Original Paper

Pediatric Neurosurgery

Pediatr Neurosurg 2004;40:51–57
DOI: 10.1159/000078908

Occult Tight Filum terminale Syndrome: Results of Surgical Untethering

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Key Words
Filum terminale syndrome · Tethered cord syndrome · Occult spinal dysraphism · Spine · Spina bifida · Enuresis · Encopresis · Scoliosis

Abstract
The entity of an occult tight filum terminale syndrome, characterized by clinical findings consistent with a tethered cord syndrome, but with the conus ending in a normal position, has been recognized recently. The indications for sectioning the filum terminale in this situation are not well characterized and are controversial. We report a retrospective review of a consecutive series of 60 children (ages 3–18 years) with a diagnosis of occult tight filum terminale syndrome who underwent section of the filum and were followed for more than 6 months (mean 13.9 months). The criteria for surgical intervention were (1) spina bifida occulta, (2) progressive bladder instability unresponsive to conservative measures, (3) urological/nephrological evaluation to confirm or rule out nonneurogenic etiology, and (4) two or more of the following: (a) bowel involvement (fecal incontinence or chronic constipation), (b) lower extremity weakness, (c) gait changes, (d) reflex/tone abnormalities, (e) sensory disturbances, (f) back/leg pain, (g) orthopedic abnormalities/limb length discrepancy, (h) scoliosis/lordosis, (i) recurrent urinary tract infections, (j) abnormal voiding cystourethrogram/ultrasound, (k) syringomyelia, and (l) neurocutaneous stigmata. Postoperatively, urinary incontinence/retention showed complete resolution in 52%, marked improvement (>95% resolution) in 35%, moderate improvement (>75%) in 6%, minimal improvement (> 50%) in 6%, and no improvement (<50%) in 2%. Fecal incontinence completely resolved in 56%, improved in 41%, and was unchanged in 3%. Weakness, sensory abnormalities, and pain improved or resolved in all patients.

Introduction

The entity of a tethered cord syndrome, resulting from a short, thickened filum terminale, associated with the conus medullaris in a low position is well recognized [1]. The conus is considered to be 'low' when the tip is below the L2 vertebral body. The clinical features of the syndrome include neurological, musculoskeletal and urological abnormalities, which are often reversed or improved by section of the filum. The concept of a tethered cord syndrome in which the conus was not low was first proposed in 1990 in a report by Khoury et al. [10]. They reported 31 patients, who had unstable bladders, unre-

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1016–2291/04/0402–0051$21.00/0

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sponsive to conservative management, in whom radiographical examination showed spina bifida occulta, but a conus in normal position. The clinical symptoms of neurogenic bladder dysfunction, similar to those of patients noted to have the conus in an abnormally low position, led to a tentative diagnosis of tethered cord syndrome. The fila were sectioned and significant postoperative resolution of symptoms occurred in the majority of the patients. Since then, there have been a number of reports of clinical series of children with clinical findings suggestive of a tethered cord syndrome, but with the imaged conus in a normal position, in whom section of the filum terminale has resulted in improvement in the clinical findings [3–7]. Almost without exception, the primary clinical abnormality in these cases has been urinary incontinence, which has been neurogenic in origin. In some series, additional manifestations of occult spinal dysraphism, such as a cutaneous abnormality or a fatty filum, have had to be present to justify surgical intervention [6, 7]. However, in other series, the presence of a neurogenic bladder alone has been enough to precipitate surgical section of the filum [3–5]. The indications for section of the filum in the context of a child with neurogenic urinary dysfunction have become an area of controversy.

The purpose of this study was to review the outcomes of a relatively large series of children presenting with neurogenic urinary incontinence, in whom the filum terminale was sectioned, despite the radiographic findings of a normally positioned conus and a normal-appearing filum.

Methods

A retrospective case review was performed in children operated on with section of the filum terminale for an 'occult tight filum terminale syndrome' between 1998 and 2000 by a single neurosurgeon (M.W.). Children with clinical features of a tethered cord syndrome, but with a conus below L2–L3 on magnetic resonance imaging (MRI) or radiographically apparent tethering entities, were excluded. Patients were referred for neurosurgical assessment after urological/nephrological evaluation had revealed a neurogenic origin of urinary instability. Conservative therapies, including behavioral incentives, strict timed voiding regimens, kegel exercises (pelvic musculature contractions), and anticholinergic therapy had failed prior to neurosurgical evaluation. Urological evaluation routinely included an abdominal radiograph (KUB), renal/bladder ultrasound with pre- and postvoid assessment, and voiding cystourethrogram (VCUG) to document urological function. Primary incontinence was defined as inability to toilet-train by the age of 5 years, and secondary incontinence was defined as that recurring after a period of at least 6 months without incontinence. Spina bifida occulta in the lumbosacral area was seen on all included patients.

Table 1. Surgical criteria

1. Progressive bladder instability (continued worsening of frequency and/or severity of symptoms over time) despite conservative and pharmacological therapy
2. Spina bifida occulta identified on plain spine radiograph and MRI
3. Urology/nephrology evaluation to rule out non-neurogenic etiology
4. Two or more of the following:
   - Bowel involvement (fecal incontinence, chronic constipation, or constipation documented on KUB)
   - Lower extremity weakness
   - Gait changes
   - Reflex/tone abnormalities
   - Sensory disturbances
   - Back/leg pain
   - Scoliosis and/or exaggerated lumbar lordosis
   - Orthopedic abnormalities (pes cavus, equinovarus) and/or limb length discrepancy
   - Recurrent UTIs
   - Abnormal VCUG/US
   - Syringomyelia
   - Neurocutaneous stigmata (dimple, hypertrichosis, cutaneous hemangioma)

Surgical criteria are listed in table 1. Urinary incontinence was assessed by the patient's and family's count of episodes of incontinence on a daily and weekly basis. In addition, the volume of urine lost with each episode of incontinence was categorized as dribbling/wet underwear, wet outer clothes, or full volume loss. The postoperative results were recorded as the percentage decrease in episodes of incontinence. Fecal incontinence was recorded as number of episodes.

Muscle strength was assessed using the Royal Medical Research Council of Great Britain strength-grading scale. Lower extremity muscle testing included extensor hallucis longus, anterior tibialis, gastrocnemius, quadriceps, hamstrings, and iliopsoas muscles, as well as ability to toe and heel walk. Gait changes were assessed by direct observation of the child walking and/or running, tandem walk, heel walk, toe walk, and history of gait/coordination problems noted by the parents/PE instructors/physical therapists.

Sensation was measured by pinwheel, light touch, and joint position sense. Back pain was assessed by character, frequency, location, and duration of complaint, as well as exacerbating and relieving factors. Scoliosis was measured on pre- and postoperative standing spine radiographs. Urinary tract infections were documented by primary care physicians, urologists, or nephrologists. Follow-up appointments were scheduled at 1 week, 1 month, 3 months, 6 months, and 1 year after surgery.

The operative procedure was consistent and involved a lumbar flavotomy and section of the filum terminale. The procedure was performed through a midline incision of <2.5 cm over the lower lumbar spinous processes, typically at L4–L5. Following the exposure of the laminae, the interspinous ligaments and the inferior portion of the spinous process of L4 were removed. After removal of the ligamentum flavum, it was often possible to perform the procedure through

the interlaminar space, with no need for a laminotomy. Using the operative microscope, a midline dural opening of approximately 8 mm was performed. The filum, which was typically located dorsally in the thecal sac, was separated from the cauda equina, tested with a low electrical current, coagulated and divided sharply. The dura was then closed in a watertight fashion. Entrance of epidural blood into the thecal sac was avoided to minimize the possibility of arachnoiditis. The layers were then closed.

A Foley catheter, placed after induction of anesthesia, was maintained in the immediate postoperative period to keep the incision site clean and dry. The child was kept flat until the first postoperative day to minimize the risk of CSF leak/pseudomeningocele and postoperative headache. The child was discharged home on the first postoperative day.

Results

There were 66 children who met the study criteria. Of these, 6 were lost to follow-up and were excluded from the evaluation of the outcome. The remaining 60 children were followed for a mean of 13.9 months (range 6–37 months). The 60 children comprised 22 males and 38 females with an age range from 3 to 18 years (mean 8.0 years). All but 3 patients were over 5 years of age at the time of surgery. One 3-year-old child had been in urinary retention with a suprapubic catheter in place for 7 months prior to surgery. Two children were between 4 and 5 years, and both presented with secondary bowel and bladder incontinence.

In keeping with the standardized criteria for surgical intervention, as outlined in the methods, trials of conservative therapies for bladder instability had proved ineffective in all cases, and radiological studies of all children indicated spina bifida occulta of the lumbosacral spine.

Neurological evaluation showed a variety of symptoms and signs (table 2), including urinary instability, bowel involvement, lower extremity weakness, back/leg pain, gait changes, reflex/tone abnormalities in lower extremities in relation to upper extremities, and sensory disturbances. In 82% of patients, there were 3 or more category 4 surgical criteria (table 3). Only one child had fewer than the 2 category 4 criteria required for surgical intervention as part of the standardized protocol. This child had radiographic evidence of neurogenic bladder on urological evaluation and complete resolution of urinary incontinence within the first postoperative week.

Intraoperatively, the filum was found to be tight, dorsal in location in all cases and often with excess tissue, frequently lipomatous material. The filum was typically visible prior to opening the dura because of its dorsal location in the thecal sac, and the tension was evident by its location and in elevating the filum with a nerve hook. Upon sectioning, the two free ends of the filum retracted out of site within the thecal sac. The filum was photographed through the operating microscope for documentation, but pathological specimens were not routinely sent. No operative complications were encountered.

Postsurgical results (table 4) are as follows: Urinary urgency/frequency resolved or improved in all affected children (58/58). Urinary incontinence/retention resolved or improved to varying degrees in 53/54 children. Urinary

Table 2. Presentation of symptoms and signs

	Patients
Symptoms	
Urinary instability	60 (100%)
Urgency/frequency	58 (97%)
Incontinence	54 (90%)
Primary	7/54 (13%)
Secondary	47/54 (87%)
Retention	1 (2%)
Chronic constipation	33 (55%)
Fecal incontinence	32 (53%)
Back/leg pain and leg cramps	30 (50%)
Nocturnal enuresis	28 (47%)
Gait changes	24 (40%)
Recurrent UTI	16 (27%)
Signs	
Reflex abnormalities	49 (82%)
LE hyporeflexia	8/49 (16%)
LE hypereflexia	41/49 (84%)
Weakness	21 (35%)
Abnormal VCUG/US	15 (25%)
Muscle tone abnormalities	14 (23%)
Sensory disturbances	9 (15%)
Cutaneous stigmata	7 (12%)
Scoliosis/lordosis	6 (10%)
Orthopedic/limb discrepancy	5 (8%)

Table 3. Category 4 criteria met at presentation

Category 4 criteria	Patients
1	1 (2%)
2	10 (17%)
3	12 (20%)
4	24 (40%)
5	10 (17%)
6	3 (5%)

Table 4. Postoperative results

Postoperative results	Patients/total
Urinary instability	
Urgency/frequency	
Resolved/improved	58/58 (100%)
Incontinence/retention	
Resolved	28/54 (52%)
Improved	25/54 (46%)
Near-complete resolution (>95%)	19/54 (35%)
Marked improvement (>75%)	3/54 (6%)
Moderate improvement (>50%)	3/54 (6%)
Unchanged (<50%)	1/54 (2%)
Fecal incontinence	
Resolved	18/32 (56%)
Improved	13/32 (41%)
Unchanged	1/32 (3%)
Nocturia	
Resolved	13/28 (46%)
Improved	7/28 (25%)
Unchanged	8/28 (29%)
Recurrent UTI	
Resolved/improved	11/16 (69%)
Unchanged	5/16 (31%)

incontinence/retention showed complete resolution in 52% (28/54); marked improvement (greater than 95% resolution) in 35% (19/54); moderate improvement (greater than 75% resolution) in 6% (3/54); minimal improvement (greater than 50% resolution) in 6% (3/54), and no significant improvement in 2% (1/54). Fecal incontinence resolved in 56% (18/32), improved in 41% (13/32), and was unchanged in 3% (1/32).

Although nocturnal enuresis was not considered one of the criteria for surgery, results were recorded. There was resolution in 46% (13/28), improvement in 25% (7/28) and no change in 29% (8/28). Recurrent urinary tract infections (UTI) resolved (no further infections during time of postoperative follow-up) in 69% (11/16), but continued to occur in 31% (5/16).

Weakness, when present preoperatively, resolved in 21/21 patients. Weakness was most commonly seen in extensor hallucis longus, hamstrings, anterior tibialis and gastrocnemius, and was typically asymmetrical. Sensory abnormalities, consisting primarily of hyperesthesia or hypesthesia to pinprick in distal lower extremities and/or saddle distribution, resolved on postoperative examination in 9/9 patients. Back/leg pain/cramps frequently awakened the child at night, requiring massage or medication. Daytime symptoms were typically worse with sitting or standing for a prolonged period of time, and occasionally exacerbated with prolonged activity. These complaints improved or resolved in all patients (30/30). On follow-up radiographs at the 3- or 6-month follow-up appointment, scoliosis improved in 1 child with syringomyelia, and was unchanged in 3 others without syringomyelia. Formal gait analysis was not performed, but pre- and postoperative clinical examination, including tandem walk, toe and heel walk, and direct observation, as well as patient reports showed improvement in gait in all affected children (24/24). No child had worsening of symptoms.

Improvements were noted as early as the first postoperative week in many children, and nearly all were experiencing improvements by 1 month. There appeared to be three patterns of recovery; (1) immediate dramatic improvement, (2) gradual steady improvement over weeks and months, (3) initial dramatic improvement, followed by exacerbation of symptoms several weeks to months postoperatively but not to the preoperative severity, followed by continued improvement. No correlation was found between age, duration, or severity of symptoms and outcome.

Only one child showed no significant improvement in urinary or fecal incontinence. This was a 10-year-old female with a 5-year history of bowel and bladder incontinence and urinary tract infections, who had toilet trained at 4 years of age. All the 3 children who had only minimal improvement (greater than 50%) in urinary incontinence had primary urinary incontinence. One of these 3, an 8-year-old male, also had fecal incontinence. The other 2, both female, aged 5 and 6 years, had recurrent urinary tract infections and constipation. No child had worsening of symptoms.

Discussion

The relationship between the syndrome of the tethered spinal cord and a variety of forms of spinal dysraphism is well described [1–3, 6, 8–10]. For patients with urological dysfunction, early surgical treatment has been associated with better outcomes [11–17]. Increasing urodynamic difficulties in older children with urinary dysfunction suggest that the problem may be progressive, and if surgery is a treatment option, this argues for early rather than delayed surgical intervention [2, 14, 18, 19]. Urinary instability may be among the first distinguishing symptoms of neurological damage caused by occult spinal dysraphism [2]. This underscores the importance of urologi-

cal evaluations to identify a neurogenic etiology, as other clinical indicators may not be apparent at early ages. Treatments for urinary instability typically include conservative managements, such as behavioral incentives, timed voiding regimens, kegel exercises, and anticholinergic therapy. If these treatments are unsuccessful, and a nonneurogenic etiology (structural anomaly, obstruction, infection, etc.) has been ruled out, a neurogenic etiology is probable, and the possibility of a tethered cord syndrome should be entertained.

In our patient population, conservative urological management had failed in all cases. These children were observed to have progression of their urinary dysfunction over time, up to the time of surgical intervention. A significant number of the children had additional neurological findings, such as motor, reflex or sensory changes. The clinical picture in these patients strongly suggests, but does not prove, that the urinary dysfunction was of neurogenic etiology. Nonetheless, it is clear that the preoperative condition represented the nadir in their continence.

In this series, because progressive urological instability was only one (and not always the most prominent) of multiple criteria used in patient selection, formal urodynamic assessments were not routinely done. The usefulness of formal urodynamics in this population remains controversial among pediatric urologists, and the ability of centers to perform them and obtain reliable information is extremely variable. While urodynamic testing may be helpful in the evaluation of children with more severe urological involvement, for example myelomeningocele, we have not found it helpful in this patient population. It has been shown that many patients who complain of urinary urge incontinence do not have unstable contractions on routine office cystometry [20]. However, when examined with ambulatory monitoring, between 40 and 84% have instability [21]. Most urologists feel that a patient reporting sudden leakage with urgency should be assumed to have detrusor instability even if it is not present on the cystometrogram [20]. Because normal urodynamics does not rule out a neurogenic bladder, we have found a careful and detailed history in conjunction with KUB, ultrasound, and/or VCUG to be satisfactory. Our current practice is to perform formal urodynamics only when a neurogenic bladder is suspected in the absence of other corroborating evidence of tethered cord, requiring that the diagnosis must be based solely on urological function.

The outcomes after section of the filum in this group of patients have been very positive. Khoury et al. [10] reported on 31 patients who had unstable bladders unresponsive to conservative management, in whom there was spina bifida occulta but a normal conus, with 72% having resolution of daytime incontinence. Warder and Oakes [6, 7] also sectioned the fila in 13 patients in whom the conus was in a normal position. They restricted operative intervention to those having clinical findings of a tethered cord syndrome and, in addition, had other indicators of spinal dysraphism, such as cutaneous lesions or a fatty filum. Surgical intervention resulted in improvement or stabilization of the majority of the presenting complaints. Three of the 4 with bladder dysfunction improved, and all 3 with bowel dysfunction improved. Nazar et al. [3] reported on 32 pediatric patients presenting with symptoms associated with tethered cord syndrome, including urinary dysfunction, but with the conus in normal position, and found that 97% of patients experienced significant relief after filum section. In these patients the diameter of the filum terminale was <2 mm and there were no other stigmata of occult spinal dysraphism. Selcuki et al. [5] also sectioned the fila in 17 children with clinical and radiological features similar to those reported by Nazar et al. [3]. In all cases the diagnosis of a neurogenic bladder was confirmed by urodynamic testing. Long-term benefit from surgery, with improvement in continence, was seen in 76.5%. Palmer et al. [22] presented 8 children with day and/or night wetting, UTIs and encopresis with spina bifida occulta and a neurogenic bladder (documented by urodynamics) but a normal neurological and urological exam (including cystoscopy) and MRI. By 6 months after section of the filum, all had complete resolution of nocturnal enuresis and stool incontinence. Diurnal enuresis was completely resolved in 57% or markedly improved in 43%.

In the past, the diagnosis of a tight filum terminale as the cause for a tethered cord syndrome required MRI findings of a low conus, below the level of L2. Our observations, and those of others, as noted above, indicate that a tight filum terminale can result in the clinical tethered cord syndrome in the absence of a low conus, and that section of the filum in these cases may be beneficial. Selcuki et al. [23] recently reported that fila in these patients contained histological features that may reflect decreased elasticity resulting in a tethering effect. The 'normal-appearing' fila in these patients had elevated amounts of connective tissue, dense collagen fibers, hyalinization, and dilated capillaries. These findings were clearly distinct from those in the control group, in whom the fila were chiefly made up of connective tissue around the ependymal canal.

Other findings on MRI, such as a thickened, and particularly a fatty filum, have been used as one of the criteria for surgical intervention in this group of patients.

Palmer et al. [22] have suggested that visualization of a normally sized and nonfatty filum terminale on axial images as a discrete midline moderate intensity signal structure might correlate with clinical improvement. Cine MRI to evaluate cord traction by measuring the degree of motion apparent with each pulsation has been used in evaluating tethering of the cord in children with lipomyelomeningocele and myelomeningocele [24].

One of the findings in our patient population was a high incidence of neurological abnormalities, including motor, sensory and reflex changes. Clinical evaluation of these children requires a meticulous and time-consuming neurological evaluation, without which neurological abnormalities may be missed [25]. In children with progressive urinary instability, the finding of any neurological or skeletal abnormalities in the lower limbs increases the probability that a tethered cord is the causal factor in the urinary dysfunction.

Daytime urinary incontinence is commonly encountered in school-aged children, with studies reporting a prevalence of 8–16% [26–28]. However, Sureshkumar et al. [27] found that, while 16.5% of 1,419 children with a mean age of 5.9 years reported one or more episodes of wetting in the past 6 months, only 2% wet twice or more per week and 0.7% wet every day. Children in our study typically suffered multiple episodes of incontinence per week, with many having one or more per day. Studies show that the prevalence decreases as children get older [12, 28], but urinary incontinence may persist into adulthood.

The psychological effects of incontinence should not be underestimated. In a study ranking fears in children and adolescents, wetting pants at school was the third most catastrophic event that could occur, surpassed only by death of a parent or becoming blind [29].

Many school-aged children with refractory daytime incontinence unresponsive to conventional conservative therapy (behavioral techniques, anticholinergics, timed-voiding, etc.) are referred by their pediatrician to a pediatric urologist. The presence of spina bifida occulta, along with either prolonged primary or secondary enuresis, bowel problems (encopresis/constipation), worsening of symptoms over time, exacerbation of symptoms during growth spurts or failure of conservative management, results in the referral of approximately 1 out of 20 of those refractory incontinent children seen by the urologist of this study (P.O.) to the pediatric neurosurgeon.

The incidence of urinary incontinence in young, healthy patients is surprising. A study of 4,200 healthy, nulliparous nursing students, 17–25 years of age, reported a 51% incidence of sporadic episodes of incontinence, with 16% having major or regular urine loss [11, 16]. Three studies dealing with incontinence in males <30 years of age reported prevalences ranging from 6.1 to 10% [11, 15, 30, 31]. In a study of 20,000 patients >5 years of age in a general medical practice, 27.6% of women and 10.8% of men surveyed reported urinary incontinence [15]. More than 15% of women and 3% of men from each decade of life were affected. Interestingly, incontinence was noted in the practice records of less than 0.1% of these patients. In a recent study, urinary incontinence was reported in 26% of Japanese women under 30 years of age and in 40% over 40 years of age [32]. However, only 4% of women with incontinence discussed this with their physicians, citing shame and resignation as major reasons. Patients tend not to discuss the issue of incontinence because of embarrassment and low expectations for treatment, even though it has a profound impact on many of them [33]. Findings of such a high prevalence of idiopathic incontinence in the young, nulliparous adult population [16] raises the question of etiology. It has been postulated in the past that 'idiopathic' urinary instability results from an occult central or peripheral nerve disorder [21]. Perhaps the persistence or exacerbation of childhood incontinence is far more pervasive than commonly believed.

In the light of the high incidence of urinary incontinence in the population and the social implications of this issue, it is important that rational and evidence-based criteria be developed for selection of those patients with urinary incontinence who might benefit from section of the filum terminale. Should any patient with neurogenic urinary incontinence be offered surgery to section the filum as per Nazar et al. [3], Selcuki and Coskun [4], and Selcuki et al. [5], or should this be offered only in the presence of other findings such as a thickened, fatty filum or cutaneous stigmata of spinal dysraphism, as per Warder and Oakes [6, 7]? Is it necessary to put children through urodynamic testing to prove the presence of a neurogenic bladder, even though it has been shown to be unreliable, if other evidence of tethered cord is present [20, 21]? Using our surgical criteria (table 1), there was improvement in 97% of patients in this series, and one might argue that these surgical selection criteria may have been too restrictive. Because there are many children with urinary incontinence, patient selection is critical. By requiring other evidence of tethered cord before proceeding with surgery, we hoped to select only children who were most likely to benefit and to avoid unnecessary surgery.

Conclusion

The tethered cord syndrome often presents with progressive urinary instability and is ultimately a clinical diagnosis. Although the radiographic finding of a low conus has traditionally been considered a necessity in the indications for surgery, lack of such findings does not rule out the syndrome. There is much controversy about the indications for surgery in patients with clinical features consistent with a tethered cord syndrome, but with a conus in normal position. The positive results of this study, in which a consistent set of clinical criteria was used to indicate which children with bladder instability might benefit from having the filum terminale sectioned, support an aggressive approach to clinical diagnosis and treatment of this population.

Acknowledgements

Many thanks to Joanne Wallis, P.N.P. and Colleen Phillips, P.A.-C. for their assistance in accumulating these patient data, Shannon Richards CRN-FA for her technical assistance, and to Kathleen Thomas for lending us her literary talents in preparing the manuscript. We also wish to thank Dr. Paul Steinbok for reviewing the manuscript and offering helpful suggestions.

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