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Myelomeningocele and Kyphosis: Natural History of the Disease and the First Results of a New Surgical Technique

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ABSTRACT

AIM: To define the natural course of kyphosis, and to evaluate the efficiency of a new technique in surgical correction of kyphosis seen in myelomeningocele(MM) patients.

MATERIAL and METHODS: We retrospectively reviewed our patients with MM. The rate of kyphosis, mean angle of progression and mean angle of surgical correction were evaluated. Surgical correction was achieved with the same technique in all patients; kyphectomy, short segment instrumentation with plate system and long segment instrumentation with screw-rod system.

RESULTS: A total of 14 patients were treated surgically and the mean age at the surgery was 39 months. The incidence of kyphosis rate was %21 in this study. The mean angle of kyphosis was 85.8°. Average angle of progression was 15.7° whereas it was 6.3° degree in patients whose kyphosis angle ≤90 and >90 degree, respectively, at birth. 14 patients were treated surgically and the mean age at the surgery was 39 months. The mean angle of correction of kyphosis was 86 degree. The most common complications were wound dehiscence and cerebro-spinal fluid leak. One patient died 3 months after surgery, and one patient was reoperated due to pull-out of screws.

CONCLUSION: Effective surgical correction of kyphosis in MM patients can be achieved with the described surgical technique even in younger ages. Prospective studies in larger study population are necessary for more accurate definition of natural history of kyphosis in MM patients.

KEYWORDS: Myelomeningocele, Kyphosis, Spinal Deformity, Kyphectomy

ABBREVIATIONS: **MM:** Myelomeningocele, **MRI:** Magnetic resonance imaging, **CSF:** Cerebrospinal fluid, **CT:** Computed tomography

INTRODUCTION

Myelomeningocele (MM) is not only a simple neural tube formation defect but also, can lead to progressive deformity of the spine (19,32). The lack of posterior bony elements and the spinal musculature may cause two types of deformities: lordoscoliosis and sharp angled kyphosis (32). Progressive neurologic compromise, skin ulceration, decreased lung capacity, sitting difficulties and osteomyelitis may occur according to the severity and the level of deformities (2,16,21,22,37).

Among these two deformities, lordoscoliosis is seen more frequently, thus the natural history of the disease, indications of surgery and surgical management have already been documented (6,13). However, the management of kyphosis, especially in severe forms, is more complex, because the natural history is still unknown. Surgical indications are not clear and there is no unique surgical approach which has been described (6,13,33,34). Published papers include only case reports and limited case series at different ages and managed

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by different techniques. Long-term follow-up results of these studies are lacking especially in younger ages.

In this study, we analyzed the rate of kyphosis in patients with MM, evaluated the natural history focusing on the progression of the kyphosis and described a new surgical technique used for deformity correction, with long-term results in younger ages.

MATERIAL and METHODS

This study was conducted in a single tertiary center. All patients diagnosed with MM between 2009-2019 were retrospectively analyzed. The aim of this study was to evaluate the natural history of kyphosis in patients with MM. Patients who had preoperative radiologic studies, at least 2 years of follow-up and radiologic documentations during the follow-up period, were included in the study. Magnetic resonance imaging (MRI) and/or X-Ray were used to evaluate kyphosis using Cobb angle.

Patients who received kyphectomy and instrumentation with our technique were reviewed to evaluate results of the surgery. Skin ulcerations over the deformity and sitting imbalance were indications of the surgery (Figure 1A, B). The main goal of the surgery was to prevent pressure on the skin and maintain an optimal alignment of the spine. All patients were operated using with the same surgical technique.

Surgical Technique

All patients were operated in prone position. For patients with skin ulceration, incision was spindle shaped to isolate the affected area. For those with no ulceration, skin incision was made in the midline. Incision length included at least two levels above the apex of the kyphosis proximally and the iliac bones distally. After skin incision kyphosis was exposed and if present in the kyphectomy area, the dural sac was transected with enclosed spinal cord and tied up proximally and distally (Figure 2A, B). Kyphectomy was performed by removing apical vertebrae, adjacent vertebrae and intervertebral disks to manipulate the spine. Based on intraoperative observation,

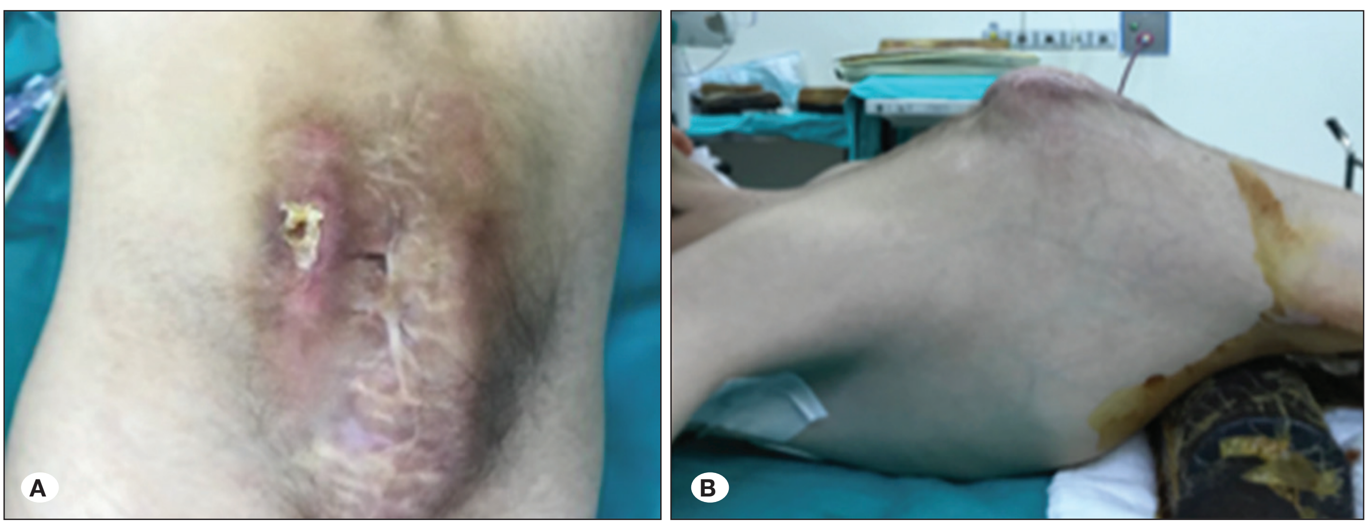

Figure 1: Skin ulceration **(A)**, and gibbus deformity **(B)** that caused sitting imbalance were indications of the surgery.

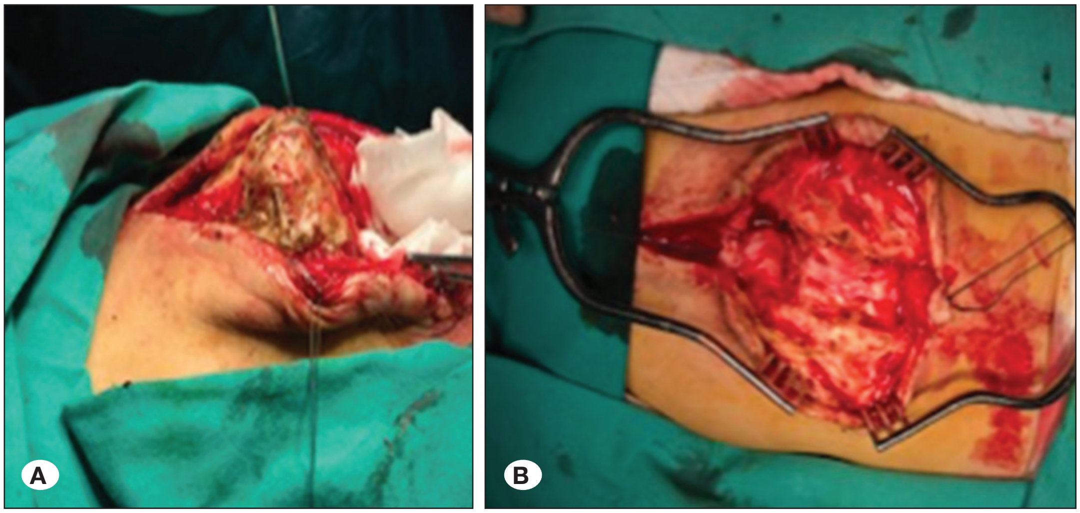

Figure 2: After skin incision kyphosis was exposed **(A)**. If there was dural sac at the kypectomy area it was transected **(B)**.

the extent of vertebrectomy in order to maintain spinal alignment adjacent to the apical vertebra was decided. When the upper and lower vertebrae around the kyphectomy area could be pushed away with fingers from posterior to anterior like a "closing bridge," no more vertebrectomy was performed (Figure 3A, B).These segments were then stabilized using a plate system (Figure 3C). To improve stabilization and alignment, vertebrae that were located cranially at the kyphectomy area were fixed with pedicle screws, and the system was completed with unshaped straight rods to the iliac screws distally (Figure 3D). Dural sac was then closed with the suture while ensuring that the spinal cord was not sutured. Figure 4A-D shows the effect kyphosis correction after surgery in a patient. Surgical technique is schematized in Figure 5A-D.

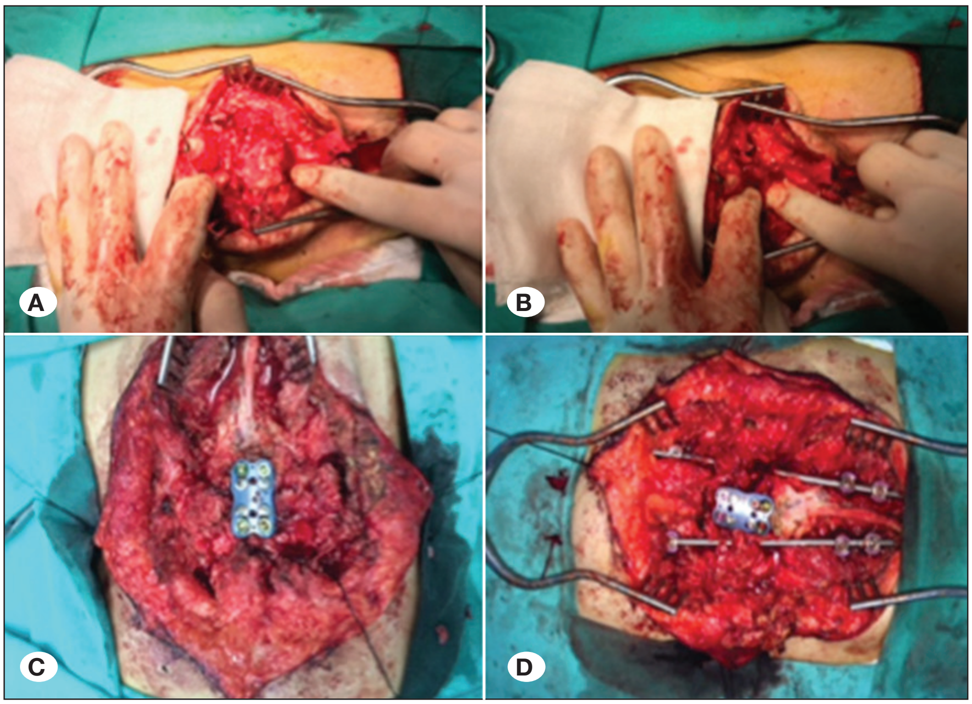

Figure 3: Proximal and distal vertebras pushed away by fingers **(A,B)** from posterior to anterior. Plate system was used to ensure short segment instrumentation first **(C)** the screw-rod system was performed from proximal vertebras to the iliac bone **(D)**.

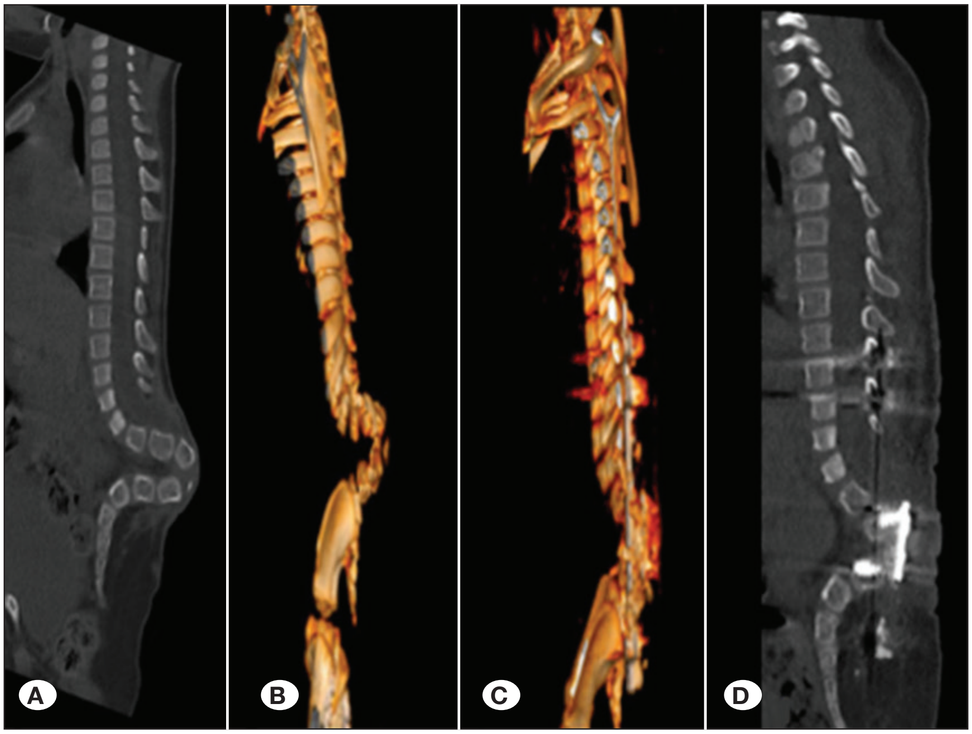

Figure 4: Preoperative reconstructed sagittal Computed Tomography (CT) scan **(A)** and Three Dimension (3D) CT scan **(B)** shows sharp angled lumbar kyphosis in 35 months old boy. Successful surgical correction was achieved with screw-rod system **(C)** and plate system **(D)** in this case.

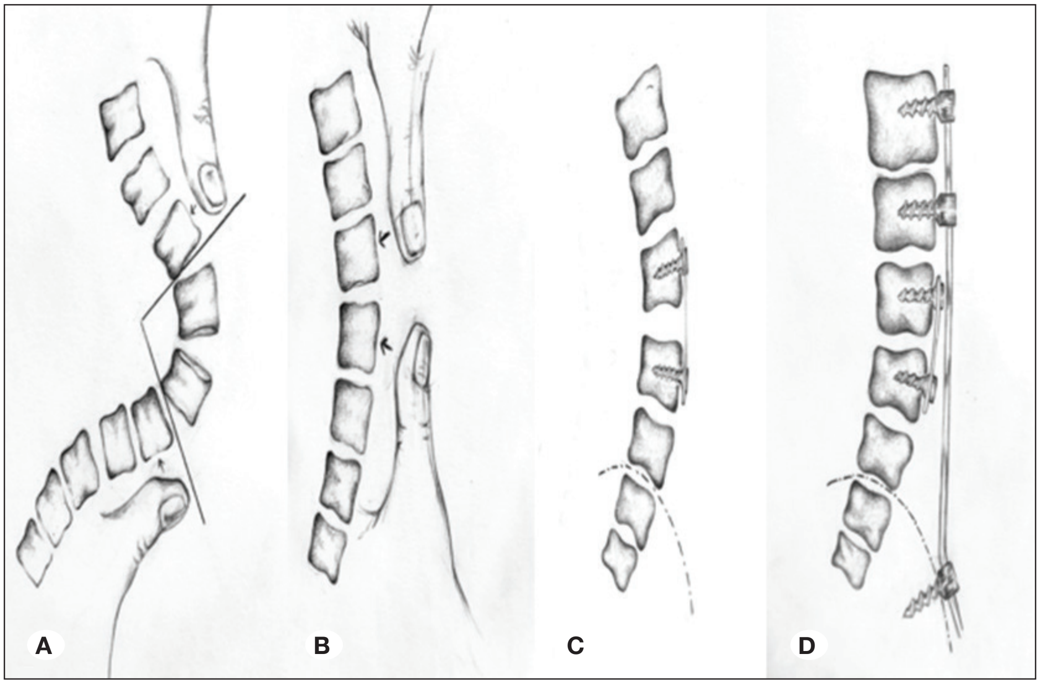

Figure 5: Schematic drawing of the surgical steps. Kyphectomy area was determined by the fingers while the spine being mobilized **(A,B).** Plate **(C)** and screw-rod systems were placed **(D)**.

RESULTS

A total of 147 patients with MM were included in the study. Thirty-two of them had kyphosis at birth. Kyphosis did not develop at the end of the follow-up period (mean 26 months) in rest of 115 patients.

Lowest and highest kyphosis angle were 26.5° and 157.1°, respectively (mean, 85.8°). Progression of the kyphosis was seen in 23 patients. In 9 patients, kyphosis angle was above 90.0° (mean, 112.4°), which was defined as sharp angle. All patients who had a sharp angle showed progression, and the mean angle of progression was 15.7°. For the remaining 14 patients, the mean angle was 67.6° (range, 42°–87°) and the mean angle of progression was 6.3°.

According to surgical criteria, surgical treatment was offered to 16 patients. However, 2 families refused surgery, thus, only 14 patients were operated upon. Patients data is summarized in the Table I.

There were 10 girls and 4 boys. The mean age of the patients at the time of surgery was 39 months (range, 21–49 months). The apex of the kyphosis was L3 in 8 patients, L2 in 4 patients and L4 in 2 patients. Skin ulceration was seen in 12 patients. Four of them had a history of long-term medical treatment for osteomyelitis. Nine patients had a gibbus deformity, and 11 had sitting imbalance.

Six of 14 patients were discharged without any complication after surgical treatment. In 7 patients, wound dehiscence was seen after surgery. Four of them also had cerebrospinal fluid (CSF) leakage, and dura repair was performed. Pneumothorax occurred in 1 patient and he died 3 months after surgery due to sepsis. In another patient, the whole system except the plate was removed due to pulling out of screws.

Deformity correction was successful in all surgically treated patients. The mean postoperative kyphosis angle was 27.1° (range, 3°–34.8°). The mean correction angle was 76.2° (range, 64.4°–126.7°). One patient experienced screw pull out. In this patient, pedicle and iliac screws were removed at second operation, but not the plate system. Following correction, there was a 14-degree angle loss from immediate postoperative to 17 months follow-up.

DISCUSSION

The incidence of kyphosis in patients with MM has been reported to be 1%–48% in the literature (8,10,13,25,30). One of the largest series was published by Carstens et al. (5). They detected 151 patients out of 719 patients (21%). In our study, the rate of incidence was 22%. Carstens et al. found that the most common location of the apex was L2 (5), in the present study it was L3.

Natural history of kyphosis in patients with MM is not well known (4,5,8,11,34). Deficiency in the posterior bony elements impairs the balance between the anterior and posterior musculature, which is the main cause for progression of kyphosis. The degree of kyphosis progression is mainly determined by the severity of imbalance, and severe displacement of the muscles act as perverted lumbar flexors and consequently cause sharp angled kyphosis (13,30,32,33). Although it has been reported that the mean progression rate ranges from 2° to 6°; depending on the severity of imbalance and the age of the patient, it may cause higher degrees of progression (7,13,32,33). In a study of 35 patients, Mintz et al. found that the progression rate was greater when the patient had a sharp angle (>90°) at < 1 year old. They also found that the angle did not affect the progression rate in those >1year old (25). In our study, all patients were evaluated according to their angle at

Table I: Preoperative and Postoperative Data of Patients is Summarized in Table

Age (Months)	Indication of Surgery	Level of Paralysis	Level of Apex	Preoperative Degree of Kyphosis	Degree of Kyphosis After Surgical Correction	Follow-up (Months)	Complications
21	SU	N/A	L4	81.4	3.0	25	Wound dehiscence, CSF Fistula
29	SU	N/A	L3	86.3	18.7	32	None
33	SU, SI	N/A	L4	88.6	15.6	41	Wound dehiscence
35	SU, GD	N/A	L2	93.2	28.8	27	None
37	SU, SI, GD	N/A	L2	157.1	30.4	31	Wound dehiscence, Screw Pull-out, CSF Fistula
38	SU, SI, GD	N/A	L3	131.7	32.8	38	Wound dehiscence
39	SU, SI, GD	T10	L3	123.4	34.8	28	None
42	SU, SI, GD	T10	L3	89.3	28.7	26	Wound dehiscence, CSF Fistula
43	SU, SI, GD	T12	L2	98.3	28.9	40	None
43	SU, SI	T12	L3	87.3	32.6	32	Wound dehiscence
45	SU, SI, GD	T8	L3	97.6	31.7	53	Pneumothorax
45	SU, SI, GD	T10	L3	106.8	32.2	27	Wound dehiscence, CSF Fistula
47	SI, GD	T12	L3	93.1	28.9	47	None
49	SI	T10	L2	110.4	31.6	43	None

***SU:** Skin ulceration, **SI:** Sitting imbalance, **GD:** Gibbus deformity, **N/A:** Not applicable.*

birth. We found that 23 of 32 patients showed progression of kyphosis. Significant progression was seen in patients who had sharp angle kyphosis (mean, 15.7°). The rest of the patients showed mild progression (mean, 6.3°), and their mean kyphosis angle was 67.6° at birth.

Progression of kyphosis does not only depend on the anatomic nature of the pathology, but also on the increased moment of the arm and physiologic load on sitting. Eventually, vertebral bodies become wedge shaped anteriorly and thoracal lordosis develops (13). As the severity of the kyphosis progresses, it may lead to gibbus deformity, sitting imbalance, skin breakdown, osteomyelitis and end up with respiratory compromise. The most common findings were skin ulceration, sitting imbalance and gibbus deformity, in our surgically treated patients. One patient had a respiratory compromise in addition to her gibbus deformity.

Non-surgical management including bracing is not the treatment of choice. There is a consensus about the surgical indications as sitting abnormality and skin ulceration (2,5,10,21). These two clinical scenarios were also indications of surgery in our cases. The timing of surgery and the optimal type of surgery is controversial. We operated our patients who had surgical indications "as soon as possible." Although some reports showed that neonatal kyphectomy provides good initial correction, it has also been shown that recurrence is common (7,32,33,34). In older children, extensive fusion and instrumentation are needed. However, no unique surgical approach has been described in the literature. Since the first description of vertebra body resection by Sharrard in 1968, numerous techniques has been described such as posterior fusion, anterior fusion, sublaminar wires, pedicle hooks, plates, cables and pedicle screws (6,13,32,33,35). However, due to rarity of the disease and the high risk of the surgery, published clinical series are scarce and no consensus has been made yet. In our series, we used plate system and pedicle screws to maintain sagittal alignment with posterior only approach. To the best of our knowledge, this combined technique has not been defined before in the literature.

With lack of consensus about the techniques to maintain the spinal alignment, the optimal age for surgery and the extent of fixation is debatable. Most surgeons agree that surgery can be performed at 5–12 years of age, to not prevent chest development (13,18,19,30,32,33). Some reports indicated that growing rods can be used for not to prevent spine growth in these cases. Alshaalan et al. performed distal intravertebral fixation and thoracic growing rod in their case with good result (1). Bas et al. used growing rod in their 10 cases and they concluded that this system is a reasonable alternative of fixation postkyphectomy (3). In these two studies the authors used high thoracic levels for proximal fixation. In the presented study, we used two vertebrae close to the kyphotic level for proximal anchoring and no proximal vertebra from the T11 was included in the instrumentation, For that reason, we were not anxious about any negative effect on lung development due to minimal intervention to thoracic area.

Both short and long segment fixation has been described in the literature (6-8,12,13,33). Short segment fixation is usually done by wires and sutures around the posterior dysplastic elements especially in younger children. However, only short segment fixation may not provide good surgical correction of kyphosis and lead to loss of initial correction (17). Many reports have described the excellent surgical correction rates with long segment fixation. In 1986, Heydemann and Gillespie published their results of long segment stabilization (14). They used posterior segmental instrumentation with Luque technique in 12 patients with satisfactory results. The mean age of patients was 11 years in their study. Loss of correction was seen in only one patient at the end of the follow-up. Kocaoglu et al. used posterolateral placed poliaxial screws in combination with segmental Luque instrumentation (20). They used this technique in 7 patients with significant reduction of the kyphosis. The mean age of their patients was 7 years. Their technique differs from the technique that was described by the Heydemann and Gillespie, by additional screws and wires to the rods around the kyphectomy site. None of their patients showed significant loss of correction at the end of the follow-up. Hwang et al. reported their experience with only long segment pedicle screw construct. They concluded that severe spinal deformity could be treated using this technique (15). However, their experience composes just two cases. Although all these studies showed excellent surgical correction rates, due to longer segments of the spine are fused (From high thoracic levels to pelvis) spinal growth is likely to be compromised and may prevent chest development in younger ages as mentioned earlier. Martin et al. published results of kyphectomy with wire fixation and spinal fusion in their 10 patients (23). Mean age of their patients was 59 months and they concluded that long segment stabilization is more likely to be used in older children (>8 years). In this study the mean age of patients was 39 months which is youngest in the available literature. As concluded by Martin et al. we believe that the surgery should be performed early before development of severe deformity (23). Our technique has two advantages as follows: plate system stabilizes the kypectomy area as short segment fixation and screw-rod system provide additional correction without including high thoracic levels that might compromise chest development.

Most surgeons include the pelvis to the fixation for better correction of deformity (12,13,33,34). An iliac bone is more frequently used than a sacral bone for distal screw fixation because of the soft nature of the sacrum (4,13,34). Niall et al. performed long segment stabilization with different techniques (Luque, Harrington, Cotrel-Dubousset, Gardner bottle screw, Hartshill rectangle) in their 24 patients (27). They included pelvis in the fixation in 10 patients and concluded that these patients had better correction rates (64% vs 52%). Altiok et al. included pelvis in the stabilization in their patients (2). The mean correction rate was 81% in their study. Mc Call reported 86% mean correction rate with including the pelvis in the stabilization (24). In our cases we used iliac screws at the caudal side to provide good surgical correction. With this technique, the mean deformity correction angle was 76.2° in our patients which is similar to that in previous reports using different techniques (8,12,13,32,34).

Surgery of the kyphosis in patients with MM has risks for serious complications such as wound dehiscence, CSF leakage, infection, implant failure and even death (4,11,12,38). These complications may be seen as high as 93% and the wound dehiscence is seen between 63%-89% (8,10,30,36). Overall complication rate was 57% in our study. The most common complications were wound dehiscence and CSF leakage as in the literature. We had more acute wound problems compared to the literature. The reason for this difference could be that the mean age of our patients is lower than that in other studies. Petersen et al. reported that 3 of their 28 patients experienced implant failure (31). They used sacrum for distal anchoring point in their surgical technique and suggested that using more anchoring point such as iliac screws would be more effective to prevent pseudoarthrosis. In our surgical technique we used iliac screws as a distal anchoring point and pull out of screws was seen only in one case. We did not remove the plate system in this case and there was no progression after 17 months follow-up. Duddy et al used only plate system in their case and had good results (9). One patient died due to pneumonia related sepsis that developed after iatrogenic pneumothorax during surgery. In this case the vertebrae fused with the ribs, which made the surgery difficult. Petersen et al. reported that their 19 of 28 patients underwent reoperation due to infection and implant failure. They used long segment stabilization from high thoracic levels to the sacrum.

Kyphectomy in MM patients can be done with or without transection of the dural sac. Mc Call and Nolden et al. showed that sacrification of the non-functional segmental lumbar nerves creates an enough lateral space for kyphectomy (24,28). Odent et al. also used same technique in their 7 cases with satisfactory results (29). Miyamato et al. described a technique for kyphectomy with surgical threadwire to preserve the entire dural sac (26). In our surgical technique, transection of the dural sac should be made to use plate fixation of vertebrae, which enables proper correction of the kyphosis. For this reason, our technique cannot be used if patient has conserved spinal cord function below the level of the kyphosis. However, most patients who need surgical intervention have severe kyphosis and already have paraplegia.

CONCLUSION

Kyphosis in patients with MM is a progressive deformity and likely to cause serious complications. Early surgical correction is mandatory for preventing complications. Our surgical procedure provides an excellent surgical correction of the kyphosis and can be used even in younger patients. To detect the real incidence of deformity and the risk factors of progression, prospective studies in large patient series are needed.

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