Filum Section for Urinary Incontinence in Children with Occult Tethered Cord Syndrome: A Randomized, Controlled Pilot Study

Paul Steinbok,*,† Andrew E. MacNeily,† Alexander R. Hengel, Koroush Afshar, Jeanne M. Landgraf, Walter Hader and Jeffery Pugh

From the Division of Neurosurgery (PS, ARH) and Department of Urologic Sciences (AEM, KA), University of British Columbia and British Columbia Children's Hospital (AEM, KA), Vancouver, British Columbia and Divisions of Neurosurgery, University of Calgary and Alberta Children's Hospital, Calgary (WH) and University of Alberta and Stollery Children's Hospital, Edmonton (JP), Alberta, Canada, and HealthActCHQ (JML), Boston, Massachusetts

Purpose: Occult tethered cord syndrome, in which there is normal neuroanatomic imaging despite clinical and urodynamic evidence of neuropathic bladder behavior, is controversial. Several uncontrolled series describe improvement in bladder function following section of the filum terminale. We performed a pilot randomized, controlled study comparing medical treatment to surgical section of the filum plus medical treatment in children with occult tethered cord syndrome.

Materials and Methods: Children refractory to standard medical management for 1 year or more with normal conus position on magnetic resonance imaging and abnormal urodynamics were randomized. Exclusion criteria included any neurological conditions, spinal dysraphism, bladder outlet obstruction and an atonic bladder. Patients were assessed at randomization and 1 year later with a standardized urodynamic score, the validated PEMQOL (Pediatric Enuresis Module on Quality of Life™) scale, and a validated bowel and bladder dysfunction score.

Results: After 8 years we accrued 21 patients. The bowel and bladder dysfunction score improved in the surgical and medical arms (20% and 24%) and the urodynamic score improved slightly (6% and 4%, respectively). The PEMQOL Child and Family Impact Scales improved modestly in both groups. All differences were nonsignificant. Interim analysis indicated that more than 700 patients in each arm would be required to demonstrate a statistical difference with respect to urodynamic score based on our preliminary data.

Conclusions: There appears to be no objective difference in urological outcome between medical management plus or minus filum section for patients with occult tethered cord syndrome. These data challenge the existence of the concept of occult tethered cord syndrome, in which bowel and bladder dysfunction score is attributed to tethering by the filum despite a normally located conus.

Key Words: urinary bladder, neural tube defects, urinary incontinence, cauda equina, questionnaires

Abbreviations and Acronyms

BBD = bladder and bowel dysfunction
BBDs = BBD score
EV = expected volume
MRI = magnetic resonance imaging
OTCS = occult TCS
QOL = quality of life
REB = research ethics board
TCS = tethered cord syndrome
UDS = urodynamics

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* Correspondence: Division of Pediatric Neurosurgery, British Columbia Children's Hospital, 4480 Oak St., Room K3-159, Vancouver, British Columbia, Canada, V6H 3V4 (telephone: 604-875-2094; FAX: 604-875-3109; e-mail: psteinbok@cw.bc.ca).

† Equal study contribution.

TETHERED cord syndrome secondary to a tight filum terminale is well recognized as a clinical problem in children and adults.[1–3] Pathogenesis is thought to be traction on the lower end of the spinal cord by a thickened

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filum, causing decreased blood flow in the conus.[4] Clinical features include neurological, musculoskeletal and urological abnormalities, which are often reversed or improved by filum transection. TCS is typically associated with a low-lying conus ending below the L2 vertebral level. Thickening of the filum is thought to result from abnormal development during secondary neurulation. Other evidence of spinal dysraphism includes common associated findings, such as midline lumbosacral cutaneous lesions.

More recently the concept of TCS in which the conus was not low, that is OTCS, has been proposed. Khoury et al first reported on 31 children who had detrusor overactivity unresponsive to conservative management and radiographic examinations that showed a normally located conus.[5] After filum section urological symptoms significantly resolved in 70% of patients. There followed a number of publications of children with urological findings suggestive of TCS and the conus in a normal position on neuroimaging (MRI or computerized tomography myelography).[6–13] Filum section resulted in clinical improvement in 60% to 97% of these children with OTCS. However, all studies were uncontrolled, lacked any objective measure of incontinence and often did not contain supporting urodynamic data.

Adding to the difficulty of drawing conclusions from the literature is the fact that urinary incontinence, considered the clinical diagnosis of BBD, is common in school-aged children, representing 20% to 40% of office visits to pediatric urologists.[14,15] These symptoms often improve gradually with time.[16]

We performed a pilot randomized, controlled study comparing medical treatment only to surgical section of the filum plus medical treatment in children with OTCS.

METHODS

Our primary hypothesis was that filum section plus standard medical therapy would result in reliable, clinically significant improvement in 2 markers of incontinence within 12 months of surgery compared to patients who received only medical therapy according to 1) physiological markers of urinary incontinence as measured by a UDS score and 2) QOL as measured by a validated, enuresis specific QOL scale. Our secondary hypothesis was that filum section would improve voiding function as measured by a decreased BBDs.[17]

Study Population

The population comprised children 5 to 18 years old who presented with refractory primary or secondary daytime urinary incontinence at least 1 year in duration. Centers were eligible to participate if they had a qualified pediatric urologist and pediatric neurosurgeon, and obtained institutional REB approval. The Appendix shows the study timeline.

Study Inclusion and Exclusion Criteria

Urological. We used certain urological inclusion criteria, including 1) primary or secondary daytime urinary incontinence that persisted during 12 months of medical treatment at the discretion of a pediatric urologist, including a combination of timed voiding, biofeedback, anticholinergics, α-antagonists, laxatives and prophylactic antibiotics. An abnormal voiding diary compiled for 3 weeks was used to confirm BBD as defined by 2 or more episodes of incontinence per week despite compliance with scheduled voids and stools, and medication/dietary recommendations.

2) A normal renal/bladder ultrasound was required. If ultrasound showed more than minimal bladder thickening (greater than 3.0 mm at 50% or greater expected capacity), a voiding cystourethrogram was required to exclude bladder outlet obstruction.[18]

3) Abnormal UDS testing was defined by any 1 of certain criteria detailed, including low bladder capacity as expected for age,[19] abnormal bladder compliance as determined by standard formulas,[20] detrusor overactivity as evidenced by an involuntary pressure increase during the filling phase associated with urgency[21] and abnormal bladder sensation with volume at first sensation less than 20% of expected bladder capacity (table 1).

UDS were performed with euthermic fluid using a fill rate of 10% of predicted capacity per minute. The study was repeated twice at the same setting and the more normal study was included in the trial. Anticholinergics and α-antagonists were discontinued 48 hours before UDS. Rectal monitoring of intra-abdominal pressures was done. Patients were awake and cooperative during study. Sedative/hypnotic agents were not administered.

Urological exclusion criteria included 1) bladder outlet obstruction demonstrated on voiding cystourethrogram and/or cystoscopy, 2) alternative diagnoses known to be associated with neuropathic bladder dysfunction (eg spinal dysraphism, spinal cord injury, cerebral palsy or other traumatic brain injury), 3) anorectal malformations, 4) alternative urological diagnoses with other defined treatment options besides further medical therapy as described, not including filum section (eg radiation cystitis, Eagle-Barrett, etc), 5) insufficient mental capacity to gain continence within 1 year, 6) noncompliance with medical management, 7) unwillingness to undergo initial and followup UDS, and 8) UDS evidence of an atonic bladder, defined as capacity greater than 125% of EV.

Radiological. Radiological inclusion criteria included 1) a normally positioned conus on spinal MRI (above the inferior L2 end plate), 2) any size filum, 3) any amount of fat in the filum, 4) terminal syringomyelia of less than 1 bony level was acceptable and 5) lumbar bifid spinal lamina was acceptable. Radiological exclusion criteria included 1) recognized spinal dysraphism such as lipomyelomeningocele, myelomeningocele, dermal