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Post-operative cerebrospinal fluid leak: single-continuous versus double-breasted dural repair of myelomeningocele

Usman Daibu^{1*} , Aliyu Muhammad Koko² and Babagana Usman¹

Abstract

Background Myelomeningocele is the most common neural tube defect in our environment. Initial surgical management involves untethering and water-tight dural closure. Single-continuous dural repair is more straightforward and faster than the double-breasted repair, even though the latter offers more strength to the reconstructed dura as the repair is in two layers. Preference was given to single-continuous repair even though the two techniques were not compared in terms of post-operative cerebrospinal fluid leak. The aim of this study was to compare the frequency of cerebrospinal fluid (CSF) leak following single-continuous versus double-breasted dural repair of myelomeningocele.

Patients and methods This was a randomized prospective study that reviewed all patients that presented to Usmanu Danfodiyo University Teaching Hospital (UDUTH) Sokoto, Nigeria, with myelomeningocele who met the inclusion criteria. Fifty-four patients were enrolled into the study randomized into two groups of 27 patients each. Group 1 had single-continuous repair, while group 2 had double-breasted technique. Post-operatively, patients were assessed for post-operative cerebrospinal fluid leak and pseudomeningocele. Data collected were analysed using the statistical package for social sciences version 22.0. The value for significance was set at 0.05.

Results The median age at presentation for both groups was 5 months. Both groups showed female preponderance with a female-to-male ratio of 1.3:1 and 1.7:1. Post-operative CSF leak occurred in 2(7.4%) patients in the single-continuous group compared to 3(11.1%) patients in the double-breasted group. Only 1(3.7%) patient in the single-continuous group developed pseudomeningocele and none in the double-breasted.

Conclusion Dural repair technique of myelomeningocele does not influence the occurrence of post-operative cerebrospinal fluid leak.

Keywords Myelomeningocele, Single-continuous, Double-breasted, Cerebrospinal fluid leak

Introduction

Myelomeningocele is the most common neural tube defect in our environment [1]. It is defined as a congenital defect in the vertebral arches with cystic dilatation of meninges and structural or functional abnormality of the spinal cord or cauda equina due to failure of the spinal cord to fuse dorsally during primary neurulation [2, 3]. The goal of surgical repair is to free the neural placode from its dural attachment (to avoid tethering),

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water-tight dural repair and skin closure to avoid CSF infection especially in ruptured myelomeningocele [2].

The average complication rate following Myelomeningocele repair has been reported to be between 7.7 and 33% [4–7]. Cerebrospinal fluid leak was among the common complications [1, 8]. Others include meningitis, seroma, hematoma, wound infection and dehiscence, skin flap necrosis and hydrocephalus [1, 9]. Shehu et al. reported a CSF leak rate of 6% using predominantly the single repair dural closure [10]. Khan et al. in a study on short-term complications of Myelomeningocele reported an incidence of 15% even though the dural repair technique was not mentioned [11]. Kural et al. reported a CSF leak rate of 6% similar to that reported by Shehu et al. and pseudomeningocele rate of 17% without highlighting the dural closure technique [12]. However, most studies did not mention pseudomeningocele as a separate complication of dural closure of myelomeningocele.

Dural closure preference has been debated in contemporary neurosurgery [13]. Among the different techniques described in the literature, single-continuous and double-breasted techniques are the most widely employed [1, 8]. Preference was to the single repair technique with double-breasted used occasionally. The added benefit of double breasting of the dura remains unclear in preventing post-operative cerebrospinal fluid leak. Therefore, this study compared the two techniques (single-continuous versus double-breasted) to determine the occurrence of post-operative cerebrospinal fluid leaks.

Patients and methods

This study was a hospital-based prospective study conducted at the Department of Neurosurgery Usmanu Danfodiyo University Teaching Hospital (UDUTH) Sokoto, Northwestern Nigeria, for one year. Ethical clearance and informed consent were obtained from the hospital ethical committee and parents/caregivers before the study’s commencement. A total of 27 patients per group arrived at, using the formula for comparative study when the endpoint is qualitative. All patients with myelomeningocele that presented to the Neurosurgery outpatient department were enrolled except for patients with co-existing symptomatic hydrocephalus that will require shunting, infected myelomeningocele, ruptured/leaking myelomeningocele and patients whose parents/caregivers did not consent to the study. A total of 54 patients for the two groups (group 1—single-continuous repair and group 2—double-breasted repair) were recruited. Computer generated random numbers were used to randomize these patients into the two groups of 27 patients each before commencement of the study (Table 1). At the time of enrolment, patients were blinded as they were

Table 1 Randomization of study participants

Total number of patients (n = 54) serial number	Computer generated random numbers	Grouping Group 1—Single-continuous Group 2—Double-breasted
50	0.681648	GROUP1
23	0.673605	GROUP1
34	0.8203	GROUP2
36	0.931182	GROUP2
27	0.870242	GROUP1
21	0.769891	GROUP2
38	0.397943	GROUP2
17	0.341132	GROUP2
20	0.18704	GROUP1
46	0.64165	GROUP2
3	0.96855	GROUP1
35	0.907612	GROUP2
13	0.374128	GROUP1
40	0.943072	GROUP2
8	0.544075	GROUP2
12	0.869858	GROUP1
14	0.665156	GROUP1
5	0.577816	GROUP2
26	0.575054	GROUP1
29	0.067453	GROUP2
48	0.633643	GROUP2
44	0.554266	GROUP2
4	0.476851	GROUP1
22	0.085918	GROUP2
52	0.034968	GROUP1
6	0.09727	GROUP1
16	0.731923	GROUP1
18	0.262094	GROUP1
32	0.64946	GROUP1
2	0.1814	GROUP1
15	0.496957	GROUP1
10	0.601016	GROUP1
42	0.343562	GROUP2
9	0.081956	GROUP1
47	0.045926	GROUP2
53	0.680607	GROUP1
33	0.896009	GROUP2
7	0.076527	GROUP1
37	0.212344	GROUP2
28	0.888529	GROUP2
45	0.710663	GROUP1
1	0.501163	GROUP1
19	0.021801	GROUP1
39	0.932092	GROUP2
25	0.511502	GROUP2

Table 1 (continued)

Total number of patients (n = 54) serial number	Computer generated random numbers	Grouping Group 1—Single-continuous Group 2—Double-breasted
54	0.932383	GROUP2
31	0.236601	GROUP2
41	0.403465	GROUP1
49	0.553887	GROUP1
24	0.737736	GROUP2
30	0.531601	GROUP2
11	0.463397	GROUP2
51	0.314646	GROUP1
43	0.323104	GROUP2

given serial number 1–54 as they present consecutively which automatically placed them into one of the two randomized groups. Patients’ information was entered into a proforma by the researcher and trained research assistants. At presentation, information that was obtained included the followings: biodata, size of Myelomeningocele among others. The researcher and research assistants performed the surgery according to the principle of repair under the supervision of training consultants.

The asepsis technique was strictly applied. The dural defect was closed with an absorbable suture (polyglactin- 910, size 3/0) in a single-continuous fashion and two layers (double-breasted) in single- and double-breasted groups, respectively. Following dural repair, water-tight closure was confirmed by the Valsalva manoeuvre. Post-operatively, patients were nursed in the paediatric neurosurgical ward and discharged on 7th post-operative day in the absence of apparent complications. Stitches were removed on the post-operative day 14 in the outpatient clinic. All patients were followed for 30 days, including the hospital stay. Follow-up visits for patients that did not develop complications before discharge was scheduled for the 14th and 30th days post-operative, while those that developed post-operative complications before discharge had a single follow-up visit on the 30th day post-operative as they were still on admission on post-operative day 14. All the patients completed the study. Phone numbers of the parents/relatives collected were used to remind them two days before the scheduled date.

During this period, they were observed by trained and blinded research assistants for possible CSF leak (discharge of clear and colourless fluid from the

wound), pseudomeningocele (an extradural collection of CSF that results from dural breach), wound infection and hydrocephalus in the two groups (single-continuous and double breasting).

Data collected were analysed using the statistical package for social sciences (SPSS 22.0). Results were presented in tables. The Chi-square test was used to test the differences between the categorical variables and Fisher’s exact was used when Chi-square test was violated. Mann–Whitney test was used to compare continuous variables that are not normally distributed. Results were subjected to a statistical significance test, and a *P* value of less than 0.05 was considered significant.

Results

Fifty-four (54) patients were recruited in this study and randomized into two groups of 27 each using computer-generated random numbers. Group 1 patients had single-continuous, while group 2 had a double-breasted dural repair. All 54 patients completed the study. In both groups, the most common age range at presentation was 1–12 months. The median age was five months for both groups (*p*=0.931). Both groups showed female preponderance with a female-to-male ratio of 1.3:1 and 1.7:1 for groups 1 and 2, respectively (*p*=0.580), as shown in Table 2.

Eighty-one point five per cent of participants in group 1 had their lesion located in the lumbosacral region compared to 66.7% in group 2 (*p*=0.700). The least common site for both groups was cervical (Table 3).

As highlighted above, there was no statistically significant difference in age at presentation, gender and location of myelomeningocele between the two groups in this study, showing that the groups were well-matched for comparison.

Post-operative CSF leak occurred in 2(7.4%) patients in the single-continuous group compared to 3(11.1%) patients in the double-breasted group. Only 1(3.7%) patient in the single-continuous group developed pseudomeningocele, and none in the double-breasted (Table 4). All the 2 patients that developed CSF leak in the single-continuous group had concomitant wound infection compared to 1 out of the 3 patients in the double-breasted group.

Two (7.4%) patients in the single-continuous repair group developed symptomatic hydrocephalus compared to 1(3.7%) in the double-breasted group (Table 4). None of these patients had concomitant CSF leak.

Four (14.8%) patients in group 1 had wound infection with 2 patients having concomitant CSF leak. Out of the 3 (11.1%) patients that developed wound infection in group 2, only 1 had concomitant CSF leak.

Table 2 Distribution of participants by age and gender

Variables	Group 1 n=27 frequency (%)	Group 2 n=27 frequency (%)	Test of significance P value
<i>Age (month)</i>			
< 1	0 (0)	0 (0)	
1–12	23 (85.2)	26 (96.3)	Fisher's exact
> 12	4 (14.8)	1 (3.7)	p=0.351
Total	27 (100)	27 (100)	
Median age (IQR) (month)	5.00 (6)	5.00 (4)	MWU=359.5 p=0.931
<i>Gender</i>			
Male	12 (44.4)	10 (37.0)	$\chi^2=0.307$
Female	15 (55.6)	17 (63.0)	p=0.580
Total	27 (100)	27 (100)	
<i>Maternal age</i>			
< 20	7 (25.9)	9 (33.3)	
20–35	20 (74.1)	18 (66.7)	$\chi^2=0.355$
> 35	0 (0.0)	0 (0.0)	P=0.766
Total	27 (100)	27 (100)	
Median age (IQR)	25 (11)	22 (8)	MWU=331.500 P=0.566

χ^2 =Pearson's chi-square, MWU=Mann-Whitney U, Group 1= single-continuous dural repair, Group 2= Double-breasted dural repair

Table 3 Distribution of site of myelomeningocele between the two groups

Variables	Group 1 n=27 frequency (%)	Group 2 n=27 frequency (%)	Test of significance P value
<i>Site</i>			
Lumbosacral	22 (81.5)	18 (66.7)	Fisher's exact
Lumbar	2 (7.4)	5 (18.5)	p=0.700
Thoracic	2 (7.4)	3 (11.1)	
Cervical	1 (3.7)	1 (3.7)	
Total	27 (100)	27 (100)	

Discussion

Complications following myelomeningocele wound closure are well described [4–6, 14]. The most common are cerebrospinal fluid leak, meningitis, seroma, hematoma, skin flap necrosis, wound infection and dehiscence [1]. Post-operative cerebrospinal fluid leaks occur despite intraoperative Valsalva manoeuvre use to achieve watertight dural closure [15]. Of all the closure techniques, single-continuous and double-breasted repair were the most widely employed [1, 8]. However, a paucity of studies compare the two techniques with regard to these complications.

Findings in this study showed that 2(7.4%) patients that had the single-continuous dural repair developed

Table 4 Distribution of individual complications between the groups

Variable	Group 1 n=27 frequency (%)	Group 2 n=27 frequency (%)	Test of significance P value
<i>Complications</i>			
<i>CSF leak</i>			
Yes	2 (7.4)	3 (11.1)	Fisher's exact p=1.000
No	25 (92.6)	24 (88.9)	
Total	27 (100)	27 (100)	
<i>Pseudomeningocele</i>			
Yes	1 (3.7)	0 (0)	Fisher's exact p=1.000
No	26 (96.3)	27 (100)	
Total	27 (100)	27 (100)	
<i>Wound infection</i>			
Yes	4 (14.8)	3 (11.1)	Fisher's exact p=1.000
No	23 (85.2)	24 (88.9)	
Total	27 (100)	27 (100)	
<i>Hydrocephalus</i>			
Yes	2 (7.4)	1 (3.7)	Fisher's exact p=1.000
No	25 (92.6)	26 (96.3)	
Total	27 (100)	27 (100)	

post-operative CSF leak compared to 3(11.1%) in the double-breasted group, which was not statistically significant (p=1.000). Our finding was similar to that of Shehu et al. in Zaria, who reported a CSF leak rate of 6% [10],

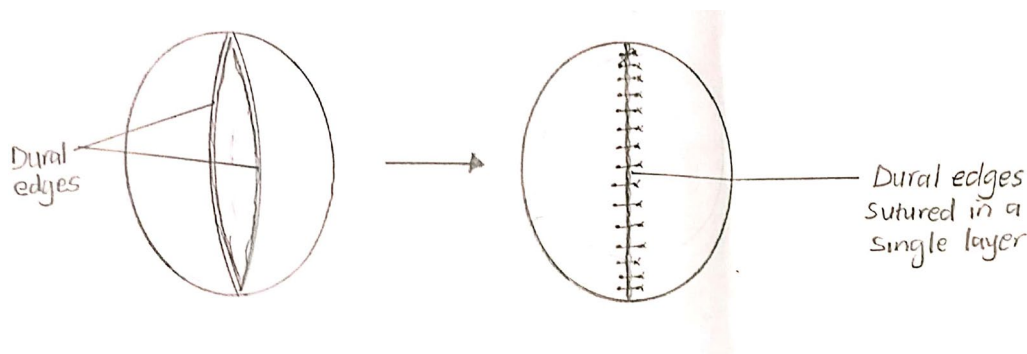


Fig. 1 Single continuous dural repair

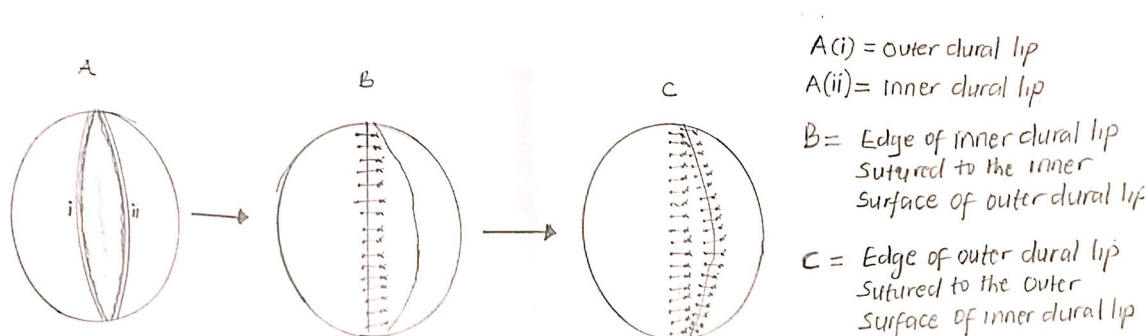


Fig. 2 Double breasted dural repair

using the single-continuous technique as highlighted in a subsequent study titled repair of myelomeningocele: how i do it [1]. It was also consistent with the CSF leak rate of 8.1% reported by Noman and Khan in Pakistan that employed a similar study design, and 6% reported by Kural et al. though the repair techniques were not mentioned [9, 12]. A CSF leak rate of 15% was reported by Khan et al. using a similar study design but without highlighting the dural closure technique [11]. It was higher than the rates reported in both groups in this study, possibly due to the higher rate of wound infection (23%) recorded in that study which is a known risk factor for CSF leak. All the patients that developed post-operative CSF leak in both groups were managed nonoperatively with acetazolamide to reduce CSF production, nursing in prone position and wound dressing. Management of post-operative cerebrospinal fluid leak remains controversial [16]. Many advocate primary repair, while others recommend a trial of cerebrospinal fluid diversion [17]. Khan et al. in a study on cerebrospinal fluid leak following repair of congenital spinal pathologies described the application of tincture benzoyl in 3 patients, skin reinforcement stitches in addition to tincture benzoyl in 9 patients and reoperation in 1 patient [18].

Pseudomeningocele is an extradural accumulation of cerebrospinal fluid in the back's soft tissue that extravasates through the dural tear [19]. It is an uncommon complication following spinal surgery [20], and myelomeningocele repair is not an exception. In this study, only one patient (3.7%) in group 1 who had repair of thoracic myelomeningocele developed this complication. This patient subsequently had wound exploration and direct dural repair which agrees with the finding of Tosun et al. [16] Lumbar-subarachnoid drain was also described as an alternative option [16]. None of the patients in group 2 had a similar complication, and the frequency of occurrence between the groups was not statistically significant ($p=1.000$). This complication may not be immediately evident as the CSF may be initially absorbed into the surrounding tissues but later less readily due to progressive reactions in the connective tissue of the surrounding, thus resulting in pseudomeningocele formation [19]. Kural et al. reported a higher pseudomeningocele rate of 17% [12], probably due to a longer mean follow-up period of 36 months compared to 30 days in our study. The short duration of follow-up in this study could explain why this complication occurred in only one patient. Most previous studies on myelomeningocele

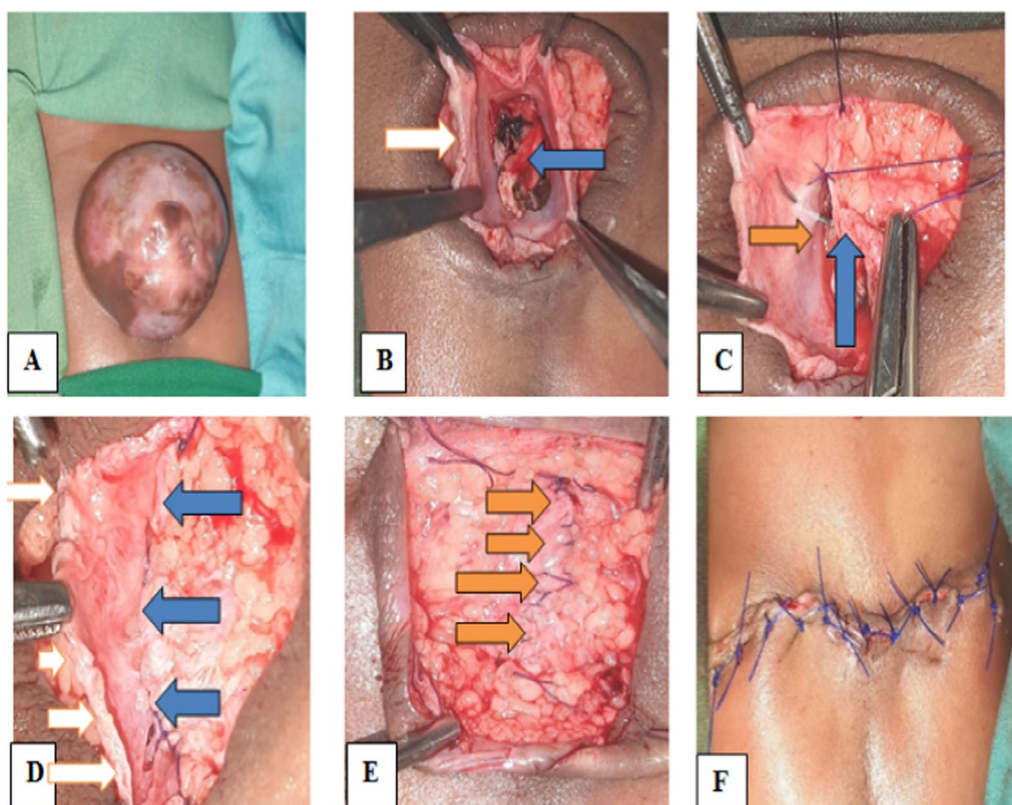


Fig. 3 Intraoperative pictures above showing a typical myelomeningocele (A), circumferentially dissected dura and neural tissue indicated by white and blue arrows respectively (B), the tip of inner dural leaflet (blue arrow) being sewn to the lower most inner surface (yellow arrow) of the outer dural leaflet (C), the first/inner breasting completed as shown in blue arrows and free edge of the outer dural leaflet held up as indicated by white arrows (D), the free edge of the outer dural leaflet sutured to the outer surface of the inner dural leaflet to complete the double breasting as shown by yellow arrows (E), wound closed in layers (F)

repair were silent about this complication, probably because is rare or was considered an occult CSF leak.

Two (7.4%) patients in the single-continuous repair group developed symptomatic hydrocephalus compared to 1(3.7%) in the double-breasted group, which was not statistically significant. These patients had ventriculoperitoneal shunt. In both groups, symptomatic hydrocephalus was the commonest complication that necessitated re-operation which agrees with the finding of Noman and Khan [9]. None of the patients with symptomatic hydrocephalus had concomitant CSF leak.

Four (14.8%) patients in the single-continuous dural repair group had wound infection with 2 patients having concomitant CSF leak compared to 3 (11.1%) patients in the double-breasted group with 1 patient having concomitant CSF leak. The frequency of occurrence of this complication between the 2 dural closure techniques was not statistically significant. Overall, our wound infection rate was 13% which is lower than 23% reported by Khan et al. [11] Generally, we encouraged parents/caregivers

to avoid wearing diaper to the patients post-operatively to prevent faecal contamination of the wound (especially lumbosacral lesions). This might have contributed to the low rate of wound infection in our study. All cases of wound infection were managed with wound dressing and antibiotics.

The fact that there was no statistically significant difference in the frequency of occurrence of post-operative CSF leak and pseudomeningocele between the two techniques in this study showed that none of the techniques is superior to the other. So also, the findings for both groups were consistent with that of authors that employed the single-continuous repair technique and most of the authors that did not mention the dural closure technique. This further supports our findings, though the single-centre nature of the study, the short duration of follow-up and the racial differences are some of the limitations that should be considered in interpreting the findings of this study.

Conclusions

Cerebrospinal fluid leak can occur following myelomeningocele repair, regardless of the dural closure technique employed. Therefore, the choice of technique of dural closure (single- or double-breasted), to a more significant extent, depends on the surgeon's preference. However, further studies are needed to validate this finding, considering the study's limitations.

We recommend that the dural closure technique of myelomeningocele can be achieved with a single-continuous or double-breasted technique with a similar outcome (Figs. 1, 2, 3).

Abbreviations

CSF	Cerebrospinal fluid
IQR	Interquartile range
MWU	Mann–Whitney <i>U</i>
SPSS	Statistical package for social sciences
UDUTH	Usmanu Danfodiyo University Teaching Hospital

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Author contributions

UD wrote greater part of the manuscript having conceived the idea and nurtured it. AMK assisted in developing the work and helped in reviewing it. BU proofread and discussed the manuscript.

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Availability of data and materials

The data that support the findings of this study are openly available.

Declarations

Ethical approval and consent to participate

Ethical approval was obtained from Usmanu Danfodiyo University Teaching Hospital Ethical and Research Committee (UDUTH/NHREC 2019/NO. 778). Informed consent was obtained from the parents/guardian of the participants before data collection.

Consent for publication

Consent to publish this research was obtained from the parents/caregivers of the study participants.

Competing interests

The authors declare that they have no competing interests.

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