Clin Orthop Relat Res
DOI 10.1007/s11999-010-1650-8

SYMPOSIUM: MYELOMENINGOCELE

Kyphectomy Improves Sitting and Skin Problems in Patients with Myelomeningocele

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Abstract

Background Progressive kyphosis occurs in up to 20% of patients with myelomeningocele. Severely affected patients can develop recurrent skin breakdown, osteomyelitis, sitting imbalance, and poor cosmetic appearance.

Questions/purposes We (1) assessed the ability of kyphectomy to restore an intact skin envelope and allow comfortable seating in a wheelchair; (2) reviewed the complications of kyphectomy and spinal fusion in myelomeningocele; and (3) determined whether patients requiring unexpected reoperation had worse correction or more ulceration compared with those patients treated with a single surgery.

Methods We retrospectively reviewed the records of 23 children with thoracic-level myelomeningocele who were treated with kyphectomy and spinal fusion since 1980. Indications for surgery included recurrent skin breakdown (15 patients) and poor sitting balance or unacceptable cosmetic deformity (three patients). We evaluated operative technique, type of sacropelvic fixation, surgical complications, radiographic correction, and skin condition at followup. The minimum followup was 2 years (median, 4.1 years; range, 2.1–10 years); 18 of the 23 children had greater than 2 years followup and are reported here.

Results Kyphectomy achieved a sitting balance and resolved in skin ulceration in 17 of 18 patients. Seven patients had complications requiring reoperation. Three patients had multiple reoperations for early deep infection and one patient each had reoperation for late infection, pseudarthrosis, implant-related sacral pressure sore, and planned extension of proximal fusion after growth. Patients requiring multiple operations had similar correction and relief of ulceration to those treated with a single procedure.

Conclusions Complications after kyphectomy are frequent and many children with myelomeningocele and severe hyperkyphosis require multiple procedures and lengthy hospital stays. Nonetheless, improved seating balance and resolution of skin problems was achieved in 17 of 18 patients.

Level of Evidence Level IV, therapeutic study. See Guidelines for Authors for a complete description of levels of evidence.

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Each author certifies that his or her institution approved the human protocol for this investigation and that all investigations were conducted in conformity with ethical principles of research.
This work was performed at the Texas Scottish Rite Hospital for Children, Dallas, TX, USA.

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Introduction

Progressive kyphosis frequently presents in children with myelomeningocele. The incidence approaches 20% and is more common in patients with a thoracic level of neurologic function [3]. Kyphosis occurs as a result of

Published online: 02 November 2010

the absence of appropriate motor function and lack of posterior bony elements and can be compounded by congenital failures of spinal formation or segmentation. Sharp kyphosis often leads to deterioration of sitting posture and recurrent soft tissue ulceration that may be associated with osteomyelitis. Severe deformity can affect bimanual function because the upper extremities cannot be used to manipulate objects when they are being used to support the trunk upright with severe kyphosis.

Nonoperative treatment with bracing and wheelchair modification is usually ineffective and may only exacerbate soft tissue problems [5, 7]. Several techniques for surgically correcting severe kyphosis have been described, including use of Harrington rods, plate fixation, the Galveston technique, Dunn-McCarthy fixation, and the Warner and Fackler technique. Newer surgical techniques have improved sagittal plane correction; however, complication rates remain high (greater than 50%) for all techniques [1, 5, 6, 8, 10, 11, 13–15].

Major complications of kyphectomy include deep wound infection, osteomyelitis, skin ulceration resulting from recurrent deformity or prominent implants, cerebrospinal fluid flow dysfunction requiring shunt revision, and death. Minor complications include delayed wound healing, superficial infection, urinary tract infection, pressure sores from casting/bracing, postoperative lower extremity fractures, and asymptomatic pseudarthrosis. Although all reported series are relatively small, major complications occur with reported frequencies of between two of nine patients and eight of nine patients while the percentage final correction ranges from 39% to 96% (Table 1).

Because of the reported high rates of complications and variability of correction we sought to: (1) assess the ability of kyphectomy to restore an intact skin envelope and allow for comfortable seating in a wheelchair; (2) review the complications of kyphectomy and spinal fusion in myelomeningocele; and (3) determine whether patients requiring unexpected reoperation had worse correction or more ulceration compared with those patients treated with a single surgery.

Patients and Methods

We conducted a computer search of medical records since 1980 to identify all patients with myelomeningocele treated with a kyphectomy and spinal fusion. Patients treated for kyphosis without vertebral body resection or patients undergoing kyphectomy without the diagnosis of myelomeningocele were excluded. Indications for surgery were either recurrent problems with skin ulceration resulting from the gibbus, inability to sit comfortably resulting from the gibbus (15 patients), or for cosmetic improvement at the request of the patient and family as comfort with the procedure increased (three patients). We identified 23 children who met the inclusion criteria. All had thoracic-level myelomeningocele. Of these, two children had surgery less than 2 years ago and three were lost to followup. This left 18 children for review. Their age at the time of surgery ranged from 6.3 to 17.9 years (average, 12 years). The group consisted of 10 boys and eight girls. The minimum followup was 2 years (median, 4.1 years; range, 2.1–10 years). No patients were recalled specifically for this retrospective study; all data were obtained from medical records. We had prior Institutional Review Board approval.

All patients had resection of the gibbus and posterior spinal instrumentation. Basic principles of safe spinal exposure in myelomeningocele were used. The dural sac was identified in the distally open posterior segments of the spine. The sac was mobilized proximally and nerve roots were tied sequentially until intact posterior elements were found. The dural sac and enclosed spinal cord were transected at a level proximal to the planned kyphectomy. The sac was then closed with a pursestring suture, taking care to ensure the spinal cord was not sutured to prevent acute hydrocephalus by central canal obstruction. Dural sac

Table 1. Summary of previous series of kyphectomy in myelomeningocele

Study	Year	Number of patients	Preoperative kyphosis	Final kyphosis	Correction	No complications	Minor complications	Major complications
Garg et al. [current study]	2010	18	147	51	65%	5/18	7/18	6/18
Warner and Fackler [15]	1993	12	101	4	96%	8/12	2/12	2/12
Thomsen et al. [14]	2000	9	152	48	68%	7/9	0/9	2/9
Odent et al. [13]	2004	9	110	15	86%	3/9	3/9	3/9
Niall et al. [11]	2004	24	121	57	53%	4/24	1/24	19/24
Ko et al. [6]	2007	9	122	38	82%	1/9	0/9	8/9
Heydemann and Gillespie [5]	1987	12	124	33	73%	7/12	3/12	2/12
Furderer et al. [4]	1999	14	128	81	39%	4/14	0/14	10/14
Akbar et al. [1]	2006	24	124	43	65%	12/24	2/24	10/24

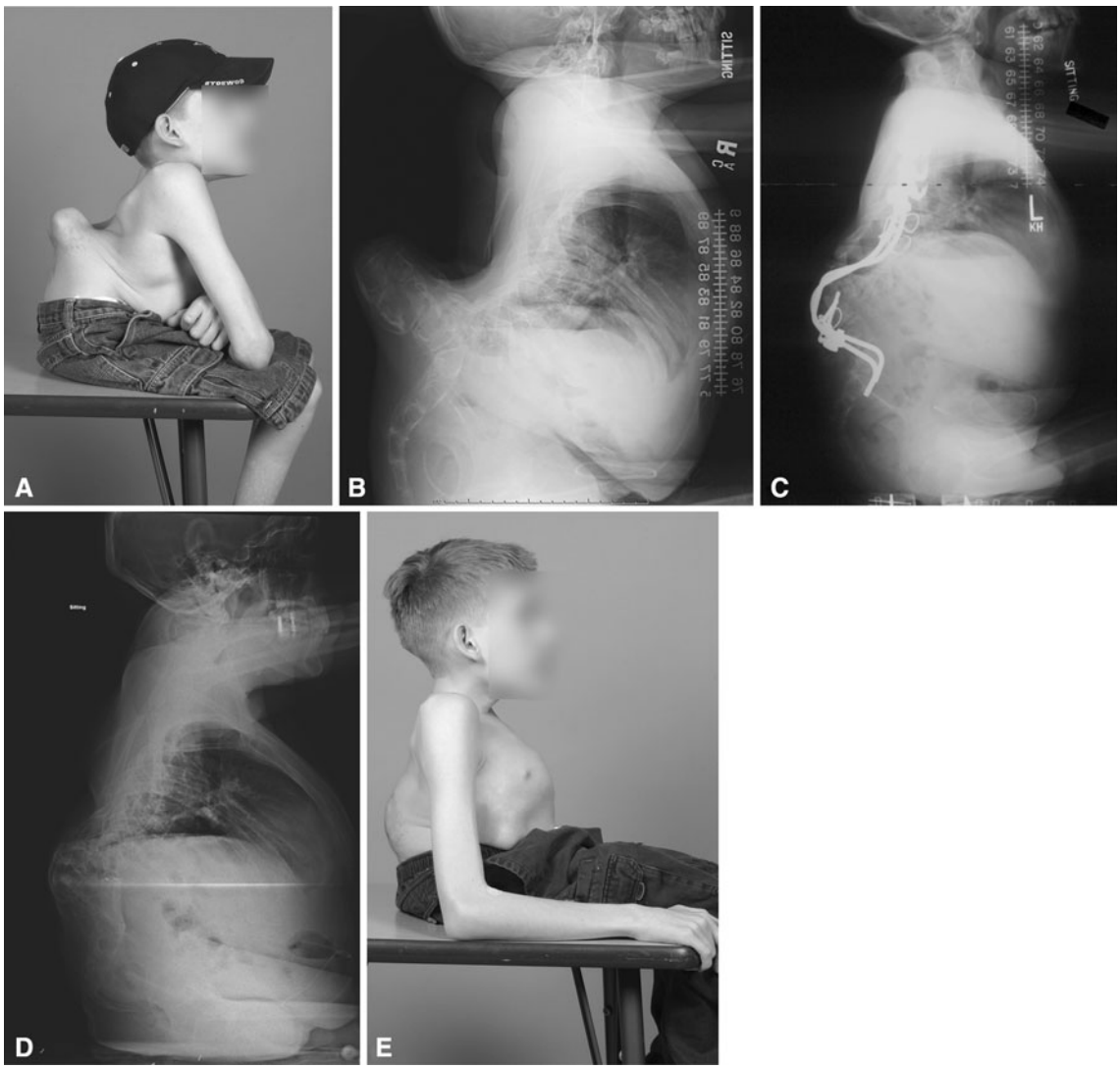

Fig. 1 A–E (**A**) A photograph and (**B**) a sitting sagittal radiograph show the preoperative clinical and radiographic appearance of a patient with thoracic-level myelomeningocele and severe rigid hyperkyphosis. (**C**) A postoperative sitting sagittal radiograph shows the patient after kyphectomy with segmental proximal spinal fixation and distal sacral fixation. The patient developed an early deep infection and was treated with an anterior fusion while the posterior infection was suppressed. Once the anterior fusion had solidified, the suppression was discontinued and the infection recurred, necessitating implant removal. (**D**) A radiograph shows the final radiographic appearance. Note the solid anterior fusion distal to the gibbus. (**E**) A photograph shows the final clinical appearance.

resection was performed to allow for improved exposure of the spine for kyphectomy. Extraperiosteal exposure of the kyphotic area of the spine was then performed to provide full anterior exposure before resection. Vertebral resection was then performed using a Gigli saw. The resected kyphotic levels were chosen based on preoperative planning to allow for restored balance when the proximal and distal segments of the spine were brought together. The technique for spinal instrumentation consisted of a combination of sublaminar wires, hooks, and pedicle screws. Four children treated before 1995 had distal fixation with Luque rods impacted into the iliac wings bilaterally. Five children treated between 1995 and 2003 had distal fixation by S-shaped rods placed anteriorly over the sacral ala as described by McCarthy et al. [9]. The remaining nine patients treated since 2003 had posterior instrumentation anchored distally with contoured rods placed through the first sacral foramen as described by Warner and Fackler [15] (Fig. 1). Local autograft from morselizing the resected vertebral bodies and supplemental allograft were used.

Postoperatively, the patients were treated with parenteral pain medication, as needed, for 2 to 3 days and then converted to oral analgesics. Unless there was concern about stability of the incision, the patients were placed upright into a bedside chair by postoperative Day 2 or 3. Subcutaneous drains were removed once the drainage was no greater than 50 to 100 mL over a 12-hour period. Patients were up in their wheelchairs within 5 days. Once

their sagittal spinal deformities were improved, the necessary adjustments to their wheelchairs were performed by the occupational therapy department before discharge. Instructions for patient transfers were performed by nursing staff or physical therapists. Because none of the patients were ambulators, there was no formal physical therapy program prescribed postoperatively. All patients had brace immobilization of their spine for 4 to 6 months postoperatively.

Patients returned to the clinic approximately 6 weeks postoperatively, 6 months postoperatively, 1 year postoperatively, and yearly thereafter. The condition of the skin of the back and lower extremities was examined carefully to check for pressure sores or incisional irritation. Sitting AP and lateral radiographs were obtained at each clinic visit.

We reviewed the clinical records to identify the indication for surgery, operative technique, estimated blood loss, operative time, complications, need for reoperation, and clinical course.

One of us (SG) reviewed all radiographs and recorded the degree of kyphosis on sitting films at three time points: last radiograph before surgery, first radiograph out of brace or cast postsurgery, and most recent radiograph. Kyphosis was measured using the Cobb method. Pencil and goniometer were used for standard hard-copy radiographs and electronic measurement was performed for digitized radiographs using Synapse® (Version 3.2.1; Fujifilm Medical Systems, USA, Stamford, CT). All radiographs were measured by a single author (SG). Carmen et al. showed 95% of the differences between multiple measurements of kyphosis are 7° or less [2]. Pseudarthrosis was inferred when broken rods were identified on radiographs or by direct observation during revision surgery.

Patients treated with one operation (n = 11) and patients requiring multiple operations (n = 7) comprised the two groups compared for differences. To compare the groups, 95% confidence intervals (CIs) for the difference in population means were computed for all studied factors (age at surgery, length of followup, estimated blood loss, operative time, kyphosis, and length of hospitalization). Conclusive differences in the means of the two groups were observed when the 95% CI for the difference did not include zero. If the 95% CI for the difference included both positive and negative values, we presumed there was no difference in the two groups. Statistical analyses were performed using SAS/STAT® software (Version 9.1 of the SAS® System for Windows®; SAS Institute Inc, Cary, NC).

Results

At final followup, 17 of the 18 children could sit comfortably in their wheelchair and had a healthy posterior skin envelope. The sole child who failed to achieve treatment goals at last followup developed an early deep infection that ultimately required implant removal. Although he initially had good correction of his deformity, once the implants were removed, the deformity recurred and his sitting balance was not improved from his preoperative position. Despite the severe deformity, he did not have skin breakdown over his kyphosis at last followup.

Major complications were defined as those requiring an unplanned surgical procedure, whereas minor complications could be dealt with nonoperatively. Thirteen of the 18 patients developed at least one complication (seven major, six minor); five had no complications (Table 2). The most frequently identified complications included deep infection (four patients), delayed wound healing not requiring surgery (three patients), pseudarthrosis (five patients), and femur fracture during brace immobilization (two patients). Seven patients required at least one additional surgical procedure, three for early (less than 30 days) deep infection and one patient each for late (greater than 12 months) deep infection, pseudarthrosis, prominent implants producing a sacral pressure sore, and a planned proximal extension. The latter was the youngest patient in the cohort (aged 6.3 years at time of surgery) and had proximal fixation initially without fusion to allow continued growth. Once the rods had disengaged from the proximal sublaminar wires as a result of growth, the patient had definitive spinal fusion proximally. All children treated for either early or late infection ultimately required removal of their posterior implants. Two patients with early deep infection underwent anterior fusion while the posterior infection was suppressed. Once the anterior fusion had solidified, the suppression was discontinued. In both patients, the infection recurred necessitating implant removal (Fig. 1). Although pseudarthrosis was detected in five children, only two patients required surgical intervention for progressive deformity. Three patients had broken implants identified on radiographs but were asymptomatic without deformity, although one of these patients developed a late deep infection and required implant removal. Because of the relatively small sample sizes, there were no statistical differences between those patients treated with one operation and those requiring multiple operations with respect to age at the time of surgery, length of followup, estimated blood loss, operative time, preoperative kyphosis, postoperative kyphosis, and kyphosis at final followup (Table 3). However, there is a trend towards smaller, final kyphosis in the single operation group. Also, the percent correction at final followup averaged 72° in the single surgery group as opposed to 56° in the multiply operated group. The method of surgical instrumentation did not affect final

Table 2. Concise patient information.

Patient number	Age (years)	Followup (years)	Estimated blood loss (mL)	Operating room time (hours)	Preoperative kyphosis (degrees)	Final kyphosis (degrees)	Length of stay (days)	Complications	Number of reoperations
1	10.8	8.6	1700	3.6	152	59	29		0
2	16.1	2.3	2800	7.5	156	59	15		0
3	13.6	3.1	1200	7.1	119	50	9		0
4	12	5.1	1730	5.4	120	0	7		0
5	13	3.3	900	7.0	143	30	9		0
6	10.4	6.8	2700	3.5	118	25	23	Urinary tract infection	0
7	11.9	3.3	2000	2.8	150	35	17	Stable, asymptomatic radiographic pseudarthrosis	0
8	14.6	3.1	3000	8.0	147	53	26	Pressure sore from brace	0
9	6.9	6.7	500	5.5	120	22	19	Delayed wound healing, not requiring reoperation	0
10	11.1	4.6	800	7.8	150	51	111	Delayed wound healing not requiring reoperation, asymptomatic radiographic pseudarthrosis	0
11	15.1	2.2	1700	4.8	179	57	8	Deep venous thrombosis, femur fracture	0
12	8	9.2	700	4.6	176	37	43	Urinary tract infection, delayed wound healing not requiring reoperation, progressive pseudarthrosis	1
13	9.1	9	750	4.0	133	51	38	Prominent implants producing pressure sore	1
14	6.3	10	1000	5.3	155	55	13	Femur fracture	1
15	17.9	2.1	3000	8.0	174	63	17	Late deep infection, progressive pseudarthrosis	1
16	14.1	4	1400	6.2	167	111	25	Early deep infection	4
17	13.9	4.1	1200	7.1	140	100	182	Early deep infection, progressive pseudarthrosis, recurrent kyphosis	8
18	11.8	2.9	800	5.1	148	58	282	Early deep infection	10

Table 3. Results between groups

Parameter	Single surgery group (N = 11) Mean (range)	Multiple surgery group (N = 7) Mean (range)	Range of 95% confidence interval of difference
Age at surgery (years)	12.3 (6.9–16.1)	11.6 (6.3–17.9)	−3.3 to +4.7
Length of followup (years)	4.5 (2.2–8.6)	5.9 (2.1–10.0)	−4.7 to +1.9
Estimated blood loss (mL)	1730 (500–3000)	1264 (700–3000)	−571 to +1502
Operative time (hours)	5.7 (2.8–8.0)	5.7 (4.0–8.0)	−2.2 to +2.1
Preoperative kyphosis	141° (118–179)	156° (133–176)	−38.2° to +8.5°
Postoperative kyphosis	38° (0–65)	60° (35–112)	−50.7° to +8.2°
Kyphosis at final followup	40° (0°–59°)	68° (37°–111°)	−55.9° to +0.3°
Percent correction at final followup	72.3% (58.0–100.0)	56% (28.6–79.0)	−2.1 to 34.8
Hospital days	25 (7–111)	86 (13–282)	−130.8 to +9.0

correction. As expected, children requiring multiple operations had a longer average duration of hospitalization than those successfully treated with a single operation (Table 3).

Discussion

Severe, rigid hyperkyphosis in patients with myelomeningocele is a challenging problem. Severe deformities can

produce complications resulting from altered sitting balance and skin pressure. Therefore, we (1) assessed the ability of kyphectomy performed through a posterior approach with vertebral body resection and posterior instrumentation to restore an intact skin envelope and allow for comfortable seating in a wheelchair; (2) reviewed the complications of kyphectomy and spinal fusion in myelomeningocele; and (3) determined whether patients requiring unexpected reoperation had worse correction or more ulceration compared with those patients treated with a single surgery.

There are a few limitations to our study. First, because this was a retrospective chart and radiographic review, there is the potential for reporting bias in the medical records. Despite the potential for reporting bias, we believe our use of objective findings (presence of skin breakdown, need for reoperation, radiographic measurements) limits the influence of this type of bias. Second, we lacked a validated patient-related outcome measurement tool to record patient outcomes after spinal deformity surgery in myelomeningocele; as such, we are unable to discuss the effect of this surgical procedure on patient-related outcomes other than those we measured. Third, radiographic review is challenging with severe spinal deformity, although to minimize interobserver variation, we used a single reviewer with a uniform technique. Although this can introduce systemic bias, the inter- and intraobserver variances of radiographic measurements are well documented [2] and do not alter the fundamental findings of our research. Fourth, although this is one of the largest case series reported on this subject, there are only 18 patients, making comparisons of patients within the series difficult as a result of the small number.

Acknowledging these limitations, our data suggest kyphectomy through vertebral body resection through a posterior approach with segmental posterior spinal instrumentation can improve sitting balance and reduce ulceration in patients with myelodysplasia and thoracolumbar kyphosis. Seventeen of our 18 patients achieved improved sitting balance with a stable spine and an intact skin envelope. These findings compare favorably with others in the literature. McMaster [10] reported two of his 10 patients (treated with a variety of surgical techniques) had superficial wound necrosis after kyphectomy, which healed with nonoperative treatment, and three other patients had ulcerations over prominent hardware requiring hardware removal for wound healing. Ko et al. [6] reported seven of nine patients with wound healing issues after kyphectomy (surgical technique not described). Eventually six of their seven patients eventually required implant removal. In one of the largest series in the literature, Niall et al. [11] reported 19 of 24 patients (treated with multiple surgical techniques) with wound complications. Thirteen of these patients had issues with primary wound healing with 11 of these patients eventually having chronic issues with hardware exposure. Six patients had delayed wound breakdown as a result of prominence of hardware and ulceration. Clearly wound healing is a major source of problems in this patient population. Obviously, the previously operated and subsequently compromised posterior skin is a risk factor for infection, but there has been no consensus as to how to best address this issue preoperatively. In general, we treated soft tissue wounds with local wound care until healing had been obtained and it was deemed safe to proceed with surgical intervention. Other authors have reported extensive preoperative hospital stays for wound management before kyphectomy, whereas others performed surgical débridement of the area before kyphectomy to optimize wound healing [11, 13].

Although wound problems are the most common postoperative complication after kyphectomy, there are other potential complications. We identified complications in 13 of 18 patients, seven of whom required a repeat operative procedure to address the complication. Nolden et al. [12] reported six complications (no mortality) in 11 patients using their technique of decancellation vertebrectomies. In the two largest series in the literature, Niall et al. [11] and Akbar et al. [1] each reported separate case series of 24 patients. Niall et al. [11] reported 20 complications in 24 patients, most consisting of wound issues, and Akbar et al. [1] reported 12 complications in 24 patients, including one perioperative death. Fortunately, we were able to achieve similar correction and relief of ulceration for patients requiring only a single operation compared with those who had an unexpected reoperation. The amount of sagittal plane correction appears to be a factor in limiting postoperative complications and achieving clinical success. We observed a trend toward improved kyphosis at last followup in patients requiring only one operation compared with those requiring multiple operations. Other authors have demonstrated the importance of maximal deformity correction in long-term success of this procedure [1, 13].

Early deep infection is perhaps the most common complication in this patient population. The management of these infections can be difficult. Our initial patient who developed early deep infection was treated with débridement and suppression of the infection in an attempt to delay removal of the implants until a solid posterior fusion had developed. When the implants were removed, we believed this patient to have a solid fusion; however, over time, he developed recurrent deformity distal to his fusion. As a result of this experience, we treated the subsequent two patients with early deep infection with an anterior fusion, whereas the posterior infection was suppressed. Both patients developed recurrent infection necessitating implant removal, but neither developed recurrent deformity (Fig. 1). Other authors have also reported substantial loss of correction after early hardware removal without

supplemental fusion to treat complications [7, 9]. A recent article advocated performing a staged anterior spinal fusion after kyphectomy with posterior spinal fusion in all of these patients. Odent et al. [15] reported nine patients using this circumferential fusion technique with excellent immediate kyphosis correction (86% correction), no loss of correction at final followup, and no complications using this combined technique. Although we agree anterior spinal fusion in this patient population is feasible and effective, we have had good success in correcting and maintaining correction of the kyphosis using only the posterior approach. We do, however, believe using an anterior spinal fusion as a salvage technique in these cases in the face of deep infection necessitating hardware removal may facilitate clearance of the infection while preventing loss of kyphosis correction.

In conclusion, we found kyphectomy with vertebral body resection and segmental posterior spinal instrumentation effective in achieving correction of deformity, healing of skin ulceration, and improving sitting balance in children with severe thoracolumbar kyphosis associated with myelomeningocele. However, this success comes with a high incidence of perioperative complications. Patients and their families should be counseled preoperatively regarding the likelihood of complications and possibility of multiple surgical procedures. Fortunately, initial preoperative goals can be achieved in patients requiring multiple operations.

Acknowledgments We thank Tara Kristof for assistance with obtaining medical records and database entry and Richard Browne, PhD, for statistical analysis.

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