Personal practice

Urge syndrome and urge incontinence

J D VAN GOOL* AND G A DE JONGE†

*Department of Paediatric Nephrology, University Hospital for Children and Youth, Utrecht, and

†Department of Paediatrics, Vrije Universiteit, Amsterdam, The Netherlands

Diurnal enuresis is still the common denominator for children who wet their pants. A closer look at children with daytime wetting, however, shows that there are two distinct categories: one with classic diurnal enuresis and normal bladder function, and the other with urinary incontinence caused by an 'urge syndrome', with bladder sphincter dysfunction, and a conspicuously high incidence of recurrent urinary tract infections.

Overactivity of the detrusor and urethral muscles is now an established cause for bladder sphincter dysfunction in children, and it is usually associated with symptoms such as urgency, frequency, daytime wetting, and urinary tract infections.^{1–5} The symptoms of dysfunction induced by overactivity have in the past been blamed on either the detrusor muscle or the urethral closure mechanism, resulting in two different concepts of diagnosis and treatment.⁶ Cystometric findings of uninhibited detrusor activity in children with recurrent urinary tract infections and urge syndrome were interpreted as persistent infantile bladder, occult neuropathic bladder, or isolated or subclinical neurogenic bladder. Accordingly, treatment consisted of rehabilitation programmes and anticholinergic drugs. Cystoscopy and voiding cystourethrography, on the other hand, showed abnormalities that were labelled as wide bladder neck anomaly or 'spinning top' urethra, distal urethral stenosis, external sphincter spasticity, or sphincter dyssynergia, popularising the concepts of urethral dysfunction and stenosis, and of urethral dilatation or urethrotomy as methods of treatment.

Urodynamic studies showing the interaction between bladder and sphincter muscles by simultaneous registration of bladder pressure and urethral sphincter activity at first added even more labels to the two controversial concepts, and widened the range of possible treatments. Recent reviews^{7,8} support the idea that most labels can be brought down to patterns of bladder sphincter overactivity: in one pattern, strong uninhibitable detrusor con-

tractions give rise to urgency and frequency early in the filling phase; in the other pattern, incomplete relaxation of striated urethral and pelvic floor muscles interrupts actual voiding and causes a *staccato* urinary flow.

As these patterns of bladder sphincter overactivity are detectable with urodynamic techniques we decided to investigate prospectively a selected group of children of school age whose presenting symptoms were persistent daytime wetting, urgency, and frequency.

Patients

Ninety girls and three boys with urge incontinence and recurrent urinary tract infections were studied. They were referred after the failure of previous attempts at treatment to a combined service of the departments of paediatric nephrology and child psychology in our University Children's Hospital. For every child a documented history of urinary tract infections was available; all the children had had full urological investigations (excretory urography and voiding cystourethrography), and all results were available for review. Our part of the comprehensive evaluation consisted of psychological interviews of children and parents, of elaborate history taking with emphasis on voiding habits and urge, and of urodynamic studies.

Each urodynamic investigation consists of simultaneous recording of the bladder pressure, abdominal pressure, electromyographical activity of the external anal sphincter, and urine flow rate. Several recordings are made, both during filling of the bladder and emptying of the bladder, without anaesthesia or sedation.

Bladder filling and registration of bladder pressure are done through one small soft transurethral catheter (Charrière size 6). The catheter has a filling lumen and a microtransducer at the tip (MTC microtip transducer, PPD Hellige). The bladder is

filled with a sterile 0.9% saline solution, the filling rate not exceeding 15 ml/minute. Abdominal pressure is registered with a Charrière size 4 catheter containing a microtip transducer inserted high into the rectum. The electromyographical signal from the anal sphincter (a measure for pelvic floor activity) is picked up with surface electrodes, amplified and monitored with an oscilloscope, and registered on a chart recorder together with the amplified pressure signals. The smallness and ease of operation of the microtip transducers (together with patience on the part of the investigator) are prerequisites for successful paediatric urodynamics, and when the procedure is properly explained, most children tolerate the investigation extremely well.

At or near the end of the filling phase the child is asked to cough, and to change from a supine to a standing or sitting position, so that detrusor instability may be detected. Bladder emptying is recorded with the child in the appropriate voiding position. Abdominal pressure is subtracted electronically from bladder pressure, to differentiate true detrusor activity from effects of abdominal straining. All events are recorded continuously without having to ask the child to use inappropriate 'holding manoeuvres' in the important transition phase between filling and micturition. Methods, definitions, and units conform to the standards recommended by the International Continence Society.

Signs and symptoms

Characteristically, children with the urge syndrome have sudden attacks of uninhabitable urge to void, inducing forceful compression of the urethra to avoid unwanted loss of urine^{2 3 7-14}. Usually the first attacks do not occur before the end of the morning. During the course of the day the attacks increase in number as well as in severity. The syndrome may have a nocturnal component too, as some children also experience sudden urge and loss of urine during the night.

The urge to void comes suddenly and unexpectedly. The sensation is so imperative that loss of urine can only be avoided by maximal compression of the urethra both by forceful contraction of all pelvic floor muscles and by external mechanical compression. Every child has his or her own method of external compression—a common one is squatting on one foot, with the heel of the foot against the perineum. This asymmetrical squat conveys a sense of elegance—it resembles an obeisance and Vincent,⁹ who first described it, termed it the 'curtsey' sign. In assuming this position children often pretend that something was dropped on to the floor, or that a shoelace had become untied. Compression is sus-

tained until the urge to void has passed, which can take up to several minutes.

Most children do not succeed completely (if at all) in countering the imperative need to void with external compression of the urethra, and more often than not some urine will escape. This loss of urine is involuntary and incomplete; it never takes the course of a complete micturition. It is a specific form of incontinence—urge incontinence.

OCCURRENCE ACCORDING TO AGE AND SEX

The urge syndrome is seldom seen in boys. The prevalence in girls aged 4 to 12 years is reported to be 1–3/1000. Parents usually state that symptoms and signs started around age 4 to 5; parental attitudes towards incontinence become less permissive at that age, however, and careful questioning may discover an earlier age for the first occurrence—often in conjunction with the first urinary tract infection.

Around puberty the prevalence starts to decrease, and in adolescence it is low. The 'female urethra syndrome' might be an adult counterpart of the urge syndrome in girls, but prospective studies are not available to prove this postulate.^{15 16}

Fig 1 shows the age distribution for 93 children with urge syndrome all of whom were referred to our department of paediatric nephrology because of incontinence and recurrent urinary tract infections.

URINARY TRACT INFECTIONS

The correlation between urinary tract infections and the urge syndrome is generally accepted, but hard to specify.^{1 2 4 8 17} A first infection may mark the beginning of a long series of recurrent infections and persisting symptoms of the urge syndrome; in other

Fig 1 Age and sex distribution of 93 children with urge syndrome and recurrent urinary tract infections who were referred for cognitive bladder training.

cases, however, recurrent urinary tract infections seem to appear only during the course of the syndrome.

Although urinary tract infections may not constitute the primary cause of the urge syndrome they invariably occur in conjunction with the syndrome, and have a key role in the persistence and severity of its symptoms. It is probably not by chance that the prevalence of recurrent urinary tract infections and urge syndrome in girls have the same age distribution.¹⁸

As a consequence of this correlation, vesicoureteric reflux and reflux nephropathy also have high prevalences in children with urge syndrome.^{6 13 14 19-21} In the 93 children shown in fig 1, vesicoureteric reflux could be documented in 40%, and reflux nephropathy in 30% (table 1). The expected correlation between the grade of vesicoureteric reflux and the prevalence of reflux nephropathy was, however, conspicuously absent.

BEHAVIOURAL PROBLEMS

In children with long standing urge syndromes a set of secondary behavioural problems will develop (table 1). These problems can be so impressive that, at first glance, a child with urge syndrome will be diagnosed as having primary behavioural problems with 'psychogenic' enuresis.⁵ Careful questioning will show that behaviour in children with urge syndrome is usually related to acceptance and understanding of their incontinence problem by peers and parents.²²

CONSTIPATION

An urge syndrome is often associated with persistent constipation, sometimes causing faecal soiling and spurious diarrhoea. Chronic constipation is often associated with urinary tract infections and should be looked for and treated in children with urge syndrome.

Table 1 Incidence of urinary tract infections, vesicoureteral reflux and reflux nephropathy, bladder sphincter function, and secondary behavioural problems in 93 children referred with urge syndrome and incontinence

	No (%)
Urinary tract infections	87 (94)
Vesicoureteral reflux	37 (40)
Reflux nephropathy	28 (30)
Detrusor overactivity	42 (45)
Urethral overactivity	28 (30)
Valsalva manoeuvre	18 (19)
Normal bladder sphincter function	6 (6)
Behavioural problems	12 (13)

It is not clear whether the constipation is caused by the habit of contracting all pelvic floor muscles each time an urge to void is experienced. The association between chronic constipation and urge syndrome, first described by Vincent, merits further investigation.^{9 23}

Urodynamic findings

Urodynamic studies in children with urge syndrome have made it clear that imperative urge to void coincides with uninhibited (unstable) detrusor contractions, occurring early in the filling phase.^{2 3 7 8 13 14} Simultaneous registration of bladder pressure with electromyographical activity of the pelvic floor shows how children suppress the sudden urge by forceful contraction of all pelvic floor muscles, to

Fig 2 Detrusor overactivity countered by pelvic floor contractions (measured by electromyography) in a 5 year old girl with recurrent urinary tract infections and urge incontinence. Detrusor contractions start at bladder volume of 55 ml and loss of urine is prevented by forceful pelvic floor contractions, resulting in high bladder pressures in the filling phase. During voiding bladder pressure remains normal, and after voiding there is a small after contraction. Detrusor pressure is bladder pressure minus abdominal pressure.

cause a reflex inhibition of the contractions of the overactive detrusor muscle (fig 2). The urge to void grows stronger with the amplitude of the detrusor contraction, and in the end (with each contraction of the overactive detrusor muscle against the forcefully closed urethral sphincter) bladder pressure will rise to a high level.

In the presence of vesicoureteric reflux these high pressure waves will be transmitted to the renal pelvis and parenchyma, where they play a crucial role in the pathogenesis of reflux nephropathy—either with or without urinary tract infections.²⁴ The correlation between high bladder pressures and reflux nephropathy in children without anatomical or neurological obstruction of the lower urinary tract has been reported previously^{8 13 14 19} and was confirmed by recent retrospective studies.^{6 21}

A separate entity in children with urge syndrome is the occurrence of urethral overactivity during micturition. Urodynamically it is similar to detrusor sphincter dyssynergia¹³ in neurogenic bladder dysfunction: urine flow is *staccato*, interrupted by short periods or bursts of pelvic floor activity with concomitant peaks in detrusor pressure (fig 3). Some children with urge syndrome have acquired

the habit of voiding with abdominal pressure using the Valsalva manoeuvre: this too generates repeated interruptions of the urine flow through reflex triggering of pelvic floor activity by each rise in abdominal pressure. In these children, cystometric bladder capacity may reach high values and they run the risk of developing a 'lazy bladder syndrome' in spite of the fact that they initially had detrusor overactivity.

Treatment

Long term chemoprophylaxis has a definite place in the management of children with urge syndrome: if recurrences of urinary tract infections can be avoided for a period of six to nine months, the symptoms of imperative urge and urge incontinence will diminish appreciably—in mild cases they may even disappear. The finding of vesicoureteric reflux is of course a primary indication for long term chemoprophylaxis.

Anticholinergic drugs offer little advantage in the long run except in mild cases of the syndrome; symptoms tend to reappear immediately after discontinuation of the drugs. Anticholinergics such as oxybutynin have a more specific action on detrusor overactivity, and they seem excellently suited for a short term trial, or as an adjunct to other forms of treatment.

Muscle relaxants (for example, baclofen), or α -adrenergic blocking agents (for example, phenoxybenzamine), may have a place in the treatment of children with urge syndrome and 'dyssynergic' voiding as they lower overall urethral resistance to flow. Long term effects are less promising, however, in neurologically normal children with urge syndrome than in children with true neurogenic detrusor sphincter dyssynergia.

A more lasting effect can be expected from cognitive bladder (re)training, as many symptoms and signs of the urge syndrome are rooted in a wrong perception of signals from the bladder, and—accordingly—on habitual non-physiological responses to these signals. Children with urge syndrome have to learn again how to recognise the first sensations of urge, and how to suppress these by central inhibition instead of resorting first of all to emergency procedures like urethral compression.

They also have to learn again how to void with a completely relaxed pelvic floor. To achieve this, urodynamic signals such as urine flow rate or pelvic floor electromyogram can be used as feedback.^{6 22 25-27} Feedback during training programmes has been termed 'biofeedback' by Miller: 'The use of modern instrumentation to give a person better moment-to-moment information about a specific

Fig 3 'Dyssynergic' voiding: short bursts of overactivity of striated urethral and pelvic floor muscles (measured by electromyography) interrupt the flow of urine in a 7 year old girl with recurrent urinary tract infections and urge syndrome. Steep rises in bladder pressure coincide with bursts of electromyographic activity and interruptions of urine flow. Detrusor pressure is bladder pressure minus abdominal pressure.

physiological process, that is under control of the nervous system, but not clearly or accurately perceived.²⁸

Since 1980 a programme for cognitive bladder training is being used at the University Children's Hospital in Utrecht,⁶ with the registration of urine flow rate and voided volume serving as the feedback (fig 4). The training is organised as a 10 day course, and children are admitted to hospital where they are supervised by a psychological assistant; the same person does the pretraining interviews, supervises the course, and carries out the follow up. The children are trained in pairs; they are instructed to void at the first sensation of urge, and to use the flow recorder for every voiding. This serves to record the number of voidings each day, and the volumes voided: the number has to decrease and the volume increase. The actual flow curve, displayed directly on a monitor and printed on paper afterwards, tells the children how to void with a smooth uninterrupted flow: 'dyssynergic' voiding or abdominal straining show immediately, and the print-out serves as proof. Medical interventions and diagnostic tests are limited to an absolute minimum during the training course, and an important part of each day is devoted to keep up with schooling.

Long term results with this programme in a group of 93 children (fig 1), all referred for long standing urge syndrome, persistent urge incontinence, and recurrent urinary tract infections, are summarised in table 2. The non-invasive flow rate recordings made during the actual training always confirmed the initial diagnosis of detrusor or urethral overactivity. In 60% of the 93 children detrusor overactivity, 'dyssynergic' voiding patterns, and concomitant urge incontinence disappeared completely without recurrences during the follow up period (3.7 (1.4) years). Nocturnal wetting, when present, also disappeared completely. In 22% of the 93 children a lasting and considerable improvement was achieved in the symptoms of the urge syndrome. Of the 18% that could not be cured, one child had structural

Table 2 Association between results of cognitive bladder training and recurrences of urinary tract infections in 93 children with urge syndrome and incontinence; mean (SD) follow up 3.7 (1.4) years

	Training results			Total
	Cured	Improved	Relapsed	
Urinary tract infections:				
Recurrences	4	12	15	31
No recurrences	52	8	2	62
Total	56	20	17	93

$\chi^2=46.7$; $df=2$; $p<0.001$.

incontinence caused by incision of the bladder neck during an earlier urethrotomy and had to be treated by colposuspension; two children had developed the 'lazy bladder syndrome' and were successfully re-trained after a period of clean intermittent self catheterisation.

In this selective group of 93 children with long standing urge syndrome, recurrent urinary tract infections had a key role, with a prevalence as high as 94%. Of the group 40% had vesicoureteric reflux, and 30% presented with signs of reflux nephropathy (table 1).

During follow up, recurrences of urinary tract infections occurred almost exclusively in conjunction with complete or partial recurrence of urge incontinence (table 2). This correlation between recurrences of urinary tract infections and urge incontinence is significant (χ^2 test, $p<0.001$). Among the children that remained free of urge incontinence during follow up, there were almost no recurrences of urinary tract infections and chemoprophylaxis could safely be discontinued—often after years of recurrent urinary tract infections and persistent daytime wetting, repeated diagnostic and therapeutic procedures such as cystoscopy, urethral dilatation or urethrotomy, and numerous patient months of antibiotic and chemotherapeutic regimens. In some cases the vesicoureteric reflux disappeared.

Conclusions

Despite the fact that urge incontinence is quite different from diurnal enuresis, most children with an urge syndrome are still being labelled as having diurnal enuresis.²⁹ With such a label these children are bound to be referred primarily to general practice, child psychology, or even child psychiatry for diagnosis and treatment. The association of the urge syndrome with urinary tract infections and vesicoureteric reflux warrants a different approach,

Fig 4 Drawing by 7 year old boy of staccato urine flow (fout=wrong) and normal urine flow (goed=good). Visual perception of urine flow helps children during cognitive bladder training, and they also record the number of times they void and the amount.

and it is worthwhile to revise the definition of diurnal enuresis accordingly: incontinence implies abnormal bladder sphincter function, enuresis denotes wetting without abnormalities of bladder sphincter function. Our results with cognitive training indicate that both detrusor overactivity and urethral overactivity are to a certain extent results of inappropriate learning processes, acutely aggravated or triggered by urinary tract infections. Prospective studies will be needed to elucidate further the exact role of urinary tract infections in the pathogenesis of the urge syndrome. The prevalence of uropathogenic *Escherichia coli* with adherence antigens in children with urge syndrome and recurrent urinary tract infections is a promising subject for research.³⁰ Detection of detrusor overactivity and urethral overactivity seems essential in all children with vesicoureteric reflux, because of the contribution of high bladder pressures to reflux nephropathy.^{6 21-24} Much could be learned from the incorporation of bladder pressure recordings in the radiological diagnosis of vesicoureteric reflux as indicated by the lack of correlation between the grade of vesicoureteric reflux at standard cystography and the prevalence of reflux nephropathy in children with urge syndrome.

Part of this study was supported by a grant (C85.564) from the Dutch Kidney Foundation.

References

- Lyon RP, Marshall S. Urinary tract infections and difficult urination in girls: long term follow-up. *J Urol* 1972;**105**:314-7.
- Van Gool JD, Tanagho EA. External sphincter activity and recurrent urinary tract infection in girls. *Urology* 1977;**10**:348-53.
- Allen TD, Bright TL. Urodynamic patterns in children with dysfunctional voiding problems. *J Urol* 1978;**119**:247-9.
- Koff SA, Lapides J, Piazza DH. Association of urinary tract infection and reflux with uninhibited bladder contractions and voluntary sphincter obstruction. *J Urol* 1979;**122**:373-6.
- Hinman F Jr. Nonneurogenic neurogenic bladder (the Hinman syndrome)—15 years later. *J Urol* 1986;**136**:769-77.
- Van Gool JD, Kuijten RH, Donckerwolcke RA, Messer AP, Vijverberg M. Bladder-sphincter dysfunction, urinary infection and vesicoureteral reflux—with special reference to cognitive bladder training. *Contributions to Nephrology* 1984;**39**:190-210.
- Bauer SB, Retik AB, Colodny AH, Hallett M, Khoshbin S, Dyro FM. The unstable bladder of childhood. *Urol Clin North Am* 1980;**7**:321-36.
- Koff SA. Bladder-sphincter dysfunction in childhood. *Urology* 1982;**19**:457-61.
- Vincent SA. Postural control of urinary incontinence: the curtsey sign. *Lancet* 1966;**ii**:631-2.
- De Jonge GA. The urge syndrome. In: Kolvin I, McKeith RC, Meadow SR, eds. *Bladder control and enuresis*. London: Heinemann, 1973:66-9.
- Fielding D, Berg I, Bell S. An observational study of postures and limb movements of children who wet by day and by night. *Dev Med Child Neurol* 1978;**20**:453-61.
- Kondo A, Kato K, Takita T, Otani T. Holding postures characteristic of unstable bladder. *J Urol* 1985;**134**:702-4.
- Griffiths DJ, Scholtmeijer RJ. 'Detrusor-sphincter dyssynergia' in neurologically normal children. *Neurology and Urodynamics* 1983;**2**:27-37.
- Borzyskowski M, Mundy AR. Videourodynamic assessment of diurnal urinary incontinence. *Arch Dis Child* 1987;**62**:128-31.
- Rees D, Whitfield HN, Islam AKMS, Doyle PT, Mayo ME, Wickham JEA. Urodynamic findings in adult females with frequency and dysuria. *Br J Urol* 1975;**47**:853-60.
- Kaplan WE, Firlit CR, Schoenberg HW. The female urethra syndrome: external sphincter spasm as etiology. *J Urol* 1980;**124**:48-9.
- Lapides J, Diokno AC. Persistence of the infant bladder as a cause for urinary infection in girls. *J Urol* 1970;**103**:243-8.
- Winberg J, Bergström T, Jacobsson B. Morbidity, age and sex distribution, recurrences and renal scarring in symptomatic urinary tract infection in children. *Kidney Int* 1976;**8**:S101-6.
- Allen TD. Vesicoureteral reflux as a manifestation of dysfunctional voiding. In: Hodson CJ, Kincaid-Smith P, eds. *Reflux nephropathy*. New York: Masson, 1979:171-81.
- Taylor CM, Corkery JJ, White RHR. Micturition symptoms and unstable bladder activity in girls with primary vesicoureteric reflux. *Br J Urol* 1982;**54**:494-8.
- Nielsen JB, Djurhuus JC, Jørgensen TM. Lower urinary tract dysfunction in vesicoureteral reflux. *Urol Int* 1984;**39**:29-31.
- Millard RJ, Oldenburg BF. The symptomatic, urodynamic and psychodynamic results of bladder re-education programs. *J Urol* 1983;**130**:715-9.
- Vincent SA. Treatment of enuresis with a perineal pressure apparatus: the irritable bladder syndrome. *Dev Med Child Neurol* 1964;**6**:23-9.
- Goldraich NP, Barratt TM. Vesicoureteric reflux and renal scarring. In: Holliday MA, Barratt TM, Vernier RL, eds. *Pediatric nephrology*. 2nd ed. Baltimore: Williams and Wilkins, 1987:647-66.
- Hellström A-L, Hjälmås K, Jodal U. Rehabilitation of dysfunctional bladder in children—method and 3-year follow up. *J Urol* 1987;**138**:847-50.
- Sugar EC, Firlit CF. Urodynamic biofeedback: a new therapeutic approach for childhood incontinence/infection (vesical voluntary sphincter dyssynergia). *J Urol* 1982;**128**:1253-8.
- Wear JB, Wear RB, Cleeland C. Biofeedback in urology using urodynamics: preliminary observations. *J Urol* 1979;**121**:464-8.
- Miller NE. Learning of visceral and glandular responses. *Science* 1969;**163**:434-7.
- Editorial. Diurnal enuresis. *Lancet* 1987;**ii**:314-5.
- Kallenius G, Winberg J. Bacterial adherence to periurethral epithelial cells in girls prone to urinary tract infections. *Lancet* 1978;**ii**:540-2.

Correspondence to Dr JD van Gool, University Children's Hospital, PO Box 18009, NL-3501 CA Utrecht, The Netherlands.