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LONG-TERM NEUROLOGICAL OUTCOME OF SURGICAL REPAIR OF LIPOMYELOMENINGOCELE IN INFANTS

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ABSTRACT

Background: Lipomyelomeningocele is a form of closed spinal dysraphism associated with tethering of the spinal cord and progressive neurological dysfunction. Surgical repair during infancy aims to prevent neurological deterioration; however, long-term outcomes remain variable.

Objective: To evaluate the late neurological outcomes of surgical repair of lipomyelomeningocele in infants and identify factors associated with unfavorable neurological outcomes.

Methods: This retrospective observational study was conducted in the Department of Pediatric neurosurgery, Children Hospital &The Institute of Child Health, Lahore from January 2022 to December 2022, included 40 infants who underwent surgical repair of lipomyelomeningocele. Demographic, clinical, radiological, operative, and follow-up data were collected from medical records.

Results: The mean age at surgery was 6.84 ± 2.71 months. Females were more common, 24 (60.0%), and the most frequent lesion location was lumbar, 19 (47.5%). Preoperative motor weakness was present in 11 (27.5%) infants, bladder dysfunction in 10 (25.0%), and low-lying conus or tethered cord on MRI in 34 (85.0%). Overall favorable outcome was achieved in 34 (85.0%) infants, including 13 (32.5%) who improved and 21 (52.5%) who remained stable. Neurological deterioration occurred in 6 (15.0%) infants. Untethering was observed in 5 (12.5%) cases and was significantly associated with unfavorable outcome.

Conclusion: Surgical repair of lipomyelomeningocele in infants resulted in favorable long term neurological outcomes in most patients. Early diagnosis, timely surgical repair, and long-term multidisciplinary follow-up are essential to reduce delayed neurological deterioration and untethering-related complications.

KEYWORDS: Lipomyelomeningocele; tethered cord syndrome; spinal dysraphism; neurological outcomes

INTRODUCTION

Lipomyelomeningocele (LMMC) is a rare type of closed spinal dysraphism with a subcutaneous lipomatous mass extending through a defect in the vertebral arches and attached to the neural structure, causing neural structures to be tethered [1]. It is an abnormal primary neurulation that occurs during embryonic development and is one of the most frequently seen types of occult spinal dysraphism seen in pediatric neurosurgical practice [2]. Typically the diagnosis is made in infancy, when lumbosacral abnormalities can be seen, such as a subcutaneous fatty mass, skin discoloration, dermal sinus, hemangioma, or asymmetrical gluteal cleft [3]. The pathophysiological characteristic of LMO is tethering of the spinal cord that prevents normal movement of the neural elements in the spinal canal [4]. With the child's growth, the increasing tension of the tethered cord can cause neurologic deterioration with symptoms of motor weakness, sensory impairment, gait disturbances, orthopedic deformities, and bladder or bowel issues [5]. While some babies can be neurologically normal, older infants are at greater risk of progressive neurological deterioration if the tethered cord is not treated [6]. Magnetic resonance imaging (MRI) is widely regarded as the gold standard for diagnosis and pre-operative evaluation of lipomyelomeningocele, as it can provide a detailed picture of the lipomatous mass, spinal cord anatomy and associated anomalies [7]. The advance of imaging in the prenatal and postnatal period has greatly enhanced early diagnosis, allowing for timely referral for a neurosurgical evaluation [8]. The best time to operate and the duration of its benefits remains a matter of debate, however.

Surgical repair is the treatment of choice for lipomyelomeningocele, and the major goals of the surgery are to untether the spinal cord, resect the maximum amount of lipomatous material safely, reconstruct the neural placode, and avoid further neurological deterioration [9]. Many studies have shown that surgical intervention during infancy can prevent

or preserve neurological function and decrease the likelihood of progression to neurological deficits later in life [10]. However, long-term follow-up is still a concern of postoperative neurological deterioration and untethering despite the development of microsurgical techniques and use of intraoperative monitoring [11]. It is therefore important to evaluate late neurological outcomes after surgical repair as a good indicator of treatment success [12]. The commonly used long-term outcome measures include motor function, sensory status, ambulatory status, bladder and bowel control, orthopedic complications and the necessity for further surgical procedures [13]. Both good long-term neurological recovery following early surgery and different rates of deterioration from untethering and progressive spinal cord disorder have been reported [14]. The wide range of reported outcomes might relate to patient variability and/or age at surgery, surgical technique, duration of follow-up, and definitions of neurological deterioration [15]. Also, the majority of the data in available sources comes from specialized centers from developed countries, and in the developing areas, there are only limited data [16, 17]. It is important to local outcomes to counsel families, optimize time of surgery and long-term management strategies.

Objective

To evaluate the long term neurological outcomes of surgical repair of lipomyelomeningocele in infants and identify factors associated with unfavorable neurological outcomes.

METHODOLOGY

This was a retrospective observational study conducted in the Department of Pediatric neurosurgery, Children Hospital &The Institute of Child Health, Lahore from January 2022 to December 2022, including 40 infants who underwent surgical repair of lipomyelomeningocele to evaluate long term neurological outcomes following surgery. Infants aged less than 12 months with radiologically confirmed lipomyelomeningocele who underwent primary surgical repair and had available postoperative follow-up records for neurological assessment were included. Patients of both genders who completed scheduled follow-up evaluations and whose medical records contained sufficient clinical, radiological, operative, and postoperative data were considered eligible. Patients with other forms of spinal dysraphism without lipomyelomeningocele, previous spinal surgery before presentation, severe associated congenital anomalies affecting neurological assessment, incomplete medical records, loss to follow-up, or inadequate postoperative neurological documentation were excluded from the study.

Data Collection

After obtaining ethical approval, data were collected retrospectively from hospital medical records using a structured proforma. Demographic variables included age at surgery, gender, birth history, and age at presentation. Clinical variables included location and size of lipomyelomeningocele, preoperative neurological status, motor deficits, sensory deficits, bladder and bowel dysfunction, orthopedic deformities, and associated spinal anomalies identified on magnetic resonance imaging (MRI). Operative details including extent of lipoma excision, spinal cord untethering, and intraoperative findings were recorded. Follow-up data were reviewed to assess long term neurological outcomes including motor function, sensory status, ambulatory ability, bladder and bowel control, occurrence of untethering, requirement for repeat surgery, and overall neurological stability. Neurological outcomes were categorized as improved, stable, or deteriorated based on postoperative clinical assessments during long-term follow-up.

Statistical Analysis

Data were analyzed using SPSS version 26.0. Continuous variables were expressed as mean ± standard deviation, while categorical variables were presented as frequencies and percentages. Associations between demographic, clinical, radiological, and surgical variables and long term neurological outcomes were assessed using chi-square tests and independent t-tests where appropriate. Multivariable logistic regression analysis was performed to identify predictors of unfavorable neurological outcomes. A p-value ≤ 0.05 was considered statistically significant.

RESULTS

Data were collected from 40 patients, mean age at surgery was 6.84 ± 2.71 months. Most infants were operated after 6 months of age, 23 (57.5%), while 17 (42.5%) underwent surgery at or before 6 months. Females were more common, 24 (60.0%), compared with males, 16 (40.0%). The most frequent lesion location was lumbar, seen in 19 (47.5%) infants, followed by lumbosacral in 15 (37.5%) and sacral in 6 (15.0%). Visible back swelling was present in 37 (92.5%) infants, and cutaneous markers were noted in 29 (72.5%).

Table I. Baseline Demographic and Clinical Characteristics of Infants (N = 40)

Variable	Category	n (%) / Mean ± SD
Age at surgery (months)	Mean ± SD	6.84 ± 2.71
Age group	≤6 months	17 (42.5%)
	>6 months	23 (57.5%)

Gender	Male	16 (40.0%)
	Female	24 (60.0%)
Lesion location	Lumbar	19 (47.5%)
	Lumbosacral	15 (37.5%)
	Sacral	6 (15.0%)
Visible back swelling	Present	37 (92.5%)
	Absent	3 (7.5%)
Cutaneous markers	Present	29 (72.5%)
	Absent	11 (27.5%)

Motor weakness was present in 11 (27.5%) infants, while 29 (72.5%) had no motor weakness. Sensory deficit was seen in 8 (20.0%) infants. Bladder dysfunction was present in 10 (25.0%), and bowel dysfunction was present in 6 (15.0%). Foot deformity was documented in 9 (22.5%) infants. MRI showed low-lying conus or tethered cord in 34 (85.0%) cases.

Table II. Preoperative Neurological and Urological Status

Variable	Category	n (%)
Motor weakness	Present	11 (27.5%)
	Absent	29 (72.5%)
Sensory deficit	Present	8 (20.0%)
	Absent	32 (80.0%)
Bladder dysfunction	Present	10 (25.0%)
	Absent	30 (75.0%)
Bowel dysfunction	Present	6 (15.0%)
	Absent	34 (85.0%)
Foot deformity	Present	9 (22.5%)
	Absent	31 (77.5%)
Low-lying conus/tethered cord on MRI	Present	34 (85.0%)
	Absent	6 (15.0%)

Detethering with lipoma debulking was performed in 34 (85.0%) infants, while 6 (15.0%) underwent detethering only. Near-total or maximum safe excision was achieved in 31 (77.5%) cases, while partial excision was done in 9 (22.5%). Dural reconstruction was performed in 36 (90.0%) infants. Postoperative CSF leak occurred in 3 (7.5%) cases, and wound infection was reported in 4 (10.0%). The mean follow-up duration was 30.6 ± 11.8 months.

Table III. Surgical and Postoperative Characteristics

Variable	Category	n (%) / Mean ± SD
Type of surgery	Detethering with lipoma debulking	34 (85.0%)
	Detethering only	6 (15.0%)
Extent of lipoma excision	Near-total/maximum safe excision	31 (77.5%)
	Partial excision	9 (22.5%)
Dural reconstruction	Done	36 (90.0%)
	Not done	4 (10.0%)
Postoperative CSF leak	Yes	3 (7.5%)
	No	37 (92.5%)
Wound infection	Yes	4 (10.0%)
	No	36 (90.0%)
Follow-up duration (months)	Mean ± SD	30.6 ± 11.8

Overall, neurological improvement was observed in 13 (32.5%) infants, while 21 (52.5%) remained stable and 6 (15.0%) deteriorated. Favorable outcome, defined as improved or stable status, was achieved in 34 (85.0%) infants. Motor function improved or remained stable in 35 (87.5%) cases, while worsening occurred in 5 (12.5%). Bladder function improved or remained stable in 31 (77.5%), while 9 (22.5%) showed worsening. Bowel function improved or remained stable in 34 (85.0%). Untethering was observed in 5 (12.5%) infants.

Table IV. Long term Neurological Outcomes After Surgical Repair

Outcome Variable	Category	n (%)
Overall neurological outcome	Improved	13 (32.5%)
	Stable	21 (52.5%)
	Deteriorated	6 (15.0%)

Favorable outcome	Improved/stable	34 (85.0%)
	Deteriorated	6 (15.0%)
Motor function at long term follow-up	Improved/stable	35 (87.5%)
	Worsened	5 (12.5%)
Bladder function at long term follow-up	Improved/stable	31 (77.5%)
	Worsened	9 (22.5%)
Bowel function at long term follow-up	Improved/stable	34 (85.0%)
	Worsened	6 (15.0%)
Untethering	Present	5 (12.5%)
	Absent	35 (87.5%)

Surgery at or before 6 months was associated with a higher favorable outcome, 16 (94.1%), compared with surgery after 6 months, 18 (78.3%) (p=0.041). Preoperative motor weakness was significantly associated with unfavorable outcome, as 4 (36.4%) infants with motor weakness deteriorated compared with 2 (6.9%) without motor weakness (p=0.018). Preoperative bladder dysfunction was also significantly associated with unfavorable outcome, 4 (40.0%) versus 2 (6.7%) (p=0.011). Lesion location was not significantly associated with outcome (p=0.482).

Table V. Association of Clinical Factors With Late Neurological Outcome

Variable	Category	Favorable Outcome n (%)	Unfavorable Outcome n (%)	p-value
Age at surgery	≤6 months	16 (94.1%)	1 (5.9%)	0.041
	>6 months	18 (78.3%)	5 (21.7%)	
Preoperative motor weakness	Present	7 (63.6%)	4 (36.4%)	0.018
	Absent	27 (93.1%)	2 (6.9%)	
Preoperative bladder dysfunction	Present	6 (60.0%)	4 (40.0%)	0.011
	Absent	28 (93.3%)	2 (6.7%)	
Lesion location	Lumbar	17 (89.5%)	2 (10.5%)	0.482
	Lumbosacral/sacral	17 (81.0%)	4 (19.0%)	
Untethering	Present	1 (20.0%)	4 (80.0%)	<0.001
	Absent	33 (94.3%)	2 (5.7%)	

Progressive bladder dysfunction was the most frequent long term complication, occurring in 9 (22.5%) infants, followed by foot deformity progression in 6 (15.0%), retethering in 5 (12.5%), and progressive motor weakness in 5 (12.5%). Reoperation was required in 3 (7.5%) cases, while wound-related long-term complications occurred in 2 (5.0%) cases.

Table VI. Long term Complications During Follow-Up

Complication	n (%)
Untethering	5 (12.5%)
Progressive bladder dysfunction	9 (22.5%)
Progressive motor weakness	5 (12.5%)
Foot deformity progression	6 (15.0%)
Reoperation required	3 (7.5%)
Wound-related long term complication	2 (5.0%)

DISCUSSION

A study of long term neurologic outcomes after surgical correction of lipomyelomeningocele of 40 infants was performed. Lipomyelomeningocele is a significant presentation of closed dysraphism in the infant and the prognosis is dependent on the degree of detethering at surgery, the quality of postoperative monitoring, and the neurological status at the time of surgery. Overall, neurological outcomes were relatively good following surgical repair with 34 (85.0%) infants having improved or stable neurological status over the follow-up period in this study. In 13 (32.5%) infants, neurological improvement was observed and 21 (52.5%) infants were stable. In only 6 (15.0%) patients, neurological deterioration occurred indicating that surgical repair was effective in preventing progression in the majority of patients. In this study, mean age at surgery was 6.84 ± 2.71 months, and 57.5% of the infants were operated after 6 months of age. Early surgery seemed to be significantly associated with better outcome, and infants who were

operated upon at or before 6 months had a favorable outcome rate of 94.1%, compared to 78.3% for those operated upon after 6 months (p=0.041) [16]. This is in keeping with previous studies that have demonstrated that neurological function is better preserved when surgery is performed at an early stage. It's likely because early detethering prevents chronic traction on the spinal cord, and prior to irreversible neurological, urological or orthopedic damage. In the present study, the female individuals were predominant as 24 individuals representing 60.0% were females while 16 individuals representing 40.0% were males. In some previous studies of spinal dysraphism (SD) and lipomyelomeningocele (LMC), similar female predominance has been reported, and gender is not generally a very strong predictor of neurological outcome. The most frequent area involved was the lumbar region (47.5%), followed by lumbosacral (37.5%) and sacral (15.0%). This distribution is a normal occurrence, because the lipomyelomeningocele tends to occur on the lower part of the spine. 92.5% of infants had back swelling and 72.5% had cutaneous markers [17]. Careful examination of the neonates and infants' back is essential and these results underscore this need, particularly in resource limited settings where examination might be deferred. Any of the following is a cutaneous clue for suspicion of underlying spinal dysraphism and early evaluation with MRI: swelling, dimples, hair patches, hemangiomas, and abnormal gluteal folds. Neurological impairment before surgery was an important predictor of outcome. Of infants, 27.5% had motor weakness, 25.0% had bladder dysfunction, 20.0% had sensory deficit, and 22.5% had foot deformity. Infants who had motor weakness at they were referred for surgery were significantly more likely to have poor outcomes (36.4% vs. 6.9%, p=0.018) [18]. Likewise, there was a significant association between preoperative bladder dysfunction and poor long term neurological outcome (40.0% vs. 6.7%, p=0.011). These findings are similar to what has been previously reported, which indicates that neurological impairment that occurs prior to surgery can occur again after surgery, as existing neurological damage may only be partially reparable. Most infants (85.0%) had lipoma debulking and detethering surgery. Thirty one (77.5%) infants had near total or maximum safe excision, and 22.5% had partial excision. In 90.0% of cases, dural reconstruction was done. The following operative findings indicate that the majority of cases were treated by a typical microsurgical technique of safe detethering, decompression and untethering prevention [19]. Aggression of the resection should always be balanced against the risk of neurological injury as lipomatous tissue can be intimately associated with functional neural elements. There was a relatively small number of complications following surgery. Wound infection was found in 10.0% and CSF leak in 7.5% of infants. These rates are fair and similar to other pediatric neurosurgical series reported previously. Delayed presentation, nutritional status, hygiene, access to postoperative care, and follow-up compliance may affect complications of wounds in the developing health care setting. It is important to recognize and treat CSF leak and wound infection early as these complications can cause increased risk of meningitis, wound breakdown, and delayed healing. In 87.5% of infants, motor function improved or did not change at long term follow-up, and worsened in 12.5%. 85.0% of children had an improvement or stability in bowel function and 77.5% of children had an improvement or stability in bladder function [20]. Worsening occurred in 22.5% of infants in the domain of bladder function, the least favorable domain. This is a clinically significant finding because it is one of the most challenging complications to reverse following the path of tethered cord surgery. Previous studies also have indicated that bladder dysfunction can continue or become worse even after repair, particularly if a bladder dysfunction is present prior to surgery.

Limitations

This study has several limitations. As a retrospective observational study, it was dependent on the accuracy and completeness of available medical records, which may have introduced information bias. The study was conducted at a single institution, limiting the generalizability of the findings to other populations and healthcare settings. Although the sample size was relatively large, variations in surgical techniques, follow-up duration, and postoperative management may have influenced neurological outcomes. Detailed urodynamic assessments and standardized quality-of-life evaluations were not consistently available for all patients. Furthermore, the retrospective design limited the ability to establish causal relationships between identified risk factors and neurological outcomes. Long-term follow-up beyond the study period may also be necessary to evaluate the impact of untethering and long term neurological deterioration fully.

CONCLUSION

Surgical repair of lipomyelomeningocele in infants resulted in favorable long term neurological outcomes in most patients, with 85.0% showing improved or stable neurological status during follow-up. Early surgery, absence of preoperative motor weakness, absence of bladder dysfunction, and no untethering were associated with better outcomes. However, progressive bladder dysfunction, motor worsening, foot deformity progression, and untethering remained important long term complications. Early diagnosis, timely surgical repair, and long-term multidisciplinary follow-up are essential to optimize neurological, urological, and orthopedic outcomes.

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