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ARTICLE

# Long-term urological outcomes of spinal lipoma after prophylactic untethering in infancy: real-world outcomes by lipoma anatomy

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## Abstract

**Study design** Cohort study.

**Objectives** Long-term urological outcomes in patients with spinal lipoma after prophylactic tethered cord release (TCR) in infancy were investigated.

**Setting** Children's hospital in Yokohama, Japan.

**Methods** Children under one year of age with spinal lipoma who underwent TCR between 1990 and 2010 were investigated. According to Arai's classification, lipomas other than filar lipoma were classified into four types: caudal, dorsal, transitional, and lipomyelomeningocele. The level of the conus medullaris was divided into three categories: L3-5, L5/S1, and sacral. Urological outcomes, including the need for clean intermittent catheterization (CIC), urinary incontinence, presence of renal deterioration, and the need for bladder augmentation, were investigated by both lipoma type and level of the conus medullaris.

**Results** Fifty-three patients met the inclusion criteria. The median follow-up period was 14.2 years (interquartile range 9.6–17.6 years). Of the 53 patients, ten (19%) were on CIC, and six (11%) were incontinent. Overall, two patients (4%) had renal deterioration detected by DMSA renal scan, and two (4%) needed augmentation cystoplasty. Of the lipoma types, transitional type showed the worst outcomes with respect to need for CIC (54%) and urinary incontinence (38%). There were no significant differences in renal deterioration and the rate of bladder augmentation by lipoma type. No urological outcomes were significantly associated with conus level.

**Conclusions** Even after prophylactic TCR in infancy in children with spinal lipoma, 19% of patients needed CIC in long-term follow-up. Of the lipoma types, transitional type showed the worst outcomes with respect to need for CIC and urinary incontinence.

## Introduction

In the long-term follow-up of patients with spinal lipoma of the conus medullaris, major urological concerns are need for clean intermittent catheterization (CIC), urinary incontinence, and renal deterioration. Several studies have reported long-term urological outcomes after tethered cord release (TCR) in children with spinal lipoma, and better urologic results are anticipated if surgery is performed in infancy [1–4]. However, few studies have clearly described the urological outcomes for each subtype of lipoma. Yerkes et al. recently reported the long-term urological outcomes after TCR in patients with spinal lipoma [5]. The outcomes were evaluated by each lipoma type, and the need for CIC was significantly associated with transitional lipoma. Although they described the details of lipoma type, conus medullaris level, and the extent of resection, the study had several weaknesses. First, they included patients with neurologic symptoms before TCR, and second, not all patients underwent TCR in infancy. If lipoma type affects future urological outcomes even in patients who underwent TCR in infancy, lipoma classification will be very useful for family counseling about bladder management and to establish the follow-up plan for each child after prophylactic TCR. The aim of this study was to investigate long-term urological outcomes by lipoma anatomy in patients who underwent prophylactic TCR in infancy.

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## Methods

After receiving institutional review board approval, the medical records of patients from 1990 to 2010 were reviewed to identify children under 1 year of age with spinal lipoma of the conus medullaris who underwent TCR in our institution. Exclusion criteria were as follows: filar lipoma or fatty filum terminale; concomitant anomalies of the lower urinary tract and anorectal region; patients with <5 years of follow-up; and no definite information on urinary continence in the medical records.

### Lipoma type and conus medullaris levels

A surgically oriented classification of intraspinal lipoma was first proposed by Chapman, who presented the following three subtypes: caudal, dorsal, and transitional types [6]. Arai et al. added another subtype, "lipomyelomeningocele", to Chapmans' classification. This subtype had a low-lying tethered spinal cord protruding externally to the spinal canal and terminating in the lipomatous mass in the subcutaneous meningeal sac [7]. Although the term lipomyelomeningocele (LMMC) has been used as a general term indicating all variations of spinal lipomas by many authors and has caused some confusion, Arai's classification of spinal lipoma has been well known in Japan and used in our institution. For this reason, based on neuroimaging and operative records, intraspinal lipomas were classified into four types in this study: caudal type; dorsal type; transitional type; and LMMC [8, 9]. As previously mentioned, filar type lipoma was excluded from this study, because in our institution, children with filar type lipomas were generally observed in infancy. The review of operative records and lipoma classification were done by pediatric neurosurgeons. The level of the conus medullaris was determined by a pediatric radiologist based on preoperative spinal MRI in our institution, and it was divided into the following three categories: low lumbar (L3-5), lumbosacral (L5/S1), and sacral (≥S1).

### Urological outcomes

To evaluate the long-term urinary condition, the following four outcomes were investigated by lipoma type and conus medullaris level: the need for CIC; urinary incontinence (w/ or w/o CIC); the presence of renal deterioration; and the need for bladder augmentation. In this study, urodynamic findings were not evaluated, because not all infants underwent preoperative urodynamic studies, and children with volitional voiding were followed-up by uroflowmetry and post-void residual urine tests without EMG. The indications for CIC were: voiding difficulty; large postvoid residual urine (PVR); urinary incontinence; worsening of the upper urinary tract including hydronephrosis or vesicoureteral reflux; and recurrent febrile urinary tract infections. The PVR was determined by video urodynamics, uroflowmetry, or several measurements by bladder ultrasound in children before toilet training. There was no strict definition of large PVR, but more than half of the total or expected bladder volume was considered large PVR. Information on urinary incontinence was obtained from the patient or caregiver and was categorized by use of CIC. Incontinence without CIC was daytime incontinence despite timed voiding or nighttime incontinence over age eight years. Incontinence with CIC was involuntary urine leakage despite appropriate use of CIC.

Renal deterioration at the latest follow-up was diagnosed by renal cortical imaging with a dimercaptosuccinic acid (DMSA) renal scan and/or renal ultrasound (US). Since 2003, renal US has been routinely performed in all patients with spinal lipoma every year. Patients on CIC and/or patients with a history of suspected febrile urinary tract infections (UTIs) underwent DMSA renal scans regularly. Renal deterioration was determined by an obvious renal parenchymal abnormality including a cortical defect and peripheral irregularity, not only by dilatation of the renal pelvis.

As for bladder augmentation, there were no clear standard surgical indications. However, this procedure was the last option for the management of patients with spina bifida in our institution. In cases of renal deterioration or refractory urinary incontinence due to a high-pressure bladder, patients were offered bladder augmentation by one of the senior urologists (YY).

### Follow-up procedure

All patients were evaluated at our urology clinic at least once a year. Besides history taking about urological symptoms, urinalysis, and renal-bladder US were routinely performed at each visit regardless of whether the patient was on CIC. Uroflowmetry was done in patients with volitional voiding after toilet training. Video urodynamics were done in patients on CIC or if symptoms such as incontinence or febrile urinary tract infections occurred.

### Statistical analysis

In the evaluations of outcome differences associated with lipoma anatomy, including lipoma type and conus level, Fisher's exact tests were performed. Kaplan–Meier (KM) curves of CIC initiation were constructed, and the log-rank test was used for comparisons. A two-tailed $p < 0.05$ was considered significant. The statistical analyses were performed using the free statistical software EZR (Version 1.37, Bone Marrow Transplantation 2013:48,452–458)

## Results

Between 1990 and 2010, a total of 82 patients with spinal lipoma of the conus medullaris underwent TCR at our institution. Of these 82 patients, 62 (76%) underwent surgery in infancy, and 53 (24 males, 29 females) met the inclusion criteria. The median follow-up period was 14.2 years (interquartile range (IQR) 9.6–17.6 years). The patients' characteristics are shown in Table 1. Overall, 43 (81%) voided volitionally, and 41 (77%) were continent without CIC at the latest follow-up. Six patients (11%) were incontinent with or without CIC. The median age at CIC initiation was 4.2 years (IQR 0.7–16.8 years). At the latest follow-up, 51 (96%) patients had undergone renal cortical imaging, including a DMSA renal scan in 36 patients and renal US in 15 patients. Two patients (4%) on CIC had renal deterioration detected by the DMSA renal scan. One patient had a history of recurrent UTI, and the cystogram showed vesicoureteral reflux (VUR), while the other had no definitive history of febrile UTI.

Bladder augmentation was performed for two patients (4%). One patient underwent the surgery at the age of seven years due to high bladder pressure and the appearance of VUR with renal scarring. The other underwent the surgery at the age of five years because he had both day and night time incontinence despite regular CIC. The parents of this patient very much wanted him to achieve urinary continence before entering elementary school.

**Table 1** Patients' characteristics

| | |
|---|---|
| Total patients | 53 |
| Sex M/F | 24/29 |
| Median age at surgery (IQR), months | 4.2 (2.2–6.2) |
| Median follow-up period (IQR), years | 14.2 (9.6–17.6) |
| Lipoma Type (*n*, (%)) | |
| Caudal | 14 (26%) |
| Dorsal | 20 (38%) |
| Transitional | 13 (25%) |
| LMMC | 6 (11%) |
| Conus level (*n*, (%))[a] | |
| L3–5 | 27 (53%) |
| L5/S1 | 5 (10%) |
| ≥S1 | 19 (37%) |
| Ambulatory status (*n*, (%)) | |
| Socially ambulatory | 51 (96%) |
| Wheelchair | 2 (4%) |
| Re-TCR (*n*, (%)) | 3 (6%) |

*IQR* interquartile range, *LMMC* lipomyelomeningocele, *TCR* tethered cord resection

[a]$N = 51$ because of no available MRI data in two patients

### Lipoma type and urological outcomes

The most common type of lipoma was dorsal (38%), followed by caudal (26%), transitional (25%), and LMMC (11%). Table 2 shows the major urological outcomes by the four types of spinal lipoma. The need for CIC and urinary incontinence were significantly associated with lipoma type. Of the four types, transitional type showed the worst outcomes with respect to both need for CIC (54%) and urinary incontinence (38%). Of the 6 patients showing urinary incontinence, 5 (83%) had transitional type. On the other hand, all patients with caudal type voided volitionally and showed urinary continence. Regarding renal deterioration and the rate of bladder augmentation, there were no significant differences by lipoma type.

As for the initiation of CIC over the entire follow-up period, KM curves showed a high initial CIC rate in infants with LMMC type. However, after pubertal age, the CIC rate increased in patients with transitional type and ultimately became the highest (Fig. 1). Details of patients with CIC are shown in Table 3.

### Conus level and urological outcomes

Of the 53 patients, 51 (96%) patients underwent MRI in our institution, and the conus level was re-evaluated. Two patients only had radiographic image interpretation at another institution. Table 4 presents the urological outcomes by conus level. None of the outcomes was significantly associated with conus level.

**Table 2** Urological outcomes by lipoma type

| | Caudal $n = 14$ | Dorsal $n = 20$ | Transitional $n = 13$ | LMMC $n = 6$ | Total $n = 53$ | *p* value |
|---|---|---|---|---|---|---|
| CIC | 0 (0%) | 1 (5%) | 7 (54%) | 2 (33%) | 10 (19%) | **0.0004** |
| Incontinence | 0 (0%) | 1 (5%) | 5 (38%) | 0 (0%) | 6 (11%) | **0.0084** |
| Incontinence w/ CIC | 0 (0%) | 0 (0%) | 4 (31%) | 0 (0%) | 4 (8%) | |
| Incontinence w/o CIC | 0 (0%) | 1 (5%) | 1 (8%) | 0 (0%) | 2 (4%) | |
| Renal deterioration | 0 (0%) | 1 (5%) | 1 (8%) | 0 (0%) | 2 (4%)* | 1 |
| Augmentation | 0 (0%) | 0 (0%) | 1 (8%) | 1 (17%) | 2 (4%) | 0.124 |

*CIC* clean intermittent catheterization

[a]$N = 51$ because of no available renal imaging (DMSA scan or US) in two patients

During the follow-up period, four (8%) patients showed re-tethered cord symptoms. These four patients primarily had transitional type, and subsequent urological deterioration, such as urinary incontinence, decreased bladder compliance, and VUR were seen in all of them; three of them underwent secondary TCR, at a median age of 15.2 years (range 12.1–17.6 years).

## Discussion

One of the main considerations in TCR in patients with spinal lipoma is long-term urological outcomes after surgery. Although overall bladder function was significantly better in non-myelomeningocele patients than in myelomeningocele patients [10], key clinical concerns in these patients were need for CIC, urinary incontinence, renal deterioration, and the need for major reconstruction surgery, represented by bladder augmentation. Few studies have reported the long-term clinical urological outcomes as described above after prophylactic TCR in infancy, though multiple studies suggest that prophylactic early intervention for lipoma is beneficial [11, 12]. Furthermore, some publications have reported the outcomes following TCR in a cohort of mixed diagnoses, including not only conus lipoma, but also fatty filum, dermal sinus tract, and VATER's syndrome [12–14]. In addition, due to the lack of a standard classification of spinal lipoma, few studies investigated these urological outcomes in each subtype of spinal lipoma [5]. If the subtype of lipoma affects urological outcomes, it is possible to establish a better follow-up schedule for each subtype. To the best of our knowledge, this is the first report of long-term clinical urological outcomes after prophylactic TCR for conus lipoma in infancy.

Of all the outcomes, the need for CIC is the primary issue after TCR, although there is no clear consensus as to when

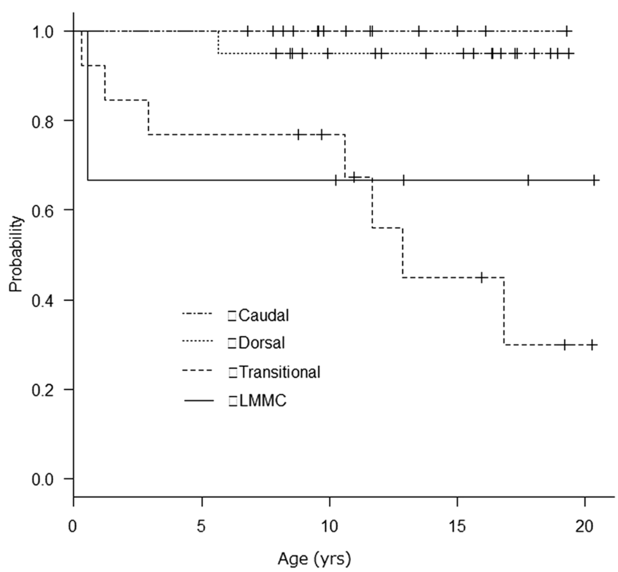


**Fig. 1** Kaplan–Meier plot showing patient age at the time of CIC initiation

**Table 4** Urological outcomes by conus level

| | L3-5 $n = 27$ | L5/S1 $n = 5$ | ≥S1 $n = 19$ | Total $n = 51$ | $p$ value |
|---|---|---|---|---|---|
| CIC | 3 (11%) | 1 (20%) | 6 (32%) | 10 (20%) | 0.204 |
| Incontinence | 2 (7%) | 0 (0%) | 4 (21%) | 6 (12%) | 0.265 |
| Renal deterioration | 1 (4%) | 0 (0%) | 1 (5%) | 2 (4%) | 1.000 |
| Augmentation | 0 (0%) | 0 (0%) | 2 (10%) | 2 (4%) | 0.322 |

*CIC* clean intermittent catheterization

**Table 3** Details of ten patients on CIC

| Pt. | Lipoma type | Sex | Starting age of CIC (y) | Indication for CIC | Concomitant use of anticholinergics at the latest follow-up |
|---|---|---|---|---|---|
| 1 | LMMC | M | 0.5 | Large PVR | None (post augmentation) |
| 2 | LMMC | M | 0.5 | Large PVR | Fesoterodine |
| 3 | Transitional | F | 0.3 | Large PVR, fUTI | None (post augmentation) |
| 4 | Transitional | F | 1.2 | fUTI | None |
| 5 | Transitional | F | 2.9 | Large PVR, voiding difficulty | Propiverine |
| 6 | Transitional | M | 7.3 | Large PVR, incontinence | None |
| 7 | Transitional | M | 10.6 | Incontinence, DSD on UDS | None |
| 8 | Transitional | F | 12.9 | Large PVR, voiding difficulty | Propiverine |
| 9 | Transitional | M | 16.8 | Large PVR | None |
| 10 | Dorsal | F | 5.6 | Large PVR | Propiverine |

*PVR* postvoid residual urine, *fUTI* febrile urinary tract infection, *DSD* detrusor sphincter dyssynergy, *UDS* urodynamic study to initiate CIC in children with neurogenic bladder dysfunction [15]. The present study showed that 19% of children eventually needed CIC for a long time even after prophylactic TCR in infancy. More importantly, the rate of CIC was significantly associated with lipoma subtype. Patients with transitional lipoma showed the highest CIC rate (54%), and none of the patients with caudal lipoma needed CIC. Although there were slight differences in the lipoma classification and the age at TCR, this CIC outcome was very similar to the recent reports from the Chicago group. Yerkes et al. reported the long-term urological outcomes after TCR in 56 patients with spinal lipoma [5]. The outcomes were evaluated with four subtypes of lipoma (Chapman's classification of three, such as dorsal, caudal, and transitional, and they added the fourth category, the chaotic lipoma described by Pang et al.) [16]. The overall need for CIC was 23%, and it was significantly associated with transitional lipoma [5]. In their study, 7 of 15 patients (47%) with transitional lipoma needed CIC. This rate was significantly the highest among their four subtypes of lipoma, as in the present study. On the other hand, conus level did not affect the CIC rate, not only in the present study, but also in the study by the Chicago group.

Regarding the status of urinary continence, the present study showed a good continence rate of 88%, and urinary incontinence was also significantly associated with the transitional type. This continence rate was also similar to the report from the Chicago group in which an overall continence rate of 86% was seen [5]. However, lipoma type was not associated with urinary incontinence in their study. The reason remains unexplained, but in the study by the Chicago group, not all patients (71%) underwent primary TCR at <1 year of age, and 32% of patients presented with symptoms before TCR.

The most critical therapeutic goal in children with spina bifida is to prevent renal deterioration. It has been widely known that upper urinary tract deterioration typified by VUR and hydronephrosis is caused by high bladder pressure in patients with spina bifida [17]. However, the key finding of renal deterioration is renal cortical loss, not the dilation of the urinary tract due to VUR and hydronephrosis. It has been shown that patients with spina bifida are at increased risk of renal cortical loss detected by DMSA scans. Shiroyanagi et al. reported that 16 of 64 (25%) patients older than 10 years having a history of CIC showed abnormal renal scans [18]. Woo et al. reported that renal cortical loss on DMSA scans was found in 43/100 (43%) of patients, and history of VUR, history of hydronephrosis, and CIC from birth were significant predictors of renal damage in children with spinal dysraphism [15]. Few reports have investigated renal deterioration specifically in patients with spinal lipoma. Yerkes et al. reported that 2 of 53 (4%) patients had significant hydronephrosis detected by renal US. They did not use DMSA renal scans to evaluate renal cortical loss. The present study showed that only 2/51 (4%) of lipoma patients showed renal deterioration on DMSA. The possible explanation for this low deterioration rate was the low CIC rate in patients with spinal lipoma who underwent prophylactic TCR compared with patients with myelomeningocele.

As for bladder augmentation, two (4%) patients underwent surgery. Compared with the surgery rate of 9% in non-myelomeningocele patients in the National Spina Bifida Patient Registry (NSBPR), the surgery rate in the present study was small [19]. This low surgery rate is possibly due to the low rates of incontinence and renal deterioration in the present study. However, the indications for this surgery are not clearly standardized, though this surgery is usually performed to protect against renal deterioration and urinary incontinence caused by high bladder pressure in patients with spina bifida. In fact, from the perspective of performance of bladder augmentation in this population, Wang et al. recently reported significant geographic variation in the USA [20].

As with retrospective studies, there are several limitations to this study other than the indications for bladder augmentation. First, incontinence was assessed from medical records based on subjective information from patients or caregivers, not by any objective scale. Second, the need for CIC was decided by the CIC status of each patient at the latest visit, not by the clinical indications. Third, to evaluate renal deterioration, not all patients underwent DMSA renal scans. Although renal US is useful to detect dilatation of the urinary tract, it is less sensitive to detect renal cortical loss than DMSA renal scans. Fourth, although the extent of the lipoma resection (complete or partial) has been reported as a critical issue in postoperative outcomes [3], it was not evaluated due to the difficulty of reviewing the operative records of more than one neurosurgeon. Last, urodynamic data were not assessed in this study. The reason was not only the low preoperative performance rate as described, but also the subjectivity of the assessment of electromyographic activity and detrusor overactivity that was confirmed in a recent study [21]. In fact, the interpretation of detrusor sphincter dyssynergism in infants with occult dysraphism is challenging due to transient detrusor sphincter dyscoordination, which is nonpathological and frequently observed in this age group [22].

## Conclusions

To the best of our knowledge, this is the first report of the long-term clinical urological outcomes, including need for CIC, urinary incontinence, renal deterioration, and need for bladder augmentation, after prophylactic TCR for conus lipoma in infancy. Even after prophylactic TCR in infancy, 19% of patients needed CIC, and 11% of patients had urinary incontinence in the long-term follow-up of children with spinal lipoma. Lipoma type was significantly associated with need for CIC and urinary incontinence. Of the four types of lipoma, transitional type showed the worst outcomes with respect to both need for CIC (54%) and urinary incontinence (38%). On the other hand, conus level did not affect any urological outcomes.

## Data availability

The datasets generated and analyzed in the current study are available from the corresponding author on request.

**Author contributions** CH and YY initiated the research question and the protocol for data analysis. CH was responsible for the data analysis and interpretation, under the supervision of YY. CH wrote the manuscript, under the supervision of YY. YK, HS, and DH contributed to the interpretation of the results. All authors approved the final version of the manuscript.

## Compliance with ethical standards

**Conflict of interest** The authors declare that they have no conflict of interest.

**Ethics** We certify that all applicable institutional and governmental regulations concerning the ethical use of human volunteers were followed during the course of this research.

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