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Study of Tethered Spinal Cord Syndrome Diagnosis Following a Close Examination of Daytime Urinary Incontinence

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Abstract: *Background:* Tethered spinal cord syndrome refers to neurological disorders caused by the lower end of the spinal cord becoming entrapped in the caudal tissue. Dysuria is one of the most common symptoms and often appears at an early stage. In this study, we investigated cases of children with tethered spinal cord syndrome who were presented to the urology department with daytime urinary incontinence. *Subjects and methods:* Eighteen children with tethered spinal cord syndrome were diagnosed between March 2011 and October 2017 after a visit to the urology department with daytime urinary incontinence as the main complaint. The reasons for the diagnosis of tethered spinal cord syndrome and the changes in clinical symptoms before and after laminectomy were investigated. *Results:* Nine boys and nine girls were selected as the study subject. The mean age at the first examination was 6.3 years (range 4–9 years). All patients underwent urodynamics and voiding cystourethrogram (VCUG) for refractory daytime urinary incontinence. Spinal MRI was performed in all patients to investigate abnormal findings on VCUG and urodynamics. Only four patients had indications of tethered spinal cord syndrome on MRI. Eight patients had lipoma of the filum terminale, four patients had potential tethered spinal cord syndrome, four patients had low-lying conus, one patient with conus lipoma, and one patient had intrasacral meningeal cyst. The mean follow-up after laminectomy was 66.3 (22–116) months, 11 patients were asymptomatic, 4 patients had residual nocturia only, and 3 patients were managed with intermittent voiding. *Conclusion:* It is necessary to treat patients with daytime urinary incontinence is one of the symptoms of the tethered spinal cord syndrome. Bladder function assessed by urodynamics may provide a diagnostic sign for spinal cord compression.

Keywords: Diurnal urinary incontinence; Spinal cord compression; Urodynamics

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1. Introduction

The most common clinical manifestation of spina bifida is tethered spinal cord syndrome. Tethered spinal cord syndrome is a condition in which the lower end of the spinal cord is fixed (entrapped) in caudal tissues (dura mater, subcutaneous tissue, etc.) due to neural tube dysplasia during the embryonic period. Continued caudal traction of the spinal cord results in increased lower limb skeletal muscle tendon reflexes and autonomic disorders. Dysuria, in particular, is considered to be one of the earliest manifestations of tethered spinal cord syndrome [1]. In the present study, we investigated a case of a child presenting with daytime

urinary incontinence and nocturia, which led to the diagnosis of tethered spinal cord syndrome.

2. Methodology

We retrospectively studied 18 children who presented to our urology department between March 2011 and October 2017 with daytime urinary incontinence and were diagnosed with tethered spinal cord syndrome upon examination. Cases in which tethered spinal cord syndrome had already been detected by MRI or other means prior to the visit to the urology department were excluded. The age, gender, voiding cystourethrogram (VCUG), reason for spinal MRI, diagnosis of spinal MRI, urodynamics before and after surgery, postoperative voiding symptoms, and urinary tract management of the patients were investigated. Besides, the patients' background, reasons for spinal MRI, and spinal MRI diagnosis were assessed descriptively terms of percentage: maximum bladder capacity at VCUG, maximum bladder capacity/expected bladder capacity [2], bladder deformity [3] or vesicoureteral reflux (VUR), urodynamic parameters (presence or absence of detrusor overactivity [DO], bladder compliance) before and after untethering were compared using the Mann-Whitney U test or Fisher's exact test. This study was reviewed and approved by the Research Review Committee within the Ethics Committee of the Tokyo Metropolitan Children's General Medical Centre (Ethics Committee No. 2019b-105).

3. Results

Nine boys and girls were selected as test subjects in this study. The mean age at the first examination was 6.3 years (range 4–9 years). The patients' background is shown in **Table 1**. All patients were referred from other hospitals for a thorough examination of daytime urinary incontinence or refractory urinary incontinence, and 13 (72%) had concomitant nocturia. All patients underwent bladder function tests, VCUG, and urodynamics at an average of 5.8 months after the initial examination for refractory urinary incontinence.

The mean maximum bladder capacity on VCUG was 133 ± 69 mL, and the mean maximum bladder capacity/expected bladder capacity was 60 ± 29%. It was found that 4 patients had grade 1 bladder deformity, 2 patients with grade 2, and 1 patient with grade 3 bladder deformity according to the Ogawa classification [3], and VUR was observed in 2 patients. Urodynamics showed DO in 12 patients (67%), with a mean compliance of 15.0 ± 15.4 mL/H_2O.

Table 1. Patient background

	All cases (*n* = 18)	**Boys (*n* = 9)**	**Girls (*n* = 9)**	***P***
Age at first consultation (years)	6.3 ± 1.6	6.4 ± 1.7	6.2 ± 1.4	0.97
Nocturia *n* (%)	13 (72)	6 (67)	7 (78)	0.88
Constipation, *n* (%)	4 (21)	3 (33)	1 (11)	0.30
Sacral dimple, *n* (%)	5 (26)	2 (22)	3 (33)	0.85
VCUG				
Maximum bladder capacity[*1] (mL)	133 ± 69	107 ± 40	159 ± 81	0.33
Maximum bladder capacity/EBC[*2] (%)	60 ± 29	49 ± 17	71 ± 35	0.62
Bladder deformity, *n* (%)	12 (67)	6 (67)	6 (67)	1.0
VUR, *n* (%)	2 (11)	1 (11)	1 (11)	1.0

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	All cases (*n* = 18)	Boys (*n* = 9)	Girls (*n* = 9)	*P*
Urodynamics				
DO+, *n* (%)	12 (67)	8 (89)	4 (44)	0.13
Bladder compliance, mL/cmH_2O	14.9 ± 15.8	17.4 ± 20.9	12.3 ± 7.0	0.66
Age at the time of surgery (years)	7.3 ± 1.6	7.0 ± 1.8	7.6 ± 1.3	0.34
Period between surgery and initial consultation (months)	16.7 ± 10.7	13.0 ± 11.0	20.4 ± 8.9	0.22
Period between final observation from initial consultation (month)	66.3 ± 23.8	67.7 ± 27.7	65.0 ± 19.0	0.92

Abbreviations: VCUG, voiding cystourethrogram; EBC, expected bladder capacity; VUR, vesicoureteral reflux; DO, detrusor overactivity. [*1]VCUG, [*2](Age +1) × 30 mL.

All patients underwent spinal MRI; one patient had a sacral dimple, which was performed at the same time as the bladder function test. The other 17 patients (95%) were screened for spinal cord disease and tethered spinal cord syndrome based on the results of urodynamics or cystography (**Table 2**). Eight patients (44%) had filum terminale lipomas, four (22%) had low-lying spinal conus, one (6%) had a conus lipoma, and one (6%) had an intrasacral meningeal cyst. Spinal MRI showed tethering of the spinal cord or abnormal spinal conus position in only 4 patients (22%); 14 patients (78%) had no anatomical tethering of spinal cord but were diagnosed with symptomatic tethered spinal cord syndrome based on urinary incontinence symptoms and urodynamic results that indicated the need for a surgery. Four patients (22%) were diagnosed with occult tethered cord syndrome (OTCS) without any abnormal findings on imaging, such as filum terminale lesions or abnormal spinal conus position. In comparison between OTCS and non-OTCS cases, all patients in the OTCS group had DO and significantly lower bladder compliance (**Table 4**).

Table 2. Reasons for spinal MRI

	***n*（%）**
DO or low compliance	16 (89)
Bladder deformity	1 (6)
Sacral dimple	1 (6)

Abbreviation: DO, detrusor overactivity.

Table 3. Diagnosis on spinal MRI scan

	***n*（%）**	**Tethering, *n***
Ulnar lipomatosis	8 (44)	0
Potential occult tethered cord syndrome (OTCS)	4 (22)	0
Low spinal cord conus	4 (22)	3
Conus lipoma	1 (6)	1
Intraspinal meningocele cyst	1 (6)	0

Abbreviation: OTCS, occult tethered cord syndrome.

Table 4. Comparison of potential OTCS with other cases

	OTCS (*n*=4)	**Other than OTCS (*n* = 14)**	***P***
Age at first consultation (years)	6.3 ± 1.5	6.3 ± 1.6	1.00
VCUG			
Maximum bladder capacity[*1] (mL)	109 ± 45	140 ± 70	0.49
Maximum bladder capacity/EBC[*2] (%)	54 ± 23	62 ± 31	0.49
Bladder deformation, *n* (%)	4 (100)	8 (57)	0.25
VUR, *n* (%)	1 (25)	1 (7)	0.41
Urodynamics DO+, *n* (%)	4 (100)	8 (57)	0.16
Bladder compliance (mL/cmH_2O)	7.6 ± 2.7	16.2 ± 17.7	*0.038
Age at laminectomy (years)	6.5 ± 1.7	7.5 ± 1.6	0.36

Abbreviation: OTCS, occult tethered cord syndrome; VCUG, voiding cystourethrogram; EBC, expected bladder capacity; VUR, vesicoureteral reflux; DO, detrusor overactivity. *$P < 0.05$

All patients underwent laminectomy, and their bladder capacity increased from 148 ± 93 mL to 182 ± 97 mL after surgery, but there was no statistically significant difference. Bladder deformity improved in six patients, VUR disappeared in one patient, and DO disappeared in seven patients (**Table 5**). Bladder compliance improved markedly from 15.0 ± 15.4 to 26.5 ± 36.3 cmH_2O ($P < 0.0001$). Daytime urinary incontinence resolved in all patients: four were treated with anticholinergics for frequent urination and oliguria, four remained nocturia only and were treated with desmopressin or anticholinergics, and three were on intermittent voiding control and anticholinergics.

Table 5. Changes in bladder capacity and urodynamics before and after laminectomy

	Before surgery	**After surgery**	***P***
Maximum bladder capacity[※1] (mL)	133 ± 69	167 ± 76	*0.021
Maximum bladder capacity/EBC[※2] (%)	60 ± 29	64 ± 27	0.090
Bladder deformation or VUR, *n* (%)	14 (78)	7 (39)	*0.049
DO+, *n* (%)	12 (67)	5 (28)	*0.049
Bladder compliance (mL/cmH_2O)	14.9 ± 15.8	26.5 ± 36.3	*< 0.0001

Abbreviation: EBC, expected bladder capacity; VUR, vesicoureteral reflux; DO, detrusor overactivity. *$P < 0.05$, [※1] VCUG, [※2] (Age + 1) × 30mL.

4. Discussion.

The standard treatment for daytime urinary incontinence in children is generally conservative treatment emphasizing on urotherapy, followed by pharmacological treatment with anticholinergic drugs [4]. In refractory cases, screening for bladder dysfunction by urodynamics and VCUG is recommended [4]. There are a number of brain and spinal cord disorders that can cause a suspected neurogenic bladder, the most common of which is spina bifida in children [5]. In general, the earlier the surgery for spina bifida is performed, the better the long-term functional prognosis, including urinary function [6-9]. In this study, children diagnosed with tethered spinal cord syndrome following a close examination for daytime urinary incontinence were included. All patients underwent laminectomy, and as a result, postoperatively daytime urinary incontinence disappeared, and urodynamic parameters improved.

Invasive tests such as urodynamics are not recommended in the early stages of the treatment of daytime urinary incontinence and nocturia [4,10]. However, in our study, all patients were children under 10 years of age, and none of them were presented with symptoms due to puberty-related changes in body size, and all presented with persistent daytime urinary incontinence since childhood. Secondary nocturia is defined as the reappearance of nocturia after more than 6 months of absence [12], and it is considered necessary to consider underlying neurogenic bladder disorders such as terthered spinal cord syndrome for secondary nocturia [10]. All patients with nocturia in the present study (13 patients: 68%) had primary nocturia and none had secondary nocturia.

In our hospital, children presenting for refractory urinary incontinence are often assessed for bladder function using urodynamics or other methods if consent is obtained. Screening for bladder dysfunction using urodynamics [13] and early diagnosis and intervention of terthered spinal cord syndrome have been reported to be effective in the prognosis of future voiding function [14-16]. At our hospital, 369 patients underwent urodynamic and VCUG screening for daytime urinary incontinence and nocturia in the same period, and spinal MRI was performed in 129 patients. Approximately 5% of children presented to our hospital with daytime urinary incontinence had tethered spinal cord syndrome. It is difficult to determine the exact frequency of spinal cord entrapment syndrome in children with daytime urinary incontinence because our clinic is often referred to by other hospitals as a refractory case. However, the possibility of terthered spinal cord syndrome being present in children with daytime urinary incontinence as the main complaint should be taken into account during treatment, and bladder function assessment should be actively considered, at least when the patient is considered refractory [4].

In this study, only 4 (21%) children had tethered spinal cord syndrome findings on MRI, and all 4 had a shallow ulcer in the sacral region. Tamura *et al.* reported that 5.8% of children with sacral dimple had tethered spinal cord syndrome [17]. Although there are differing opinions as to whether all cases of sacral dimple should be investigated by MRI or other methods, it should be noted there is a certain possibility of terthered spinal cord syndrome [18]. Tamura *et al.* reported that 7.8% of their patients were diagnosed with terthered spinal cord based on abnormal findings in urodynamics, although anatomical indications was not observed [18]. In the present study, 14 patients (78%) had no anatomical spinal cord involvement, and four of them did not even have spinal cord lesions such as a filum terminale lipomas, leading to a diagnosis of OTCS [19-21]. The four OTCS patients also had no skin abnormalities, including sacral dimple. Although there is controversy regarding surgical intervention and timing of surgery in the absence of anatomical spinal cord involvement on imaging studies, children with clinical symptoms of neurogenic bladder, with or without anatomical spinal cord involvement, are indicated for laminectomy as symptomatic patients [22-23]. It is important to recognize that some children with daytime urinary incontinence or nocturia may be candidates for spinal deactivation and an accurate assessment of bladder function should be performed using urodynamics.

In our hospital, the main focus is on children with spina bifida who often require voiding care, including the patients in this study, and conferences are held in the departments of urology, gastroenterology and neurosurgery. In some cases, the patient has bladder dysfunction on examination but no abnormal findings on MRI, and Hinman syndrome [24] is suspected based on the history and examination, but after a conference. In these cases, laminectomy is not performed. The patients who underwent laminectomy as OTCS often had DO and findings of low bladder compliance. As shown in **Table 5**, abnormal findings in urodynamics that could not be explained in OTCS cases played a role in determining the need for surgery. The patient's bladder was not found to have a low bladder compliance. However, if neurogenic bladder is strongly suspected based on urodynamic results, the possibility of OTCS should be considered, and the indication for laminectomy should be discussed with the neurosurgeon. There is no difference in surgical findings between OTCS and non-OTCS cases; pathological investigation of terminal

thread lesions in OTCS may lead to a better understanding of the pathogenesis of OTCS and warranting more studies in the future.

5. Conclusion

In this study, we report on a child diagnosed with terthered spinal cord syndrome are complaints of daytime urinary incontinence was reported. It is necessary to treat cases in which daytime urinary incontinence is one of the symptoms of terthered spinal cord syndrome. A detailed bladder function assessment should be performed in cases of refractory daytime urinary incontinence, and screening for terthered spinal cord syndrome should be considered.

Disclosure statement

The authors declare no conflict of interest.

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