



Original Article

Incidence of Post-Operative Cerebrospinal Fluid Leak in Patients Operated for Spinal Dysraphism

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ABSTRACT

Objective: The purpose of the study is to find the incidence of cerebrospinal fluid leak in patients operated for spinal dysraphism.

Materials and Methods: 80 patients who underwent surgery for spinal dysraphism were enrolled in this study. This study was conducted at Neurosurgery Unit, JPMC, Karachi. Patients from 1 month to 3 years of age were included in the study. Patients were followed for 2 months post-surgery. The incidence of cerebrospinal fluid leak among patients operated for spina bifida was determined.

Results: Mean age was $1.5 \text{ years} \pm 5 \text{ months}$. There were 68.7% males and 31.3% females. 47.5% of patients had myelomeningocele, 41.3% of patients had meningocele, and 11.25% of patients had Tethered cords. Among 80 patients, 16.3% developed CSF leaks. 6.3% of patients were those who developed postoperative hydrocephalus and their CSF leak resolved after the VP shunt. In 8.8% of patients, CSF leaks resolved after suturing the leak area. 1.3% of patients with CSF leak developed meningitis; he was kept on antibiotics and was discharged on the 10th day of readmission. 5% of patients developed wound infections, they were kept on antibiotics, and daily dressings and were discharged within a week.

Conclusion: Most common location for myelomeningocele and meningocele is the lumbosacral area followed by the thoracic and cervical regions. Overall, 28.6% developed complications. 16.3% of patients developed CSF leaks. CSF leak was seen more in myelomeningocele repair patients as compared to meningocele and tethered cord. Most of the leaks can be managed with simple suturing and VP shunts in patients complicated by postoperative hydrocephalus.

Keywords: Myelomeningocele (MMC), Meningocele, Cerebrospinal fluid leak (CSF), VP Shunt.

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INTRODUCTION

Congenital malformations of the Spine are generally described under the term Spinal

dysraphism or Spina Bifida (SDs). The term dysraphism is derived from the Greek words dys (bad) and raphe (suture). There is abnormal cell migration and differentiation during neural tube formation in the first trimester of pregnancy which results in Spinal Dysraphism/Bifida. The most complicated type of spina bifida is myelomeningocele which results from incomplete closure of the neural tube during primary neurulation.¹

After congenital heart disease, 2nd most common birth defects are spina bifida/neural tube defects. Prevalence is 1 – 3/1000 live births². The most common site is the lumbosacral spine (90%), followed by a dorsal spine (6 – 8%), and cervical spine (2 – 4% cases).³ Worldwide, its prevalence varies in each country. Spinal Dysraphism is divided into Spina Bifida occulta and Cystica (meningocele and myelomeningocele). Occulta type results in congenital absence of posterior elements including spinous processes and lamina, with no exposure of meninges and neural tissues. Meningocele results in a defect in vertebral arches with protrusion of meninges, but no neural tissue content or abnormality. Myelomeningocele results in the same defects as seen in meningocele along with abnormality of the spinal cord or cauda equina.⁴

The etiology of Spinal dysraphism is multifactorial, comprising nutritional, genetic, and environmental components.^{5,6} The most important factor in spinal developmental errors is genetics. But despite huge development in genetics over the past few years, little is known about the spinal developmental process. Variations in some specific genes and variants of chromosome 6q T locus have been related to the development of SDs. There is a higher prevalence of spinal dysraphism in females as compared to males.⁷ Nutritional deficiencies, particularly folate deficiency are associated with neural tube defects. All childbearing-age females should take 0.4 mg of folic acid daily.⁸ Some antiepileptics are

considered teratogenic (carbamazepine and valproic acid) and are associated with a 1% chance of having a child with spinal dysraphism.⁹

Hydrocephalus is commonly associated with spinal dysraphism, and if present, it requires treatment in 90 to 98% of patients which improves survival rate. Prenatal counseling is important for parents to understand that their child will need multidisciplinary care. Myelomeningocele repair should be performed within the first 72 hours after birth. Delayed repair is associated with an increased risk of complications which can result in mortality. CSF leak is a very common complication after myelomeningocele repair and is considered a deadly complication if it leads to meningitis. The incidence of CSF leaks after surgeries varies between 18% to 30%.¹⁰ We will determine local post-spinal bifida repair statistics from this study.

MATERIALS AND METHODS

Study Design and Setting

It is a prospective study. This study was carried out at Jinnah postgraduate medical Centre at their neurosurgery department in Karachi, for six (6) months from the 10th of July to the 10th of January 2022.

Number of Patients

84 patients with spinal dysraphism who were operated on for myelomeningocele, meningocele, and tethered cord were included. 4 patients did not return for follow-up. So, 80 patients who were operated for spina bifida were included.

Inclusion Criteria

All those patients were operated on after admission to a public sector for spinal dysraphism. All patients aged between 1 month and 3 years were included in the study.

Exclusion Criteria

Patients who did not return for follow-up.

Collection of Data

Operated patients with spinal dysraphism were followed for 2 months post-surgery. We collected data on baseline demographics like age, gender, and patients who also had hydrocephalus along with spinal dysraphism and postoperative complications including CSF leak, wound infection, hydrocephalus, and meningitis.

Data Analysis

Data analysis was done using SPSS version 25.0. Data is represented in the form of percentages and the chi-square test is applied and p-values are calculated as well.

RESULTS

Age and Gender Distribution

80 patients who underwent surgery for spina bifida were included in the study. The mean age was 1.5 years $\pm$ 5 months. There were 55 (68.7%) males and 25 (31.3%) females with male to female ratio of 2.2:1. Gender distribution is shown in Table 1.

Table 1: Gender distribution of patients.

Variables	Number of Patients	% Age
Males	55	68.7%
Females	25	31.3%
Total	80	100%

Table 2: Location of Myelomeningocele and Meningocele.

Variable	Number of Patients	Lumbosacral Region	Thoracic Region	Cervical Region	P value
Myelomeningocele	38	27 (71.05%)	6 (15.7%)	5 (13.1%)	0.986
Meningocele	33	24 (72.7%)	5 (15.2%)	4 (12.1%)	
Total	71	51 (71.8%)	11 (15.5%)	9 (12.6%)	

Location of Myelomeningocele and Meningocele

38 (47.5%) patients had myelomeningocele, 33 (41.3%) patients had meningocele, and 9 (11.25%) patients had Tethered cords. 27 (71.05%) out of 38 patients of myelomeningocele and 24 (72.7%) out of 33 patients of meningocele were located in the lumbosacral area, 6 (15.7%) patients of myelomeningocele were located in thoracic and 5 (13.2%) in the cervical region, while 5 (15.2%) patients of meningocele were located in thoracic and 4 (12.1%) in cervical region. The location of different pathologies is shown in Table 2.

Other Associated Pathologies with Spinal Dysraphism

52 (65%) patients had associated hydrocephalus preoperatively, 5 (6.25%) developed hydrocephalus after surgery, and 69 (86.3%) patients had Chiari 2 malformation.

Complications and Their Management

Among 80 patients 23 (28.75%) patients developed postoperative complications. 13 (16.3%) developed CSF leak, and 5 (6.3%) patients were those who developed postoperative hydrocephalus and their CSF leak resolved after VP shunt. In 7 (8.8%) patients, the CSF leak resolved after suturing the leak area. 1 (1.3%) patient developed meningitis, he was admitted, kept on antibiotics, and was discharged on the 10th day of readmission. 4 (5%) of our patients developed wound infections but there was no CSF leak, they were kept on antibiotics, and daily dressings and were discharged within a week. Complications of specific pathologies are shown in Table 3.

Table 3: Complications According to Specific Pathology.

Complications	Myelomeningocele N = 38	Meningocele N = 33	Tethered Cord N = 9	Total N = 80	P value
Post-Operative hydrocephalus	3 (3.75%)	1 (1.25%)	1 (1.25%)	5 (6.35%)	0.984
CSF leak	7 (8.75%)	4 (5%)	2 (2.5%)	13 (16.3%)	
Wound Infection	2 (2.5%)	1 (1.25%)	1 (1.25%)	4 (5%)	
Total	12 (15%)	5 (6.25%)	4 (5%)	23 (28.7%)	

DISCUSSION

The burden of spinal dysraphism is more in developing countries and it is challenging to address this issue. The main cause is a nutritional deficiency in these underdeveloped countries and our main focus should be on counseling families regarding the nutritional status of pregnant women. Surgery is one of the many components of the multidisciplinary approach needed for these babies. The major complications of repair of these congenital conditions are infections and CSF leaks.

In our study, the most common location of myelomeningocele and meningocele is at the lumbosacral area (71.8%) followed by thoracic (15.5%) and cervical region (12.6%). Similar to our results, a study conducted by Ntimbani et al, concluded that the most common type of spina bifida is myelomeningocele and it comprises up to 86.8% of spina bifida patients.¹¹ According to a study by Kumar R et al, among 155 cases of spina bifida in their study, myelomeningocele was found in 72% of cases and meningocele in 2% of patients.¹²

According to our study, the male-to-female ratio was 2.2:1. According to Anegbe et al, the male: female ratio was 1.3:1.¹³

In our study, the reported rate of postoperative complications is 28.7%. Among these CSF leak is seen in 16.3% which responded to plain sutures around leak areas and VP shunts in patients who developed hydrocephalus after myelomeningocele and meningocele repairs. VP shunt was performed in 5 (6.3%) patients. The CSF leak was more frequent in myelomeningocele repair (8.75%) patients as compared to

meningocele (5%) and tethered cord (2.25%). One of our patients developed meningitis after a CSF leak. 4 (5%) of our patients developed wound infections but no CSF leak. According to Reynolds et al, after surgery for myelomeningocele, wound infection is the most common postoperative complication.¹⁴ According to Cherian et al, CSF shunts were performed in 8% of the cases after surgery for myelomeningocele.¹⁵ In a study done by James HE et al, 10% of patients underwent VP Shunt, 20% developed post-operative wound infection and 14% developed CSF leak.¹⁶

One of the studies conducted by Khan et al, among their 73 patients, CSF leak was seen in 17.8%, which is near to our result. Their study also showed an increased rate of CSF leaks in patients with myelomeningocele repair as compared to other conditions. They employed VP shunts and simple suturing to control CSF leaks similar to ours but they also employed a tincture of benzoyl which was not used in our study.¹⁷ Another study by Baldia et al, in which he noted CSF leaks in 16/305 patients operated for the tethered cord (4.5%). In our study, the incidence of CSF leak in tethered cord patients was 2/9 (2.5%).¹⁸

In the Hungarian study which included 352 cases of myelomeningocele and meningocele, the most common location was cervical (1.8%) followed by thoracic (4.2%), lumbar (16.8%), sacral (34.5%), and at junctions; Cervicothoracic (0.9%) and Lumbosacral (22.3%).¹⁹ In the Nigerian study which included 106 cases, the most common location was lumbar (55.7%).²⁰

Nearly 40% of patients who developed CSF leak had postoperative hydrocephalus and the leak resolved as soon as the VP shunt was placed

in these patients. We did a radiological investigation after 2 weeks to see the development of hydrocephalus and early investigations were done in only those who developed CSF leaks. We need to perform early radiological investigations to observe the development of postoperative hydrocephalus. Studies are needed to see how early hydrocephalus develops in post-operative spinal dysraphism patients to avoid these complications through early interventions.

CONCLUSION

We concluded that the most common location for myelomeningocele and meningocele is the lumbosacral area followed by the thoracic and cervical region. Overall, 23 patients (28.6%) developed complications. 13 (16.3%) patients developed CSF leaks. The CSF leak was seen more in myelomeningocele repair patients as compared to meningocele and tethered cord. Most of the leaks can be managed with simple suturing and VP shunts in patients complicated by postoperative hydrocephalus. Approximately 40% of patients with CSF leaks had postoperative hydrocephalus for which a VP shunt was done and the CSF leak was resolved. So, early postoperative radiological investigations can help detect this problem, and studies are needed to see how early we can detect hydrocephalus after spinal dysraphism surgeries.

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Additional Information

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Ethical Review Board Approval: The study was conformed to the ethical review board requirements.

Human Subjects: Consent was obtained by all patients/participants in this study.

Conflicts of Interest:

In compliance with the ICMJE uniform disclosure form, all authors declare the following:

Financial Relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work.

Other Relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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AUTHORS' CONTRIBUTION

Author's Full Name	Intellectual Contribution to Paper in Terms of:
Raheel Gohar	Study design and methodology.
Iram Bokhari	Literature review and referencing.
Lal Rehman	Final review and approval.
Tanveer Ahmed	Data collection and calculations.
Sagheer Ahmed	Interpretation of results.
Daniyal Ahmed	Analysis of data.