The spectrum of musculoskeletal problems in lipomyelomeningocele

Lee S. Segal · Wojciech Czoch · William L. Hennrikus ·
M. Wade Shrader · Paul M. Kanev

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Abstract

Purpose Patients with lipomyelomeningocele (LMMC) represent a unique population within the spectrum of spinal dysraphism. The natural history of LMMC remains poorly defined. The description and prevalence of the presenting orthopaedic clinical signs and symptoms for LMMC have been infrequent and often documented only in general terms. The goal of this study is to define the patterns and prevalence of presenting clinical musculoskeletal signs and symptoms in LMMC patients.

Methods This study was a retrospective review of charts of all patients identified as having LMMC in our spina bifida clinic. Patient charts with incomplete data or diagnoses other than LMMC were excluded from the analysis. Data collected included age at initial tethered cord release (TCR); repeat TCR; limb length discrepancy; foot deformities; asymmetry of motor and sensory deficits; presence of scoliosis; orthotic needs; assistive devices; functional status.

Results We identified 32 patients with LMMC (21 female and 11 male patients). The majority of patients had their primary TCR by ≤ 1 year of age (59 %), with 22 and 19 % having primary TCR at ages 1–15 and >15 years,

respectively. Fifteen patients had at least one repeat TCR, with ten of these having more than one repeat TCR. A significant relationship was noted between low back/radicular pain and repeat TCR ($p < 0.001$). Ten patients (31 %) had a limb length discrepancy of >2.5 cm, and 53 % of patients had asymmetric involvement. Nine patients (28 %) had scoliosis of whom only one required operative treatment. Fifteen patients had foot deformities. Thirteen patients (41 %) had two or more orthopaedic procedures in addition to other neurologic or urologic procedures.

Conclusion The presenting musculoskeletal clinical signs and symptoms in patients with LMMC are uniquely different in terms of both pattern and frequency compared to myelomeningocele and other forms of spinal dysraphism. We noted a high prevalence of asymmetrical involvement, a high operative burden, and a high rate of repeat symptomatic tethered cord syndrome requiring TCR. As previously noted by others, TCR in LMMC does not prevent long-term functional deterioration. These findings may be important to our colleagues providing counsel to their patients with LMMC and to their families.

Keywords Lipomyelomeningocele · Spinal dysraphism · Spina bifida · Myelomeningocele · Tethered cord

L. S. Segal (✉) · M. Wade Shrader
Division of Pediatric Orthopaedics, Phoenix Children's Hospital,
1919 East Thomas Road, Phoenix, AZ 85016, USA
e-mail: lsegal@phoenixchildrens.com

W. Czoch · W. L. Hennrikus
Department of Orthopaedics and Rehabilitation, Penn State
Hershey Medical Center, The Pennsylvania State University
College of Medicine, Hershey, PA, USA

P. M. Kanev
Division of Pediatric Neurosurgery, Connecticut Children's
Medical Center, Hartford, CT, USA

Introduction

Patients with lipomyelomeningocele (LMMC) represent a unique population within the spectrum of spinal dysraphism. LMMC is defined as a subcutaneous lipoma occurring in the lumbosacral region and connected by a fibro-fatty stalk to an intramedullary lipoma. The

malformation tethers the spinal cord. With increasing patient age, signs and symptoms characteristic of tethered cord syndrome (TCS) may develop [1, 2]. Numerous authors have noted that the natural history of LMMC remains poorly defined, and adding to this confusion are the various classifications that LMMC have been grouped under, including spinal lipomas, closed neural tube defects, and occult spinal dysraphism [2–5].

Since its initial description by Hoffman et al. [6], TCS has been defined as a collection of signs and symptoms in patients with spinal dysraphism that may represent a tethered spinal cord. The orthopaedic literature comprises a limited number of studies evaluating the presenting signs and symptoms of TCS, and specifically the prevalence of musculoskeletal (MSK) sequelae in LMMC has received little attention [4, 7–9]. In a study comparing patients with myelomeningocele (MMC) and LMMC re-operated for a tethered cord, Herman et al. [7] described six presenting signs and symptoms and noted that they were similar in both groups of patients. It has been our impression that the clinical presentation of TCS in patients with LMMC differs from the presenting MSK signs and symptoms associated with other types of spinal dysraphism.

The reported incidence of LMMC is 1/4,000 live births [2, 4, 10]. It has been proposed that LMMC may become more common than MMC in the future [2]. This may be due to increased recognition and awareness of this closed neural tube defect and the increased awareness and use of folic acid supplementation to decrease the incidence of MMC and other open neural tube defects [2]. Interestingly, the incidence of LMMC is not affected by folic acid supplementation [11].

The goal of this study is to define the prevalence and pattern of clinical presenting musculoskeletal signs and symptoms in LMMC patients seen at an academic, multi-disciplinary spina bifida clinic. Determining these presenting musculoskeletal clinical signs and symptoms in patients with LMMC will help us in counseling our patients. This counseling includes advising patients and their families of the potential operative burden that patients may incur over their lifetime, the increased risk of re-tethering, and the need for continued surveillance for TCS over a longer period of time.

Methods

The Institutional Review Board of the Pennsylvania State University College of Medicine approved this study. A retrospective review of charts in patients identified as having LMMC in our Spina Bifida clinic was completed. Patient charts with incomplete data or diagnoses other than LMMC were excluded from the study. A data collection

Table 1 Modified Hoffer functional classification used in this study to describe the functional status of patients with myelomeningocele

Functional classification	Designation of classification	Description
Normal ambulator	N	Walking without crutches or braces
Community ambulator	C	Walking with or without crutches or braces, may use wheelchair for long distances
Household ambulator	H	Walk only indoors and with an apparatus, may use wheelchair for some indoor activities at home and school, and for all activities in the community
Therapy ambulator	T	Walk only in a therapy session, afterwards they use their wheelchair
Wheelchair ambulator	W	Patients who use a wheelchair only

The original Hoffer classification [12] was modified according to Schoenmakers et al. [13]

form was used to obtain demographic data which included the age at initial tethered cord release (TCR); repeat TCR procedures; musculoskeletal problems before and after TCR; limb length discrepancy (LLD); foot deformities such as cavovarus or equinovarus deformities; trophic ulcers; asymmetry of motor and sensory deficits; presence of scoliosis; orthotic needs; assistive devices; functional status [12, 13]. We used a modified Hoffer classification, which includes a normal ambulatory subtype, to determine the functional status of patients. The normal ambulatory subtype classification is defined as patients who can walk in the community without the need for crutches or braces (Table 1). We determined the prevalence of presenting musculoskeletal signs and symptoms in those patients enrolled in the study and examined the relationship between low back pain/radicular pain symptoms and the number of tethered cord releases using a chi-squared test. Statistical significance was set at $p = 0.05$.

We performed a PubMed peer reviewed literature search for the term “lipomyelomeningocele,” similar to a recent review performed by Tubbs et al. [4], to identify those studies that specifically addressed presenting musculoskeletal signs and symptoms related to lipomyelomeningocele. Data from other studies were also evaluated using Fisher’s exact test analysis to determine statistical significance with 2×2 contingency tables.

Results

We identified 38 patients with lipomyelomeningocele. Six patients with incomplete data were excluded. Of the

Table 2 Clinical data on the 32 patients with lipomyelomeningocele enrolled in the study

Case	Gender	Neurology level	Age at follow-up (years)	TCR ^a	Age at TCR	Repeat TCR ^a	MSK issues	Spine deformity ^a	PSH	Functional status ^b	Orthotics	Assistive device
1	Female	Left L3	14	Y	2 months	N	Equinovalgus foot ETT, LLD (2.5 cm) hallux valgus	N	L TAL × 2, vulpius calcaneal lengthening	C	DAFO #4 lift for LLD	None
2	Male	Symmetric L5/S1 weakness	11	Y	6 months	Y × 11	B/L Eq-valgus foot deformities	N	B/L calcaneal length vulpius, PL/PB length	C	DAFO #4	None
3	Male	Left L4/5	13	Y	<1 year	N	LLD (5 cm), Left ETT left calcaneovalgus	N	Left PMR, TAL, SPLATT tibia rotational osteotomy; right tibia epiphysiodesis	C	B/L AFO's	None
4	Female	Symmetric LE weakness	29	Y	24 years	Y	Scoliosis	Y	Multiple foot procedures	C	NR	Cane
5	Male	Right LE weakness	61	Y	46 years	Y	None noted	N	No orthopaedic procedures	W	None	WC
6	Female	Motor intact decreased distal sensation	16	Y	6 months	Y × 2	B/L cavovarus foot LLD (1 cm), trophic ulcers	Y	PT transfer, B/L PFR and 1st MTO	C	B/L UCBL	None
7	Male	Left LE weakness	29	Y	6 months	Y × 4	LLD (5 cm)	N	Left TAL	C	Left AFO	None
8	Female	Normal motor left decreased sensation	36	Y	30 years	Y × 2	None noted	N	No orthopaedic procedures	N	None	None
9	Female	Right L4/L5 weakness	9	Y	2 months	N	Cavus deformity	N	LLD (2.5 cm)	N	None	None
10	Female	Normal	8	Y	7 years	Y × 1	Scoliosis (10°)	Y	No orthopaedic procedures	N	None	None
11	Female	Left L5–S1	31	Y	12 years	Y × 2	Left cavus, clubfoot, scoliosis (45°), LLD (4 cm)	Y	Left triple arthrodesis, midfoot osteotomy long LD	C	Left AFO, Walker for long LD	None
12	Male	Symmetric distal weakness and sensation	53	Y	25 years	Y × 2	Cavus and trophic ulcers	N	No orthopaedic procedures	C	B/L AFO's	None
13	Female	Normal	14	Y	2 months	N	LLD (2.5 cm), grade II spondylolisthesis	Y	Right TAL, calcaneal lengthening	N	Right shoe lift	None
14	Female	Right LE weakness	20	Y	16 years	N	Scoliosis (62°)	Y	No orthopaedic procedures	C	Right FRAFO, Lofstrand WC for LD	None
15	Male	Symmetric L4	16	Y	4 months	Y × 2	Scoliosis (19°)	Y	No orthopaedic procedures	N	None	None
16	Male	Left L3/L4	25	Y	20 months	Y × 3	LLD (2.5 cm), cavus trophic ulcers	N	Left calc lengthening, PFR, multiple tendon releases left foot	C	Left AFO	Cane

Table 2 continued

Case	Gender	Neurology level	Age at follow-up (years)	TCR ^a	Age at TCR	Repeat TCR ^a	MSK issues	Spine deformity ^a	PSH	Functional status ^b	Orthotics	Assistive device
17	Male	Left LE distal weakness	18	Y	<6 months	N	LLD (>2 cm), left cavus, scoliosis (14°)	Y	Right tibia epiphyseodesis left calcaneal osteotomy, PFR	C	Left AFO	None
18	Female	Left LE weakness	16	Y	1 months	Y × 2	LLD (2.5 cm), ETT left hip dysplasia, left FA right calcaneus	N	Right tibia epiphyseodesis B/L VDRO and muscle transfers	C	B/L AFO's	Lofstrand WC for LD
19	Male	Right LE weakness	9	Y	<6 months	N	LLD (1 cm), right cavus, claw toes	N	No orthopaedic procedures	N	None	None
20	Female	Normal	13	Y	3 months	N	Left equinus	N	No orthopaedic procedures	N	None	None
21	Female	Right LE weakness	16	Y	4 months	Y × 1	Right cavovarus foot, trophic ulcer	N	Right PFR, Jones procedure cuneiform osteotomy	N	None	None
22	Female	Left LE weakness	32	Y	NR	N	LLD (4 cm), right hip dysplasia	N	Right VDRO, HWR	N	None	None
23	Female	Right LE decreased sensation	14	Y	9 months	N	None noted	N	No orthopaedic procedures	N	None	None
24	Female	Right LE	33	Y	NR	Y	Low back pain	N	Placement spinal cord stimulator for back pain	C	B/L AFO's	None
25	Male	Normal	15	Y	2 years	N	None noted	N	No orthopaedic procedures	N	None	None
26	Female	Normal	30	Y	NR	Y × 3	Chronic low back pain	N	No orthopaedic procedures	N	None	None
27	Female	Normal	46	Y	39 years	Y × 1	Claw toes	N	No orthopaedic procedures	N	None	None
28	Female	Left LE weakness	23	Y	NR	Y × 1	LLD (3 cm), Left FA ETT, cong scoliosis	Y	Femoral and tibial rotational osteotomies ASF + PSF	C	B/L AFO's	None
29	Female	L5/S1 weakness	15	Y	5 years	Y × 2	Left hallux valgus	N	No orthopaedic procedures	N	None	None
30	Female	Normal	9	Y	<6 months	N	Scoliosis (17°)	Y	No orthopaedic procedures	N	None	None
31	Male	Normal	8	Y	2 months	N	None noted	N	No orthopaedic procedures	N	None	None
32	Female	Left L3 weakness	19	Y	7 months	Y × 1	LLD (2.5 cm)	N	Right femoral epiphyseodesis	C	Right KAFO	None

AFO ankle foot orthosis, ASF anterior spine fusion, B/L bilateral, DAFO Dynamic Ankle Foot Orthosis (Cascade, Ferndale, WA), ETT external tibial torsion, FA femoral anteversion, FRAFO floor reaction ankle foot orthosis, HWR hardware removal, KAFO knee ankle foot orthosis, L lumbar, LD long distances, LE lower extremity, LLD limb length discrepancy, MSK musculoskeletal, MTO metatarsal osteotomy, NR not recorded, PFR plantar fascia release, PL/PB peroneus longus/brevis tendons, PMR posteromedial release, PSF posterior spine fusion, PT posterior tibial tendon, S sacral, SPLATT split anterior tibial tendon transfer, TAL tendoachilles lengthening, TCR tethered cord release, UCBL University California Berkeley Lab orthosis, VDRO varus derotational osteotomy, WC wheelchair

^a Y, Yes; N, no

^b See Table 1

Table 3 Comparison of presenting symptoms for tethered cord syndrome

Presenting symptom	Myelomeningocele	Lipomyelomeningocele	<i>p</i> value
Scoliosis	51/100	6/53	<0.0001
Pain	32/100	30/53	0.005
Orthopaedic deformity	11/100	17/53	0.002
Motor weakness	55/100	25/53	0.397
Gait abnormality	54/100	23/53	0.237
Urinary incontinence	6/100	11/21	0.0125

Data presented as the number of patients exhibiting the symptom, were extracted from Herman et al. [7]

remaining 32 patients, 21 were female and 11 were male patients (Table 2). The mean age of the patients at last follow-up was 21.9 years (range 8–61 years). The majority of patients had their TCR by ≤ 1 year of age (59 %), with 22 and 15 % having primary TCR at ages 1–15 and >15 years, respectively. Fifteen (47 %) patients had at least one repeat TCR, with ten of these having more than one repeat TCR. A significant relationship was noted between low back/radicular pain and repeat TCR ($p < 0.001$). Thirteen patients (40 %) had a LLD, with the LLD being >2.5 cm in ten of these patients. Four patients underwent epiphysiodesis to equalize limb lengths. Of the 32 patients, 53 % had asymmetric involvement. Nine patients (28 %) had scoliosis; of these, one patient had operative treatment for a congenital scoliosis, and the other eight patients were observed and did not require operative intervention. Two patients were identified as having hip dysplasia; both have a LLD, but the hip dysplasia involved the longer lower extremity in only one patient. Fifteen patients had foot deformities; the majority of whom had cavovarus deformities. Thirteen patients (41 %) underwent two or more orthopaedic procedures, in addition to other neurologic or urologic procedures. Sixteen patients were normal ambulators, 15 were community ambulators, and one was a wheelchair ambulator.

Our literature search of PubMed peer reviewed publications revealed 148 papers, of which we excluded 16 that were case reports or editorial comments. Upon further analysis of the remaining 132 papers, we found seven papers that addressed presenting clinical signs or symptoms [2–4, 7, 9, 14, 15]. We extracted the comparative data from the study by Herman et al. [7] and found statistically significant differences in the presenting clinical signs and symptoms for scoliosis, pain, and orthopaedic deformity between MMC and LMMC (Table 3).

Discussion

The first report of LMMC was published by Johnson in 1857, as cited by Hoffman et al. [6]. Despite the passage of

150 years, the natural history of LMMC remains unclear. The goals of the primary surgery for LMMC are preservation of all neural elements, debulking of the intramedullary lipoma, release of spinal cord tethering, watertight dura reconstruction, and resection of the subcutaneous lipoma mass with tension-free skin closure. The most challenging primary repairs occur with transitional lipoma attachment and the caudal malformation as nerve roots frequently traverse the lipoma within the distal thecal sac. It was initially thought that most patients have normal function at birth and subsequently demonstrate progressive loss of neurologic function with associated tethering of the spinal cord. This notion has been subjected to debate in the literature, as has the role of early intervention to debulk the lipomatous mass, decompress and untether the cord before apparent clinical signs and symptoms of TCS, and the extent or degree of resection [1, 2, 5, 10, 16–18].

The description and prevalence of the presenting orthopaedic clinical signs and symptoms of LMMC and TCS have been infrequent and often documented only in general terms [2–4, 7, 9, 14, 15]. Herman et al. [7] described and compared six primary clinical signs and symptoms, including scoliosis, orthopaedic deformities, gait deterioration, motor weakness, and urinary incontinence after re-operation for tethered cord in both MMC and LMMC patients. When we analyzed these two patient groups using the data provided in their report, we found that scoliosis was more common in the MMC group of patients, whereas pain and orthopaedic deformity were significantly more common in the LMMC group (Table 3). Compared to MMC, it is difficult to assign a neuro-segmental level in LMMC, and in the majority of patients with LMMC, the asymmetry of neurologic involvement is predominant.

Our experience noted a low prevalence of scoliosis in our small cohort of patients, with only one of nine patients requiring operative intervention. Other studies have also reported similar findings. Kanev et al. [2] noted a prevalence of 5 % of scoliosis (8/177) in their analysis of LMMC patients. The respective authors of three series evaluating the prevalence of scoliosis and TCS in

myelomeningocele noted that the prevalence of scoliosis was greater [19–21]. Sarwark et al. [21] found an incidence of new or progressive scoliosis of 60 % in patients with low level (L3 and below) MMC. In their study of children with lumbar level MMC, McLone et al. reported on 43 children (47 %) who had scoliosis as one of their signs of deterioration. Of these 43 patients, 30 had a tethered cord alone without syrinx or Arnold–Chiari malformation compression of the hindbrain [20]. Bowman et al. [19] found a 40 % incidence of progressive scoliosis requiring a TCR in their long-term study of 114 patients with MMC. The small number of patients in our series with scoliosis precludes our ability to support or comment on the presence of scoliosis as an indication for TCR in LMMC.

A high percentage of patients (53 %) in our series demonstrated asymmetric involvement. A LLD was noted in 13 patients (40 %), and the majority of the patients had a LLD of more than 2.5 cm. Asymmetric motor weakness in the involved extremity was also found. Many other studies have commented on LLD, primarily due to neurologic causes, being common sequelae of LMMC [2, 4, 7–9]. The lower extremity asymmetry in LMMC also extends to foot deformities, foot length, and muscle mass discrepancies, and these morphologic changes in the extremities and feet tend to present early and are progressive [15, 22]. Pierre-Kahn and colleagues [15] described the collection of musculoskeletal presenting signs and symptoms as the “neuro-orthopedic” syndrome and that these are often unilateral and at least asymmetric. Progression of MSK deformities is often due to further growth in a skeletally immature child. Re-tethering of the cord is always a concern in LMMC as noted below. In addition, after skeletal maturity is reached, biomechanical forces often influence progression of the MSK deformities. Despite the progression of the deformities, 31 of our patients (97 %) were either normal or community ambulators at last follow-up.

The asymmetrical pattern of involvement seen in LMMC is best described and explained in the neurosurgical literature. Chapman and Davis [23] described three different forms of LMMC that include transitional, dorsal and caudal forms. Pang et al. [17] have recently described a fourth type of spinal lipoma, the chaotic type—so named since it “does not follow the rules of either the dorsal or transitional lipoma”. Cochrane has written extensively on the transitional form of LMMC and has noted that patients with asymmetric malformations may exhibit unilateral functional neurologic or orthopaedic abnormalities [3, 16]. Hakuba et al. [24] proposed a more extensive classification of four types and six subtypes. Asymmetric malformations were found to be associated with deterioration on the ipsilateral side in nine of 11 patients, with these nine patients also showing a more rapid return of signs (mean 22 months) in the post-operative period compared to those

with symmetric malformations (range 50–65 months) [3]. Technically, the rotated asymmetric lipoma–neural interface in these forms of transitional LMMC are more difficult to debulk and untether [3]. Other authors have noted other anatomical differences in LMMC, including short nerve roots, syrinx rostral to the lipoma, and split cord malformations [3, 15, 24, 25]. Tubbs et al. [26] found a high percentage of LMMC patients with Chiari malformation type I (13 %) compared to the general population. At primary repair and during re-operations, complete untethering of the spinal cord is not always possible. With asymmetric lipomas and cord rotation, the roots are quite short and truncated on the lipoma side, restricting the relaxation and mobility of the spinal cord, and they are often dysfunctional.

The operative burden on patients with LMMC has not been emphasized in the literature. In our series of patients, 13 patients (41 %) had undergone two or more orthopaedic procedures—in addition to their other neurosurgical and urologic procedures. In his study, Cochrane noted that five of the 21 patients (22 %) required three to six procedures per patient for foot deformities [3]. Tubbs et al. [4] noted that 50 % of their patients had orthopaedic complications. Interestingly, these authors found that the caudal form of LMMC was more likely to be associated with orthopaedic complications, which is in contrast to the conclusions reached by Cochrane [3, 4]. No correlation was found between the location or size of the lipoma and neurologic function [2, 4]. We did not specifically address the urologic issues as these fall outside the scope of our study, but these issues are important to consider and add to the operative burden of these patients.

We noted a high incidence (47 %, 15/32 patients) of repeat TCR in our small cohort of patients with LMMC, with ten of these patients having more than one repeat TCR. We also found a significant relationship between repeated TCR and low back/radicular pain. Huang et al. [10] noted that the incidence of re-tethered cord ranged from 3 to 20 %. Colak and colleagues [27] reported a 20 % incidence of re-tethered cord, but cautioned that the frequency was dependent on the duration of follow-up and that their actuarial risk increased to 40 % at an 8-year follow-up endpoint. The transitional type of LMMC has been reported to have a higher rate of symptomatic re-tether than the other subtypes [16, 28]. Maher et al. [28] also found that an increased number of previous TCR procedures were associated with a worse result for pain relief and a greater chance of significant morbidity, including cerebrospinal fluid leak, pseudomeningocele, and severe dysesthetic leg pain likely from inadvertent sectioning of nerve roots. Schoenmakers et al. noted that late deterioration of motor or ambulatory level was only seen in the LMMC patients after revision of an initial TCR [12].

The limitations of our study include the retrospective nature of the study, the small number of patients in our cohort, and the number of patients excluded from the study due to incomplete data. We did not specifically identify the subtypes of LMMC as described by Chapman and Davis [23]. Our cohort of patients spanned a long period of time, and they were treated at other institutions and by different orthopaedic surgeons and neurosurgeons which precluded standardization of care such as indications for TCR. As noted earlier by Tubbs et al. [4], this wide disparity in the historical, geographic, and quality of care provided could confound the conclusions of this study.

In summary, the results of our study of patients with LMMC suggest that the presenting clinical signs and symptoms of TCS may differ in terms of both pattern and frequency from those of MMC and other forms of spinal dysraphism. We noted a high prevalence of asymmetrical involvement, a high operative burden, and a high rate of repeat symptomatic TCS requiring TCR. As other authors have noted, TCR in LMMC does not prevent long-term functional deterioration. These findings may be important to our colleagues providing counsel to their patients with LMMC and their families.

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