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Case Report

A Case Report on Sacral Lipomyelomeningocele

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ABSTRACT

Lipomyelomeningocele (LMMC) is a neural tube defect characterized by the presence of a spinal lipoma that incorporates neural elements, leading to various symptoms like skin lesions, musculoskeletal issues, sensory deficits, and urinary dysfunction. It typically occurs in the lumbosacral region with a prevalence of 0.3-2.5 per 10,000 live births. A 1.4-year-old boy presented with a gradually enlarging swelling on the left lower back since birth, without any skin changes or discharge. Developmentally, he showed delays in mobility and increased urinary frequency compared to his sibling. Neurological examination revealed limb length discrepancy and absent anal wink reflex, suggesting potential tethered cord syndrome. Imaging confirmed a large subcutaneous lipoma with intradural extension at the S1-2 level. The patient underwent L5 to S3 laminectomy and subtotal excision of the lipoma. Postoperative management included dexamethasone and antibiotics. At discharge, he was afebrile, ambulant, and voiding normally, with stable neurological status. LMMC is a significant form of closed spinal dysraphism, often presenting with skin and musculoskeletal signs. Early surgical intervention is crucial for alleviating neurological deficits and preventing complications. This case underscores the importance of awareness and timely treatment in managing spinal dysraphism to optimize long-term outcomes.

Keywords: Lipomyelomeningocele, Case report, Neural tube defects, 1.4-year-old boy.

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INTRODUCTION

A neural tube abnormality called Lipomyelomeningocele (LMMC) occurs when a spinal lipoma incorporates neural factors. A variety of symptoms can accompany LMMC, including minor skin lesions, musculoskeletal disorders, sensory impairments, and urinary dysfunction.[1] The unfused ventral neural ectoderm enters the periaxial mesoderm intact, and the neural ectoderm splits from the cutaneous ectoderm. After that, the mesoderm develops into fatty tissue, preventing the posterior part of the spine and the neural canal from fusing together.[2] The prevalence ranges from 0.3 to 0.6 per 10,000 live births.[3] LMMCs are lipomas that extend dorsally through a spina bifida and are firmly affixed to the dorsal surface of a neural placode, where they meet the subcutaneous fat. In the extradural area, the lipoma is located directly outside of the connective tissue. A 2.5 LMMC rate per 10,000 newborns has been estimated. The illness usually affects females before the age of six months, while it can also infrequently linger undiagnosed until childhood. The cord is typically tethered at the level by LMMC, which typically occur in the lumbosacral area. Lipomas that are subcutaneously positioned often behave benignly and only cause cosmetic issues; but, depending on where they are, they may occasionally produce pressure complaints.[4-6] Here we presented a case with a lump over the back region since birth.

CASE DESCRIPTION

1 year and 4-month-old boy was brought by his parents with complaints of a progressively enlarging swelling over the left side of the lower back region that had been present since birth. There is no history of discharge from the swelling or any excoriation of skin or other skin changes. His mother gave a history of being able to stand alone and uses two finger pincer grasps. There was a history of increased frequency of micturition compared with her first son. He has been noted to be unable to take more than a few steps with support for his age as compared to the older sibling, who was able to walk without support at the same age. There was no past history of seizure, loss of consciousness, extensor posturing, vomiting, or motor or sensory deficit. He is the second-born child of a non-consanguineous marriage, born via full-term Caesarean delivery. His sibling has no similar history of neural tube defects. He had episodes of apnoea preceded by crying with unresponsiveness six to seven months ago.

CLINICAL DIAGNOSIS AND ON EXAMINATION

His length is 76 cm, weight is 9 kg, and BMI is 15.6 kg/m2. His pulse rate was 120/min and blood pressure was 90/54 mm hg. General and systemic examination was normal. Head circumference was 45 cm and midupper arm circumference was 14 cm. The anterior fontanelle is closed. There is nevus over

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the left back; no other neurocutaneous markers. He is able to track objects at 4 feet. He was not cooperative for the fundus examination. Pupils were 2 mm equal and reactive. Extraocular movements were normal in all directions. There was limb length discrepancy with the left lower limb measuring 34 cm and the right lower limb measuring 36 cm. Tone was normal in the lower limbs with power of at least grade 4 over both hips, knees, and ankles. Up to S1 dermatome, withdrawal was present for sensory stimulus. His anal tone was lax, and the anal wink reflex was absent bilaterally. Perianal sensations could not be reliably commented upon. Deep tendon reflexes were normal in lower limbs. There were no signs of meningeal irritation. There was a 10 cmx10 cm large ovoid-shaped soft fluctuant swelling at the left lower lumbar region. The skin overlying the swelling was normal with nevus and without any evidence of erythema, discharge, or warmth. On the lumbar spine, X-ray AP and lateral view MRI spine with whole screening report show there is a large subcutaneous lipoma in the sacral region. More to the left of midline communicating with the intradural compartment through a large defect in the fascia and posterior elements of sacrum to extend into the low lying conus at S1-2 level, more on the left side. The conus is pushed towards the right due to the lipoma. There is a small synx above the level of the lipoma-cord interface. CT brain plain shows no hydrocephalus.

LABORATORY INVESTIGATIONS [PRE-OPERATIVE]

Biopsy: gliotic nervous tissue, lobules of mature adipose tissue, scanty meningothelial nodules, and few nerve fascicles. CSP culture (collected from thecal sac during re-exploration) No growth.

OPERATION DONE AND FINDINGS

L5 to S3 laminectomy and subtotal excision of lipoma and repair of dural and fascial defects in prone position with MEP and BCR monitoring done on 06/06/2022.

FINDINGS

There was a large subcutaneous lipoma more to the left of midline in the sacral region that was extending through a defect in the fascia and dura into the intradural compartment. There was no thecal sac below the level of the intradural lipoma. The lipoma extended into the low-lying conus, which was seen at S1-2 level. The lipoma was asymmetric, transitional type, and more towards the left half of the cord. In addition to the lipoma along the interface with the placode, there was a subpial lipoma covered by intact arachnoid superiorly for two vertebral levels, and roots were seen to exit through this component of the lipoma. This part of the lipoma was left behind. Deeper into the intramedullary fat was gliotic nervous tissue, which was firm and greyish. MEPs were recorded from EDB and AH muscles bilaterally at the beginning of the procedure. At the end of the procedure, MEPs were obtained from all lower limb muscles bilaterally along with sphincter muscles. BCR was never obtained at any time during the procedure.

Re-exploration, primary closure of dural defect, and closure of fascial defect with PTFE graft in prone position.

On Pre-operative laboratory investigation, Serum Creatinine, PCV, MCV, RDW were decreased than normal. On post-operative laboratory investigation, PCV is decreased (Table 1).

OPERATIVE FINDINGS

There was a large collection of CSF beneath the subcutaneous plane. There was a defect in the lower part of the fascia where part of the fat used previously for closure appeared to have undergone necrosis. There was a defect in the dura at the lowermost part of the previous closure from where CSF was seen to egress. No CSF leak was observed from the remaining part of the dura. Once the dural defect was closed, no CSF was seen to egress on Valsalva manouevre.

TREATMENT

Pre-Operative Medications

Tab. Junior lansoprazole 7.5 mg once a day.

Tab. Dexamethasone 0.5 mg 4 times daily on the preoperative day.

Syp. Paracetamol 70 mg as and when needed for pain or fever.

Inj. Ceftriaxone 500 mg intravenously at induction of Anaesthesia.

Post-Operative Medications

Inj. Dexamethasone 0.5 mg 4 doses intravenously on the first postoperative day.

Inj. Pantoprazole 10 mg 1 dose intravenously on the first postoperative day.

Inj. Paracetamol 90 mg 4 doses intravenously on the first postoperative day.

Tab. Junior lansoprazole 7.5 mg once a day.

Tab. Dexamethasone 0.5 mg 4 times daily on the preoperative day.

Syp. Paracetamol 90 mg as and when needed for pain or fever.

Inj. Ceftriaxone 500 mg once daily for five days intravenously after second surgery.

Condition at Discharge

A child was afebrile, ambulant, voiding normally and tolerating a normal diet. The rest of the neurological examination was the same as at admission. The surgical wound was healing well.

Table 1: Laboratory investigation.[7]

Sl. No.	Parameters	Patient value	Reference value
1.	Sr. Creatinine	0.39 mg%	0.5-1.4 mg%
2.	PCV	36.7%	45-65%
3.	MCV	78.1 fl	80-100 fl
4.	RDW	13.3%	15-20%
	Postoperative Investigation		
1.	PCV	31.4%	45-65%

Discharge Medications

1. Syp. Paracetamol 90 mg as and when needed for pain.

DISCUSSION

A subcutaneous adipose tumour covering the intergluteal cleft is the main symptom of lipomeningomyelocele, a type of closed spinal dysraphism. There is uncertainty about the lipomeningomyelocele predisposing factors. The spinal cord is dorsally divided with a lipoma connected to it. Tethered cord syndrome is a possible manifestation of lipoma tethering to the superficial dermal factors.[4] A hair tuft, a dimple, swelling, a subcutaneous lipoma, or dermal sinus can all be signs of spinal dysraphism on the skin.[5] Infrequently, they can include skin appendages or hypertrichosis. Asymmetry of the foot or leg, aberrant Gyration, unilateral or bilateral foot abnormalities, and scoliosis are examples of musculoskeletal findings. Incontinence, frequency, urgency, and urinary tract infections are examples of urological findings. Tethered cord syndrome causes neurological symptoms such as weakness, aberrant gait patterns, sensory difficulties, and back or leg pain that gets worse with movement.[1] In these children, the tethered cord resulting from the LMMC or a disfigurement during embryogenesis causes poor innervation of the urinary system, which in turn causes secondary urinary problems. There are three possible causes of urinary dysfunction: dyssynergy of the detrusor sphincter, paresis, and malfunction of the external sphincter. Urine dysfunction can manifest as symptoms including urgency, repeated UTIs, incontinence, and, in more severe situations, damage to the upper urinary tract due to hydronephrosis or pyelonephritis. Alterations in micturition pattern are often the first signs of a neurogenic bladder. Testing for urological dysfunction using urodynamics may be helpful in determining the extent of performing. Likewise, the primary indication of neurological impairment in these childrens can be abnormalities related to bladder function.[6] The severity determines how the lesion should be managed clinically. Patients who don't exhibit any symptoms can be managed conservatively with routine monitoring. Patients who exhibit symptoms such as sensory impairments or a progressive deterioration of musculoskeletal problems are scheduled for surgical surgery. Those connected to multisystem disorders make management more difficult.[1] Neurological impairments can be avoided in asymptomatic patients by undergoing early surgery.[7] Surgery is performed with the intention of removing the fibroadipose mass, relieving the spinal cord's tethering effect, protecting neural structures, and preventing the spinal cord from retethering. While each variety of lipoma requires a distinct level of surgical difficulty, the fundamental surgical techniques are the same.[8]

CONCLUSION

In this case report, we presented a case with sacral lipomyelomeningocele, a rare but significant spinal dysraphism. The clinical presentation, including neurological deficits and imaging findings, underlined the significance of early diagnosis and surgical intervention in managing such cases. Our case's outcome highlights the critical part of multidisciplinary care in optimizing functional and neurological issues. This case underscores the need for heightened awareness among clinicians regarding the different manifestations of spinal dysraphism and the significance of timely intervention to help or alleviate long-term complications.

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CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

ABBREVIATIONS

LMMC: Lipomyelomeningocele; **MEP:** Motor evoked potential; **BCR:** Bulbocavernosus reflex; **EDB:** Extensor digitorum brevis; **AH:** Abductor halluces; **PTEF:** Polytetrafluoroethylene; **CSF:** Cerebrospinal fluid.

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