Surgical treatment of severe congenital scoliosis with unilateral unsegmented bar by concave costovertebral joint release and both-ends wedge osteotomy via posterior approach

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Abstract

Introduction Congenital scoliosis with unilateral unsegmented bar has remained a surgical challenge. If it is treated with a traditional release of the convex side and an apical wedge osteotomy, there is a risk of bony bridge fracture on the concave side and spine translation during correction maneuvers, which may then result in spinal cord injuries. The authors developed a technique that consists of a concave-side costovertebral joint release followed by both-ends wedge osteotomy via a posterior-only approach. In this article, we describe the technique in detail, and present the results of ten patients treated with this technique.

Methods A total of ten patients with congenital scoliosis with unilateral unsegmented bar, who had undergone a concave-side costovertebral joint release followed by both-end wedge osteotomy via a posterior-only approach were followed up for a mean of 34 months (range 26–48 months). The radiographic parameters and clinical records were all reviewed and analyzed.

Results Body height increased by a mean of 7.3 cm (range 6.0–9.0 cm). The preoperative coronal Cobb angle was 102° (range 83°–139°) with a mean flexibility of 14%. At the most recent follow-up visit, the mean Cobb angle was 35° (range 12°–53°) and the mean correction rate was 66%. The coronal imbalance improved from 3.4 cm (range 0.8–6.3 cm) preoperatively to 1.1 cm (range 0.6–1.8 cm) postoperatively, a 67% correction. There were no definite pseudarthroses, no implant failure, and no obvious loss of

correction in the follow-up period. Complications included one patient with hemopneumothorax and another patient with incomplete paralysis of the left lower extremity caused by a pedicle screw violating the spinal canal at the T5 level. The screw was removed 4 h after the initial operation, and the patient fully recovered after 3 months.

Conclusion We have had good results with our technique of concave-side costovertebral joint release and both-end wedge osteotomy. It has the advantage of remnant anulus fibrosus, the ligamentum flavum, and the facet joints on the concave side serving both as a hinge and to minimize translation of the spine ends. It can provide excellent three-dimensional curve correction for patients with severe rigid congenital scoliosis with unilateral unsegmented bar.

Keywords Congenital scoliosis · Unilateral unsegmented bar · Concave-side release · Both-ends wedge osteotomy · Posterior approach

Introduction

Congenital vertebral anomalies are caused by failures of formation and failures of segmentation. A complete failure of segmentation results in a so-called block vertebrae, which results in shortening of the length of the spine but does not cause scoliosis. Partial failure of segmentation creates a bar which, depending on its location, results in scoliosis or kyphoscoliosis due to the asymmetrical growth of the spine. The most serious deformity occurs when a unilateral unsegmented bar (UUB) is opposite fully segmented hemivertebrae.

The surgical management of congenital scoliosis with UUB is challenging and controversial. The pathological anatomy is complex, often including neural axis anomalies

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Fig. 1 **a** Traditional single apical vertebra wedge osteotomy. AO and BO are osteotomy lines. **b** The single apical vertebra wedge osteotomy of the spinal column segments with UUB. **c** The bony bridge is fractured as the osteotomy gap is closed by compression. Shifting of the fracture ends results in shear injury to the spinal cord

such as diastematomyelia and tethered cords, and rib cage malformations. Many surgical techniques have been proposed over the years, with the most dramatic corrections being obtained by utilizing some form of vertebral column resection. From a purely biomechanical standpoint, the most efficient method of correction is a wedge osteotomy at the apex of the non-segmented zone. Unfortunately, that is also where the anatomy is usually most distorted. Closing of the wedge necessitates breaking of the UUB (Fig. 1), which has been known to cause translation of the ends of the spine at the fracture site and shear injury to the spinal cord (personal experience and unpublished case reports).

The authors have developed a surgical technique where the wedge osteotomy is not performed within the non-segmented zone, but instead at both the cranial and caudal ends. The main advantage is that the osteotomy site anatomy is closer to normal and much more familiar to the surgeon. A concurrent concave-side costovertebral joint release removes the external fixator like effect of the ribs and allows for even more correction. The technique will be described in detail and the results will be presented.

Materials and methods

This is a retrospective case series conducted at a single institution. The senior surgeon (C.L.) performed all surgeries. The study was approved by the institutional ethics review board. No external funding was obtained.

The study group consisted of ten patients with congenital scoliosis with UUB who underwent surgery using the concave-side costovertebral joint release (CCJR) and both-ends wedge osteotomy (BEWO) technique between November 2004 and June 2008. There were six males and four females. The median age at surgery was 17.1 years old (range 14–22 years of age).

Clinical records were reviewed including demographic data, operating time, blood loss, functional improvement and complications. Measurements of curve magnitude and coronal balance were made on preoperative standing anteroposterior and lateral radiographs. The deformity correction, spinal balance, complications related to the instrumentation and fusion status were assessed beginning 2 weeks after surgery and at intervals till the most recent follow-up. Apical vertebral translation for thoracic curves was measured based on the distance between the C7 plumb line and the center of the apical vertebral body or disc. The measurement of apical vertebral translation for thoracolumbar and lumbar curves was based on the distance between the central sacral vertical line and the center of the apical vertebral body or disc. The coronal curve was measured using Cobb's method. Coronal balance was measured based on the distance between the C7 plumb line and the center sacral line. Thoracic kyphosis was measured using Cobb's method from the superior endplate of T5 to the inferior endplate of T12. Sagittal balance was measured based on the distance between C7 plumb line and the posterior superior corner of S1.

CT scans of the spine with 3D reconstructions were obtained in all patients. All ten patients had UUBs in the thoracic spine, and one patient also had unsegmented lumbar vertebrae. The pathological anatomy was very complex. Six of the ten patients had an unsegmented bar at one site, two at two sites and two at three sites. One patient had associated hemivertebrae and another one had associated diastematomyelia. In four patients the convexity of the curve was to the left and in the remaining six the convexity was to the right. The UUBs were always on the concave side.

The preoperative mean coronal Cobb angle was 102° (range 83° – 139°) with a mean flexibility of 14% (range 7.8–20.1%). Two patients had associated thoracic hyperkyphosis, measuring 68° and 101° respectively. Another

Table 1 Demographics and surgical data of patients

Patient no.	Sex	Age (years)	Scoliosis direction	Unsegmented levels	Osteotomy levels	Fusion levels	OR time (h)	EBL (ml)	Follow-up (months)
1	F	18	L	T5–T7	T5, T7	T2–L2	7.3	3,600	34
2	M	15	R	T6–T8	T6, T8	T2–T12	8.1	3,400	32
3	M	14	L	T9–T12	T9, T12	T3–L4	7.4	3,300	26
4	F	16	R	T5–T6, T7–T9	T7, T9	T2–L3	7.6	4,100	48
5	F	17	R	T4–T5	T4, T5	T3–L2	7.8	3,800	34
6	F	18	L	T6–T7, T8–T11, L1–2	T8, T11	T3–L4	11.2	4,500	46
7	M	16	L	T6–T8, T9–T11	T9, T11	T2–L3	10.6	4,100	38
8	F	22	R	T7–T9	T7, T9	T4–L3	8.2	3,900	26
9	F	18	R	T3–T4, T5–T7, T8–T11	T5, T7	T2–L5	14.1	4,400	28
10	M	17	R	T9–T12	T9, T12	T4–L3	7.2	3,200	32

F female, L left, M male, R right, OR operation, EBL estimated blood loss

Table 2 Radiographic data

Patient	Flexibility (%)	Kyphosis	Scoliosis cobb (°)			Thoracic kyphosis cobb (°)			Apical vertebral translation (cm)			Coronal imbalance C7PL (cm)		
			Preop	Posop	F/U	Preop	Posop	F/U	Preop	Posop	F/U	Preop	Posop	F/U
1	12.8	N	86	28	29	0	21	21	5.5	1.2	1.3	3.6	0.8	0.9
2	13.3	N	83	21	22	20	22	22	8.2	1.6	1.6	1.2	0.6	0.6
3	10.4	N	93	44	46	10	24	25	12.9	3.9	4.0	0.8	0.7	0.7
4	20.1	Y	139	51	53	101	47	48	13.2	2.5	2.5	5.1	1.7	1.8
5	16.5	N	94	11	12	20	23	25	7.8	0.7	0.8	2.4	1.2	1.2
6	14.7	N	102	38	40	35	27	27	11.8	2.2	2.4	4.8	2.5	2.6
7	12.6	Y	114	46	48	68	20	21	13.6	4.2	4.4	3.4	1.2	1.3
8	17.5	N	92	32	34	22	28	31	6.3	2.3	2.5	2.5	0.6	0.6
9	14.5	N	132	32	44	0	26	27	13.3	3.8	3.9	6.3	1.1	1.2
10	7.8	N	85	22	24	22	22	22	6.3	1.8	1.8	4.2	0.5	0.5

C7PL C7 plumb line, F/U follow-up

two patients had associated thoracic hypokyphosis, with both patients measuring 0°. The remainder were deemed normokyphotic. The mean coronal imbalance was 3.4 cm (range 0.8–6.3 cm) (Tables 1, 2).

Description of surgical technique

3D-CT reconstructions are invaluable in the pre-surgical planning for congenital scoliosis. Locations for pedicle screw placement are marked. It is often possible to place screws in the unsegmented zone. Note that the pedicles on the concave side of the unsegmented spine section are often small and without clear anatomic landmarks. The spinal cord is shifted to the concave side. We believe it is safer to insert pedicle screws on the convex side.

All surgeries were performed with neuro-monitoring using somatosensory evoked potentials. Intraoperative

radiographs were taken as needed for localization. Computer aided navigation was not available at our hospital.

The patients were placed in the prone position on a four-poster frame. The spine was exposed via subperiosteal dissection, and pedicle screws were placed according to plan.

Concave-side costovertebral joint release

The transverse processes on the concave side were resected. Depending on the length of the curve, this usually works out to be four to six segments. The costovertebral joints were exposed, the periosteum and underlying parietal pleura were bluntly detached from the proximal end of the ribs, and then the rib heads were resected. If the rib heads were too close together to be resected due to severe scoliosis, or even partially fused, an osteotome was used to disarticulate the costovertebral joint, so as to detach the ribs from the vertebrae.

Fig. 2 **a** Both-ends wedge osteotomy of the spinal column segments with UUB. AO, BO, CO and DO are osteotomy lines. At both O points, a portion of the ligamentum flavum and anulus fibrosus was preserved to function as the hinge for osteotomy closure; **b** double wedge osteotomy on both ends of the UUB; **c** the osteotomy space was closed

Both-ends wedge osteotomy

The concept is given in Fig. 2. The cranial wedge resection is usually done first. The posterior elements were removed and the neural elements were exposed by a wedge resection with the base on the convex side. The corresponding transverse process, the rib head and a segment of the proximal rib on the convex side were also removed to expose the lateral wall of the vertebral bodies. Dissection was carried out subperiosteally along the lateral wall and the anterior surface of the vertebral bodies, which could be palpated. The pedicles and the lateral portion of the vertebral body on the convex side were removed using osteotomes and a power bur. The vertebral bodies and the disc were removed in piecemeal fashion towards the mid-line and over to the concave side of the vertebral body. A thin rim of posterior vertebral wall adjacent to the dura should be kept intact, in order to avoid epidural bleeding. The anterior wall of the vertebral body was also removed in piecemeal fashion, always keeping in mind the mediastinal structures anteriorly. It is important to remove as much vertebral body and disc as possible at this stage. If necessary the exiting nerve root may be sacrificed.

A temporary safety rod can be placed on the convex side if needed. The osteotomy is continued from the concave side, and finally the posterior vertebral wall is removed. Care is taken to remove all bony and disc fragments that may compress the dura during osteotomy gap closure. Portions of the annulus fibrosus, the ligamentum flavum and the facet joint on the concave side were preserved to serve as a hinge.

The wedge osteotomy procedure is repeated at the caudal end of the unsegmented zone. The dura should be watched at all times and contact with it is avoided. When

closing the gap, keep in mind that the osteotomy space is three dimensional in nature.

Deformity correction was performed with the techniques of compression, translation and cantilever bending using pre-contoured rods. It is important to avoid inadvertent distraction of the spinal cord. The vertebral column was shortened by gradually closing the osteotomy gap along the convex side rod. We have found it safe to compress until we see wrinkling of the dura mater. With the convex side rod locked in place, some more correction can be obtained by gentle distraction along the pre-contoured rods on the concave side. With the instrumentation in place, meticulous decortication was done, and autogenous bone graft obtained from the vertebrectomy and the excised rib segments were placed over the instrumented levels. The wound was closed in layers over suction drainage.

Post-operative management

The drains were removed approximately 36 h after surgery. The patients were placed in a posterior shell brace and instructed to wear the brace when upright for 1 year.

Results

The mean follow-up time for this study was 34.4 months (range 26–48 months). All ten patients were seen at clinic and had standing radiographs at last follow-up.

Patients' results are listed in Table 1. The mean operating time was 8.9 h (range 7.1–14.1 h). Mean blood loss was 3,830 ml (range 3,200–4,500 ml). A mean of 5.2

(range 4–6) costovertebral joints were released from the concave side. Mean fusion extent was 13 vertebrae (including the unsegmented zone). There were no infections that required further surgery.

At the most recent follow-up visit, patient's body height increased by a mean of 7.3 cm (range 6.0–9.0 cm). The mean Cobb angle improved from the preoperative 102°–35° (range 12°–53°) and the mean correction rate was 66% (range 51–87%). In the six normokyphotic patients, the post-operative thoracic kyphosis (sagittal Cobb angle) was

between 21° and 48°, the mean correction rate being 61% (range 53–70%). In the two patients with preoperative hypokyphosis, this was corrected to 21° and 27° from the previous 0°. In the two patients with preoperative hyperkyphosis, the kyphosis correction was respectively from 68° to 21° and from 101° to 48°.

The coronal imbalance (distance between the C7 plumb line and the central sacral vertical line) improved from 3.4 cm (range 0.8–6.3 cm) preoperatively to 1.1 cm (range 0.6–1.8 cm) postoperatively, a 67% correction.

Fig. 3 15-year-old female presenting with severe scoliosis due to an UUB, who underwent CCJR and BEWO. **a, b** Preoperative photographs; **c** preoperative coronal Cobb angle was 83°; **d** bending radiograph Cobb angle was 72°, with rigid deformity; **e** preoperative thoracic kyphosis was 20°; **f** 3D-CT showed that the UUB was located at vertebrae T6–8, forming a fan-shaped deformity; **g** postoperative

coronal Cobb angle was 21°, with a correction rate of 74.7% and balanced coronal plane; **h** post-operative thoracic kyphosis was 22°; **i, j** the postoperative photographs showed good trunk balance; **k, l** no loss of correction was found in either the coronal or sagittal plane 2 years after the operation

At the most recent follow-up visit, there were no breakage or loosening of the internal fixation screws. No measurable loss of correction or add-on curve progression (decompensation) was observed. While the use of only plain films and the presence of multiple implants preclude any definitive statements regarding fusion, no definite pseudarthroses were noted (Figs. 3, 4; Table 2).

Complications were encountered in two patients. One patient experienced incomplete paralysis of the left lower extremity due to spinal cord compression at the T5 level by a pedicle screw that violated the spinal canal. The offending screw was removed in an emergent second

surgery, 4 h after the initial operation. The patient recovered full neurologic function after 3 months. Another patient suffered from hemopneumothorax secondary to pleura laceration sustained during the costal head resection. This was successfully managed with closed drainage of the pleural cavity.

Discussion

Congenital scoliosis by definition is caused by asymmetrical vertebral anomalies. Hemivertebrae are thought to be

Fig. 4 18-year-old male presenting with severe scoliosis due to UUB at multiple locations, who underwent CCJR and BEWO. **a, b** Preoperative photographs showed scoliosis with trunk tilt to left; **c** preoperative coronal Cobb angle was 102°; **d** bending Cobb angle was 87°; **e** preoperative thoracic kyphosis was 35°; **f, g** 3D-CT reconstruction showed that the UUB was located at vertebrae T6–7, T8–11 and

L1–2; **h** postoperative coronal Cobb angle was 38°, with a correction rate of 63% and balanced coronal plane; **i** postoperative thoracic kyphosis was 27°; **j, k** the postoperative photographs showed good trunk balance; **l, m** no loss of correction was found in either the coronal or sagittal plane 2 years after the operation

failures of formation. There have been a number of reports on the surgical management of scoliosis due to the presence of hemivertebrae and most authors have reported good clinical results [1, 9, 10].

The incidence of scoliosis due to failure of segmentation is not known. Although not commonly reported in either the English or Japanese language literature, the authors have heard many such cases presented at specialty conferences, mainly by Turkish and Chinese authors. The pathogenesis of scoliosis with UUB is complicated, and it would be fair to say that no two cases are exactly alike. The curves are usually severe and preoperative curve flexibility is very low (only 14% in our series). The deformed ribs act as an external fixator of the spine, further limiting motion.

Many techniques have been described in an effort to improve the spinal flexibility in patients with severe and rigid scoliosis. Li et al. [6] described a single-stage anterior release followed by posterior instrumentation and fusion for the treatment of severe scoliosis (average 98°) in 31 cases, with a 48.6% rate of correction. Shen et al. [11] described a single-stage anterior release followed by instrumentation and fusion for the treatment of 12 cases of severe scoliosis (average 98.5°) with a 59.5% rate of correction. However, these procedures are traumatic and many complications such as pneumonia, atelectasis, sympathetic nerve injury and chylothorax have been reported [3, 4, 7, 8]. Hempfing et al. [2] stated that better flexibility of the spine could be obtained by resection and release of the ribs on the concave side, whereas the release of the spine by resection of intervertebral discs would only lead to slight improvement in flexibility. This is in agreement with Voos et al. [13], who reported that for patients with preoperative flexibility less than 20%, simple anterior release surgery had little effect in improving postoperative correction.

Suk et al. [12] described the technique of posterior-only vertebral column resection. In his series of 16 patients with a mean preoperative Cobb angle of 109°, the post-operative Cobb angle was 46° (59% correction). However, complications were encountered in four patients, including one with complete permanent paralysis. The authors have some experience with this technique, and we believe the danger occurs when all three spinal columns are transected. There is an unavoidable translation motion at the osteotomy site, and the spinal cord sustains a shear and compressive force. Placement of so-called safety rods helps but does not fully eliminate the problem. The situation is aggravated when there are UUBs. If the traditional single apical vertebral osteotomy is used, the surgeon starts with a disadvantage by working in a zone of distorted, unfamiliar anatomy. As the wedge osteotomy proceeds to the concave side, and when the osteotomy gap is closed, the UUB may suddenly fracture and lead to a displacement of the vertebral ends, which may result in a shear injury to the spinal cord (Fig. 1).

We designed our osteotomy procedure to avoid the dangers presented above. We call it the “Both-ends osteotomy”, because it is done at both cranial and caudal ends of the unsegmented zone, instead of at the apex. It has the advantages of being performed in an area of relatively normal anatomy, where there is safety in familiarity. The spinal column is never completely transected. The remnant anulus fibrosus, the ligamentum flavum, and the facet joints on the concave side serve both as a hinge and to minimize translation of the spine ends. Having two osteotomy sites rather than one single site means the amount of correction needed at each site is less, potentially decreasing the incidence of spinal cord kinking. The 66% coronal Cobb angle correction we achieved in this series compares well with the results of series using posterior-only vertebral column resection (51–59%) [5, 12].

One patient sustained incomplete paralysis of the left lower limb due to a malpositioned screw, and another suffered from hemopneumothorax resulting from pleura laceration during the costal head resection. Both are technique related and not unique to our osteotomy technique. With our limited case numbers we cannot state whether the complication rate of our technique is higher or lower compared with other techniques based on posterior osteotomies.

This study is a retrospective practice audit of a surgical series by a single experienced surgical team, and thus suffers from all the limitations of this type of study. Patients with UUB present with greatly different curve patterns and magnitude, so individualized surgical planning is mandatory for each patient. There is a learning curve with this technique, so our results should be extrapolated with caution. Even with experience, we are reporting 9 h surgery time and 3,800 ml range blood loss, which is not trivial.

We have had good results with our technique of concave-side costovertebral joint release and both-ends wedge osteotomy. It is not required for the management of the routine idiopathic curve, but for patients with severe congenital scoliosis with UUB and Cobb angle >80° and a flexibility of <20%, it can provide excellent three dimensional curve correction.

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Conflict of interest None.

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