



Research

Determining Hydrocephalus and V-P Shunt Requirements After Repair of Myelomeningocele Defects in Infants

İnfanlarda Miyelomeningosel Defektlerinin Onarımı Sonrası Hidrosefali ve V-P Şant Gereksinimlerinin Belirlenmesi

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ABSTRACT

Objective: Myelomeningocele (MM) is an important developmental defect that requires surgical treatment, and hydrocephalus is an important complication that may develop after surgical closure of the defect. To analyze the factors that may determine the need for shunting in patients with MM defects and hydrocephalus.

Methods: A retrospective analysis of 103 patients (63 females and 40 males) who were treated for MM between 2016 and 2023 at our institution was conducted. The infants were divided into two groups: Group 1; those who underwent V-P shunt surgery following MM repair surgeries (n=81) and Group 2; those who did not receive V-P shunt surgery following MM repair (n=22). Parameters such as head circumference, MM sac integrity, MM sac location, and birth weight were examined. The results were analyzed to identify any potential differences between the two groups.

Results: The study included 103 patients with MM abnormalities. The rate of V-P shunt insertion was significantly higher in infants with preoperative hydrocephalus, those with an open MM sac structure, and those with abnormalities in the thoracic/thoracolumbar region.

Conclusion: Development of hydrocephalus and the need for V-P shunt placement after defect repair are crucial in infants born with MM. This evaluation helps in planning the management of these patients with the aim of minimizing complications and improving the overall prognosis.

Keywords: Myelomeningocele, hydrocephalus, frontal and occipital horn ratio

ÖZ

Amaç: Miyelomeningosel (MM) cerrahi tedavi gerektiren önemli bir gelişimsel defekt olup, defektin cerrahi olarak kapatılmasından sonra gelişebilecek komplikasyonlar arasında hidrosefali önemli bir yer tutmaktadır. Hidrosefali ile birlikte MM defekti olan hastalarda postoperatif dönemde şant ihtiyacını belirleyebilecek faktörlerin analiz edilmesidir.

Gereç ve Yöntem: Kliniğimizde 2016-2023 yılları arasında MM nedeniyle tedavi gören 103 hastanın (63 kadın, 40 erkek) retrospektif analizi yapıldı. Bebekler iki gruba ayrıldı: Grup 1; MM onarımı ameliyatı sonrası V-P şant ameliyatı geçirenler (n=81) ve Grup 2; MM onarımı sonrası V-P şant ameliyatı geçirmeyenler (n=22). Baş çevresi, MM kesesi bütünlüğü, MM kesesinin yeri, doğum ağırlığı gibi parametreler incelendi. Sonuçlar, iki grup arasındaki potansiyel farklılıkları belirlemek için analiz edildi.

Bulgular: Bu çalışmaya MM anormallikleri olan 103 hastayı dahil ettik. Preoperatif hidrosefalisi olan, MM kesesi yapısı açık olan ve torasik/torakolomber bölgede anormalliği olan bebeklerde V-P şant takılma oranı anlamlı olarak daha yüksekti.

Sonuç: MM ile doğan bebeklerde hidrosefali gelişimi ve defektin onarımı sonrası V-P şant yerleştirilmesi ihtiyacı önemlidir. Bu değerlendirme, komplikasyonları en aza indirmeyi ve genel prognozu iyileştirmeyi hedefleyerek bu hastaların yönetiminin planlanmasına yardımcı olur.

Anahtar Kelimeler: Miyelomeningosel, hidrosefali, frontal ve oksipital boynuz oranı

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INTRODUCTION

Myelomeningocele (MM) is the most common and severe form of open neural tube defects (ONTDs) and may cause significant morbidity and mortality due to hydrocephalus, lower extremity paralysis, and neurogenic bowel and bladder dysfunction. These conditions require multidisciplinary management, such as neurosurgical, orthopedic, and nephrourological treatments (1).

One of the most important hypotheses regarding the formation of neurological deficits in infants with ONTD is the "double hit hypothesis" proposed by Heffez et al. (1), which suggests a two-stage effect. A neural tube defect initially occurs during neurulation, followed by further damage due to continuous exposure to amniotic fluid. Additionally, the skin defect leaves neural tissue vulnerable to mechanical trauma from the uterine wall and the neurotoxic effects of amniotic fluid, resulting in additional neurological injury. The interruption of simultaneous normal neural tube development can lead to lifelong disabilities, including paralysis, incontinence, and cognitive impairments (1,2).

The incidence of hydrocephalus in patients with ONTD varies from 60% to 90%, and 80% of those who develop hydrocephalus require surgical diversion of Cerebrospinal Fluid (CSF) (3). Ultrasonography (USG) and magnetic resonance imaging (MRI) are widely used for the anatomical assessment of intracranial structures, especially ventricular sizes, in the follow-up and treatment of hydrocephalus. There are many techniques to assess hydrocephalus; however, the frontal occipital horn ratio (FOHR) has been shown to be a reliable index for clinical diagnosis and follow-up of infantile ventriculomegaly (4).

The aim of our study was to identify risk factors for determining the need for CSF diversion surgery in the postoperative period through preoperative evaluation of patients with MM.

METHODS

A retrospective analysis of 103 patients (63 females and 40 males) who were treated for MM and MS between 2013 and 2023 at our institution was conducted. The goal of this study was to identify factors influencing the decision to perform CSF diversion surgeries, which are ventriculo-peritoneal shunt (V-PS) surgeries, at our clinic. The infants were divided into two groups: Group 1; those who underwent V-P shunt surgery following MM repair surgeries (n=81) and Group 2; those who did not receive V-P shunt surgery following MM repair (n=22). Parameters such as head circumference (HC), MM sac integrity, MM sac location, and birth weight

were examined. Additionally, preoperative ventricular volumes were measured using transfontanel USGs (TFUSG) and cranial MRIs to calculate the FOHR. The results were analyzed to identify any potential differences between the two groups.

Human subjects: All participants in this study provided consent or waived consent. This study University of Health Sciences Türkiye, Ümraniye Training and Research Hospital Clinical Research Ethics Committee (decision no: 557, date: 21.12.2023).

Animal subjects: All authors have confirmed that this study did not involve animal subjects or tissue.

Imaging Technique

For USG, a radiology specialist used a 5-8 MHz curved array EPIC system probe (Philips Healthcare) through the open anterior fontanel. This method was used to measure the sizes of the frontal, occipital, and temporal horns, as well as the biparietal diameters, across the coronal, sagittal, and oblique planes.

MRI was performed using a 1.5- Tesla system (Magnetom Avanto, Siemens Healthcare). The routine MRI protocol included spin-echo T2-weighted images (slice thickness, 2 mm; interslice gap, 2.2 mm), 3D T1-weighted magnetization-prepared rapid gradient-echo imaging (MP-RAGE, Siemens Healthcare), axial 2D T2-weighted fluid-attenuated inversion recovery (FLAIR) imaging, axial susceptibility-weighted imaging, and axial echo-planar DWI. The fast protocol comprised axial, coronal, and sagittal plane T2-weighted imaging with a slice thickness of 4 mm and an interslice gap of 5 mm, axial echo-planar FLAIR imaging, axial echo-planar DWI, and axial T2* and sagittal T1* volumetric interpolated breath-hold examination (VIBE) imaging techniques. In particular, ventricular volumes were measured using coronal and axial T2-weighted images.

Evaluation of Images

The USG were reviewed by two neurosurgeons and one pediatric neuroradiologist, each with a minimum of 10 years of experience in their field. Coronal USG images were examined to determine the bifrontal horn size, maximum bitemporal horn size, maximum bioccipital horn size, and maximum biparietal calvarial size. The same team analyzed the MRI ventricular volumes using analyze software (version 12.0, Analyze Direct). In MRI, T2-weighted image signal intensities were normalized within a range of 0 to 1 and segmented using a region-growing technique, as described in previously published studies (5,6). For the FOHR, measurements were made of the bifrontal horn diameter at approximately the largest frontal horn size at the level

of the foramen of Monro, bilateral occipital horn diameter at the largest occipital horn size at the level of the atria of the lateral ventricles, and the biparietal size at the largest

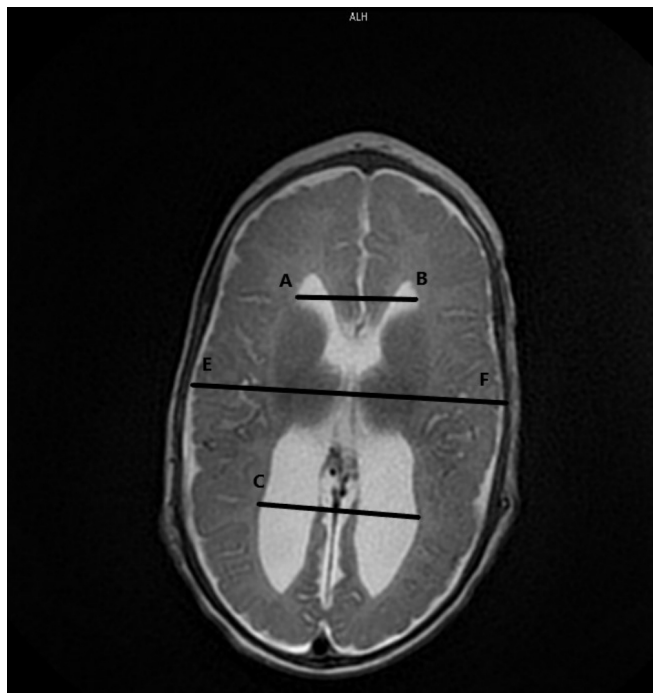


Figure 1. Axial MRI image showing linear measurements used to calculate the frontal occipital horn ratio: Bifrontal horn dimension (arrow AB), bioccipital horn dimension (arrow CD), and biparietal dimension (arrow EF)

MRI: Magnetic resonance imaging

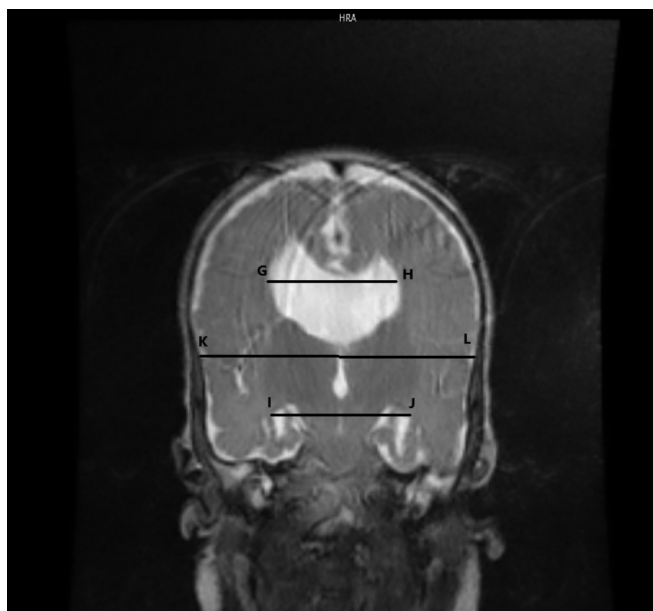


Figure 2. Coronal MRI image showing linear measurements used to calculate the frontal temporal horn ratio: Bifrontal horn dimension (arrow GH), bitemporal horn dimension (arrow IJ), and biparietal dimension (arrow KL)

MRI: Magnetic resonance imaging

transverse calvarial dimension; all measurements were conducted on axial (Figure 1) and coronal (Figure 2) images.

The FOHR was calculated as follows: (bifrontal horn size+bioccipital horn size) / (2×biparietal calvarial size).

Statistical Analysis

The assumption of normality for continuous variables was tested using the Kolmogorov-Smirnov test. Categorical variables are presented as frequencies (n, %), whereas continuous variables are presented as mean and standard deviation. Comparisons between two groups for continuous variables were conducted using the Independent sample t-test. The relationship between two continuous variables was examined using the Pearson correlation test. For qualitative comparisons between groups, chi-square tests (Pearson chi-square test and Fisher's exact test) were used. Results were considered statistically significant at a confidence interval of 95% if $p < 0.05$. All statistical analyses were performed using SPSS software version 26 (IBM Corp., Armonk, NY, USA).

RESULTS

We included 103 patients with MM abnormalities. The average weight of infants by the time of MM/MS surgery was 3163.60 ± 624.99 grams. According to routinely performed preoperative TFUSGs and MRIs on newborns with MM, 81 were diagnosed with hydrocephalus and 22 were found to be normal. During the postoperative follow-up of 81 infants with hydrocephalus, 74 required V-P shunt placement and 7 did not. Conversely, among the 22 infants without evident hydrocephalus, 9 required a V-P shunt insertion in the postoperative period.

When considering the location of the MM; 22 infants had the lumbar region, 10 had the sacral region, 8 had the thoracic region, 23 had the lumbosacral level, and 40 had the thoracolumbar level. Other characteristics of the infants were as follows: average HC was 36.56 ± 4.95 cm, lesion diameter was 5.68 ± 1.90 cm, and the FOHR was 0.52 ± 0.13 . We determined that 77 infants had an open MM sac structure and 26 had an unruptured sac structure. Additionally, 67 infants were found to have complete paralysis, whereas 28 had partial neurological deficits (Table 1).

The lesion diameter ($t=2.606$; $p=0.011$) and the FOHR ($t=10.007$; $p<0.001$) were significantly higher in 81 infants undergoing V-P shunt procedure. There was no statistically significant difference in the weight of the infants based on whether or not they required a V-P shunt surgery ($p>0.05$). The rate of V-P shunt insertion was significantly higher in infants with preoperative hydrocephalus (91% vs. 37%;

$p < 0.001$), those with an open MM sac structure (95% vs. 35%; $p < 0.001$), and those with abnormalities in the thoracic/thoracolumbar region (92% vs. 71%; $p = 0.009$). Among the infants who underwent V-P shunt surgery, 75% showed complete motor and sensory loss, whereas 21% had partial deficits ($p < 0.001$) (Table 2).

DISCUSSION

MM is known as the most common neural tube defect (NTD); however, a study by Schindelmann et al. (7) showed that myeloschisis (MS) is not as rare as it is believed, but rather a common NTD. Contrary to the literature, their results showed that MS (31%) occurred more frequently than MM (23%) (7). Therefore, the distribution and classification of NTDs differ significantly between studies. The reported incidence of MS is often lower than the actual incidence, likely due to the

need for a more detailed neuropathological examination to ascertain the presence of a neural placode (7). Although the presence of a membrane (MM sac) and cyst surrounding the defect is generally considered a distinguishing feature between MM and MS, the clinical presentation of these two forms of ONTDs remains the same (8). Our findings indicate that the presence of a MM sac covering the lesion or evidence of CSF leakage can be determinant factors for postoperative hydrocephalus development.

Numerous studies have acknowledged that the absence of a MM sac significantly increases the risk of developing postoperative hydrocephalus requiring a V-P shunt (9). In our study, the need for a V-P shunt placement was found in 34% of cases where the sac was notably intact, compared with 94% in cases where it was open, which was statistically significant.

Another important issue in infants with MM is the diagnosis of hydrocephalus and the timing of treatment. In historical reports published until 1990, many opted for V-P shunt implantation at birth even if the HC was within normal limits (10). Consequently, numerous studies reported high rates of V-P shunt placement, such as Januschek et al. (11) reported an 85% V-P shunt rate, Laskay et al. (12) 84.6%, and Marreiros et al. (13) 70%. However, by tolerating larger ventricles and applying better postoperative wound care, few experienced centers have managed to reduce the V-P shunt rates to 55-65% (14).

Although V-P shunt surgeries are the most effective and widely used treatment method for hydrocephalus, alternative treatments have also come to the fore as a result of recent technological advances. Warf (15) summarized in their study that techniques such as endoscopic third ventriculostomy and choroid plexus cauterization can be an alternative for hydrocephalus in non-developing countries that cannot support shunt equipments.

The primary concern in the management of infants with detected preoperative ventriculomegaly is the progression, regression, or stability of ventriculomegaly. The long-term neurocognitive impact of allowing larger ventricles is unknown, but it appears to be insignificant in short-term evaluations. On the other hand, these patients may be spared the morbidity associated with repeated operations and complications due to V-P shunt insertion (14).

To monitor the hydrocephalus and evaluate the timing of V-P shunt placement, knowing the volume of the ventricles (VV) is crucial. Data obtained from routine neonatal TFUSGs for identifying intracranial pathologies or from MRIs for a more detailed understanding can be used to create VV indices. In their study, Radhakrishnan et al. (4) found that the FOHR,

Table 1. Characteristics of babies with myelomeningocele

Variables (n=100)	n (%)
Baby weight (gr), mean±SD	3163.60±624.99
Hydrocephalus	
Yes	81
No	22
Myelomeningocele pouch structure	
Open	77
Closed	26
Ventriculoperitoneal (VP) shunt requirement	
Yes	81
No	22
Lesion diameter (cm), mean±SD	5.68±1.90
Baby head circumference (HC) (cm), mean±SD	36.56±4.95
Fronto-occipital horn ratio (FOHR), mean±SD	0.52±0.13
Lesion location	
Lumbar region	22
Sacrum region	10
Thoracic area	8
Lumbosacral region	23
Thoracolumbar area	40
Deficit existence	
Yes	67
Partial	25
No	8
Transfontanel ultrasonography (TFUSG)	
Yes	94
No	9

SD: Standard deviation

Table 2. Ventriculoperitoneal (V-P) shunt-related variables in infants with myelomeningocele

Variables	Total, n	V-P Shunt requirement		p-value
		Yes (n=81) n (%)	No (n=22) n (%)	
Baby waight (gr), mean±SD	100	3181.51±638.35	3088.16±575.18	0.561 ^a
Hydrocephalus[#]				< 0.001 ^{c*}
Yes	81	74 (91.4)	7 (8.6)	
No	22	9 (36.8)	13 (63.2)	
Myelomeningocele pouch structure[#]				< 0.001 ^{c*}
Open	77	73 (94.8)	4 (5.2)	
Closed	26	9 (34.8)	17 (65.2)	
Lesion diameter (cm), mean±SD		5.93±1.88	4.67±1.68	0.011^{a*}
Baby head circumference (cm), mean±SD		37.01±5.06	34.56±3.98	0.057
FOHR, mean±SD		0.55±0.12	0.37±0.05	< 0.001 ^{a*}
Lesion localion[#]				0.009^{b*}
Sacrum/lumbar/lumbosacral region	52	37 (71.2)	15 (28.8)	
Thoracic / tracolubar area	51	46 (91.7)	5 (8.3)	
Deficit existence^{**}				< 0.001 ^{c*}
Yes	67	61 (75.3)	6 (31.6)	
Partial	25	17 (21)	8 (42.1)	
No	11	4 (3.7)	7 (26.3)	

*p<0.05, **Column percentage given, ^aIndependent sample t-test, ^bPearson chi-square test, ^cFisher's exact test, [#]Line percentage was given, SD: Standard deviation

a VV index derived from TFUSG and MRI, showed a strong correlation between both imaging methods, and a clinical FOHR threshold of 0.55 demonstrated high sensitivity in identifying infantile hydrocephalus (4). Particularly, FOHR can be used to measure the severity of ventriculomegaly (16,17). We obtained similar results from preoperative TFUSG and MRI examinations concerning FOHR results. A close relationship was found between values exceeding 0.55 and the likelihood of requiring a V-P shunt insertion.

Another significant factor in determining the need for hydrocephalus treatment and V-P shunt placement is the routine measurement of HC. The study conducted by Vonzun et al. (18) showed that preoperative and/or postoperative HC and ventricular measurements were determinants of the need for a V-P shunt in the first year of life. They found that a HC above the 95th percentile predicted an 80% likelihood of needing a V-P shunt due to late hydrocephalus (18). We found this rate to be approximately 81% in infants with a HC of 37 cm or more, which is similar to the findings of Protzenko et al. (19), who showed that a birth HC of 38 cm or more was a significant factor for V-P shunt requirement.

We observed that regardless of the morphology of the defect, there is a greater need for V-P shunt placement in patients with lesions located in the thoracic level, larger than 5 cm in diameter, and those with more severe deficits in our

study. Some studies have indicated that the development of hydrocephalus is generally not related to the anatomical level or size of the lesion but rather to MS (1-20). Another study showed that hydrocephalus and the need for a V-P shunt placement were more common in higher spinal-level lesions (19). We believe that a possible reason for these differing results might be not fully understanding the distinctions between MM and MS. Additionally, similar to our findings, no significant impact of birth weight and gender has been identified on the need for a V-P shunt (18).

Considering all results, it was observed that knowing parameters such as the size and level of the lesion, the integrity of the sac, HC, and FOHR can play a role in determining the risk and the need for a V-P shunt due to hydrocephalus that may develop after MM repair.

Study Limitations

This study is limited by its single-center nature and relatively small number of patients. The findings could be more definitive when considered by larger groups and various centers, leading to the development of guidelines or scales.

CONCLUSION

In infants born with MM, determining the development of hydrocephalus and the need for V-P shunt placement

after defect repair is crucial. The shunt procedure, while life saving, is associated with numerous complications. Therefore, evaluating the lesion and ventricular condition in the preoperative period is essential to understand which cases carry a higher risk and to make certain predictions regarding the outcome. This evaluation helps in planning the management of these patients with the aim of minimizing complications and improving the overall prognosis.

ETHICS

Ethics Committee Approval: This study University of Health Sciences Türkiye, Ümraniye Training and Research Hospital Clinical Research Ethics Committee (decision no: 557, date: 21.12.2023).

Informed Consent: Animal subjects: All authors have confirmed that this study did not involve animal subjects or tissue.

FOOTNOTES

Authorship Contributions

Surgical and Medical Practices: L.Ş., S.I., M.U.E., M.S.S., Design: L.Ş., M.U.E., Data Collection or Processing: L.Ş., G.G.Ö., Analysis or Interpretation: L.Ş., S.I., Literature Search: L.Ş., S.I., G.G.Ö., Writing: L.Ş., S.I., M.U.E.

Conflict of Interest: No conflict of interest was declared by the authors.

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