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Comprehensive analysis of the clinical and urodynamic outcomes of primary tethered spinal cord before and after spinal cord untethering

O. Alsowayan [a,b], A. Alzahrani [a], J.-P. Farmer [a], J.-P. Capolicchio [a], R. Jednak [a], M. El-Sherbiny [a]

[a]Department of Pediatric Surgery, Division of Pediatric Urology and Neurosurgery, The Montreal Children's Hospital and McGill University Health Center, Shriners Hospital of Montréal, QC, Canada

[b]Department of Urology, College of Medicine and King Fahd Hospital of the University, University of Dammam, Saudi Arabia

Correspondence to:
O. Alsowayan, Department of Urology, College of Medicine and King Fahd Hospital of the University, University of Dammam, PO Box 40086, Alkhobar 31952, Saudi Arabia.
Tel.: +966 556855003; fax: +966 138966770

sowayan81@gmail.com
(O. Alsowayan)

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Summary

Introduction
Primary tethered spinal cord (TSC) refers to a group of abnormalities associated with a posterior bony spinal defect that develops beneath an intact dermis and epidermis. There is relative agreement that patients with symptomatic TSC will require surgical intervention. However, it is still debatable as to how to approach asymptomatic patients with primary TSC.

Objective
To study the clinical and urodynamic (UDS) outcomes of patients with primary TSC after spinal cord untethering (SCU).

Study design
Charts of patients with primary TSC between 1998 and 2010 were retrospectively reviewed. Patients that underwent before and after SCU clinical and UDS evaluation with minimum of 5-years follow-up were included. Continence status was assessed in children ≥5 years. Patients with dry intervals of ≥4 h were considered continent. Urologic and neuro-orthopedic manifestations, as well as UDS parameters, were compared before and after SCU.

Categorical data were compared using Fisher-Exact test and continuous variables were compared using Wilcoxon-Signed-Rank test. A *P*-value <0.05 was considered significant.

Results
Twenty-two patients met the inclusion criteria. The median age at time of SCU was 11.5 months (range 3–211). The median age at time of follow-up UDS after SCU was 22 months (range 9–218). The median age at time of last follow-up was 153.5 months (range 65–228). The median follow-up time was 71 months (range 60–192). A total of 14/22 patients had clinical manifestation before SCU, while 8/22 were asymptomatic and diagnosed based on magnetic resonance imaging/UDS findings. Of the symptomatic patients, 86% had symptom improvement after SCU. The UDS parameters showed statistically significant improvement in the median percentage of change of actual bladder capacity ($P = 0.01$), median intravesical pressure for patients with pre-operative pressure ≥40 cm/H2O at total cystometric bladder capacity ($P = 0.012$), and median bladder compliance at 75% bladder capacity ($P = 0.01$) (Table).

Discussion
Tethered spinal cord syndrome (TSCS) is a clinical entity that presents with neurological, urological, and/or orthopedic symptoms caused by primary or secondary tethering of the spinal cord, which may result in ischemic damage of the neural tissue and symptom development.

While some authors believe that surgical management should be reserved for symptomatic patients, others prefer prophylactic surgery to avoid possible irreversible neurological damage. The present study provides detailed discussion of the clinical and UDS outcomes for patients with primary TSC that underwent SCU.

Conclusion
For patients with primary TSC, spinal cord untethering is beneficial in terms of clinical and UDS outcomes. A prospective long-term study with large numbers could further highlight outcomes for this particular group of patients.

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Table Urodynamic outcomes.

Parameter	Median	Range	*P*-value
Percentage of change of EBC after SCU	18.5	2.2—33.3	0.01
Percentage of change of actual bladder capacity after SCU	40.9	−18 to 230	
Pressure at TCBC Before SCU (cmH2O)	37.5	9—100	0.1
Pressure at TCBC After SCU (cmH2O)	25	5—62	
Compliance at 75% capacity before SCU (ml/cmH2O)	3.5	0.4—17.9	0.01
Compliance at 75% capacity after SCU (ml/cmH2O)	7.9	2.3—60.4	

EBC, expected bladder capacity; SCU, spinal cord untethering; TCBC, total cystometric bladder capacity.

Introduction

Tethered spinal cord syndrome (TSCS) is a clinical entity that presents with neurological, urological and/or orthopedic symptoms caused by primary or secondary tethering of the spinal cord [1]. In 1976, Hoffman et al. first introduced the concept of tethered spinal cord (TSC) [2]. There is a relative agreement that patients with symptomatic TSCS will require surgical intervention [3,4]. However, it is still debatable as to how to approach asymptomatic patients with primary TSC [4]. While some authors believe that surgical management should be reserved for symptomatic patients, others prefer prophylactic surgery to avoid possible irreversible neurological damage [5—10]. The present study discusses the clinical and urodynamic (UDS) outcomes for patients with primary TSC who underwent spinal cord untethering (SCU) between 1998 and 2010.

Materials and methods

Charts of patients with primary TSC who underwent SCU between 1998 and 2010 were retrospectively reviewed. Patients that underwent before and after SCU clinical and UDS evaluation were included. All patients were followed up for a minimum of 5 years for proper clinical outcomes assessment.

Primary tethered cord was suspected due to an abnormal back exam. Diagnosis was confirmed with magnetic resonance imaging (MRI). The procedure of SCU was carried on as described by Vernet et al. [11]. A comprehensive clinical UDS using artificial filling cystometrogram, and upper tract evaluation with ultrasound were performed before and after SCU. The UDS were repeated 6—12 months after SCU and annually thereafter, if clinically warranted. VCUG was performed when Grade 3—4 hydronephrosis, according to the Society of Fetal Urology grading system, was detected [12]. Continence status was assessed in children aged $\geq$5 years. Patients with a dry interval of $\geq$4 h were considered continent. Urologic and neuro-orthopedic manifestations were compared before and after SCU.

Clinical improvement was defined as resolution of symptoms that derived SCU. Stability was considered when the patient showed neither improvement nor deterioration post SCU, and worsening when the patient had progression of the main symptom and/or developed a new symptom.

Pre-operative and 6—12-month postoperative UDS data were collected. The following parameters were examined: total cystometric bladder capacity (TCBC), intravesical pressure at TCBC, detrusor leak point pressure (DLPP), compliance at TCBC and at 75% bladder capacity, uninhibited bladder contractions (UC), detrusor sphincter dyssenergia (DSD), and the percentage of change of bladder capacity before and after SCU. The percentage of change of expected bladder capacity was compared to the percentage of change of actual bladder capacity before and after SCU in order to eliminate the false impression of improvement related to the natural growth of the bladder. The expected bladder capacity (EBC) was calculated using this formula [13]:

$$(24.5\ (\text{age}) + 62)$$

Categorical data were compared using the Fisher exact test and continuous variables were compared using the Wilcoxon Signed Rank test. A *P*-value of <0.05 was considered significant.

Results

Of the 72 patients reviewed with TSCS, 22/72 (eight males and 14 females) met the inclusion criteria. The median age at time of SCU was 11.5 months (range 3—211). The median age at time of first follow-up UDS after SCU was 22 months (range 9—218). The median age at time of last follow-up was 153.5 months (range 65—228). The median follow-up time was 71 months (range 60—192). Of the 50 excluded patients, 26 had secondary TSCS, 22 had missing pre and/or post SCU UDS, and two were lost to follow-up.

The indications for SCU were: urological in 3/22 (13.6%) and neuro-orthopedic in 11/22 (50%). Prophylactic SCU was performed in 8/22 (36.4%) asymptomatic patients with unfavorable UDS in the form of small capacity, poor compliance and high pressure at TCBC. Patients without clinical symptoms or 'unfavorable' UDS were not considered for SCU.

The brain and spine MRI showed that 16/22 (72.7%) had lumbosacral and 6/22 (27.3%) had thoracolumbar spinal cord dysraphism. Cord tethering was due to lipomyelomeningocele in seven, cord lipoma in seven, lipomatous thick filum in five, and diastematomyelia in three patients. Among these, 3/22 (13.6%) had a Chiari type-2 malformation, and two of them required a ventriculoperitoneal shunt. A total of 5/22 (22.7%) had spinal cord syrinx, of whom, 2/5 (40%) required syrinx drainage.