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Clinical characterization of post-surgical outcomes and associated risk factors of lipomyelomeningocele in Eastern India

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ABSTRACT

Background: Lipomyelomeningocele (LMMC), the most complex spinal lipoma and a common spinal malformation, can cause complications ranging from bladder dysfunction to significant neurological deficits. Therefore, prompt diagnosis and treatment are essential. **Aims and Objectives:** The study aims to determine post-operative complications, functional outcomes, and risk factors in LMMC patients. **Materials and Methods:** This prospective observational study included 40 LMMC patients who underwent surgery, regardless of age and sex. Demographic, clinical, pre-operative, radiological, intraoperative, and post-operative data were recorded. Patients were followed for 4 months to assess complications (early and late) and risk factors associated with them. **Results:** Of 40 patients, most were 0–3 years with male predominance (57.5%). The lumbosacral region (47.5%) was the most common anatomical site, and transitional lipoma was the most frequent type (40%). Detethering was performed in all 40 patients, with total lipoma excision achieved in 67.5%. Post-operative complications occurred in 42.5% (17 patients): 13 experienced early complications (cerebrospinal fluid leak [7], infection [4], wound dehiscence [2]), and four experienced late complications (neurological deficit [1], pseudomeningocele [3]). Clinically, 7.5% of patients had developed new neurological deficits. Factors associated with complications included: Age 3–6 years ($P=0.02$), A-positive blood type ($P=0.01$), lumbosacral/sacral location ($P=0.014$), dorsal/chaotic/terminal lipoma type ($P<0.001$), and partial lipoma excision with iatrogenic injury ($P=0.001$) using a patching technique ($P=0.001$). Post-operative proning reduced the risk of complications ($P<0.001$). **Conclusion:** Our study demonstrates that detethering with complete lipoma excision, dural closure using glue, and post-operative proning improve outcomes and reduce post-operative complications in LMMC patients.

Key words: Cerebrospinal fluid; Lipomeningomyelocele; Neural tube defect; Spina bifida; Tethered cord; Wound infection

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INTRODUCTION

Neural tube defects are congenital birth deformities when the neural tube does not close properly.[1] Spina bifida is the most common neural tube closure defect that occurs when the spinal column does not close completely during pregnancy.[2] In India, spina bifida affects 1–3 of every 1,000 live births.[3] Lipomyelomeningocele or lipomeningomyelocele (LMMC), a rare but severe form of spina bifida, affects 8–25% of children with this condition.[4] LMMC – typically found in the lumbosacral region – is characterized by a subcutaneous fatty tumor (lipoma) extending through a

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spinal defect, causing protrusion of cerebrospinal fluid (CSF) and meninges.[5] LMMC occurs in 3–6/100,000 live births, and these patients mostly experience neurological or bladder-related symptoms, which worsen as the condition progresses.[6] Timely diagnosis and treatment are essential to prevent further complications.

Surgical procedures for lumbosacral lipomas provide temporary relief or reduce symptoms by releasing tension in the spinal cord. The procedure involves removing the fibroadipose mass while preserving neural tissues to prevent retethering of the spinal cord. Total or near-total resection offers long-term progression-free survival, whereas partial resection causes exuberant scarring at the lipoma-cord interface, worsening the prognosis.[7,8] While surgery can alleviate these symptoms, the type of procedure results in post-surgical complications, including but not limited to wound infections, CSF leaks, neurological deterioration, and tethered cord recurrence.[6,7] However, there is a risk of retethering, with rates ranging from 5% to 50%.[9]

The prevalence of post-surgical complications following LMMC repair depends on several factors such as age, sex, morphology, presence and severity of neurological symptoms, type of lipoma and surgical technique, and absence or presence of an associated spinal cord syrinx.[9] Studies evaluating the clinical profile of LMCC and the impact of various factors on repair outcomes in Eastern India are scarce; therefore, this study aimed to address this gap. The influence of the age at which deterioration occurs on long-term outcomes has not been widely reported. In addition, no studies have compared outcomes in children who developed new deficits beyond 1 year of age with those who had symptoms at or near birth. This study thus aims to determine the complications, functional outcomes, and risk factors for post-operative outcomes in patients with LMCC.

Aims

Aims To determine the complications, functional outcomes and risk factors for outcomes following surgery for lipomyelomeningocele in Eastern India.

Objectives

1. To determine the functional outcomes and risk factors for outcomes following surgery for lipomyelomeningocele in Eastern India
2. To assess various clinical, radiological and surgical factors responsible for better surgical outcome in neural tube defects.

MATERIALS AND METHODS

This prospective observational study, conducted from July 2022 to June 2024 at IPGMER and SSKM Hospital in Kolkata, included 40 patients with LMCC who underwent neurosurgery. The study recruited patients from those with spinal dysraphism who visited the neurosurgery outpatient department of this tertiary care center. Ethical clearance was obtained from the institute's ethical committee (IPGME and R/IEC/2023/823). Before beginning the study, all participants provided written informed consent in accordance with the Declaration of Helsinki principles. We included patients of any age and sex with LMCC, as confirmed by Magnetic Resonance Imaging, who required surgery. We excluded patients who had undergone previous surgery for LMCC, had been diagnosed with bleeding diathesis, or were unsuitable for anesthesia.

Data were collected using a pre-tested, pre-designed form. Demographic and clinical details, pre-operative symptoms and radiological findings, intraoperative procedures, and post-operative complications were recorded. Post-operative findings were recorded at <3 months for the early outcome, whereas late outcomes were recorded at >3 months. All patients were followed for 4 months to assess early and late complications of LMCC surgery and the risk factors associated with it.

Surgical procedure

All the patients underwent surgery employing different surgical approaches and techniques. Detethering was done in all patients. The lipoma and the conus were detached from the dura all around, including inferiorly, to completely untether the cord. A radical or near total excision of the lipoma was defined when the entire placode and roots could be visualized, and only small bits of lipoma densely adherent to these structures were left *in situ*. Wherever there were larger residues which obscured visualization of the entire placode, the extent of resection was recorded as partial.

Statistical analysis

Reynolds et al. study found a post-operative wound infection rate of 8% (six of 75 children) among children undergoing myelomeningocele surgery.[10] Expecting similar results, we estimated a required sample size of 113 for our study, based on a 95% confidence level and a 5% margin of error, and included this sample size. However, we included only 40 patients in our study.

All statistical analyses were performed using the IBM SPSS 20. Categorical data were presented as frequency or proportion and analyzed by Chi-square. Statistical significance was set at $P<0.05$.

RESULTS

Table 1 presents the characteristics of the 40 patients who underwent LMMC surgery. Most patients (90%) were above

Table 1: Patient characteristics

Variables	n (%)
Age (years)	
0–3	22 (55)
3–6	14 (35)
6–18	3
>18	1
Male sex	23 (57.5)
Folic acid supplementation	13 (32.5)
Blood group	
A Negative	2 (0.05)
A Positive	15 (0.37)
AB Positive	3 (0.07)
B Positive	8 (0.2)
O Negative	2 (0.5)
O Positive	10 (25)
Symptoms	
Asymptomatic	7 (17)
Symptomatic	33 (83)
Anatomical level	
Cervical	2 (5)
Sacral	9 (22.5)
Lumbar	4 (10)
Lumbosacral	19 (47.5)
Thoracolumbar	6 (15)
Type of Lipoma	
Transitional	16 (40)
Dorsal	10 (25)
Caudal	6 (15)
Chaotic	5 (12.5)
Terminal	3 (7.5)
Neurological deficit	
Motor-Lower limb weakness	26 (65)
Sensory-foot numbness	17 (42.5)
Loss of reflexes	11 (27.5)
Bladder dysfunction	8 (20)
Bowel dysfunction	3 (7.5)
Sexual dysfunction	1 (2.5)
Radiological finding	
Chiari malformations	3 (8.6)
Syringomyelia (Syrinx)	11 (31.4)
Sacral agenesis	6 (17.1)
Dandy–Walker malformation	1 (2.9)
Tethered cord	27 (67.5)
Skin condition	
Type	
Discoloration	5 (12.5)
Scar	3 (7.5)
Dimple	9 (22.5)
Normal	23 (57.5)
Surface	
Discharging sinus	17 (42.5)
Cerebrospinal fluid leak	7 (17.5)
Neck-Lipoma	
Broad-based	11 (27.5)
Pedunculated	6 (15.0)
Operative factors	
Detethering	40 (100)
Total Lipoma excision	27 (67.5)
Partial Lipoma excision	13 (32.5)
Drainage placement	40 (100)
Iatrogenic injury	2 (5)
Flap used	11 (27.5)
Release of paraspinal muscle	9 (22.5)
Dural closure (Patch/Glue)	11 (27.5)/29 (72.5)
Post-operative details	
Post-operative proning	27 (67.5)
Post of complications	17 (42.5)

6 years, with a male predominance (57.5%). Most mothers did not receive antenatal folic acid supplementation (77.5%). The lumbosacral region was the most common location of LMMC (47.5%), with transitional lipoma being the most frequent type (40%). Most patients (83%) who were symptomatic at the time of admission reported neurological deficits. Skin stigmata were observed in 17 patients, with discharging sinus and CSF leaks observed in 17 and seven patients, respectively.

Detethering was performed in all 40 patients, with total lipoma excision achieved in 67.5%. Dural closure required glue in 72.5% of cases, followed by post-operative proning in 67.5%. Post-operative complications developed in 42.5% of patients, including early complications (CSF leak, infection, and wound dehiscence) and late complications (neurological deficit and pseudumeningocele formation).

Several factors showed significant associations with post-operative complications. These included: Age 3–6 years (P=0.02); A-positive blood type (P=0.01); LMMC located at sacral and lumbosacral (P=0.014); dorsal, chaotic, or terminal type of lipoma (P<0.001); partial lipoma excision with iatrogenic injury (P=0.001), and the use of a patch technique (P=0.001). Post-operative proning decreased the risk of complications (P<0.001).

Of the 17 patients with post-operative complications, 13 experienced early complications: CSF leak (7 patients), infection (4 patients), and wound dehiscence (2 patients). Clinically, 7.5% of patients had developed new neurological deficits.

Table 2 shows factors associated with early post-operative complications. Post-operative CSF leaks were prevalent among children aged 6–18 years (P=0.01), who presented with symptoms at admission (P=0.04) and had lumbosacral defects (P=0.006) with dorsal or caudal lipomas (P<0.001). Patients with tethered cords or syringomyelia (P=0.001) or pre-operative neurological deficits, especially with lower limb weakness (P=0.002), were more prone to CSF leaks. Partial lipoma excision with patch dural closure (P=0.05) was associated with an increased risk of post-operative complications. Post-operative proning decreased the risk of early complications (P=0.001).

Of the 17 patients with post-operative complications, six experienced late complications: Neurological deficit (one patient) and pseudomeningocele (five patients).

Table 3 shows factors associated with late post-operative complications. Children aged 3–6 years (P<0.001) who were presented with symptoms at admission (P=0.018) had a higher risk of late complications. Sacral, cervical, and thoracolumbar LMMCs were significantly associated

Table 2: Risk factors associated with the incidence of early post-operative complications

Variables	CSF leak, (n=7) (%)	Infection, (n=4) (%)	Wound dehiscence, (n=2) (%)	P-value
Age (years)				
0–3	2 (28.6)	3 (75.0)	0 (0.0)	0.01*
3–6	1 (14.3)	1 (25.0)	1 (50.0)	
6–18	3 (42.9)	0 (0.0)	1 (50.0)	
>18	1 (14.3)	0 (0.0)	0 (0.0)	
Male sex	3 (42.9)	1 (25.0)	2 (100.0)	0.07
Folic acid supplementation	1 (14.3)	1 (25.0)	1 (50.0)	0.28
Symptoms				
Symptomatic	7 (100.0)	4 (100.0)	2 (100.0)	0.04
Anatomical level				
Cervical	1 (14.3)	0 (0.0)	0 (0.0)	0.006*
Sacral	0 (0.0)	3 (75.0)	1 (50.0)	
Lumbar	0 (0.0)	0 (0.0)	0 (0.0)	
Lumbosacral	6 (85.7)	1 (25.0)	1 (50.0)	
Thoracolumbar	0 (0.0)	0 (0.0)	0 (0.0)	
Type of Lipoma				
Transitional	0 (0.0)	0 (0.0)	0 (0.0)	<0.001
Dorsal	4 (57.1)	3 (75.0)	0 (0.0)	
Caudal	3 (42.9)	1 (25.0)	0 (0.0)	
Chaotic	0 (0.0)	0 (0.0)	1 (50.0)	
Terminal	0 (0.0)	0 (0.0)	1 (50.0)	
Neurological deficit				
Motor-Lower limb weakness	4 (57.1)	2 (50.0)	0 (0.0)	0.002
Sensory-foot numbness	1 (14.3)	0 (0.0)	1 (50.0)	
Loss of reflexes	2 (28.6)	0 (0.0)	0 (0.0)	
Bladder dysfunction	0 (0.0)	1 (25.0)	1 (50.0)	
Bowel dysfunction	0 (0.0)	1 (25.0)	0 (0.0)	
Sexual dysfunction	0 (0.0)	0 (0.0)	0 (0.0)	
Radiological finding				
Chiari malformations	1 (14.3)	0 (0.0)	0 (0.0)	0.003
Syringomyelia (Syrinx)	2 (28.6)	1 (25.0)	0 (0.0)	
Sacral agenesis	0 (0.0)	0 (0.0)	1 (50.0)	
Dandy–Walker malformation	0 (0.0)	0 (0.0)	0 (0.0)	
Tethered cord	4 (57.1)	2 (50.0)	1 (50.0)	
Operative factors				
Detethering	4 (57.1)	0 (0.0)	0 (0.0)	
Total lipoma excision	0 (0.0)	1 (25.0)	0 (0.0)	
Partial lipoma excision	4 (57.1)	0 (0.0)	0 (0.0)	
Drainage placement	0 (0.0)	2 (50.0)	0 (0.0)	
Iatrogenic injury	0 (0.0)	0 (0.0)	1 (50.0)	
Flap used	2 (28.6%)	1 (25.0))	1 (50.0)	
Release of paraspinal muscle	1 (14.3)	0 (0.0)	0 (0.0)	
Dural closure				
Patch	5 (71.4)	1 (25.0)	2 (100.0)	0.05
Glue	2 (28.6)	3 (75.0)	0 (0.0)	
Post-operative details				
Post-operative proning	2 (28.6)	1 (25.0)	0 (0.0)	0.001

with pseudomeningocele formation, whereas lumbosacral LMMC was associated with a neurological deficit (P=0.001). Transitional lipomas were also significantly associated with late complications (P<0.001). Partial lipoma excision combined with patch dural closure increased the risk of post-operative complications. Post-operative proning decreased the occurrence of late complications (P=0.003).

Over 4 months of follow-up, the frequency of complications (infection or seroma) decreased over time, with no infections reported beyond the 1st month.

DISCUSSION

Identifying risk factors associated with the incidence of post-operative complications helps to plan preventive strategies. This report analyzes risk factors associated with post-operative complications among patients with LMMC. Of 40 patients who underwent the surgery, 17 (42.5%) showed post-operative complications, either in the early stage <3 months (32.5%), or late stage >3 months (12.5%). Symptomatic children aged 3–6 years with A-positive blood group and dorsal-type lipomas at the lumbosacral region were more prone to complications.

Table 3: Risk factors associated with the incidence of late post-operative complications

Variables	Neurological deficit, (n=1) (%)	Pseudomeningocele, (n=5) (%)	P-value
Age (years)			
0–3	0 (0.0)	1 (20.0)	<0.001*
3–6	1 (100.0)	1 (20.0)	
6–18	0 (0.0)	3 (60.0)	
>18	0 (0.0)	0 (0.0)	
Male sex	1 (100.0)	3 (60.0)	0.71
Folic acid supplementation	0 (0.0)	1 (20.0)	0.67
Symptomatic	1 (100.0)	4 (80.0)	0.018*
Anatomical level			
Cervical	0 (0.0)	1 (20.0)	0.001*
Sacral	0 (0.0)	3 (60.0)	
Lumbar	0 (0.0)	0 (0.0)	
Lumbosacral	1 (100.0)	0 (0.0)	
Thoracolumbar	0 (0.0)	1 (20.0)	
Type of Lipoma			
Transitional	1 (100.0)	4 (80.0)	<0.001*
Dorsal	0 (0.0)	0 (0.0)	
Caudal	0 (0.0)	0 (0.0)	
Chaotic	0 (0.0)	0 (0.0)	
Terminal	0 (0.0)	1 (20.0)	
Neurological deficit			
Motor-lower limb weakness	0 (0.0)	2 (40.0)	0.001*
Sensory-foot numbness	0 (0.0)	1 (20.0)	
Loss of reflexes	1 (100.0)	0 (0.0)	
Bladder dysfunction	0 (0.0)	0 (0.0)	
Bowel dysfunction	0 (0.0)	1 (20.0)	
Sexual dysfunction	0 (0.0)	1 (20.0)	
Radiological finding			
Chiari malformations	0 (0.0)	1 (20.0)	0.005*
Syringomyelia (Syrinx)	0 (0.0)	3 (60.0)	
Sacral agenesis	1 (100.0)	0 (0.0)	
Dandy–Walker malformation	0 (0.0)	0 (0.0)	
Tethered cord	1 (100.0)	2 (40.0)	
Operative factors			
Detethering	1 (100.0)	0 (0.0)	
Total lipoma excision	0 (0.0)	1 (20.0)	
Partial lipoma excision	1 (100.0)	3 (60.0)	
Drainage placement	0 (0.0)	1 (20.0)	
Iatrogenic injury	0 (0.0)	1 (20.0)	
Flap used	0 (0.0)	2 (40.0)	
Release of paraspinal muscle	1 (100.0)	0 (0.0)	
Dural closure			
Patch	1 (100.0)	4 (80.0)	
Glue	0 (0.0)	1 (20.0)	
Post-operative proning	0 (0.0)	1 (20.0)	0.003*

Partial lipoma excision using the patch technique also increased the risk.

Our findings align with previous evidence from a comprehensive review of five studies (403 patients) showing significantly higher neurological deterioration in conservatively managed patients (25.81%, 32/124 patients) compared to those undergoing prophylactic surgery (10.75%, 30/279 patients; P≤0.05). This outcome shows that early identification and appropriate surgery better protect against neurological decline.[11]

CSF leak was the most common post-operative early complication (54%). In our study, symptomatic conditions, lumbosacral or cervical lesions, dorsal lipoma, pre-operative neurological deficit (especially motor weakness and loss of reflexes), specific radiological findings (Chiari malformations, tethered cord, and syringomyelia), partial lipoma excision increased the incidence of CSF leak. Glue skin closure and post-operative proning reduced the chances of CSF leak and other complications. Baldia and Rajshekhar reported CSF leak (4.5%) and wound dehiscence (4.8%) following tethered cord surgery.[12] The study also found proning reduced early complications, and primary closure without flaps yielded better outcomes. In another study, percutaneous drains resolved recurrent CSF leaks and pseudomeningoceles within 2–3 days without any

recurrence or complications later.[13] Sidram et al. study found post-operative CSF leaks (10.3%), infections (10.3%), and paraparesis (10%) in spinal dysraphism/LMMC patients, and among those two of them died.[14] Patil et al. reported that among asymptomatic infants undergoing LMMC repair, post-operative infection occurred in one patient and seroma in eight. Their study showed improved lower limb motor function in three patients (17.6%), whereas three experienced decreased lower limb power.[6]

Pseudomeningocele was the most common post-operative late complication (83.3%). In our study, age (3–6 years), symptomatic condition, sacral, cervical, and thoracolumbar lesions, transitional lipoma, and partial lipoma excision increased the incidence of pseudomeningocele formation. Glue skin closure and post-operative proning reduced the chances of late complications. Vora et al. studied 109 children undergoing LMMC repair, finding post-operative neurological deterioration in 6 (3 recovered). The study found that early surgery correlated with improved recovery in 31 of 109 children who developed symptoms after age one. The average time to late deterioration requiring retethering was 51.3 months, with initial syrinx on MRI and partial lipoma resection identified as risk factors.[15] Patil et al. analyzed immediate and long-term outcomes of LMMC repair in 17 asymptomatic infants and found two patients developed altered sensation and lower limb weakness 2.5–3 years post-operatively, requiring repeat detethering. While two had fecal incontinence, one had constipation, and two (5.8% each) had post-operative urinary incontinence (one lumbosacral and one sacral LMMC).[6]

This research found that by the end of the follow-up, clinically, 7.5% of patients had developed new neurological deficits. In a study by Kang et al., patients who underwent LMMC surgery showed improvements in the pre-operative deficits after surgery in 29 (39%), remained stable in 28 (37%), changed slightly in 13 (17%), and worsened in 5 (7%), the cord with neurological deterioration occurred in 4 (5.3%) of the 75 patients.[7] Iqbal et al. conducted a similar study in 50 infants, where complete excision of lipoma was achieved in 96% of patients. Of 15, surgical site infection was seen in 9/40 patients. The 6th month outcome analysis revealed significant neurological improvement in 75% of the infants post-surgery.[16]

Limitations of the study

While the study is time-bound, the sample size is small and lacks the scope of neuromonitoring. Since the follow-up period was limited, the other common late complications were not assessed, such as retethering. The results of the study might help in addressing the gaps related to prognostic factors in post-operative LMCC patients.

CONCLUSION

The study results demonstrate that the surgical approach for LMMC is safe and effective when performed with the appropriate technique. We recommend detethering and total excision of lipoma with dural closure using glue and post-operative proning to reduce the risk of post-operative complications in LMMC patients. These findings emphasize the importance of timely intervention and inform surgical strategies to optimize outcomes in LMMC management. We also recommend counseling and encouraging parents regarding early repair in diagnosed cases and identifying early signs of post-operative complications.

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Authors' Contributions:
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