

ORIGINAL ARTICLE

Comparison of Synthetic Dura and Autologous Dura in Terms of Complication Development in Children Aged 0-1 Years Who Underwent Surgery for Meningocele and Myelomeningocele

Meningosel ve Miyelomeningosel Nedeni ile Opere Edilen 0-1 Yaş Çocuklarda Sentetik Dura ile Otolog Dura Kullanımının Komplikasyon Gelişimi Açısından Karşılaştırılması

¹Emir Kaan İzci , ²Fatih Keskin , ²Densel Araç 

¹Konya City Hospital, Department of Neurosurgery, Konya/TURKIYE
²Meram Faculty of Medicine Hospital, Department of Neurosurgery, Konya/TURKIYE

Correspondence

Emir Kaan İzci, Konya City Hospital, Department of Brain and Nerve Surgery, Konya/TURKIYE

E-Mail: ekaanizci@hotmail.com

How to cite ?

İzci E. , Araç D. , Keskin F. Comparison of Synthetic Dura and Autologous Dura in Terms of Complication Development in Children Aged 0-1 Years Who Underwent Surgery for Meningocele and Myelomeningocele. Genel Tıp Dergisi. 2023; 33(4): 412-420.

ABSTRACT

Aim: The aim of this study is to compare the usage of synthetic and autologous dura mater in terms of complication risk in 0-1-year-old children who were operated for meningocele and myelomeningocele.

Materials and Methods: This cross-sectional observational study was conducted with 44 children aged 0-1 years who were operated for meningocele and myelomeningocele in a university hospital neurosurgery clinic between November 2010 and December 2016. Patient data were extracted retrospectively from hospital records. The demographics and gestational and clinical features of the mothers and babies, and the need for secondary surgery and the presence of postoperative infection, necrosis, wound dehiscence, and/or neurological deficit were compared between the cases in whose surgery synthetic dura mater was used and the cases in whose surgery autologous dura was used.

Findings: In total, 86.4% of the 44 infants were preterm, and the predominant neurological problem was plegia in the vast majority. While the defect was located in the lumbar region in more than half of them, myelomeningocele was detected in 77.3% of all cases. The median defect size detected in the patients was 20.0 cm². Primary closure was performed in 30 patients, limberg flap procedure in 14 patients, however, autologous dura mater and synthetic dura mater were used equally in the patients. The defect size was larger in patients using synthetic dura, furthermore, hydrocephalus was found more frequently in these patients. While primary closure was applied in all patients using autologous dura and in one third of the patients using synthetic dura. Limberg flap procedure was applied in two thirds of synthetic dura group. The need for secondary surgery developed more frequently in synthetic dura group, and all postoperative complications were observed more frequently in these patients. In addition, the need for secondary surgery and postoperative necrosis, wound dehiscence, and neurologic deficit were more frequent in patients who underwent Limber flap compared to primary closure. However, the need for secondary surgery and the risk of postoperative complications were similar between primary closure and Limberg flap procedures in synthetic dura group.

Conclusion: Although the synthetic dura mater was used in more severe patients, it had a higher need for secondary surgery and a higher risk of complications compared to autologous dura. In patients using synthetic dura, on the other hand, primary closure and Limberg flap had similar efficacy and safety.

Keywords: Spinal Dysraphia, Myelomeningocele, Neurosurgery, Dura Mater

ÖZ

Amaç: Bu çalışmanın amacı meningosel ve miyelomeningosel nedeni ile opere edilen 0-1 yaş çocuklarda sentetik dura ile otolog dura kullanımının komplikasyon gelişimi açısından karşılaştırılmasıdır.

Gereç ve yöntemler: Kesitsel tipte gözlemsel araştırma tasarımına sahip bu araştırma, bir üniversite hastanesi beyin ve sinir cerrahisi kliniğinde Kasım 2010 ve Aralık 2016 meningosel ve miyelomeningosel nedeni ile opere edilen 0-1 yaş arası 44 çocuk ile yürütülmüştür. Hasta verileri hastane kayıtlarından retrospektif olarak taranmıştır. Anne ve bebeklerin demografik özellikleri, gestasyonel ve klinik özellikleri, ameliyat sonrası sekonder cerrahi ihtiyacı ve postoperatif enfeksiyon, nekroz, yara açılması ve nörolojik defisit gelişme durumları, sentetik dura ve otolog dura kullanılan hastalar arasında karşılaştırılmıştır.

Bulgular: Toplamda, 4 bebeğin %86,4'ü preterm iken, çoğunda baskın nörolojik durum plejidir. Yarısından fazlasında defekt lomber bölgede izole iken, %77,3'ünde miyelomeningosel saptanmıştır. Hastalarda saptanan defekt boyutu medyan değeri 20,0 cm² iken, 30 hastada primer kapama, 14'ünde Limberg flebi prosedürü uygulanmış, hastaların yarısında otolog diğer yarısında ise sentetik dura kullanılmıştır. Sentetik dura kullanılan hastalarda defekt boyutu daha fazla iken, bu hastalarda daha fazla sıklıkta hidrosefali saptanmıştır. Otolog dura kullanılan hastaların hepsinde primer kapama uygulanmışken, sentetik dura kullanılan hastaların üçte birinde primer kapama, üçte ikisinde ise Limberg flebi prosedürü uygulanmıştır. Sentetik dura kullanılan hastalarda daha fazla sıklıkta sekonder cerrahi ihtiyacı gelişmişken, bu hastalarda tüm postoperatif komplikasyonlar daha fazla sıklıkta gözlenmiştir. Ayrıca, primer kapatmaya göre, Limber flebi uygulanan hastalarda da sekonder cerrahi ihtiyacı ve postoperatif nekroz, yara açılması ve nörolojik defisit daha fazla sıklıktadır. Ancak sentetik dura kullanılan hastalarda primer kapatma ve Limberg flebi prosedürleri arasında sekonder cerrahi ihtiyacı ve postoperatif komplikasyon riski benzer bulunmuştur.

Sonuç: Sentetik dura kliniği daha ağır hastalarda kullanılmakla beraber, otolog dura ile karşılaştırıldığında daha fazla sekonder cerrahi ihtiyacı ve daha fazla komplikasyon riskine sahiptir. Sentetik dura kullanılan hastalarda ise primer kapama ve Limberg flebi benzer etkinlik ve güvenliğe sahiptir.

Anahtar kelimeler: Spinal Disrafi, Miyelomeningosel, Sinir Cerrahisi, Dura Mater.

Introduction

Spina bifida is a congenital malformation that occurs International Clearinghouse Birth Defects Center in as a result of failure in the embryonic closure of the which 2012 data were processed, the prevalence of neural tube (1). According to the 2014 report of the spina bifida varies between 0.24 and 8.67 per 10,000

births in the world (2). This rate varies according to race, geographical area and regions (1, 2). Although there are four types as occulta, closed spinal dysraphism, meningocele and myelomeningocele, myelomeningocele cases, the most common and most severe form, are characterized by different neurological symptoms depending on the severity of the lesion and the location of the sac (1). Myelomeningocele is clinically associated with orthopedic problems such as urinary and fecal incontinence, Chiari II malformation that often requires shunting, hydrocephalus and talipes, contractures, hip dislocation, scoliosis, and kyphosis, as well as motor and sensory neurologic deficits under the lesion (1, 3).

Although it varies according to the clinical characteristics of the patient and the size of the defect, there are prenatal and postnatal options for surgical treatment. Although prenatal surgery reduces the need for shunt after birth and provides better motor function and mental development results compared to postnatal surgery, it also brings risks such as oligohydramnios, preterm labor and uterine separation (4-6). In postnatal surgery, a multidisciplinary approach has gained value according to the patient's clinic in recent years, and neurological management as well as orthopedic, urological and gastrointestinal treatments are also applied according to the needs of the patients (7). Postnatal spinal surgery modalities include repair of the neural placode with the help of autologous dura or closure by synthetic dura mater, shunt revision for hydrocephalus, endoscopic third ventriculostomy and/or choroid plexus coagulation, Chiari II Malformation, and modalities for tethered cord syndrome (3). Synthetic dura mater is used in cases where the defect size is large and autologous repair of the dura mater is not possible (8, 9). However, while the defect can be closed with primary closure in three-quarters of the cases, different surgical techniques such as rotation flaps and transposition flaps can be applied in one-quarter (3, 8, 10).

In spite of the studies regarding different skin closure techniques in the literature, an ideal synthetic material and surgical method recommendation for the treatment of meningocele and myelomeningocele by directly comparing the use of autologous and synthetic dura mater has not been included in the guidelines yet (1, 3, 9, 11). It has been reported that there is a need for studies in which medium and long-term clinical results can be evaluated (11).

In the light of all this in literature, the aim of this study is to evaluate the postoperative complications between the use of autologous graft and synthetic graft during surgical treatment and to compare the efficacy and safety of the two treatment options in order to help develop new strategies for the treatment of spinal dysraphism and to deal with its complications.

Materials and Methods

Study design and location

In this cross-sectional study, which has a surgical

technique comparison design in the form of a retrospective file design, the relationship between the use of autologous and synthetic dura in the surgical treatment of patients admitted to our neurosurgery clinic between November 2010 and December 2016 with the diagnosis of meningocele or myelomeningocele and the complications developed after surgery has been researched. In the writing of this article, the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) checklist criteria were used (12).

Study population

Patients aged 0-1 years admitted to our neurosurgery clinic between November 2010 and December 2016 with the diagnosis of meningocele or myelomeningocele and who were operated on were included in this study. However, patients with missing patient records including birth records, disease history, physical examination, radiological evaluation results, surgery records and epicrisis report were excluded from the study.

Surgical procedure

The patients were placed in the prone position under general anesthesia, with the chest and abdomen free. After making an elliptical incision around the sac, the epitheliosis was incised into the thin area around the sac with a full-thickness skin incision. The incision was advanced towards the subcutaneous tissues, and it was observed that the fascia and dura mater opened outwards. The thecal sac base was dissected medially and mobilized, and its insertion through the fascia defect was found. The skin around the neural plate was excised circularly. The liberated neural plate was sutured to remain inside the dura using the microscope. In this way, the central canal was closed by suturing the neural plate with non-absorbable sutures from one side to the opposite side to form a tube. After the repair of the dural defect, the skin was dissected to mobilize and the wound was closed primarily.

The Limberg flap procedure was applied to patients who were determined to be unsuitable for primary closure. In the Limberg flap procedure, which is a transposition flap, the edges of the defect are cut as rhombuses with 120° and 60° angles, then a line equal to the length of one side of the rhombus and perpendicular to the vertical line and a second line parallel to the selected side of the rhombus are determined. A skin incision was made over this line, and then the Limberg flap was dissected over the muscular fascia. After dissection, the Limberg flap was transposed into the myelomeningocele defect. Synthetic dural tissue was used in patients whose defects could not be closed with dural repair in the Limberg flap procedure (Figure 1).



Figure 1. Schematic representation of the Limberg flap

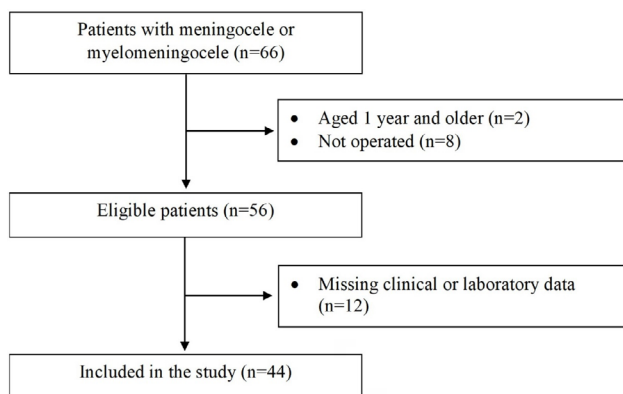


Figure 2. Flow-diagram of the study

Variables of the study

The infants' maternal age, gestational age, gender, preoperative neurological status, level of lesion, type of sac (meningocele or myelomeningocele), size of the defect, presence of chiari malformation and/or hydrocephalus, and time of shunt insertion if performed, timing of surgery, type of surgery (primary closure or Limberg flap), duration of operation (minutes), type of dura used (autologous or synthetic) were recorded. The patients were evaluated in terms of secondary surgery need and postoperative complications for 6 months after surgery, and the need for secondary surgery and postoperative complications were recorded during this period.

Defect size was obtained by multiplying the vertical and horizontal longest diameters and was calculated as square centimeters (cm²).

Five primary result variables of the study were identified. The first is the need for secondary surgery, which is used to evaluate the efficacy of the surgical procedure. Others are the presence of four different postoperative complications (postoperative infection, necrosis, wound dehiscence, and neurological deficit) that assess the safety of surgery.

Ethical issues and permissions

The study was carried out within the principles of the Declaration of Helsinki, and before the study, the ethics committee of Necmettin Erbakan University Non-Pharmaceutical and Medical Device Researches was applied and ethics committee approval was obtained (Ethics Committee Permission No: 2022/3894 Date: 22/07/2022). Since the study had a retrospective design, informed consent form could not be obtained from the parents or guardians of the patients.

Statistical analysis

The data of the study were analyzed using SPSS version 20 statistical software (IBM Corp. in Armonk, NY). The distribution of numerical variables was evaluated using the Kolmogorov-Smirnov test. Descriptive statistics of numerical variables that do not fit the normal distribution are shown with median, minimum and maximum values, and frequency (n) and percentage (%) for categorical variables. The Mann-Whitney U Test was used to compare the non-normally distributed numerical variables between the two groups. Pearson Chi-Square Test or Fisher's Exact Test was used to compare categorical variables. Statistical significance cut-off value was accepted as $p < 0.05$.

Findings

During the study, which was accepted with the approval of the ethics committee, 66 patients were admitted to the hospital with the diagnosis of meningocele or myelomeningocele. Two of these patients were 1 year old or older, and 8 were unoperated. After these patients were excluded, 56 patients were eligible to participate in the study. Twelve of these 56 patients were excluded from the study due to lack of clinical or laboratory data, and as a result, the data of 44 patients were included in the study and analyzed (Figure 1).

While the median maternal age of 44 patients included in the study was 24.5 years, more than half of the mothers were between the ages of 21-30. While the median gestational age was 36.0 weeks, the majority of babies (86.4%) were preterm. While 23 (52.3%) of the babies were boys and 21 (47.7%) were girls, and approximately two thirds of them had plegia. On the other hand, partial loss of muscle strength was found in 10 patients and paresis in 6 patients. While the lesion was isolated in the lumbar region in more than half of the patients, it was located in the lumbosacral region in 9 patients (20.5%), thoracolumbar region in 6 patients

(13.6%), and thoracic region in 5 patients (11.4%). Ten patients (22.7%) had meningocele and 34 (77.3%) had myelomeningocele. While the median value of the defect size detected in the patients was 20.0 cm², the association of Chiari malformation was found in 4 patients (9.1%), and the presence of hydrocephalus in 24 patients (54.5%). The median timing of shunt surgery was day 5 in 24 patients with hydrocephalus (Table 1).

Table 1. Demographic characteristics and basic clinical characteristics of the patients

Features (n=44)	
Maternal age (years), Median (Min-Max)	24,5 (18,0-41,0)
Aged 20 and below, n (%)	8 (18,2)
Between 21-30 years old, n (%)	24 (54,5)
Above 30 years, n (%)	12 (27,3)
Gestational age (weeks), Median (Min-Max)	36,0 (30,0-39,0)
30-37 weeks	38 (86,4)
38-42 weeks	6 (13,6)
Sex, n (%)	
Boy	23 (52,3)
Girl	21 (47,7)
Preoperative neurological status, n (%)	
No loss of muscle strength	10 (22,7)
Paresis	6 (13,6)
Plegia	28 (63,6)
Lesion level, n (%)	
Lumbar	24 (54,5)
Lumbosacral	9 (20,5)
Thoracolumbar	6 (13,6)
Thoracic	5 (11,4)
Sac type, n (%)	
Meningocele	10 (22,7)
Myelomeningocele	34 (77,3)
Defect size (cm ²), Median (Min-Max)	20,0 (4,0-100,0)
Presence of Chiari malformation, n (%)	4 (9,1)
Presence of hydrocephalus, n (%)	24 (54,5)
Shunt insertion time (days), Median (Min-Max)	5,0 (1,0-40,0)

Min: Minimum, Max: Maximum.

Surgery was performed on the first day in most of the patients, but was operated on the 26th day at the latest. The primary closure procedure was performed in 30 of the patients, and the Limberg flap procedure was performed in 14 patients. While the median operative time was 90 minutes, autologous dura was used in half of the patients and synthetic dura was used in the other half (Table 2).

A comparison of the demographic and clinical characteristics of the patients according to the dura type used is presented in Table 3. Accordingly, patients who used synthetic and autologous dura materials were similar in terms of maternal age, gestational age, gender, preoperative neurological status, lesion level, type of sac, presence of chiari malformation, surgery

timing, and operation time. On the other hand, the size of the defect was statistically significantly larger in patients using synthetic dura (24 cm² and 18 cm², respectively, $p=0.044$). However, hydrocephalus was detected in 90.9% of patients using synthetic dura and in only 18.2% of patients using autologous dura ($p<0.001$). In addition, primary closure was performed in all patients who used autologous dura, while only 36.4% had primary closure and 63.6% had Limberg flap procedure. This difference was found statistically significant ($p<0.001$) (Table 3).

Table 2. Characteristics of the operations of the patients

Features (n=44)	
Surgery timing (days), Median (Min-Max)	1,0 (1,0-26,0)
1st day	23 (52,3)
2nd day	9 (20,5)
3rd day and later	12 (27,2)
Type of surgery, n (%)	
Primary closure	30 (68,2)
Limberg flap	14 (31,8)
Operation time (minutes), Median (Min-Max)	90,0 (90,0-140,0)
Dura type, n (%)	
Synthetic	22 (50,0)
Autologous	22 (50,0)

Min: Minimum, Max: Maximum.

The secondary surgical need and complication status of the patients according to the dura type used are given in Table 4. Secondary surgery was required in the vast majority (81.8%) of patients using synthetic dura and in only 31.6% of patients using autologous dura. This difference is also statistically significant ($p<0.001$). Furthermore, postoperative infection developed in 31.8% of the patients using synthetic dura, and postoperative necrosis in 63.6%, while postoperative infection or necrosis did not develop in any of the patients using autologous dura. This difference in terms of these two complications was statistically significant ($p<0.001$ and $p<0.001$, respectively). Similarly, both postoperative wound dehiscence (72.7% vs. 4.5%) and postoperative neurologic deficit (54.5% vs. 100%) were statistically significantly more frequent in patients using synthetic dura ($p<0.001$ and $p<0.001$, respectively) (Table 4).

In Table 5, the need for secondary surgery and complication status in patients according to the type of surgery are presented. While the need for secondary surgery developed in 11 (36.7%) patients who underwent primary closure, secondary surgery was required in 10 (71.4%) patients who underwent Limberg flap procedure. This difference is statistically significant ($p=0.032$). However, the risk of developing postoperative infection was statistically similar in both surgical procedures. On the other hand, postoperative necrosis, wound dehiscence, and neurological deficit were statistically more frequent in patients who underwent Limberg flap ($p<0.001$, $p=0.002$ and $p=0.018$, respectively) (Table 5).

Table 4. Secondary surgery need and complication status in patients according to the dura type used

Features	Synthetic dura group (n=22)	Autologous dura group (n=22)	p
Secondary surgery need, n (%)	18 (81,8)	3 (13,6)	<0,001 ^a
Postoperative infection, n (%)	7 (31,8)	0 (0,0)	0,009 ^b
Postoperative necrosis, n (%)	14 (63,6)	0 (0,0)	<0,001 ^a
Postoperative wound dehiscence, n (%)	16 (72,7)	1 (4,5)	<0,001 ^a
Postoperative neurological deficit, n (%)	22 (100,0)	12 (54,5)	<0,001 ^a

a Pearson Chi-Square Test was used.

b Fisher's Exact Test was used.

Table 3. Comparison of the demographic and clinical characteristics of the patients according to the dura type used

Features	Synthetic dura group (n=22)	Autologous dura group (n=22)	p
Maternal age (years), Median (Min-Max)	26,0 (18,0-41,0)	24,5 (18,0-40,0)	0,832 ^a
Gestational age (weeks), Median (Min-Max)	35,0 (30,0-39,0)	36,0 (30,0-39,0)	0,497 ^a
Sex, n (%)			
Boy	11 (50,0)	12 (54,5)	0,763 ^b
Girl	11 (50,0)	10 (45,5)	
Preoperative neurological status, n (%)			
Loss of muscle strength	4 (18,2)	6 (27,3)	0,762 ^c
Paresis	3 (13,6)	3 (13,6)	
Plegia	5 (68,2)	13 (59,1)	
Lesion level, n (%)			
Lumbar	15 (68,2)	9 (40,9)	0,209 ^c
Lumbosacral	4 (18,2)	5 (22,7)	
Thoracolumbar	1 (4,5)	5 (22,7)	
Thoracic	2 (9,1)	3 (13,6)	
Sac type, n (%)			
Meningocele	4 (18,2)	6 (27,3)	0,472 ^b
Myelomeningocele	18 (81,8)	16 (72,7)	
Defect size (cm ²), Median (Min-Max)	24,0 (9,0-100,0)	18,0 (4,0-63,0)	0,044 ^a
Presence of Chiari malformation, n (%)	4 (18,2)	0 (0,0)	0,108 ^c
Presence of hydrocephalus, n (%)	20 (90,9)	4 (18,2)	<0,001 ^b
Surgery timing (days), Median (Min-Max)	24,0 (9,0-100,0)	18,0 (4,0-63,0)	0,346 ^a
Type of surgery, n (%)			
Primary closure	8 (36,4)	22 (100,0)	<0,001 ^b
Limberg flap	14 (63,6)	0 (0,0)	
Operation time (minutes), Median (Min-Max)	90,0 (90,0-140,0)	90,0 (90,0-110,0)	0,374

Max: Maximum, Min: Minimum.

a The Mann-Whitney U Test was used.

b Pearson Chi-Square Test was used.

c Fisher's Exact Test was used.

Table 5. Secondary surgery need and complication status in patients according to the surgery type

Features	Primary closure (n=30)	Limberg flap (n=14)	p
Secondary surgery need, n (%)	11 (36,7)	10 (71,4)	0,032 ^a
Postoperative infection, n (%)	4 (13,3)	3 (21,4)	0,662 ^b
Postoperative necrosis, n (%)	4 (13,4)	10 (71,4)	<0,001 ^b
Postoperative wound dehiscence, n (%)	7 (23,3)	10 (71,4)	0,002 ^a
Postoperative neurological deficit, n (%)	20 (66,7)	14 (100,0)	0,018 ^b

a Pearson Chi-Square Test was used.

b Fisher's Exact Test was used.

The subgroup analyzes of the secondary surgical need and complication status according to the type of surgery and the type of dura used in the patients revealed interesting results. There was no statistically significant difference in the need for secondary surgery and postoperative infection, necrosis and wound dehiscence in patients using synthetic dura according to the type of surgery. In addition, postoperative neurological deficits developed in all patients in this group in both types of surgery. On the other hand, since the primary closure procedure was used in all patients with autologous dura, the types of surgery could not be compared in these patients (Table 6).

Table 6. Subgroup analysis of secondary surgery need and complication status in patients according to the type of dura used

Type of dura used	Features	Primary closure	Limberg flap	p
Synthetic dura	Secondary surgery need, n (%)	8 (100,0)	10 (71,4)	0,254 ^a
	Postoperative infection, n (%)	4 (50,0)	3 (21,4)	0,343 ^a
	Postoperative necrosis, n (%)	4 (50,0)	10 (71,4)	0,386 ^a
	Postoperative wound dehiscence, n (%)	6 (75,0)	10 (71,4)	0,999 ^a
Autologous dura	Postoperative neurological deficit, n (%)	8 (100,0)	14 (100,0)	b
	Secondary surgery need, n (%)	3 (13,6)	-	b
	Postoperative infection, n (%)	0 (0,0)	-	b
	Postoperative necrosis, n (%)	0 (0,0)	-	b
	Postoperative wound dehiscence, n (%)	1 (4,5)	-	b
	Postoperative neurological deficit, n (%)	12 (54,5)	-	b

a Fisher's Exact Test was used.

b The test is not applicable.

Discussion

This study was conducted to compare the efficacy and safety of autologous graft and synthetic graft use during surgical treatment in patients aged 0-1 years who were operated on with the diagnosis of meningocele or myelomeningocele between November 2010 and December 2016 in a university hospital neurosurgery clinic. In total, while 86.4% of 44 infants included in the study according to the inclusion

and exclusion criteria were preterm, the predominant neurological condition was plegia in most of them. While the defect was isolated in the lumbar region in more than half of them, myelomeningocele was detected in 77.3% of them. While the median value of the defect size detected in the patients was 20.0 cm², primary closure was applied in 30 patients, Limberg flap procedure was applied in 14 patients, and autologous dura was used in half of the patients and synthetic dura was used in the other half. While the size of the defect was larger in patients using synthetic dura, hydrocephalus was found more frequently in these patients. While primary closure was applied in all patients with autologous dura, primary closure was performed in one-third of the patients in whom synthetic dura was used, and Limberg flap procedure was performed in two-thirds.

While the need for secondary surgery developed more frequently in patients who used synthetic dura, all postoperative complications were observed more frequently in these patients. In addition, the need for secondary surgery and postoperative necrosis, wound dehiscence, and neurologic deficit are more frequent in patients who underwent Limber flap compared to primary closure. However, the need for secondary surgery and the risk of postoperative complications were similar between primary closure and Limberg flap procedures in patients using synthetic dura. In other words, although the efficacy and safety of the Limberg flap were seen to be worse at first glance, it was found that the efficacy and safety of Limberg flap and primary closure were similar in patients who needed the use of synthetic dura, had larger defect and were more frequently associated with hydrocephalus.

Autologous repair of the dura mater of the spinal cord may not be possible, especially in the surgical treatment of complicated myelomeningocele and meningocele cases with large defect size. The reason for this is the increased risk of increased intracranial pressure with primary repair, and therefore the increased risk of cerebrospinal fluid (CSF) leakage (13, 14). Similarly, the same risk can occur with primary closure of the skin (14). In such cases, different flap turning techniques are used (8, 10, 15-20). As a result of these techniques, treatment results differ. Formentin et al. (15) used a keystone design perforator island flap in their study with 7 patients with myelomeningocele, in whom they evaluated that primary closure would not be possible. The authors reported that one patient died due to neonatal sepsis, none of the other six patients developed necrosis, detachment, infection, or CSF leakage, and ventriculoperitoneal shunt was required in 5 patients (15). Rankin et al. (16), in their 2-year follow-up study of 7 patients with myelomeningocele using quadruple V-Y advancement (butterfly) flaps, concluded that the mean defect width corresponded to 52% of the dorsal width, and the defect area corresponded to 21% of the total dorsal area. They reported that all patients had skin detachment on the 12th day on average, four patients needed secondary surgery, but no dural leakage or flap loss was

experienced. Lien et al. (17) performed defect repair using a combination of muscle and fascia flaps in a series of 45 patients in which they reported their 15-year experience in a single center. They used lumbosacral fascial flap in 30 patients (with paraspinous muscle in 12 cases), facial closure with pedicular periosteum in four cases, and combinations of these techniques with latissimus dorsi flap in other cases. In conclusion, it was reported that no CSF leakage was observed in any patient, and only one patient needed secondary surgery due to flap necrosis. In a four-year prospective non-randomized study conducted in a tertiary hospital with 27 newborns, bilateral V-Y advancement pedicle flap was applied to 7 infants and 1 child who could not undergo primary closure. It was reported that the mean duration of flap reconstruction after dural repair was 38.6 minutes in patients with a mean defect width of 8.5 cm, fat necrosis in two patients, flap necrosis in two patients, hematoma in one patient, CSF leak in one patient, and wound infection and subsequent meningitis in one patient (18). In another study, it was indicated that in 9 newborns with myelomeningocele with a mean defect size of 9 to 12 cm, the skin was closed with an O-S advancement flap after primary repair of the placode, and the patients were followed for an average of 9.2 months, and only one patient had CSF leak and the patient underwent secondary surgery. Apart from this, no complications such as wound dehiscence and ischemia have been reported (19). Kattan et al. (20) examined 10 cases of myelomeningocele defect closure with transverse-oblique advancement flap in their three-year retrospective study in two centers. They stated that the defect was most commonly located in the lumbosacral region (50%), the mean defect area was 22 cm², the most common preoperative neurological condition was plegia, venous congestion in 8 patients after the operation, wound dehiscence in 7 patients, flap necrosis more than 2 cm in 2 patients, and seroma in 1 patient and the complications were resolved with medical treatment and the need for secondary surgery did not develop.

Lobo and Nayak (21) compared 9 cases who underwent primary repair due to myelomeningocele and 13 cases who used V-Y advancement flap, and found that the average operative time for primary closure was 120 minutes, and for advancement flap 190.7 minutes. They reported CSF leakage in all 9 patients who underwent primary closure, hydrocephalus in 3, wound dehiscence in 6, neurological deficit in 3, and death in one case. In addition, they reported that CSF leakage developed in 3, hydrocephalus in 1, neurological deficit in 5 of 13 infants who underwent V-Y advancement flap, and there was no wound dehiscence or death in this group. Atalay et al. (22) compared the data of 11 patients in whom dorsal intercostal perforator artery flap was used for the closure of myelomeningocele defects and 13 patients who underwent primary closure, and found that most of the patients had the sac in the lumbosacral region, and the mean defect area was 14.2 cm² and 18.4 cm² in the primary closure and flap groups, respectively.

These researchers concluded that three patients in the primary closure group developed wound necrosis, two patients developed wound dehiscence, and four patients developed CSF leakage, while in the flap group, one patient developed wound necrosis and one patient developed CSF leakage. Afterwards, they reported that the difference was statistically significant, and that large defect size, kyphotic deformity and presence of hydrocephalus were risk factors for postoperative complications. Shim et al. (8) used primary closure in 12 of 14 patients in their 10-year myelomeningocele surgery series, in a similar design to our study, and used synthetic dura mater in one patient who underwent Limberg flap and Limberg flap in two cases. They stated that infection and wound dehiscence developed in one of 12 patients who underwent primary closure, and local advancement flap was applied with secondary surgery in this patient, and infection developed in one patient and was corrected with debridement. In one of the two patients who underwent Limberg flap, infection developed and therefore, the synthetic dura was removed with secondary surgery and autologous dural repair was performed. In the other Limber flap case, it was revealed that dehiscence and necrosis developed, and therefore, rotational flap and full-thickness skin graft were applied with secondary surgery although all wounds healed completely. In a more recent study, in which the Limberg flap was applied in 11 patients, it was found that wound dehiscence developed in only one patient and this case also improved with conservative treatment, and the findings were found satisfactory (10). The findings of our study show similarities and differences with the literature. Although no statistical comparison was made in the study of Shim et al. (8), our results were better than this study but worse than the other study (10). However, it should not be forgotten that the choice of surgical modality is closely related to the size of the defect and the patient's clinic (3, 23). If the vertical and horizontal diameter of the defect and the defect area are large, flap techniques are used instead of primary repair (23).

In postnatal surgery, the choice of surgical procedure and flap use and the type of flap used depend on the experience and choice of the surgeon, as well as patient characteristics (1, 3). Kemaloğlu et al. (24) performed primary repair in 30 patients, Limberg flaps in 17 patients, and bipediculated flaps in 3 patients in a study conducted with 50 patients with myelomeningocele. The authors aimed to develop a guideline to decide on the choice of surgery and purposely indicated that primary closure should be performed if the defect height/defect width < 1.5cm or the defect height/defect width ≥ 1.5cm and the posterior axillary line length/defect width ≥ 3cm. However, they argued that flap closure is appropriate if the defect height/defect width is ≥ 1.5cm and the posterior axillary interline length/defect width is < 3cm. As a result of these, they reported that successful one-stage tension-free closure was achieved in all patients, and no complications developed except for 4 patients who underwent flap reconstruction with

partial flap necrosis or minimal flap tip necrosis (24). Despite these results, the answers to the questions of which patients should be applied flaps and what type of flap should be used are still not included in the treatment guidelines (1, 3).

In our study, except for a few patients, the majority of the cases were operated within the first 3 days. Myelomeningocele surgery should be performed as early as possible if the general condition of the patient is good and there are no signs of infection (14). In general, surgery is mostly performed in the first 48 hours (3, 14). Delaying surgery for more than 72 hours increases the risk of complications that may develop in the patient (14).

Basically, the aims of spina bifida surgery are to resect the malformed sac, to eliminate the risk of infection by creating a barrier between the spinal canal and the outside of the spinal canal, and to restore the normal CSF environment by repairing the deformed spinal cord. These aims are achieved by reconstruction of the neural placode and closing the meninges, facial, subcutaneous and skin layers (14). Another important factor affecting patient outcomes as well as wound closure techniques in spina bifida surgery is the use of autologous dura, that is, the choice of primary repair use or synthetic dura use for closure of the neural placode (1, 14). In the presence of a large defect, a very large placode, or a very straight spinal canal, primary repair of the dura mater may cause compression of the spinal cord after reconstruction (14). Therefore, different types of artificial materials are used for dural repair in these cases (3, 11, 14). In the literature, among the artificial but non-synthetic materials used in postnatal surgery, those of human origin are frozen amniotic membrane (25), dermal matrix (26), autologous amniotic membrane (27), and cadaveric dura (28). As for animal source materials, horse achilles tendon-based collagen foil (29), bovine tendon-based collagen (26), bovine dermal matrix (30), bovine pericardium (31) are included. In addition, artificial materials of human origin, animal and vegetable origin are used in prenatal surgery (11).

In addition to artificial patches of animal and human origin, there are also synthetic production dura materials of different materials used in postnatal spina bifida surgery. Some of these materials are silicone coated polyester fabric (32, 33), silicone elastomer (34) and polytetrafluoroethylene (teflon) (31, 35). In the majority of studies using these materials, the study designs are case reports or small patient series. In addition, the success of the artificial or synthetic materials used can vary greatly depending on factors such as the patient's clinic, the presence of additional anomalies, and the variability of the skin closure techniques used (11).

In our study, we compared the use of synthetic dura mater with the use of autologous dura mater and found that the results were worse in patients with synthetic dura mater use as the size of the defect was larger and the presence of hydrocephalus was

more frequent in this patient group. Because, both the increase in the width of the defect size and the coexistence of hydrocephalus negatively affect the surgical success (1, 3, 6, 13, 14, 17). Moreover, in the comparison of the primary repair and Limberg flap we performed in our study at first glance, we found that both surgical techniques produced similar results in the subgroup analysis we performed according to the dura type used, although the results of the Limberg flap application were worse in the first analysis. Based on this, it can be concluded that the choice of autologous or synthetic dura mater also affects the success of the surgical closure technique.

Limitations

Our study has some limitations. The first of these is that it was carried out in a single center and with a relatively small number of patients. Therefore, the generalizability of the findings to the entire patient population is limited. Patients using synthetic dura actually consist of patients with more severe defect size and clinic, and therefore a higher risk of post-treatment complications. Advanced analysis methods such as regression analysis, in which more than one confounding factor can be evaluated, could not be used due to insufficient sample size. On the other hand, only subgroup analyzes were performed according to dura type, and the chance of success and complication rates between surgical methods were compared. In addition, the defect area was not measured using a topographic method, but was calculated only by the multiplication of the vertical and horizontal longest axis. This calculation may have resulted in an overestimation of the defect size. However, since the study data were collected retrospectively, the defect area could be calculated in this way in a standardized manner. These limitations should be considered when interpreting the results of our study.

Conclusion

Although synthetic dura clinic is used in more severe patients, it has more secondary surgery need and more complication risk compared to autologous dura. In patients using synthetic dura, primary closure and Limberg flap have similar efficacy and safety. It is necessary to compare the findings of our study with the findings of prospective studies to be conducted in larger patient groups.

Conflict of interest

There is no conflict of interest between the authors.

Financial support

No financial support was received from any person, institution or organization during the design, planning, execution and reporting stages of the present study.

Author Contributions

Conception: E.K.İ., F.K., Data Collection and Processing: F.K., D.A., Design: E.K.İ., Supervision: E.K.İ., F.K., Analysis and Interpretation: E.K.İ., F.K., D.A., Literature Review: E.K.İ., Writer: E.K.İ., Critical Review: F.K., D.A.

References

- Copp AJ, Adzick NS, Chitty LS, et al. Spina bifida. *Nature reviews Disease primers*. 2015;1:15007.
- International Clearinghouse for Birth Defects Surveillance and Research (ICBDSR), Annual Report 2014. Rome, Italy: International Centre on Birth Defects-ICBDSR Centre; 2014.
- Blount JP, Bowman R, Dias MS, et al. Neurosurgery guidelines for the care of people with spina bifida. *Journal of pediatric rehabilitation medicine*. 2020;13(4):467-77.
- Adzick NS, Thom EA, Spong CY, et al. A randomized trial of prenatal versus postnatal repair of myelomeningocele. *The New England journal of medicine*. 2011;364(11):993-1004.
- Johnson MP, Bennett KA, Rand L, et al. The Management of Myelomeningocele Study: obstetrical outcomes and risk factors for obstetrical complications following prenatal surgery. *American journal of obstetrics and gynecology*. 2016;215(6):778.e1-.e9.
- Farmer DL, Thom EA, Brock JW, 3rd, et al. The Management of Myelomeningocele Study: full cohort 30-month pediatric outcomes. *American journal of obstetrics and gynecology*. 2018;218(2):256.e1-.e13.
- Shobeiri P, Presedo A, Karimi A, et al. Orthopedic management of myelomeningocele with a multidisciplinary approach: a systematic review of the literature. *Journal of orthopaedic surgery and research*. 2021;16(1):494.
- Shim JH, Hwang NH, Yoon ES, et al. Closure of Myelomeningocele Defects Using a Limberg Flap or Direct Repair. *Archives of plastic surgery*. 2016;43(1):26-31.
- Banda CH, Narushima M, Ishiura R, et al. Local Flaps with Negative Pressure Wound Therapy in Secondary Reconstruction of Myelomeningocele Wound Necrosis. *Plastic and reconstructive surgery Global open*. 2018;6(12):e2012.
- Sharma MK, Kumar N, Jha MK, et al. Experience with various reconstructive techniques for meningocele defect closure in India. *JPRAS open*. 2019;21:75-85.
- Miyabe MM, Murphy KP, Oria M, et al. Dural substitutes for spina bifida repair: past, present, and future. *Child's nervous system : ChNS : official journal of the International Society for Pediatric Neurosurgery*. 2022;38(5):873-91.
- von Elm E, Altman DG, Egger M, et al. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement: Guidelines for Reporting Observational Studies. 2007;18(6):800-4.
- Nejat F, Baradaran N, Khashab ME. Large myelomeningocele repair. *Indian journal of plastic surgery : official publication of the Association of Plastic Surgeons of India*. 2011;44(1):87-90.
- Caldarelli M, Rocco CD. Myelomeningocele Primary Repair Surgical Technique. In: Memet Özek M, Cindilli G, Maixner WJ, editors. *The Spina Bifida: Management and Outcome*. Milano: Springer Milan; 2008. p. 143-55.
- Formentin C, de Andrade EJ, Matias LG, et al. Using the keystone design perforator island flap in large myelomeningocele closure. *Neurosurgical focus*. 2019;47(4):E19.
- Rankin TM, Wormer BA, Tokin C, et al. Quadruple Perforator Flaps for Primary Closure of Large Myelomeningoceles: An Evaluation of the Butterfly Flap Technique. *Annals of plastic surgery*. 2019;82(6S Suppl 5):S389-s93.
- Lien SC, Maher CO, Garton HJ, et al. Local and regional flap closure in myelomeningocele repair: a 15-year review. *Child's nervous system : ChNS : official journal of the International Society for Pediatric Neurosurgery*. 2010;26(8):1091-5.
- Kesan K, Kothari P, Gupta R, et al. Closure of large meningocele wound defects with subcutaneous based pedicle flap with bilateral V-Y advancement: our experience and review of literature. *European journal of pediatric surgery : official journal of Austrian Association of Pediatric Surgery [et al] = Zeitschrift für Kinderchirurgie*. 2015;25(2):189-

94.

19.Emsen IM. Closure of Large Myelomeningocele Defects Using the O-S Flap Technique. *The Journal of craniofacial surgery*. 2015;26(7):2167-70.

20.Kattan AE, Alsufayan FA, Alammam AK, et al. Extended Transverse-oblique Back Flap for Myelomeningocele Defect Closure: A Case Series of 10 Patients. *Plastic and reconstructive surgery Global open*. 2020;8(9):e3095.

21.Lobo GJ, Nayak M. V-Y Plasty or Primary Repair Closure of Myelomeningocele: Our Experience. *Journal of pediatric neurosciences*. 2018;13(4):398-403.

22.Atalay T, Oktay K, Guzel E, et al. Comparison of dorsal intercostal perforator artery flap and primary closure in myelomeningocele repair. *Child's nervous system : ChNS : official journal of the International Society for Pediatric Neurosurgery*. 2021;37(1):229-33.

23.Mohamed M, Ragae M, Ali W, et al. Standard protocol for closure and repair of post-meningocele and meningomyelocele back skin defect. 2019;4(1):104-8.

24.Kemaloğlu CA, Özyazgan İ, Ünverdi Ö F. A decision-making guide for the closure of myelomeningocele skin defects with or without primary repair. *Journal of neurosurgery Pediatrics*. 2016;18(2):187-91.

25.Pignatti M, Feletti A, Sapino G, et al. Myelomeningocele Repair Combining a Double Cryopreserved Amniotic Membrane Homograft and the Keystone Flap in a 3-Year-Old Child: A Case Report. *Pediatric neurosurgery*. 2020;55(2):106-12.

26.Grigoryants V, Jane JA, Jr., Lin KY. Salvage of a complicated myelomeningocele using collagen (Duragen) and dermal (Alloderm) matrix substitutes. Case report and review of the literature. *Pediatric neurosurgery*. 2007;43(6):512-5.

27.de Weerd L, Sjøvik K, Pedersen LK, et al. Triple Use of Autologous Amnion Graft in the Treatment of Meningomyelocele and Split Cord Malformation. *Plastic and reconstructive surgery Global open*. 2020;8(1):e2539.

28.Hoffman HJ, Taecholam C, Hendrick EB, et al. Management of lipomyelomeningoceles. Experience at the Hospital for Sick Children, Toronto. *Journal of neurosurgery*. 1985;62(1):1-8.

29.Pettorini BL, Tamburrini G, Massimi L, et al. The use of a reconstituted collagen foil dura mater substitute in paediatric neurosurgical procedures—experience in 47 patients. *British journal of neurosurgery*. 2010;24(1):51-4.

30.Susarla SM, Hauptman J, Ettinger R, et al. Acellular Dermal Matrix as a Definitive Reconstructive Option for Management of a Large Myelomeningocele Defect in the Setting of Severe Lumbar Kyphosis. *World neurosurgery*. 2019;129:363-6.

31.Samuels R, McGirt MJ, Attenello FJ, et al. Incidence of symptomatic retethering after surgical management of pediatric tethered cord syndrome with or without duraplasty. *Child's nervous system : ChNS : official journal of the International Society for Pediatric Neurosurgery*. 2009;25(9):1085-9.

32.Thompson DN, Taylor WF, Hayward RD. Silastic dural substitute: experience of its use in spinal and foramen magnum surgery. *British journal of neurosurgery*. 1994;8(2):157-67.

33.Bartal AD, Heilbronn YD, Plashkes YY. Reconstruction of the dural canal in myelomeningocele. Case report. *Plastic and reconstructive surgery*. 1971;47(1):87-9.

34.Venes JL. Surgical considerations in the initial repair of meningomyelocele and the introduction of a technical modification. *Neurosurgery*. 1985;17(1):111-3.

35.Nakazawa H, Kikuchi Y, Honda T, et al. Successful management of a small infant born with a large meningomyelocele using a temporary artificial dermis. *Scandinavian journal of plastic and reconstructive surgery and hand surgery*. 2005;39(1):53-6.