Hemivertebrae resection for unbalanced multiple hemivertebrae: is it worth it?

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Abstract

Purpose To assess the correction effect of hemivertebra resection for unbalanced multiple hemivertebrae by measuring corresponding parameters in both coronal and sagittal planes on series posteroanterior and lateral radiographs and report the related complications.

Methods Twelve children with unbalanced multiple hemivertebrae were operated on by hemivertebra resection through a combined anterior and posterior approach or a posterior-only procedure. Mean age at time of surgery was 9.8 years (range 2–14 years). They were retrospectively studied with a mean follow-up of 48.7 months (range 30–60 months).

Results The mean Cobb angle of the main curve was 65.3° (range 45°–92°) before surgery and 13.8° (range 4°–30°) at the last follow-up. The correction rate was 80.0 % (range 65.5–92.4 %). The compensatory cranial curve was corrected from 25.8° (range 5°–53°) to 11.7° (range 0°–34°) with a correction rate of 65.9 % (range 33.3–100 %), and the compensatory caudal curve was corrected from 32.4° (range 17°–57°) to 7.1° (range 0°–20°) with a correction rate of 81.4 % (range 53.1–100 %). The angle of segmental kyphosis was 41.3° (range 12°–76°) before surgery and 17.0° (range –12° to 45°) at the final follow-up. The coronal imbalance was –1.0 cm (range –3.5 to 3 cm) before surgery and 0.0 cm (range –1.0 to 1.5 cm) at the most recent follow-up. The sagittal imbalance was

0.9 cm (range –3.2 to 3 cm) before surgery and 0.6 cm (range –3.0 to 3.5 cm) at the most recent follow-up. Complications including pedicle fractures, and pseudarthrosis were found in two patients (20 %).

Conclusions In the patients with unbalanced multiple hemivertebrae, hemivertebra resection allows for excellent correction in both the coronal and sagittal planes, and great care should be taken to reduce the rate of complications.

Keywords Congenital scoliosis · Hemivertebra · Hemivertebra resection · Posterior fusion · Posterior instrumentation

Introduction

The natural history of congenital scoliosis has been well documented [1–3]. Hemivertebra is the most frequent cause of congenital scoliosis. If left untreated, hemivertebra may lead to substantial spinal deformity [1–3]. Although hemivertebra excision for congenital scoliosis due to a single hemivertebra has been widely reported [4–24], we are unaware of previous reports of the treatment of unbalanced multiple hemivertebrae, and could not find any reference to it in a computerized search utilizing MEDLINE [1–24].

Unbalanced multiple hemivertebrae can be expected to cause progression of scoliosis at a rate of 2°–5° per year, which was higher than a single hemivertebra (1°–3.5° per year) [2]. Nasca et al. described the natural history of 19 patients with unbalanced multiple hemivertebrae. The rate of progression based on periods of observation from 1 to 11 years ranged from 1.5° to 12° per year [3].

In the surgical treatment of unbalanced multiple hemivertebrae, two or more hemivertebrae should be removed,

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which brings greater technical difficulties. In addition, more severe coronal and sagittal unbalance may be encountered, which brings greater challenges to the surgeons.

There is no previous reports of the treatment of unbalanced multiple hemivertebrae. Whether hemivertebra resection for unbalanced multiple hemivertebrae can achieve a good result is still unknown. From July 2006 through October 2010, 12 patients who had congenital scoliosis due to unbalanced multiple hemivertebrae received hemivertebra resection through a combined anterior and posterior approach or a posterior-only procedure at our institution. The aim of this study was to assess the correction effect of hemivertebra resection for unbalanced multiple hemivertebrae by measuring corresponding parameters in both coronal and sagittal planes on series posteroanterior and lateral radiographs. In addition, the related complications were reported.

Materials and methods

All surgeries were performed by the corresponding author. There were seven boys and five girls. The mean age of the patients at the time of surgery was 9.8 years. The details of the patients, such as gender, age, resected hemivertebra, and associated congenital abnormalities are listed in Table 1.

Standard preoperative imaging and testing included MRI and CT scans of the spinal column and intrathecal

structures. Also renal and cardiovascular systems were investigated for associated anomalies and syndromes. Operative reports were reviewed to determine the presence of any intraoperative complications. Medical records were reviewed to identify any complications in the peri-operative and follow-up periods.

Surgical technique

Once unbalanced multiple hemivertebrae was found, hemivertebra resection was suggested to perform as early as possible, except the patient younger than 5 years with unbalanced multiple hemivertebrae located in thoracic region who need extensive thoracic fusion. As we know, extensive thoracic fusion in younger children is associated with diminished pulmonary function.

Spinal cord monitoring was performed in all operations. The approach was selected according to the progression of surgical methods and skill. The combined anterior and posterior approach was used in patients operated earlier, and the posterior-only approach in patients operated more recently.

Ten patients were treated by hemivertebra resection through a combined anterior and posterior approach. In the anterior approach, the patient was laid in lateral decubitus with the concave side of the scoliosis facing downward. The hemivertebra was first identified and its vertebral body and upper and lower discs were excised. After hemivertebra resection, if a big gap was found, especially in the

Table 1 Patient characteristics

Case	Sex	Age (years)	Resected hemivertebra	Associated congenital abnormalities	Surgery	Fused segment	Blood loss (mL)	Op-time (min)
1	F	9	T5, 9, 11-R	Left 6th and 7th rib synostosis. Butterfly vertebrae T6 and T10	APS	T3-L2	750	350
2	M	11	T11, L1-R		APS	T9-L3	800	255
3	M	7	T10, 12-L	L1-R (incarcerated). Butterfly vertebrae T1, T2, and T3	APS	T7-L2	1,500	365
4	F	12	T5, 7-R		APS	T3-9	600	260
5	M	11	T11, L1-L		APS	T9-L3	900	310
6	M	14	T8, 10 -L	Butterfly vertebrae L5	APS	T6-L2	800	270
7	M	14	L1, 2-L		APS	T9-L5	1,200	350
8	F	12	T12, L2-L		APS	T10-L4	500	240
9	M	11	T12, L2-L		APS	T10-L4	600	270
10	F	9	T11, L1-L	T1-R, T7-L, T9-R (incarcerated). Butterfly vertebrae T5	APS	T9-L3	800	300
11	M	6	L1, 2-R		PS	T11-L4	300	150
12	F	2	T12, L2-R		PS	T11-L3	300	265
Average		9.8					754.2	282.1

R hemivertebra located on the right side, L hemivertebra located on the left side, APS Anterior and posterior surgery, PS posterior surgery

patient with two or more adjacent hemivertebrae, titanium mesh cage was inserted into concave side to avoid excessive shortening of vertebral column. In addition, titanium mesh cage was also used in the patient with severe segmental kyphosis to create a fulcrum to achieve lordosis.

One week after the anterior approach, the patient received posterior surgery. The patient was placed in prone position, and a standard midline incision was made. After the exposure of the posterior elements of affected levels, pedicle screws were placed in the adjacent vertebrae and their position was checked with fluoroscopy. The posterior part of the hemivertebra, including the pedicle was excised. Then compression was utilized at the convex side and distraction was applied at the concave side. Local bone from the excised hemivertebra was used for fusion.

Two patients were treated by posterior hemivertebra resection and fixation with pedicle screws. The patient was placed in the prone position, and a standard midline incision was made. The posterior elements of the spine were carefully exposed at the affected levels. Pedicle screws were inserted into the normal upper and lower vertebrae and their position was checked with fluoroscopy. In some patients with structural compensatory cranial and caudal curves, more pedicle screws were implanted and long fusion was performed. The posterior elements of the hemivertebra were subsequently removed, including the lamina, the facet joints, the transverse process, and the posterior part of the pedicle. In the thoracic spine, the rib head and the proximal part of the surplus rib on the convex side were also removed. Then the lateral cortex of the hemivertebra was exposed by blunt dissection. Before removing the vertebral body, a safety rod was applied on the concave side to stabilize the spine. Then the vertebral body and the upper and lower discs, including the cartilage endplate, were completely removed. Another rod was applied on the convex side. Gradual compression was applied while leaving the concave rod unlocked until the gap was closed. Decortication of the posterior elements was performed, and autogenous bone from the hemivertebra was used for fusion.

When the patient started to walk after surgery, a custom-made orthosis was used. The orthosis was kept for 6 months and removed if radiograph after 6 months did not show any sign of pseudarthrosis.

Postoperative assessment

Standing posteroanterior and lateral radiographs of the full spine were evaluated postoperatively just before the patient was discharged and at the time of the most recent follow-up.

The parameters measured in the coronal plane were the segmental scoliosis curve, the total main scoliosis curve,

the compensatory cranial curve, the compensatory caudal curve, and the coronal balance. The segmental scoliosis curve was measured between the two vertebrae immediately adjacent to the hemivertebra, whereas the total main scoliosis curve measured was the maximum scoliosis angle between the two most tilted vertebrae. The coronal balance was measured as the distance between the C7 plumb line and the center sacral line.

The parameters measured in the sagittal plane were the segmental kyphosis, the thoracic kyphosis, the lumbar lordosis, and the sagittal balance. The segmental kyphosis was measured between the two vertebrae adjacent to the hemivertebra, the thoracic kyphosis was measured by the Cobb method from the superior endplate of T5 to the lower endplate of T12, and the lumbar lordosis from the superior endplate of T12 to the endplate of S1. The sagittal balance was measured as the distance between C7 plumb line and the posterior superior corner of S1.

Statistical analysis

All parameters measured in the coronal and sagittal plane were analyzed. The preoperative, postoperative, and final follow-up results were analyzed statistically with use of the paired Student's *t* test, with the level of significance set at $P < 0.05$.

Results

Ten patients received a combined anterior and posterior surgery, and two patients were treated by a posterior-only procedure. The interval between anterior and posterior surgery averaged 7.2 days (range 5–9). In total, 25 hemivertebrae were excised. Three hemivertebrae were excised in one patient, and two hemivertebrae were excised in remaining 11 patients.

The mean operating time was 282.1 min (range 150–365 min), and the mean blood loss was 754.2 mL (range 300–1,500 mL). The mean follow-up time was 48.7 months (range 30–60 months).

Correction of the coronal plane

The segmental curve averaged 63.0° before surgery (range 43°–90°), 12.8° after surgery (range 3°–26°), and 13.1° at the latest follow-up (range 5°–29°), for an average correction of 80.4 %. The main curve averaged 65.3° before surgery (range 45°–92°). After surgery the mean angle measured 13.4° (range 3°–27°). At the last follow-up, it was 13.8° (range 5°–30°). Thus, the mean correction was 80.0 % (Table 2; Fig. 1).

Table 2 Correction of the coronal plane and sagittal plane

	Preoperative	Postoperative		Final follow-up			
	Value ^a	Value ^a	Improvement (%)	P value	Value ^a	Improvement (%)	P value
The coronal plane							
Segmental curve (°)	63.0 ± 16.7 (43–90)	12.8 ± 8.1 (3–26)	80.6	<0.001	13.1 ± 8.7 (5–29)	80.4	<0.001
Main curve (°)	65.3 ± 16.2 (45–92)	13.4 ± 8.5 (3–27)	80.5	<0.001	13.8 ± 9.1 (5–30)	80.0	<0.001
Compensatory cranial curve (°)	25.8 ± 16.6 (5–53)	10.6 ± 11.7 (0–33)	70.8	<0.001	11.7 ± 12.3 (0–34)	65.9	<0.001
Compensatory caudal curve (°)	32.4 ± 11.9 (17–57)	6.4 ± 6.0 (0–15)	82.7	<0.001	7.1 ± 7.2 (0–20)	81.4	<0.001
Coronal balance (cm)	−1.0 ± 1.7 (−3.5 to 3)	0.0 ± 1.0 (−1.0 to 1.9)		0.064	0.0 ± 0.9 (−1.0 to 1.5)		0.060
The sagittal plane							
Segmental kyphosis (°)	41.3 ± 24.5 (12–76)	16.8 ± 16.5 (−10 to 45)		<0.001	17.0 ± 16.8 (−12 to 45)		<0.001
Thoracic kyphosis (°)	34.2 ± 15.6 (15–61)	28.6 ± 7.7 (16–42)		0.199	30.7 ± 8.6 (18–43)		0.404
Lumbar lordosis (°)	−48.8 ± 24.0 (−71 to −5)	−44.2 ± 12.5 (−60 to −25)		0.328	−46.9 ± 13.1 (−67 to −29)		0.685
Sagittal balance (cm)	0.9 ± 1.9 (−3.2 to 3)	0.6 ± 1.4 (−2.8 to 2.7)		0.531	0.6 ± 1.8 (−3.0 to 3.5)		0.517

^a The values are given as the mean ± standard deviation, with the range in parentheses^b According to the paired Student's *t* test, with the level of significance set at *P* < 0.05

The compensatory cranial curve averaged 25.8° before surgery (range 5°–53°), 10.6° after surgery (range 0°–33°), and 11.7° at the last follow-up (range 0°–34°). This resulted in a correction of 65.9 %. The compensatory caudal curve averaged 32.4° before surgery (range 17°–57°), 6.4° after surgery (range 0°–15°), and 7.1° at the latest follow-up (range 0°–20°), for an average correction of 81.4 % (Table 2; Fig. 1).

The preoperative coronal imbalance of −1.0 cm (range −3.5 to 3) was improved to 0.0 cm (range −1.0 to 1.9) at immediate postoperative assessment and 0.0 cm (range −1.0 to 1.5) at the most recent follow-up (Table 2; Fig. 1).

Correction of the sagittal plane

The segmental kyphosis averaged 41.3° before surgery (range 12° to 76°). After surgery the mean angle measured 16.8° (range −10° to 45°). At the last follow-up, it was 17.0° (range −12° to 45°).

The thoracic kyphosis averaged 34.2° before surgery (range 15°–61°), 28.6° after surgery (range 16°–42°), and 30.7° at the last follow-up (range 18°–43°). The lumbar lordosis averaged −48.8° before surgery (range −71° to −5°), −44.2° after surgery (range −60° to −25°), and −46.9° at the latest follow-up (range −67° to −29°).

The preoperative sagittal imbalance averaged 0.9 cm (range −3.2 to 3.0). After surgery it measured 0.6 cm (range −2.8 to 2.7). At the last follow-up, it was 0.6 cm (range −3.0 to 3.5).

Complications and additional operations

There were no neurologic complications. There was one pedicle fracture. Patient 1, who had one right-sided hemivertebra at T5, T9 and T11 level, respectively, encountered the fracture of right T4 pedicle during the posterior surgery when compression was applied to close the gap resulting from the resection of hemivertebra at T5 level. In this patient, fusion segments were extended to T3 (Fig. 1). Revision surgery was performed in one patient 3 years later. Patient 3, who had one left-sided hemivertebra at T10 and T12 level, respectively, received hemivertebra resection through a combined anterior and posterior approach and posterior fixation with pedicle screws. Two years after surgeries, no sign of implant failure or pseudarthrosis was found, and the implant was removed. One year later, progressive kyphosis was found in surgical region, and the revision surgery was performed. Pseudarthrosis was detected during the revision surgery.

Fig. 1 The radiographies and clinical pictures of patient 1. A 9-year-old female with three right-sided hemivertebrae at T5, 9 and 11 levels received hemivertebrae resection through a combined anterior and posterior approach. Besides hemivertebrae, the patient had left-sided sixth and seventh rib synostosis, and butterfly vertebrae at T6 and 10 levels. During the anterior surgery, the vertebral bodies of T9 and T11 were excised. In the posterior surgery, the appendix of T9 and T11, and the hemivertebrae at T5 were excised. The rib synostosis was severed. Right pedicle of T4 fractured when compression was applied to close the gap resulting from the resection of hemivertebra at T5

Discussion

The results of this study indicate that hemivertebra resection for unbalanced multiple hemivertebrae allows for excellent correction in both the coronal and sagittal planes, and great care should be taken to reduce the rate of complications.

Our study has several limitations. First is the small sample size. Second, patients receiving different surgeries, which may bring potential bias, were included. Both of these limitations are related to the extremely rare and unusual patients with such conditions. Third, many patients of the series are immature by the final follow-up, so the risk

of curve progression still exists. It is quite difficult to follow the patients for such long time. Although congenital scoliosis due to a hemivertebra may show spontaneous improvement [25], early surgical intervention for hemivertebra is advocated by most surgeons [1–24]. Given the rapid progression of unbalanced multiple hemivertebrae, surgery should be performed as early as possible to avert the development of severe local deformities.

The three main surgical procedures used to treat congenital scoliosis are fusion in situ, convex-side growth arrest (epiphysiodesis), and hemivertebra resection. During the last decade, the predominant form of surgery for

hemivertebra has been hemivertebra resection [1–24]. On the basis of hemivertebra resection, different fixation techniques were developed [21–23]. In addition, fusionless posterior hemivertebra resection was reported [24].

For a single hemivertebra, hemivertebra resection can be performed through anterior and posterior approaches or through the posterior approach alone [4–24]. When it comes to unbalanced multiple hemivertebrae, hemivertebra resection through a combined anterior and posterior approach or a posterior-only procedure is also feasible. In our study, the first ten patients received anterior and posterior surgeries, and the last two patients received a posterior-only procedure.

For unbalanced multiple hemivertebrae, the combined approach can offer a good visualization of neural structures and is less demanding. By the combined approach, it is easy to remove the whole hemivertebra and growth plates. Although the combined approach has such merits, some disadvantages, such as additional incision, longer operation time, and decreased pulmonary function in patient with thoracotomy, were reported. If the location of hemivertebra is far away from each other, combined anterior and posterior hemivertebra resection is unfeasible. In addition, the combined approach is not a good choice for the patients with hemivertebra located above T4, because large amount muscle needs to be dissected to achieve satisfactory exposure. In recent years, more and more studies have reported on hemivertebra excision from the posterior approach only. Posterior-only procedure, which can avoid anterior approach-related complications, has been the mainstream surgery for hemivertebra. But posterior-only procedure is technically more demanding, and should be performed by experienced surgeons.

Although there is no similar study, we compare our results with some studies about hemivertebra excision. Ruf et al. reported the results of 28 patients with congenital scoliosis treated by posterior hemivertebra resection. The main curve was corrected from 36.1° to 6.8° with a correction rate of 81 %. The compensatory cranial curve was corrected from 14.6° to 2.8° with a correction rate of 81 %, and the compensatory caudal curve was corrected from 17.3° to 2.7° with a correction rate of 84 % [18]. Bollini et al. reported the results of 21 patients with lumbar hemivertebra treated by hemivertebra resection through a combined posterior and anterior approach. The main curve was corrected from 34.1° to 12.3° with a correction rate of 63.9 %. The compensatory cranial curve was corrected from 15.6° to 8.1° with a correction rate of 48.1 %, and the compensatory caudal curve was corrected from 7.1° to 1.9° with a correction rate of 73.2 % [10]. In our study, the main curve was corrected from 65.3° to 13.8° with a correction rate of 80.0 %. The compensatory cranial curve was corrected from 25.8° to 11.7° with a correction rate of

65.9 %, and the compensatory caudal curve was corrected from 32.4° to 7.1° with a correction rate of 81.4 %. Our results were similar to Ruf's study and better than that of Bollini. It may have some relations with the different internal fixation system. In our and Ruf's study, pedicle screws were used, while in Bollini's study, laminar hooks were applied.

In patients with unbalanced multiple hemivertebrae, it is critical to decide which hemivertebra should be excised. Generally speaking, the hemivertebrae, which have obvious influence on scoliosis, should be excised. On the contrary, the hemivertebrae, such as incarcerated hemivertebrae, which have no influence on scoliosis, should not be excised. In addition, some patients have balanced multiple hemivertebra in one site and additional hemivertebra in another site. Great care should be taken to decide whether the balanced multiple hemivertebra should be excised.

In most patients with a single hemivertebra, short segmental fusion can be performed effectively and safely. But in the patients with unbalanced multiple hemivertebrae, long segmental fusion is usually needed to achieve satisfactory correction. The principle is to fuse all structural curves and preserve motion segments as much as possible. In young children with hemivertebrae located close to each other, short segmental fusion may be possible. And in young children with hemivertebrae located far away each other, respective short segmental fusion corresponding to the hemivertebra may be a good choice. In all other patients, long segmental fusion is usually necessary, which brings concern about the growth deficit after spinal fusion in young children. According to DiMeglio [26], we may expect a growth deficit at the end of growth after fusion five 5 vertebrae of 22 mm when surgery is performed at the age of 10 years. With fusion of ten segments (18 growth plates), the expected growth deficit is 49 mm (surgery at 10 years). In our study, the mean age of the patients at the time of surgery was 9.8 years, and the fusion segments averaged 7.4. So the estimated growth deficit in our study was among 22–49 mm. But according to Ruf, residual growth was observed in most cases of hemivertebra resection despite the instrumentation [18]. So the actual growth deficit is hard to estimate and a further observation is needed.

Pedicle fracture, which is the most common complication, was found in one patient. Except for technical error and improper instrumentation, the soft bone cortex and small pedicle diameter of these patients are another main cause [21]. Revision surgery was performed in one patient for progressive kyphosis resulting from pseudarthrosis. In such patients, it is critical to restore the normal sagittal balance in the first surgery. In addition, good decortication should be performed and enough bone should be grafted.

It is quite important to perform long-term follow-up in patients with unbalanced multiple hemivertebrae,

especially in immature patients with complicated deformities. In such patients, curve progression may occur even after a solid posterior spinal fusion. Progression depends on the amount of unbalanced growth potential and growth remaining. For young children who receive surgeries before puberty, long-term follow-up is mandatory because curve progression may accelerate when the patients go into the adolescent growth period of puberty. If obvious curve progression is found during follow-up, proper intervention should be given to avoid secondary deformity.

Conclusion

In the patients with unbalanced multiple hemivertebrae, hemivertebra resection allows for excellent correction in both the coronal and sagittal planes, and great care should be taken to reduce the rate of complications.

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