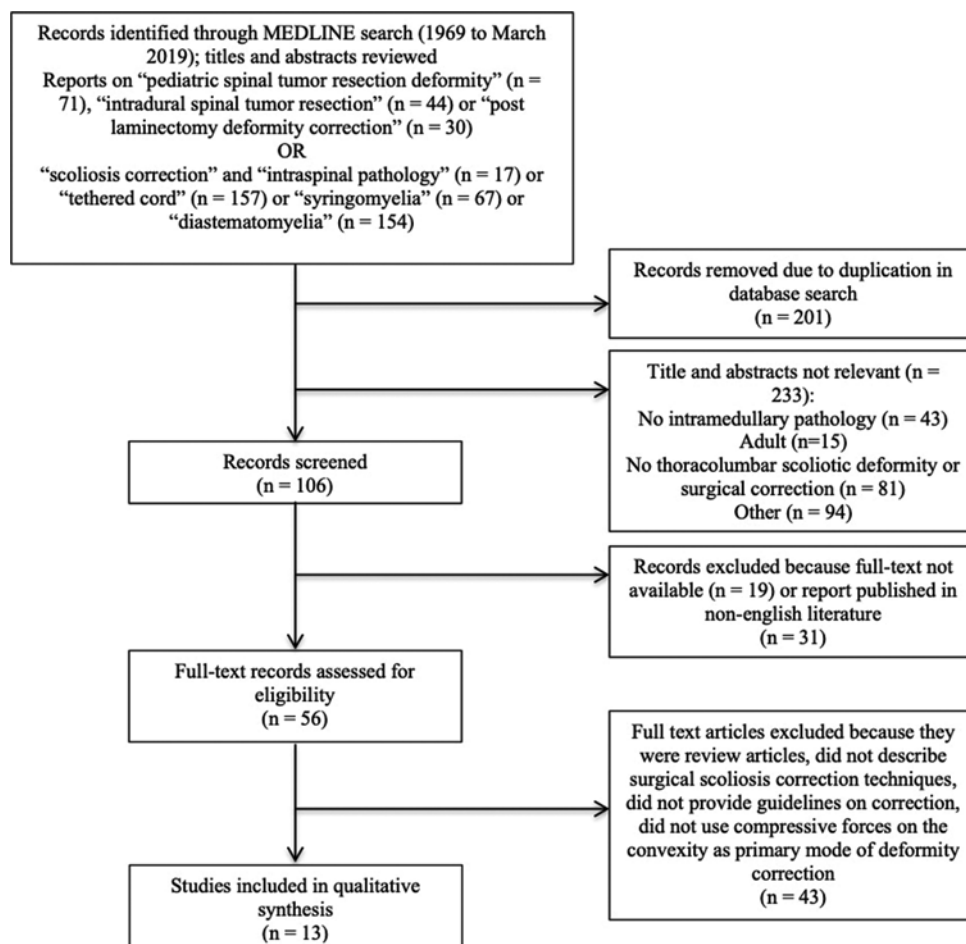


Figure 1: (a) PRISMA flow diagram of citations identified and evaluated on literature review of scoliotic deformity correction in patients after spinal cord tumor resection. (b) PRISMA flow diagram of citations identified and evaluated on literature review of scoliotic deformity correction in patients with other intraspinal pathology.

Table 1: Systematic review inclusion and exclusion criteria.

Inclusion criteria	<ul style="list-style-type: none"> • Age \leq21 years old • Patients with a history of IMSCT resection with kyphoscoliosis • Patients with congenital scoliosis and intraspinal pathology (tethered cord, diastematomyelia, Chiari malformation with syrinx, and syringomyelia) in which compressive forces were applied on the convexity or the authors provided guidelines for surgical correction of kyphoscoliosis • Patients who underwent posterior approach for correction of kyphoscoliosis
Exclusion criteria	<ul style="list-style-type: none"> • Patients >21 years of age at the time of presentation or surgery • Surgical techniques employed during correction of kyphoscoliosis were not described • Cases in which the patient does not have a three-dimensional deformity • The article is not published in the English literature • There is not full article to review

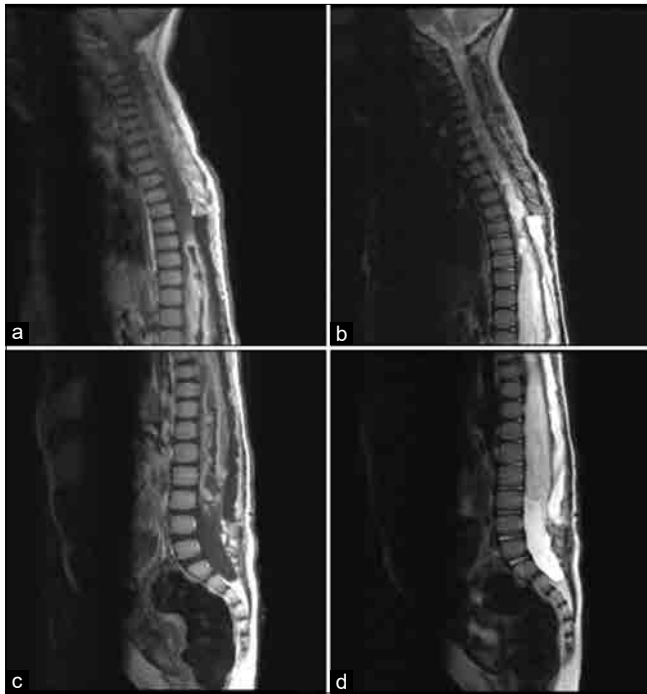


Figure 2: T1 postcontrast thoracic (a) and lumbar (b) and T2 noncontrast thoracic (c) and lumbar (d) sagittal midline views of patient's postoperative MRI demonstrate an expansile, intramedullary astrocytoma extending from T7 to L3 is demonstrated with associated syringohydromyelia extending from C2 to T7 with a remodeled and expanded spinal canal after decompressive laminectomies were performed from T7 to L3.

X-rays demonstrated interval progression of her deformity (i.e., with levoscoliosis of her T spine measuring 48.3°

(previously 38.9° from T4-T11), dextroscoliosis of the L spine measuring 43° (previously 31.8° from T11-L5), and thoracic kyphosis (now 81°). Having reached skeletal maturity [Figure 3e], the patient underwent surgical correction of her progressive kyphoscoliosis, in December 2016. Preoperative standing scoliosis and lateral bending X-rays were used to plan the extent of her fusion [Figure 5], extending posteriorly from T3-L2.

Operation

Laminectomies and facetectomies from T3-T12 ensured mobility of thoracic spine and extensive Smith-Petersen type osteotomies were undertaken from T5 to T11 to aid with correction of the kyphotic deformity. Pedicle screw fixation was performed and 5.5 mm CoCh rods, placed bilaterally from T3 to L2. Correction of the patient's coronal and sagittal deformity was accomplished utilizing a combination of cantilever and *in situ* compression maneuvers. The patient's coronal deformity was primarily fixed through compression of the convex aspect of her curve [Figure 6]. Some degree of deformity was retained due to concerns that overcorrection may result in traction on the abnormally enlarged spinal cord. During this process, there was no noted change in the patient's SSEPs or MEPs. A routine wake-up test was performed immediately following the deformity correction with the patient demonstrating satisfactory movement of both legs. An arthrodesis was performed using autograft mixed with morselized allograft. A plastic surgery team assisted with closure of the surgical wound. Following recovery from anesthesia, the patient was confirmed to have a stable neurologic exam with no new deficits.

Postoperative course

The patient's postoperative standing scoliosis radiographs demonstrated significant reduction of her kyphoscoliosis [Figure 7]. Correction of the patient's thoracic kyphotic deformity to within physiologic parameters was accomplished, as well as reduction of her thoracic levoscoliosis from 48° to 26°, her lumbar dextroscoliosis from 43° to 32°, and her kyphosis from 81° to 58°. Her postoperative course was uncomplicated and she was eventually discharged on postoperative day 4. On outpatient follow-up, the patient has recovered well from surgery and her pain has been well controlled with over 2 years of postoperative follow-up.

LITERATURE REVIEW

Our review of the literature yielded four articles that described intraoperative approaches and techniques for the management of postlaminectomy spinal deformity in pediatric patients after IMSCT resection [Table 2].^[2,4,5,26] An additional nine articles were included, describing spinal



Figure 3: Serial standing scoliosis radiographs. (a,d) Preoperative AP and lateral radiographs before laminectomy and tumor resection. (b,e) First postoperative standing long-cassette thoracolumbar radiographs, AP and lateral, obtained in 2010, treated with external bracing. (c,f) Interval AP and lateral standing long-cassette thoracolumbar radiographs obtained in 2015 demonstrating a progressive kyphoscoliosis. (g) Continued progression of scoliosis seen on subsequent AP long-cassette thoracolumbar radiographs obtained in 2016. (h) Hand radiographs obtained to evaluate patient's bone age in 2015, which was determined to be advanced of her chronologic age.

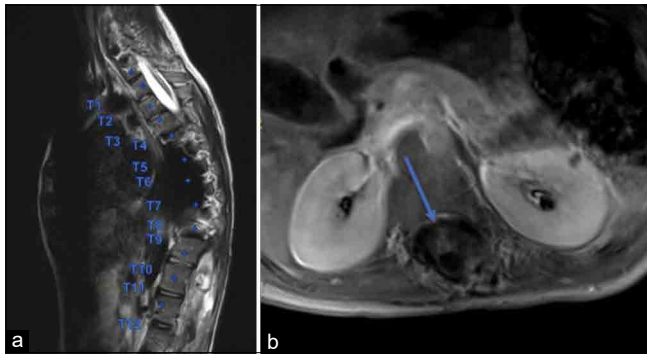


Figure 4: Interval postoperative MRI obtained to evaluate for tumor recurrence or progression revealed new enhancing 6 mm nodule at level of T11/12, deemed clinically insignificant in context of progressive kyphoscoliosis and stable in appearance on subsequent MRIs. (a) T2 noncontrast image of thoracic spine in sagittal plane, with evidence of significant coronal deformity as well as T2 hyperintense cystic lesions in the lower thoracic spine. (b) T1 postcontrast image of the thoracic spine at the inferior aspect of T11 with evidence of enhancing nodule (blue arrow).

deformity correction in patients with congenital scoliosis with intraspinal pathology, such as diastematomyelia, tethered cord, and syringomyelia with or without Chiari malformation [Table 3].^[1,3,8,11,19-21,23,27]

Surgical approaches described varied between single-stage posterior surgery versus staged deformity correction procedures with anterior release followed by posterior

fusion. Nine articles described the use of osteotomies or posterior vertebral column resection to correct kyphoscoliotic deformities without lengthening the spinal column.^[1,2,8,11,19,20,23,26,27] Of all the articles reviewed, seven described the use of compressive forces on the convex side of the deformity as the primary mode of correction.^[5,12,27]

Neuromonitoring, with SSEPs or MEPs, was used in all but two articles reviewed.^[3,23] Wake-up tests were used as the sole mode of neuromonitoring in one article,^[1] due to unavailability of other modalities,^[1] as a routine adjunct in four articles.^[2,20,21,27] Changes in neuromonitoring signals were reported in three patients during intraoperative corrective maneuvers, but did not result in permanent neurologic sequelae.^[2,21] Neurologic complications were reported in six patients (of 203 patients reported total; 2.9%) – three were transient not requiring further intervention,^[1,19,26] while two had permanent new motor deficits,^[3,8] and one whose deficits resolved only after reoperation and revision of instrumentation.^[1]

DISCUSSION

The reported incidence of progressive spinal deformity after childhood resection of IMSCs ranges from 16 to 80%, with increased incidence after laminectomies in the cervical or thoracic spine, and in younger patients.^[6,24,25] Similarly, in cases of congenital scoliosis, early-onset scoliotic deformity may occur in 25–80% of patients with DM, TC, or SM, often with Chiari malformation.^[16,22]



Figure 5: Preoperative standing scoliosis radiographs including right lateral bending (a), upright AP (b), left lateral bending (c), and upright lateral (d) views. These films show a Lenke type 3+ scoliotic curve – levoscoliosis of the thoracic spine with Cobb angle 46° and apex at T8, as well as a dextroscoliosis of the lumbar spine with a Cobb angle of 41° and apex at L2. There was minimal improvement of the patient's coronal deformity on lateral bending. The patient also has a thoracic kyphosis of 80 degrees. The optimal lowest instrumented vertebra was determined by the neutralization of the L2-3 disc space on left-sided bending radiographs (c).

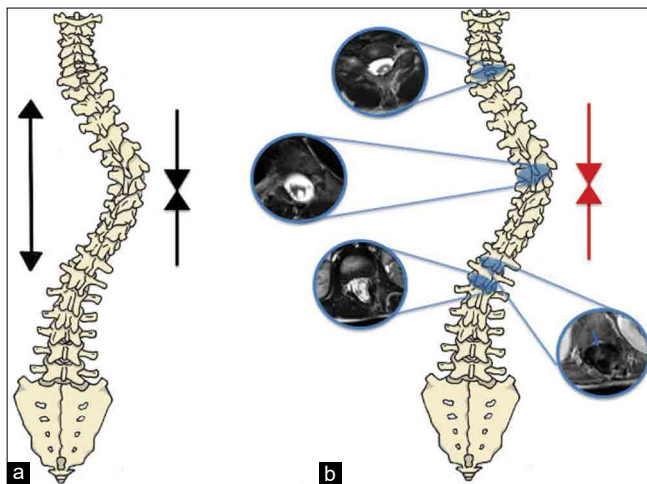


Figure 6: (a) Typical distraction and compressive forces (black arrows) placed along apex of coronal deformity in the absence of intraspinal pathology. (b) Utilization of primarily compressive forces (red arrows) along convexity of the curve to prevent overlengthening the spinal canal in the presence of intraspinal pathology, as was the case in our patient. Select axial T2-weighted noncontrast and one T1-weighted postcontrast MRI cross-sections of her spine highlight progressive intradural tumor (blue circles), cysts, and syringomyelia.

Given the authors' experience, as illustrated in the presented case, and the prevalence of postlaminectomy spinal deformity requiring fusion, the authors sought to review the English literature and discuss surgical techniques currently used, to guide recommendations for safe maximal spinal deformity correction. The authors focused the review primarily on articles

that discussed *in situ* deformity correction.^[14] Overall, there is a paucity of literature describing best surgical technique for surgical fusion and deformity correction in the setting of IMSCT [Table 2]. The majority of articles available in the English literature focus on incidence and risk factors for the development of postlaminectomy kyphoscoliosis in IMSCT patients. Addition of articles inclusive of other intraspinal pathologies, such as diastematomyelia, tethered cord, or syringomyelia with or without Chiari malformation, was still unable to provide substantial insight or guidelines on techniques of surgical correction of pediatric SDCIP [Table 3]. These articles do not explore intraoperative approaches or corrective maneuvers in detail and focus their commentary predominantly on whether there is a need for neurosurgical intervention for varying spinal pathologies before deformity correction.

Having extensively reviewed the literature, the authors of this article would agree with recommendations favoring incomplete correction, rather than overcorrection, due to the increased risks of symptomatic spinal cord pathology. Expansile intraspinal pathology should be decompressed before deformity correction. Notably, a portion of articles in our literature review described rod rotation and translation primarily from the concave side^[2,3,15] or primary use of distracting forces.^[7,10,12] These techniques, which may be routinely employed in surgical correction of idiopathic scoliosis,^[9,13,17,18] may not be as safe for surgical correction of SDCIP.^[3,18] We believe that in the setting of abnormal expansile intraspinal pathology, such as residual low-grade glioma or syringomyelia as in the present case, applying distracting forces [Figure 6] as the primary maneuver for

Table 2: Systematic review of kyphoscoliosis correction in pediatric patients with intraspinal pathology – IMSCT.

Author and year	n	Mean age at initial surgery (range)	Intervention for IMSCT	Spinal deformity	Neurologic status	IOM	Deformity correction	Deformity correction	Complications
De Jonge et al., 2005 ^[4]	46/76	4 y 7 m (2 m-16 y)	Laminectomy or laminoplasty +/- RT, EOR NS	Kyphosis with mild scoliosis (n=15) Scoliosis with hyperrotatory paradoxical kyphosis (n=4) Other/kyphosis (n=48)	NS (ranged from normal to spastic paraplegia)	NS	1-3 w of preoperative halo-traction Surgical fusion in patients without moderate (<70°), nonangular and flexible kyphotic deformities: • Anterior release and tibial strut grafting with 1 w of continuous halo-traction followed by posterior instrumentation • anterior approach at level of concavity of scoliosis • in hyperrotatory paradoxical kyphosis, anterior fusion with bone chips or rib inlay grafting approached from convex side of scoliosis Surgical fusion in less severe, nonangular, flexible thoracic hyperkyphosis: posterior fusion Postoperative external immobilization (6-12 m) Stage 1: anterior thoracolumbar tumor debulking, anterior release, and instrumentation with halofemoral traction Stage 2: posterior instrumentation for correction of kyphoscoliosis 1 level asymmetric wedge t-PSO at apex of curve followed by posterior segmental pedicle screw instrumentation using cantilever techniques, translational techniques, and concave rod rotation. Postoperative TLSO bracing Single stage IMSCT resection (EOR NS) with posterior instrumentation. • VCR (n=1) • SPO (n=1) Preoperative halogravimetry traction for 12 w (n=1; Cobb >100°)	Preoperative scoliosis Cobb 45° (SD 19.8) to postoperative 25° (SD 20.2) Preoperative kyphotic Cobb 76° (SD 22.9) to postoperative 33° (SD 20.1)	Neurological: none Other: delayed infection requiring removal of instrumentation (n=4)
Drake et al., 2010 ^[5]	1	4 y	Laminectomy, STR	Kyphoscoliosis	Bilateral dorsiflexor weakness	SSEP, MEP, EMG	Stage 1: anterior thoracolumbar tumor debulking, anterior release, and instrumentation with halofemoral traction Stage 2: posterior instrumentation for correction of kyphoscoliosis 1 level asymmetric wedge t-PSO at apex of curve followed by posterior segmental pedicle screw instrumentation using cantilever techniques, translational techniques, and concave rod rotation. Postoperative TLSO bracing Single stage IMSCT resection (EOR NS) with posterior instrumentation. • VCR (n=1) • SPO (n=1) Preoperative halogravimetry traction for 12 w (n=1; Cobb >100°)	Deformity correction NS	None
Bakaloudis et al., 2011 ^[2]	1	12.6 y (9-16 y)	Laminectomy, GTR with persistent spinal cord cavitation	Kyphoscoliosis	Paraparesis	SSEP, MEP, EMG, Wake-up test	Stage 1: anterior thoracolumbar tumor debulking, anterior release, and instrumentation with halofemoral traction Stage 2: posterior instrumentation for correction of kyphoscoliosis 1 level asymmetric wedge t-PSO at apex of curve followed by posterior segmental pedicle screw instrumentation using cantilever techniques, translational techniques, and concave rod rotation. Postoperative TLSO bracing Single stage IMSCT resection (EOR NS) with posterior instrumentation. • VCR (n=1) • SPO (n=1) Preoperative halogravimetry traction for 12 w (n=1; Cobb >100°)	Cobb angle correction of 61° (62.3%)	Neurological: intraoperative loss of MEPs during tPSO with prompt loosening of correction and negative wake-up test
Zhang B et al., 2017 ^[26]	6	13	Single stage, EOR NS	Cobb angle 69.57° +/- 20.44° (range: 45-101°) 1/5	RLE weakness	SSEPs, MEPs	Stage 1: anterior thoracolumbar tumor debulking, anterior release, and instrumentation with halofemoral traction Stage 2: posterior instrumentation for correction of kyphoscoliosis 1 level asymmetric wedge t-PSO at apex of curve followed by posterior segmental pedicle screw instrumentation using cantilever techniques, translational techniques, and concave rod rotation. Postoperative TLSO bracing Single stage IMSCT resection (EOR NS) with posterior instrumentation. • VCR (n=1) • SPO (n=1) Preoperative halogravimetry traction for 12 w (n=1; Cobb >100°)	Postoperative correction to 29.14 +/- 9.87° (range: 18-47°) Average deformity correction 55.05% +/- 18.75%	Neurological: transient strength decrease with full recovery (n=1) Other: CSF leak (n=1)

EMG: Electromyography, EOR: Extent of resection, GTR: Gross total resection, IDED: Both intradural and extradural, IDEM: Intradural extramedullary, IMSCT: Intramedullary spinal cord tumor, IOM: Intraoperative monitoring, IQR: Interquartile range, L: Lumbar, m: Months, MEP: Neuro-motor evoked potentials, N/A: Not applicable, NS: Not specified, RT: Radiation therapy, SD: Standard deviation, SSEP: Somatosensory evoked potentials, SPO: Smith-Petersen osteotomy, STR: Subtotal resection, T: Thoracic, TL: Thoracolumbar, TLSO: Thoracolumbosacral orthosis, PFD: Posterior fossa decompression, PSO: Pedicle subtraction osteotomy, VCR: Vertebral column resection, w: Weeks, y: Years; →: Indicates sequential events

Table 3: Systematic review of kyphoscoliosis correction in pediatric patients with intraspinal pathology – DM, SM, and TC.

Author and year	n	Mean age (range)	Intraspinal pathology	Spinal deformity	Neurologic status	IOM	Neurosurgical intervention and deformity correction	Outcome	Complications
*Matsumoto et al., 2009 ^[11]	1	12 y	TC, TL SM, small caudal lipoma	Cobb angle of major curve 103° (T5-L1) with 22.3% flexibility Kyphosis 90° (T3-T12)	Paresthetica pain of BLE, perianal numbness, urinary retention	MEPs	TC release performed 3 months before deformity correction Posterior VCR (Suk technique) <ul style="list-style-type: none"> • Performed at apex (T9) • Freehand placement of pedicle screws with multilevel SPO • Permanent contoured rod placed on convex side and compressive forces applied to close gap between T8-T10 • Procedure performed carefully and slowly with visual observation of neural elements (dural buckling) • Segmental correction with in situ benders and distractive priers 	Cobb angle of major curve 23°, correction 75.7% Kyphosis 36°, correction 60%	None
*Ayvaz et al 2007 ^[1]	22	12 y (7-18 y)	DM type 1 (n=15; staged intervention n=12, single stage n=3) DM type 2 (n=5) SM with TC (n=2)	Cobb angle of major curve 71+/-16.6° (range: 52-114°) Segmental T kyphosis (n=3): 80°, 68°, 53°	8 patients: hyporeflexia in LE, monoparesis	Wake-up test	Posterior only (n=11) Anterior release and fusion followed by posterior instrumentation (n=9) Anterior release and fusion followed by halotraction before posterior instrumentation (n=2) Posterior correction techniques: <ul style="list-style-type: none"> • Shortening of spinal column through osteotomies (chevron n=8, decancellation n=4) and compression rather than distraction • Translation and cantilever correction • Avoid intrusion of hooks or wires into spinal canal at anomalous segments • Combination of hooks and screws • Sublaminar wires (n=4), spinous process wiring (n=9) 	Cobb angle of major curve 40+/-16.3° (range: 12-65°); correction rate 43.6% Segmental T kyphosis (n=3): 40°, 32°, 30°	Neurological: 9% transient (n=2) – one due to misplaced instrumentation who underwent revision, another possibly due to stress on L hemicord during surgery Other: 31% (n=7): superficial infection (n=2), instrumentation removal for infection (n=1), pseudoarthrosis and rod breakage (n=1), late decompensation (n=1)

(Contd...)

Table 3: (Continued).

Author and year	n	Mean age (range)	Intraspinal pathology	Spinal deformity	Neurologic status	IOM	Neurosurgical intervention and deformity correction	Outcome	Complications
*Xie et al., 2011 ^[23]	12	15 y (12–19 y)	CM + SM (cervicothoracic)	Cobb angle of major curve average 88.2° (range: 52–110°; apex: T7-T12) Kyphosis 71.4° (range: 0–128°)	3 patients: hypermyotonia (n=2), anal sphincter relaxation (n=1)	None	Posterior VCR (modified Suk and Lenke technique) <ul style="list-style-type: none"> • VCR after pedicle screw placement • Compression on cephalad and caudad sides • Deformity correction based on exchanged rods technique (in situ bending, opening, closing, additional compression forces, and derotation) • Posterior pedicle instrumentation alone • Release of apical region of deformity with compression of convex side of curve before rod on concave side being locked in place • Other 3D correction forces used for deformity correction then applied • Instrumentation: T2 (T2-4) to L4 (L2-5) 	Cobb angle of major curve average 33.9° (range: 11–55°), correction 63.1% (range: 43.9–78.6%) Kyphosis 26.1° (range: 8–37°), correction 57.1% (range: 0–82.2%)	Neurological: None Other: Pleura perforated (n=2), pneumonia postoperative (n=1), abnormal coagulation intraoperatively (n=1)
*Cecen et al., 2013 ^[3]	1	14 y	T SM	Lenke 3A	None reported	None reported	Posterior segmental pedicle screw instrumentation <ul style="list-style-type: none"> • Posterior facetectomies and bilateral pedicle screw placement • Rod first placed on concave side and “pulled” • Rod then placed on convex side • Segmental derotation performed with apical compression on convex side and distraction of concave side 	NS	Neurological: delayed presentation of epilepsy (postoperative day 10)

(Contd...)

Table 3: (Continued).

Author and year	n	Mean age (range)	Intraspinal pathology	Spinal deformity	Neurologic status	IOM	Neurosurgical intervention and deformity correction	Outcome	Complications
*Huang et al., 2015 ^[8]	19/21	14.3y (10–18 y)	TC in all patients SM (n=2), DM (n=4), DM + SM (n=3), DM + + CM (n=2), DM + sacral canal cyst (n=1), DM + SM + sacral canal cyst (n=1), DM + lipomyelomeningocele (n=1), lipomyelomeningocele + SM + sacral canal cyst (n=1)	Cobb angle of major curve average 89.9° (range 61–154°) Kyphosis 75.5° +/- 20° (range 50–60°) all patients Apex T4-T12	Sensory disturbance (n=1), sensory and motor disturbance (n=1), motor disturbance (n=3), sensory and motor disturbance and urologic dysfunction (n=1), motor and bowel dysfunction (n=1)	SSEPs, MEPs	Posterior VCR for severe and rigid curves (Cobb > 90°, flexibility <30%) PSO for remaining curves • Performed at apical vertebrae • Temporary rods placed before lateral wall resection of VCR • Compression of precontoured temporary rods in alternate fashion • Great care taken to prevent distraction, instability, traction • Scoliosis correction through compression and shortening, more on convex side • Final rods precontoured to normal sagittal curve • <i>In situ</i> benders used, when necessary, to correct scoliosis	Cobb angle of major curve average 35.3° +/- 13.3° (range 11–62°) all patients Kyphosis 31.6° +/- 9.9o (range 11–47o) all patients	Neurological: numbness (n=1) Other: UTI (n=2), CSF fistula (n=1), blood loss > 5 L
*Tao et al, 2015 ^[20]	1	14 y	TC	Cobb angle of major curve 103° Kyphosis 93° Apex T7	Unable to walk unassisted with 3 to 4-/5 strength in BLE, bowel and bladder incontinence, perianal sensory loss	SSEPs, wake-up test	Posterior segmental pedicle screw instrumentation, posterior VCR at apex of deformity • Temporary rod placed on convex side with compressive forces in conjunction with mild distraction on concave side to close osteotomy and correct deformity • Placement of permanent contoured rod on convex side first	Cobb angle of major curve 67°, correction 54.4% Kyphosis 35°, correction 62.4% Regained neurologic functions by 6m postoperatively	None

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Table 3: (Continued).

Author and year	n	Mean age (range)	Intraspinal pathology	Spinal deformity	Neurologic status	IOM	Neurosurgical intervention and deformity correction	Outcome	Complications
Tao et al., 2019 ^[9]	15	10 y-20 y	TC in all patients Type II DM (n=11) SM (n=5) Sacral canal cyst (n=2) Lipomyelomeningocele (n=1)	Cobb angle of major curve average 99 +/- 26° with average 20% +/- 10% flexibility Kyphosis average 74o +/- 23°	Hypoesthesia in LE (n=2), weakness in BLE (n=1), weakness and hypoesthesia of LE (n=1), muscle atrophy and weakness of the right LE with sensory deficits (n=1)	SSEPs, MEPs	Posterior VCR <ul style="list-style-type: none"> Performed at apex with pedicle screws and rods placed before lateral wall resection Compressive forces applied Precontoured rods (contoured to "normal sagittal curve") applied after temporary rods removed In situ bending used to correct scoliotic deformity 	Cobb angle of major curve average 43 +/- 15° with average 56% +/- 12% flexibility Kyphosis average 33 +/- 7°, correction rate 53% +/- 16%	Neurological: transient numbness LE (n=1) Other: UTI (n=1), blood loss >5L (n=2), CSF Leak resolved (n=1)
Wang et al., 2015 ^[21]	21	15.1 y (11-19 y)	CM + SM (n=4) SM only (n=17) Unilocular with maximal syrinx/cord ratio <0.5 (n=9) Multilocular with ratio >0.5 (n=12)	Single major curve: T (n=14), TL (n=7); levoscoliosis (n=9), dextroscoliosis (n=12) Cobb angle major curve 68.05 +/- 20.1° Flexibility 45 +/- 15.8% T kyphosis 47.2 +/- 15.5°	Stable deficits (n=16): wasting of intrinsic hand muscles (n=3), numbness of left arm (n=1), absent abdominal reflex (n=12)	SSEPs, MEPs, wake-up test	<ul style="list-style-type: none"> Correction of cobb angle to <15° more than on bending films Use of fluoroscopy during correction (AP and lateral) to monitor correction No placement of screw in apical region to decrease correction ratio 	Cobb angle major curve 12.19 +/- 14.14° T kyphosis 36.38 +/- 9.06°	Neurological: intraoperative MEP signal losses (n=2) corrected by decreasing deformity correction ratio Other: postoperative infection requiring antibiotics and debridement (n=1)

(Contd...)

Table 3: (Continued).

Author and year	n	Mean age (range)	Intraspinal pathology	Spinal deformity	Neurologic status	IOM	Neurosurgical intervention and deformity correction	Outcome	Complications
Zhao et al., 2019 ^[27]	57	14.1 +/- 2.8 y (10–20 y)	DM type 1 (n=10), DM type 2 (n=13), TC (n=14), DM type 1 + TC (n=4), DM type 2 + TC (n=7), DM type 2 + SM (n=4), TC + SM (n=3), DM type 2 + TC + SM (n=2)	Location of major curve: T (n=31), TL (n=6), L (n=17), other (n=3) Cobb angle = 62.5 +/- 14.3° Flexibility = 37.3 +/- 9.1%	No deficit (n=47), sensory deficit (n=5), extremity weakness (n=6), abnormal reflexes (n=9)	SSEPs, MEPs, wake-up test	Single stage posterior correction and fusion with pedicle screw instrumentation with ponte osteotomies as needed (no PSO)	Postoperative Cobb angle = 29.9 +/- 14° (53.5% correction) →49.8% at final follow-up	Neurological: none Other: CSF leak (n=2), distal adding-on phenomenon (n=1)

BLE: Bilateral lower extremities, C: Cervical, CM: Chiari malformation, CT: Cervicothoracic, DM: Diastematomyelia, DNEP: Descending neurogenic evoked potentials, ED: Extradural, EMG: Electromyography, IOM: Intraoperative monitoring, IQR: Interquartile range, L: Lumbar, m: Months, MEP: Neuromotor evoked potentials, N/A: Not applicable, NS: Not specified, OS: Overall survival, PL: Posterolateral, pts: Patients, S: Sacral, SM: Syrinxomyelia, SSEP: Somatosensory evoked potentials, T: Thoracic, TC: Tethered cord, TL: Thoracolumbar, PFD: Posterior fossa decompression, PSO: Pedicle subtraction osteotomy, VCR: Vertebral column resection, w: Weeks, y: Years; →: Indicates sequential events. *Reports describing primary use of compressive forces on the convex side of the deformity for correction

Table 4: Recommendations/guidelines for surgical correction of kyphoscoliosis in pediatric patients with intraspinal pathology.

Author and Year	Recommendations
Bakaloudis et al., 2011 ^[2]	Corrective measures based on curve flexibility: >40%: wide posterior release with ponte osteotomies or Smith-Peterson osteotomies 20–40%: asymmetric pedicle subtraction osteotomies <20%: posterior-only approach with vertebral column resection
Huang et al., 2015 ^[8]	Posterior VCR for severe and rigid curves (Cobb > 90°, flexibility <30%) and PSO for remaining curves – performed at apical vertebrae • No interbody used in order to fully shorten the spinal column • Temporary precontoured rods attached to pedicle screws and compressed in in alternate fashion Great care taken to prevent distraction, instability, traction Scoliosis correction through compression and shortening, more on convex side. <i>In situ</i> benders used, when necessary, to correct scoliosis Final rods precontoured to normal sagittal curve
Wang et al., 2015 ^[21]	Correction of Cobb angle should be limited to <15° more than the correction obtained on lateral bending radiographs. Fluoroscopy in both the anteroposterior and lateral planes should be used intraoperatively to monitor correction. Avoid screw placement in the apical region to decrease the correction ratio.
Tao et al., 2019 ^[19]	All patients received detailed neurological exams All asymptomatic patients received a bending test and a traction test simulating the postcorrection condition to evaluate their tolerance for correction The treatment algorithm involved one of three approaches: posterior spinal fusion (PSF), pedicle subtraction osteotomy (PSO), or vertebral column resection (VCR) • PSF: patients with moderate curves (Cobb angle < 80°) and intact neurological status both before and during a bending and traction test • PSO: patients with neurological symptoms (in daily life or during the traction/bending test) and magnitude of the curve less than 80° • VCR: patients with magnitude of the curve >80° and/or flexibility <20%, with/without neurological symptoms
Zhao et al., 2019 ^[27]	Gradual and controlled correction within a defined range. Surgical goals: halt curve progression and neurologic deterioration rather than drastic scoliotic correction
Present authors	Correction should be preceded by decompression in the setting of expansile intraspinal pathology Corrective maneuvers should be primarily compressive versus distractive. Achieving an incomplete correction is favored over rather than overcorrection.

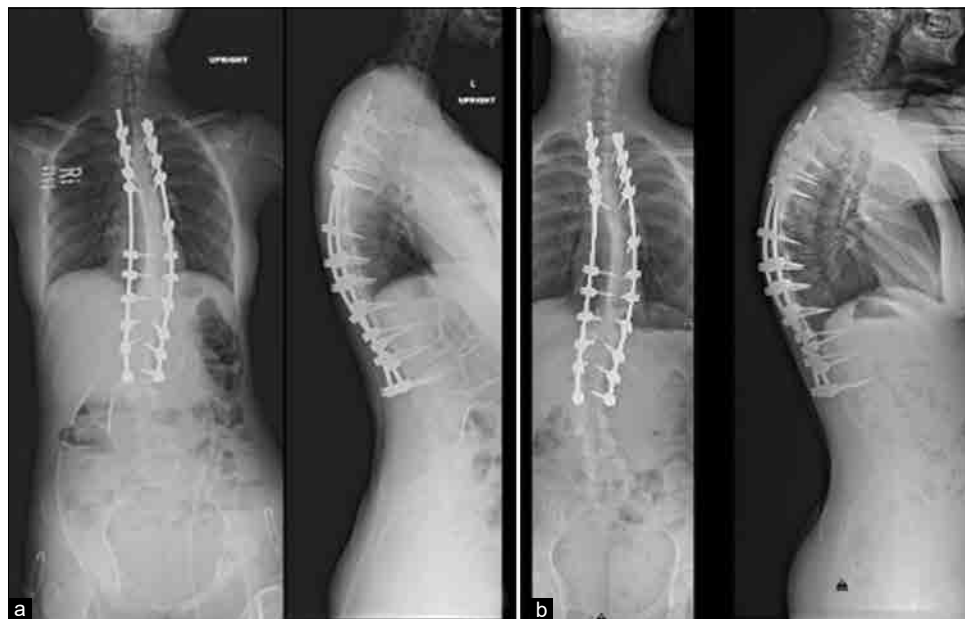


Figure 7: Standing thoracolumbar radiographs immediately (a) and 2-year postoperative (b) demonstrating significant improvement in patient's coronal deformity as well as restoration of a physiologic sagittal alignment.

correction of scoliotic spinal deformities may pose potential neurovascular risks.

The authors acknowledge that the literature review summary provided in Tables 2-3 is not streamlined, however, we believe that this speaks to the lack of literature addressing guidelines or recommendations for safe and effective surgical corrective techniques for this patient population. Few authors in the literature have made specific recommendations or proposed guidelines for the safe surgical correction of pediatric kyphoscoliosis in the setting of intraspinal pathology, primarily encouraging conservative and gradual deformity correction [Table 4].^[2,7,20,21,27] Although the surgical techniques employed by the authors in this case are not intrinsically novel, we believe that the provision of guidelines and points of consideration for surgical correction of spinal scoliotic deformities specific to patients with intraspinal pathology are a significant addition to the body of literature.

CONCLUSION

As illustrated in the present case, SDCIP can be safely and effectively surgically corrected using compressive maneuvers of the convexity of the curve with the goals of gradual and conservative correction to reduce the increased neurovascular risks secondary to an expanded and abnormal spinal cord.

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Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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