



POSTERIOR VERTEBRAL RESECTION OSTEOTOMY IN A PATIENT WITH MYELOMENINGOCELE – CASE REPORT AND LITERATURE REVIEW

MYELOMENİNGOCELLİ BİR HASTADA POSTERİOR VERTEBRA REZEKSİYON OSTEOTOMİSİ – OLGU SUNUMU VE LİTERATÜR İNCELEMESİ

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SUMMARY

Aim: The term myelomeningocele is used to define a neural tube defect affecting the spine and spinal cord. It is commonly found with spinal deformations, such as kyphosis and lordosis (62%), as well as other neurological problems. Scoliosis is seen at a rate of 50–90%, and kyphosis in the lumbar region is seen at a rate of 8–20% with myelomeningocele. Most of the curvature is rigid, often greater than 80° at birth, and it is generally progressive, progressing 6–12° per year. Surgery for the deformation carries high complication risks. The aim of this study is to evaluate the inclusion of the skin slipping process in the same session as posterior lumbar vertebral resection osteotomy without damaging the neural structures, in a myelomeningocele patient with kyphosis.

Methods: Stabilization together with posterior vertebral resection osteotomy was administered to a 6-year-old male patient with myelomeningocele (myelodysplasia or spina bifida) deformation with rigid kyphosis and skin problems, with rigid lumbar kyphosis deformation and scoliosis accompanying the posterior vertebral elements. Stabilization was performed using thoracic-lumbar-pelvic pedicle screw fixation (T9–sacroiliac), resection osteotomy from the L3 vertebral disc levels, and skin slipping with plastic surgery for sufficient scar closing in the same session. A postoperative brace was administered for six months.

Results: While the preoperative lumbar kyphosis angle was 82° (T12–S1), 26° of lumbar lordosis was obtained postoperatively. While the preoperative scoliosis angle was 21° (T8–L4), this reduced to 6°. The clinical and radiological results in the early postoperative period and after a six month follow-up were found to be very satisfactory. There were no complications in the early postoperative follow-up. The patient was able to sit without support.

Discussion: In spinal deformations seen with myelomeningocele, especially for rigid kyphotic deformations, a satisfactory result can be obtained in the early period with scar protective treatment and simultaneous deformation correction, for appropriate cases using recent approach methods. With this treatment, we obtained a better lordosis angle by protecting the neural tissues, a balanced and straight seat in the sagittal plane, and good skin closure in the surgical area.

Key words: Myelomeningocele, Kyphosis, Posterior vertebral osteotomy, skin flap

Level of Evidence: Case report, Level IV

ÖZET

Amaç: Myelomeningosel terimi, yaygın olarak omurga ve spinal kordu etkileyen nöral tüp defektini tanımlamak için kullanılır. Diğer ortopedik problemlerin yanısıra skolyoz, kifoz ve lordoz gibi omurga deformiteleri ile birlikte sıklıkla bulunur (% 62). Myelomeningosel ile birlikte skolyoz % 50-90, lomber bölgede kifoz % 8-20 oranında görülür. Çoğu eğrilik; rijit, sıklıkla doğumda 80° olur ve genellikle ilerleyicidir, yılda 6-12° ilerleme gösterebilir. Deformiteye yönelik cerrahi müdahale yüksek komplikasyon riskleri taşır. Çalışmanın amacı kifoz ile birlikte olan myelomeningosel hastasında nöral yapılara zarar vermeden posterior lomber vertebra rezeksiyon osteotomisi ile birlikte aynı seansta cilt kaydırma işleminin değerlendirilmesidir.

Metod: Rijit lomber kifoz deformitesi ve skolyozun eşlik ettiği posterior vertebral elemanların yokluğu ile birlikte rijit kifoz deformitesi ve cilt problemleri olan myelomeningosel (myelodisplazi veya spina bifida) deformiteli 6 yaşında erkek hastada posterior vertebra rezeksiyon osteotomisi ile birlikte stabilizasyon uygulandı. Torakal-lomber-pelvik pedikül vida fiksasyonu ile stabilizasyon (T9-Sakroiliak), L3 vertebra disk seviyelerinden rezeksiyon osteotomisi ve aynı seansta yeterli yara örtümü için plastik cerrahi ile cilt kaydırma yapıldı. Postop 6 ay korse uygulandı.

Sonuç: Preoperatif lomber kifoz açısı 82° (T12-S1) iken postop 26° lomber lordoz elde edildi. Skolyoz açısı preoperatif 21° (T8-L4) iken postop 6° oldu. Erken postop ve 6 aylık takip sonucunda klinik ve radyolojik sonucu çok iyi bulundu. Cerrahi sırasında gelişen dura yırtığı primer tamir edildi. Erken postop takibinde komplikasyon gelişmedi. Hasta desteksiz rahat oturur hale geldi.

Tartışma: Myelomeningosel ile birlikte görülen omurga deformiteleri ve özellikle rijit kifoz deformitelerinde güncel yaklaşım yöntemleri arasında uygun olgularda eş zamanlı deformite düzeltilmesi ile birlikte yara koruyucu tedavi ile erken dönemde başarılı sonuç elde edildi. Bu tedavi ile erken dönemde nöral dokuları koruyarak daha iyi bir lordoz açısı, sagittal planda dengeli ve dik oturma yanısıra cerrahi bölgede iyi bir cilt örtümü sağlanabilir.

Anahtar kelimeler: Myelomeningosel, kifoz, posterior vertebra osteotomisi, cilt kaydırma.

Kanıt düzeyi: Olgu sunumu, Düzey IV.

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INTRODUCTION:

Myelomeningocele (myelodysplasia or spina bifida) is defined as a neural tube defect affecting the spine and spinal cord. This defect is due to incorrect development of the neighboring mesodermal and ectodermal structures and neuropore during embryogenesis. Myelomeningocele can be classified as open or closed, according to the state of the neural tissues². Myelomeningocele might result in spontaneous abortion of the fetus in the early stages of embryogenesis¹³. Developments in prenatal screening techniques have led to an apparent increase in therapeutic abortion^{13,24}. According to epidemiological studies, although the numbers of spontaneous and therapeutic abortions are not exactly known and are difficult to detect, the prevalence of myelomeningocele shows alterations according to factors such as ethnicity, race, and geography¹³. Over 4,500 pregnancies in European Union countries, and approximately 3,000 pregnancies in the United States of America, are affected by this malformation each year. The prevalence of myelomeningocele was found to be 18.0 in every 100,000 births in a study conducted in the USA in 2005².

An apparent decrease has been seen in the myelomeningocele prevalence in industrialized societies in the last four decades, due to developments in ultrasonographic imaging techniques, diagnostic serum alpha-fetoprotein measurement, elective termination of affected pregnancies, and folic acid supplement¹³. The description of environmental risk factors such as diabetes mellitus, obesity, smoking, gene polymorphism and some drugs, has also contributed to this apparent decrease².

Myelomeningocele is a complex deformation which can cause a number of musculoskeletal problems, such as pes equinovarus, paralytic pes cavus, spinal deformations, breaks, and some ulcers². While these problems were more rarely seen before the 1950s, due to the death of myelomeningocele patients before the age of 2¹⁷, nowadays the expected lifespan and life quality have significantly increased, due to progression of the medical treatment². However, the problems caused by the spinal deformations in a growing child are a significant problem. This study aims to present the clinical and radiological results obtained in the early period with posterior vertebral osteotomy, which we administered from the posterior protecting the neural tissues and the skin, and the inclusion of skin slipping of the surgical field in the same session, administered to a patient with

a rigid kyphosis deformation with myelomeningocele, together with a literature review.

CASE PRESENTATION:

A 6-year-old spina bifida patient was brought to our clinic due to complaints that he could not sit up straight, and a structural abnormality in his back. In the patient history, the skin on the myelomeningocele sac in the lumbar region was primarily closed after birth, and a shunt was administered for hydrocephalus when he was 4.5 months old. When he was examined clinically, there was a rigid lumbar kyphosis deformation and scoliosis in the posterior, a poor skin appearance on the previously-closed myelomeningocele sac, and hair growth like a horse's mane cranial to this region. He was not able to stand still even with support, his sitting balance was disrupted in the sagittal plane towards the front, and the abdomen was collapsed inward.

The muscle strength in the lower extremities was 1/5 on the right and 2/5 on the left. He was using below-knee orthosis for the pes planus deformity in both feet (Figure-1).

There was agenesis in all lumbar and sacral posterior areas radiologically. In an X-ray taken while sitting, 82° of lumbar kyphosis (T12-S1) in the sagittal plane, 24° of thoracic lordosis (T4-12), and 21° of coronal plane scoliosis deformation (T8-L4) were measured (Figure-2).

Whole spinal examinations were performed with Magnetic Resonance Imaging (MRI). By MRI, Chiari malformation was detected, due to the herniation of the cerebellar tonsils at the level of the craniocervical junction to the level of the C2 vertebral corpus plateau, and also due to the tight and elongated view of the fourth ventricle in the posterior fossa.

A syringohydromyelic cavity was observed, starting from the C7 level of the cervical medulla spinalis, throughout the dorsal medulla spinalis up to the T12 level. There was fusion between the T12-L1 vertebral corpuses and posterior elements.

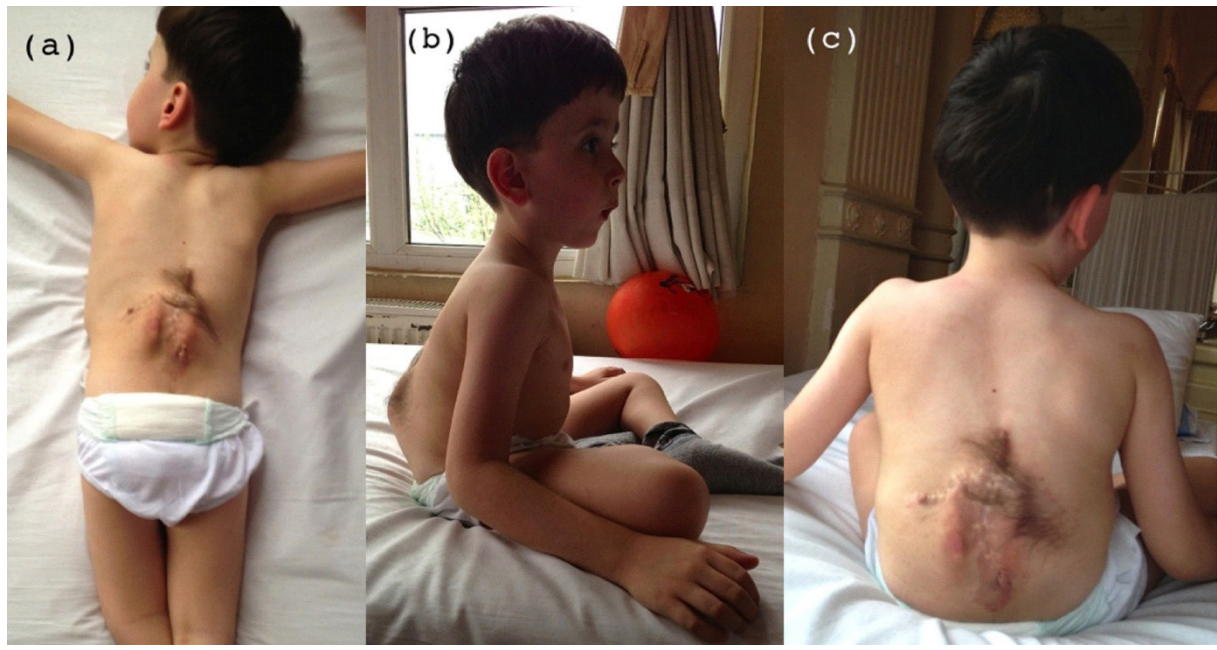


Figure-1 (a-e). Clinical view while lying down and sitting.

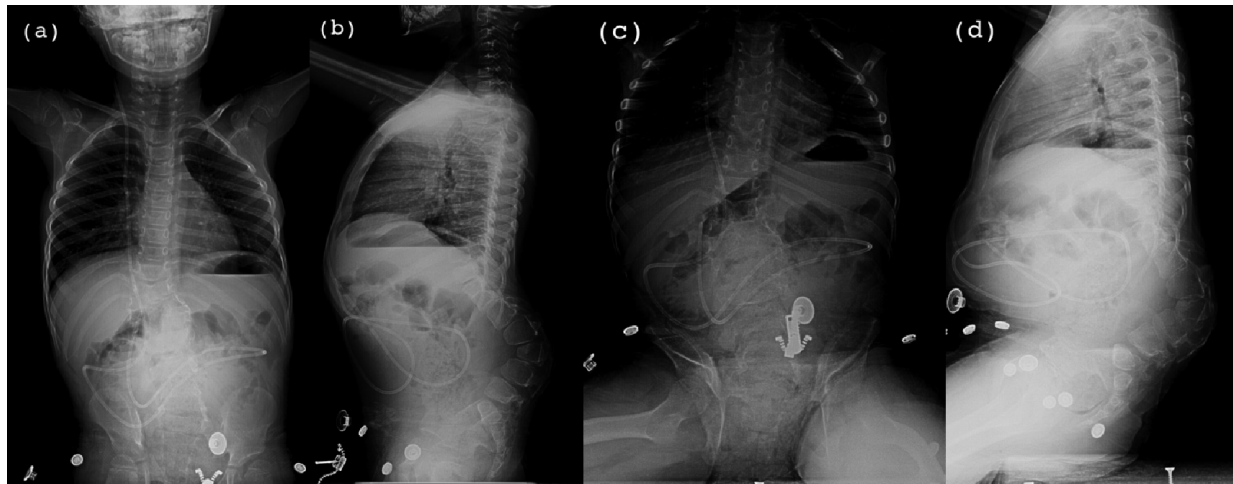


Figure-2 (a-d). X-ray view in AP-LAT plane while lying down and sitting.

Large openings in the posterior vertebral elements throughout the lumbar area, a calibration increase starting from the L1 level of the medulla spinalis in this area, and a 26×10 mm cystic semisolid lesion showing elongation towards the L2 vertebral corpus level were observed.

There were, again, syringohydromyelic alterations caudal to the lesion. The neural tissue was apparently deformed in the lumbar area, and the conus ending could not be detected accurately (Figure-3).

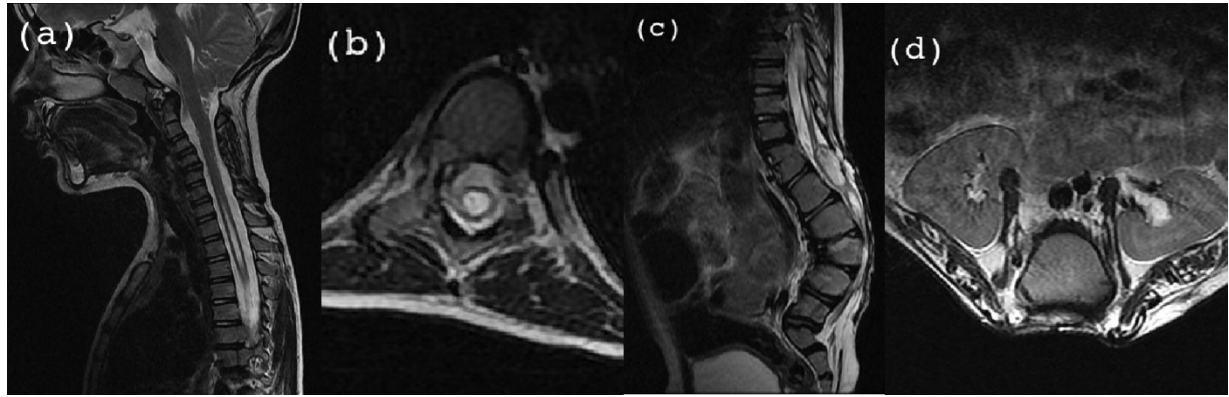


Figure-3 (a-d). Magnetic resonance imaging showing Chiari malformation, cervicothoracic syringomyelia and myelomeningocele.

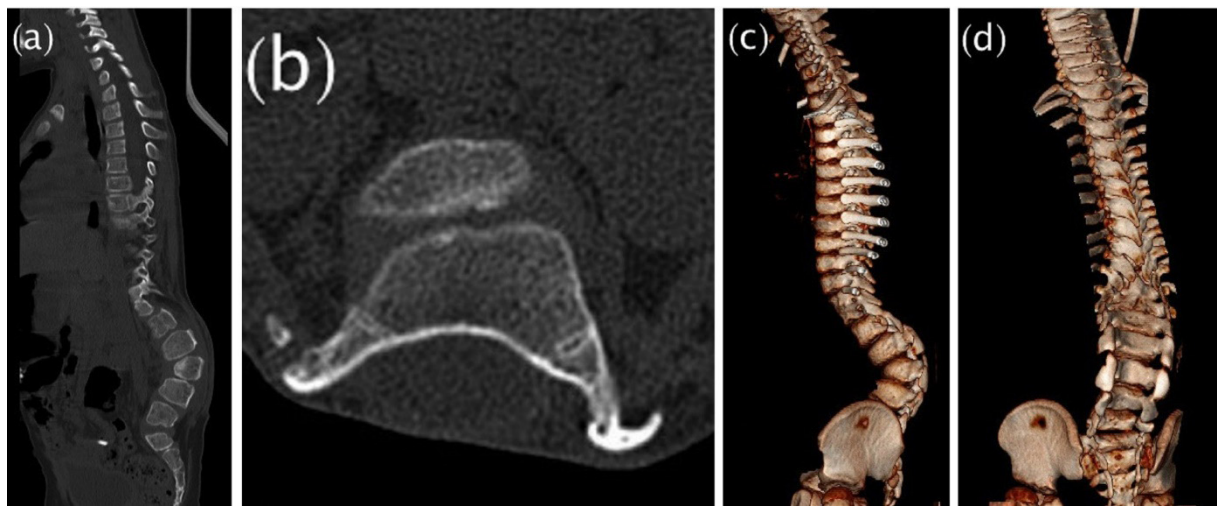


Figure-4 (a-d). Computerized tomography showing kyphosis deformation of the lumbar vertebrae, and posterior lumbosacral vertebral bone defects.

The whole spinal column was analyzed with computerized tomography (CT). Normal and 3D CT were used, especially for determination of the pedicular traces and planning of the osteotomy line (Figure-4).

SURGERY:

To plan the surgery, calculations were first done on the radiological data with Surgimap (Depuy, Synthes), to determine the level and amount of correction to be done in the sagittal plane. It was determined that sufficient correction would be achieved in the sagittal plane after osteotomy of the L3 vertebra (Figure-5). When the patient was in a prone position, without touching the area of agenesis in the posterior vertebral element, the upper skin was opened paravertebally and the area from the T9 vertebra up to the iliac region was

fixed using screw fixation from the lateral part of the structures. With neuromonitorization, without harming the neural structures, osteotomy and vertebrectomy were administered to the L3 vertebra in the kyphotic region. Lumbar lordosis was provided with compression after placing rods to the pedicle screws, giving an appropriate sagittal contour. An autogenous graft, obtained by corpectomy after facet joint osteotomy and decortication, and a 30 cc allograft fusion, were placed into the fusion area (Figure-6). By loosening the skin on the myelomeningocele sac from abdominal lateral vertical incisions prepared by plastic surgery two weeks prior to the operation, the healthy skin tissue was closed without any problems, by peeling the skin in the area of the defect from beneath the dural tissues in the same session (Figure-7).

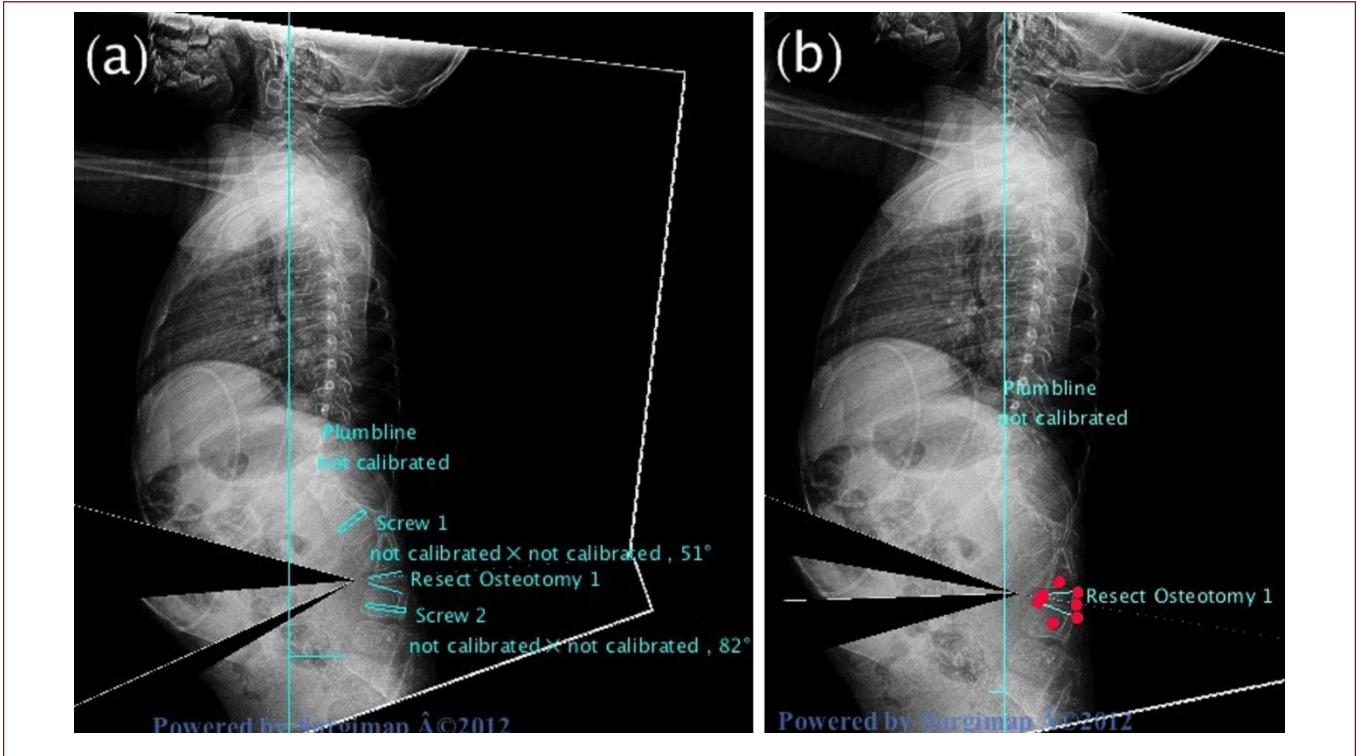


Figure-5 (a-b). Calculation of the deformation correction amount and level with the help of preoperative Surgimap.

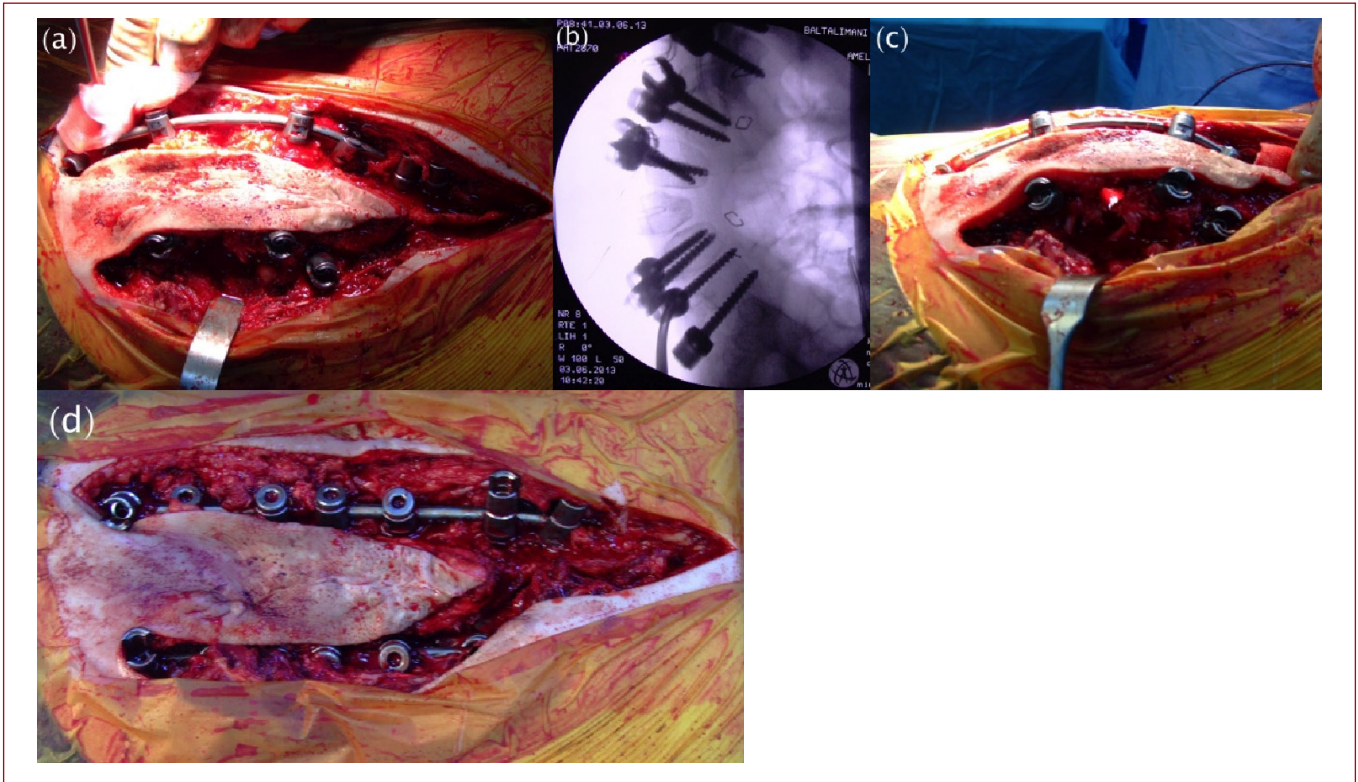


Figure-6 (a). Paravertebral approach protecting the skin in the area with a posterior vertebral defect in the middle plane, **(b).** Pedicle screws in fluoroscopy, **(c-d).** Osteotomy and L3 vertebral resection.



Figure-7 (a-b). To be able to close the skin tissue sufficiently in the operation area during surgery, the skin was prepared with loosening from both sides of the abdomen, days before the operation, by plastic surgery, **(c-d)**. The closing process of the surgical area after vertebrectomy and instrumentation, **(e)**. The view of the surgical area one week after surgery.

One of the skin graft slipping areas, opened longitudinally on both abdominal sites, was sewn primarily after the operation, while the other one was covered with a skin graft taken with a dermatome knife from the front side of the femur, to prevent problems due to scarring. The operation lasted for 5 hours and 45 minutes. During surgery, transfusion of 3 units of

erythrocyte suspension and 1 unit of freshly frozen plasma were performed. The only complication was the formation of cerebrospinal fluid, due to injury while removing the old skin tissue in the area just underneath the dural sac. This was closed with the primarily repaired and slipped skin graft. No other complications developed in the patient, who was laid down for 15 days postoperatively in a prone position. The patient was allowed to sit with a postoperative thoracolumbar brace. He was discharged from hospital with no problems on the twentieth day after surgery. No complications developed in the early follow-up period (six months).

While there was 82° of lumbar kyphosis in the sagittal plane preoperatively, 26° of lumbar lordosis was obtained postoperatively, a correction of 98°. While the angle of scoliosis in the coronal plane was 21° preoperatively, this was 6° postoperatively, a correction of 15°. The clinical and radiological results were found to be quite satisfactory (Figure-8).

DISCUSSION:

Myelomeningocele (myelodysplasia) is found in children with scoliosis, kyphosis and lordosis at a very high rate¹⁰. The prevalence of scoliosis with myelomeningocele has been reported as between 62% and 90%^{19,27,30}. Kyphotic deformation of the lumbar vertebrae is seen in 8–20% of myelomeningocele patients^{1,5,14}. Most of the curvatures consist of quite rigid components, greater than 80° at birth and increasing approximately 12° each year^{1,3,7,11,12,15,16,22,31}.

Raycroft and Trivedi reported the prevalence of spinal deformation in myelomeningocele patients as 62%^{28,37}. The accompanying spinal deformations are generally progressive and affect the prognosis of children with myelomeningocele. Vertebral abnormalities causing skeletal deformations make the clinical picture complex. The characteristic problems are segmentation, formation, or both, in any part of the vertebral ring².

The most apparent and commonly-encountered congenital abnormality is insufficiency of the posterior arch of the lumbosacral vertebrae. These abnormalities affect the treatment of the scoliosis and kyphosis by various aspects. Other congenital malformations include hemivertebrae, butterfly vertebrae, diastematomyelia, and unsegmented bar²⁶. Spinal curvatures are more commonly seen in young myelomeningocele patients compared to other developmental abnormalities²⁶. They can form at 2 or 3 years of age, and may become quite

serious by 7 years of age^{10,15,30}.

Pelvic obliquity and hip deformations affect the spinal balance. They are two important factors causing spinal deformation correction for those patients to be difficult and complex. For instance, asymmetrical hip contractures may lead to lumbar scoliosis, pelvic obliquity and abnormal lordosis in standing and sitting positions. Similarly, spinal correction as part of the treatment of scoliosis may lead to lower extremity positions inhibiting sitting and standing functions. The prevalence of scoliosis in myelomeningocele patients has been shown to be 52–89%. In some studies, this has been found to be 80–90%^{22,27,30}. Trivedi et al. defined the scoliosis Cobb angle in myelomeningocele patients as $>20^\circ$. While most of the curvatures develop in the early period of life for these patients, they develop after 9 years of age in 40%, and it has been reported that sometimes it is not clear until the age of 15³⁷.



Figure-8 (a-f). Early postoperative clinical view, **(g-h).** Early postoperative radiological view.

Kyphotic deformation of the lumbar vertebrae is seen in 8–20% of patients with myelomeningocele^{1,5,14}. Insufficient development of the neural tube in those patients results in deformation of the dorsal spinal column. In addition to harmful effects on the neural tissues in the spinal canal, the extensor muscles are repositioned towards the anterior. In this position, there are no structures allowing the vertebrae to stand upright against the iliopsoas muscle, and so these forces lead to kyphosis in the lumbar vertebrae⁵. In general, deformation increases during childhood for many of the patients, and is expected even after completion of the increase in kyphosis^{12,31–35}. Deformation is very obvious in patients with high neurological lesions^{5,22}. Carstens et al. found kyphosis in the lateral lumbar X-rays of 20% of approximately 700 patients with myelomeningocele⁵. Kyphosis deformation is especially common in patients with paraplegia in the thoracic and upper lumbar levels. Kyphosis deformation can be described as congenital or paralytic. The paralytic type of kyphosis is more common, and is subdivided into two types: kyphosis with collapse, and rigid sharp-angled kyphosis. Most of the curvatures have a number of rigid components, and generally the curvature is greater than 80°, progressing 6–12° each year^{1,3,7,11,12,15,16,22,31}.

McMaster²⁰ treated ten patients with serious kyphosis and myelomeningocele deformation with kyphosis resection, internal fixation and spinal fusion. While the mean kyphosis preoperatively was 131°, this decreased to 44° after surgery. This procedure was described as a major surgical operation with a number of complications. Harrington rods and AO plaques were used for internal fixation. It was stated that long posterior fusion from the middle thoracic area to the sacrum is necessary, to provide long-term stabilization and to prevent thoracic lordosis. It was seen that the patients were able to sit without using their arms for support, and had a flat back with no decubitus ulcers when they reached skeletal maturity²¹.

According to the experiences of Akbar et al., patients with lumbar kyphosis generally show posterior arch deformation, starting from the lower thoracic area up to the sacrum. Protection of the neurological functions is rare below the T10 and T12 levels, and there might be compensatory hyperlordosis in the neighboring vertebrae and sacrolumbar passage area. When left untreated, since the respiratory muscles cannot function effectively due to changes in the sagittal profile, this

may lead to respiratory distress, and the internal organs might be compressed. The increased flexion of the body might also make urinary drainage along the urethra difficult due to compression. As a result, the lower rib cage encounters the anterior of the femur. In addition to the limited functional capacities of the patients, serious postural problems, including insufficiency of the supine position, constrain the patient. Pressure scars may develop, especially at the apex of the kyphosis, which carries a risk of infection and meningitis, in particular, and also has the potential to cause deep infection after surgical correction². Options are limited for conservative treatment. The use of a brace is difficult, rarely effective, and impossible in the later periods¹.

Different surgical techniques, anterior strut-grafting and spinal osteotomy with anterior vertebral body excision using Luque rods have been described to stabilize and correct the lumbar kyphosis^{13,30,38}. Firstly, by Sharrard³², Lindseth and Stelzer¹⁴, vertebral body resection, short stabilization, and postoperative long-term immobilization were described for the treatment of kyphosis. Good results of long instrumentation with Luque rod fixation and distal fixation in the sacrum and pelvis were published by Heydemann and Gillespie in 1987¹⁰. Although fixation with pedicle screws led to a solution, the pedicle structure is small, dysplastic, and in an improper position in these patients. According to Rogers et al., pedicular screw fixation results in the correction and protection of lumbar lordosis. By this method, anterior approaches might be avoided²⁸.

Hook systems, on the other hand, are effective for patients with a relatively normal spinal structure. Sacral and pelvic fixation are generally thought to be necessary for patients with deformity together with fixed pelvic obliquity. However, Wild et al. reported that the pelvic obliquity can spontaneously repair after anterior and posterior spinal fusion³⁹. Cartens et al. published the results of 33 kyphectomies for patients with myelomeningocele⁵. The best results were obtained with congenital rigid-type kyphosis. Apparent postoperative derotation was observed in the flexible paralytic types of kyphosis with short fusion⁵. In the same year, Warner and Fackler published the modified sacral fixation technique³⁸. It was seen in their study that better stabilization was provided for 12 patients who had the distal end of the semi-curved rod passed through the first sacral foramina. Similarly, Akbar et al. stated that they used the Warner and Fackler technique as standard for patients with lumbar kyphosis

due to myelomeningocele^{1,2}.

Providing alignment of the sagittal plane, and obtaining better postural stability and sitting balance, are suggested for patients with serious lumbar kyphosis deformation with myelomeningocele. 11 patients with myelomeningocele and kyphosis received vertebrectomy and posterior instrumentation with a lumbar sacropelvic subtraction (decancellation) kyphectomy technique by Nolden et al. Patients, who were followed up for a minimum of two years, received subtraction (decancellation) vertebrectomy with limited arthrodesis, and posterior transpedicular lumbosacral instrumentation by protecting the thecal sac. While the mean preoperative kyphosis was 88°, immediately after surgery there was 3° of lordosis, giving a mean correction of 91°, on average, in the sagittal plane. In the final follow-up, 66° were found, and the mean kyphosis that was lost after two years of follow-up was shown to be 24°. There were no vascular or deep scar infections. The morbidity was reported to be lower for subtraction (decancellation) and stabilization compared to excision, and this was shown to provide sagittal balance. This has been suggested for the treatment of patients with myelomeningocele and accompanying kyphosis deformations²⁵.

Akbar et al. performed lumbar kyphectomy with the Warner and Facker technique for 28 pediatric patients, and performed an analysis retrospectively. The mean operation time was 3.7 (1.5–7.6) hours and the mean blood loss was 1160 (300–3000) ml. The lumbar kyphosis was corrected from 131° (90–170°) to 45° (15–90°) after surgery. The mean correction was 86° (40–130°) and an average of 2.5 (1–4) vertebrae were resected. In the final follow-up, the mean kyphosis was 45° (15–100°), and the spinal correction loss was 5° (8–55°). Spinal growth was observed in the thoracic area that received Luque wire instrumentation, and crankshaft phenomenon were encountered in one patient, to whom thoracic stabilization was administered. The functional results were found to be satisfactory. All of the patients were able to sit without hand support or a brace. There was implant deficiency in nine patients as a long-term complication, and five patients required revision surgery. After correction, for four of those patients, the rods penetrated into the sacrum, and the rods broke for five patients. Implant deficiency was not found to be correlated with age or the starting level of kyphosis. However, a relationship was shown between implant deficiency and the remaining deformation after surgery. The loading forces on the

connecting parts of the rods depend on the residual distance between the apex of the kyphosis and the plumb line, and both of them were found to be significantly related to implant deficiency¹. No correlations were found between implant deficiency and the predictive weight of the body, the length of the rods, or the lower or upper parts of the apex. Additionally, no correlation was seen for the length of the spine, representing the body length. On the basis of these results, the aim should be to establish the sagittal profile as much as possible, to decrease the risk of implant deficiency after correction^{1,2}.

Necrosis due to the incision and scar infection are seen as complications for more than half of patients. Ward et al. stated that necrosis due to the incision does not cause a long-term problem Ward et al³⁸. When the infection rates were taken together with repetitive urinary tract infection, this was seen to be 43% or more^{8,37}. The Foley catheter should be removed as soon as possible when the patient is stable. Intravenous antibiotics should be continued until the patient is discharged from hospital. The neurological deficit rate is low^{19,37}. Cerebrospinal fluid leakage might occur as a result of surgical dissection or tethering of the spinal cord. Progression of the curvature might occur above or below the fusion level, if the fusion level is chosen at an inappropriately short level. The surgical approach for more than 76% of pseudoarthrosis patients depends on the type of instrumentation, or the posterior approach alone^{4,34}. The pseudoarthrosis rate has been reported as 0–50% with isolated anterior arthrodesis, 26–76% with isolated posterior arthrodesis, and 5–23% with combined anterior and posterior arthrodesis. Secondary pseudoarthrosis due to implant deficiency was shown to occur in 65% of cases^{18,23,37}. Extremity breaks secondary to osteoporosis resulting from immobilization are common. Shunt malfunction might occur after the acute correction of large curvatures³⁵.

However, in our case, no postoperative scar problems, infection, or deterioration in the neurological deficit or instrumentation were encountered in the early period.

Dura leakage formed during sliding of the skin graft after the osteotomy. Correction was completed during surgery to primarily repair this, and no additional problems developed. In the early postoperative period, it could be seen that the correction of the deformation in the sagittal and coronal planes was protected. Removing the apical kyphotic vertebra with osteotomy from the

posterior alone, to obtain sufficient lumbar lordosis and sagittal balance, with pedicular screw fixation after careful preoperative planning, provided effective correction. It was seen that closing the area of surgery with plastic surgery was crucial, to prevent commonly encountered scarring problems and to increase the success of the surgery.

CONCLUSION:

Important developments have been obtained in the treatment of children with congenital spinal deformations in the last three decades, resulting in increased patient lifespan². Thus, orthopedic problems in the hips and lower extremities, and decubitus ulcers accompanying the complex spinal deformations that accompany myelomeningocele, are commonly encountered. Surgery for the progressive and rigid lumbar kyphosis is also still considered a problem, due to the high complication rates. Osteotomy for kyphosis correction with a posterior approach might be included with the fusion process and treatments, to protect from scarring. However, due to our short follow-up period and application to only a single case, there is a need for long-term results for a greater number of patients, in order to accurately evaluate the administered technique. Thus, surgical treatments for the spinal problems of patients with myelomeningocele should be well planned, and should be evaluated using a multidisciplinary approach with orthopedics, neurosurgery and plastic surgery.

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