

Pollakiuria in Children with Tic Disorders

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Background: To describe the rarely reported characteristics of pollakiuria in children with tic disorders (TD).

Methods: During a period of one year, all patients visiting our Tourette Syndrome (TS) Clinic with TD were studied for presentation of pollakiuria.

Results: We found that nine (1.9%) out of 467 children with TD had pollakiuria: 407 boys and 60 girls, with a mean age of 8.7 years. Of these nine children, there were eight boys and one girl, and their mean age was 7.4 years. Five had transient TD, one had chronic motor TD and the remaining three had TS. Their pollakiuria recovered within two to six months, except for two male patients aged six and 19 years, whose pollakiuria persisted for more than a year. Factors precipitating anxiety were identified in only three children. Obsessive compulsive behaviors could be traced in eight patients.

Conclusions: Pollakiuria occurring in children with TD is not frequently encountered. It usually has a benign course, as it does in those without tics. However, sometimes the pollakiuria waxed and waned over years, as do tics. Pollakiuria could be a manifestation of tics or a compulsion, instead of just an anxiety disorder.

(*Chang Gung Med J* 2005;28:773-8)

Key words: pollakiuria, tic disorder, tourette syndrome.

Frequent urination in children is a hallmark of possible underlying urinary diseases. Pollakiuria, defined as daytime urinary frequency without dysuria, is a distinctly different entity with almost no pathological situations, and it usually resolves spontaneously.⁽¹⁾ We have treated children with pollakiuria who also have active tic disorders (TD). Children, especially those who have suffered years of pollakiuria, are extremely concerned by this problem, as well as by the tics. They may have no normal social life and most of the time must stay at home, or more specifically stay close to the toilet, for they may have an urge to urinate every couple of minutes. However, we could find almost no articles in the literature discussing pollakiuria in children with TD

(either chronic or transient), except for articles about pediatric autoimmune neuropsychiatric disorder associated with streptococcal infection (PANDAS),⁽²⁾ which is an abrupt and explosive onset of behavioral problems (obsession, compulsion, tic, attention deficit, hyperactivity) with the presence of group A beta-hemolytic streptococcal (GABHS) infection. It was our aim to describe the possible association between pollakiuria and children with TD, after excluding PANDAS, and arouse clinicians' awareness of this specific issue.

METHODS

During a period of one year, all patients visiting

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Received: Feb. 25, 2005; Accepted: Sep. 23, 2005

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our Tourette Syndrome (TS) Clinic with TD were confirmed to be free from PANDAS by having no history of acute onset and no evidence of GABHS infection. Then they were asked about urination problems. Those that said they had problems were asked for further details. The etiologies were scrutinized by taking a detailed history including urination patterns; any prescription of neuroleptics, psychotropics or any other medicines; physical examination including neurological; laboratory examinations including urinalysis, clean-catch midstream urine culture, spot urine for calcium excretion (calcium to creatinine ratio); ultrasonography of the urinary tract and magnetic resonance imaging (MRI) of the spinal cord to rule out spinal dysraphism. Voiding cystourethrography and urodynamic studies were not done routinely unless patients had positive results from the above examinations. The criteria to confirm pollakiuria was set as increasing frequency of urination during daytime, many times greater than normal, that greatly concerned the patient and their caregivers.

Patients with pollakiuria were closely monitored, for frequency and urine amount at each urination, for at least one year, to find out the short-term outcome. Meanwhile, co-morbidities of attention deficit hyperactivity disorder (ADHD) fulfilling the Diagnostic and Statistical Manual of Mental Disorders - Fourth Edition (DSM-IV) criteria,⁽³⁾ self-injurious behavior (SIB) and obsessive compulsive behavior (OCB) were studied. The final classification of the patients' TD as either transient TD, chronic motor or phonic TD, or TS, was based on the diagnostic criteria proposed by the Tourette Syndrome Classification Study Group.⁽⁴⁾ A questionnaire for screening the possible precipitating factors included the following items: fear of school, fear of death, fear of ghosts, perceiving life threat, parents quarrelling, parents divorce, home burglary, arrival of a newborn and death of a parent. Triggering factor for pollakiuria indicated that the patient urinated immediately after words about urination, toilet, water or rain.

RESULTS

We found that nine (1.9%) out of 467 children with TD had pollakiuria: 407 boys and 60 girls, with a mean age of 8.7 years. Of the nine children, there were eight boys and one girl, aged between four to

19 years, with an average age of 7.4 years. Five had transient TD, one had chronic motor TD and the remaining three had TS. Four (44%) had co-morbid ADHD (1 transient TD, 1 chronic TD, 2 TS), four (44%) had minor SIB such as nail biting or skin peeling, and eight (89%) had OCB. No PANDAS was diagnosed in this series of patients with or without pollakiuria. Actually, we have experienced only one case of PANDAS and that was many years ago.

The eight boys urinated once every three to 10 minutes at the peak of their pollakiuria. The urine amount at each voiding may be just a few drops forcefully squeezed out. The only female patient kept wetting herself about every 10 to 30 minutes. Precipitating factors could be found in only three (33%) of our nine patients: one was quarrelling between parents and two had a fear of school (Table 1). Triggering of urination by word suggestion was evident in six boys. No enuresis or other urination disorders could be found in the study group. Physical examinations including neurological examinations found no positive signs. No abnormalities on urinalysis were found. All the clean-catch midstream urine cultures were negative for significant growth of bacteria. The results of the calcium-to-creatinine ratio of spot urine were all below 0.21. Ultrasonography of the kidneys, ureter and bladder were normal. The one spinal cord MRI, asked for by anxious parents, was negative for spinal dysraphism. Voiding cystourethrography and urodynamic studies performed on two boys, due to caregiver' concerns, were negative too.

The pollakiuria recovered within two to six months in seven cases. The only exceptions were

Table 1. Possible Precipitating Factors for Pollakiuria in Children with Tic Disorders

Precipitating Factor	Patient Number
Fear of school	2
Fear of death	0
Fear of ghosts	0
Perceiving life threat	0
Parents quarrelling	1
Parents divorce	0
Home burglary	0
Arrival of a newborn	0
Death of a parent	0

two male patients of six and 19 years of age, whose pollakiuria waxed and waned for several years. The first boy had recurring pollakiuria three times for two to four months duration each time, since the onset of tics two years previously. His tics and pollakiuria were not synchronized in severity patterns. The second boy had onset of tics at eight years of age. His pollakiuria waxed and waned, never completely resolving, for 11 years and affected his whole school life, and went on to affect his life as a soldier after graduating from school. When it was mild, he had to go to the toilet every one to two hours, and spent at least 10 to 20 minutes repeatedly voiding until his bladder was definitely empty. When it was severe, he had to hold a bottle in his hand to collect his urine every one to five minutes. He had no normal social life and had to stay at home, or more specifically, stay near the toilet, for his urge to void occurred every couple of minutes.

Psychotherapy did not bring about any dramatic or immediate cessation of pollakiuria in our 9 patients, including the three with positive precipitating factors. The five children with transient TD ceased their tics within two to six months. The patient with chronic motor TD and the three with TS had tics that waxed and waned, and also continued to suffer co-morbid ADHD, OCB and SIB at the last follow-up in this study.

DISCUSSION

The initial investigation of increased frequency in urination includes careful history taking and detailed physical examinations. The clinicians should look for whether other symptoms accompany the increased frequency. Particular attention should also be directed toward potential spinal dysraphism. In a study by Basar et al., 30% of their patients with a tethered spinal cord had pollakiuria.⁽⁵⁾ Complete urinalysis may exclude most of the possibilities of urinary tract infection, hematuria, glycosuria and renal tubular dysfunction. Hypercalciuria, with or without urolithiasis, is a special entity which may also present with frequency, urgency, microscopic hematuria or enuresis.⁽⁶⁾ Simple spot urinary calcium excretion can be measured easily and abnormality should be suspected when the value of calcium-to-creatinine ratio is over 0.21. Ultrasonography is a noninvasive and convenient diagnostic tool in the detection of

anatomical abnormalities of the renal collecting system. Voiding cystourethrography and urodynamic studies were indicated only in severe cases with associated problems. No abnormalities should be found in any of the above examinations in order to reach a final diagnosis of pollakiuria.

The word 'pollakis' is a Greek word which means 'often or many times'. Pollakiuria is defined as an increase in the frequency of urination at intervals varying from five to 20 minutes, with small urine volume, and only occurring during waking hours.⁽¹⁾ There is no association with dysuria, nocturia, daytime incontinence or hematuria. Pollakiuria commonly occurs at four to seven years of age, when the children begin to attend preschool or kindergarten. It is also called extraordinary daytime urinary frequency.⁽⁷⁾ Pollakiuria was first described by Asnes and Mones in 1973 with the radiological review of four patients.⁽⁸⁾ A similar manifestation was reported by Stephens et al. as a sham syndrome.⁽⁹⁾ They thought that psychological factors might be the principal cause of this disorder, as did other authors.^(7,10,11) Fear of school, death or ghosts, perceiving life threat, parents divorce, home burglary, arrival of a newborn or death of a parent were thought to be principal precipitating factors.^(7,8) However, associated tics or obsessive-compulsive disorders were rarely mentioned.

In our series, the precipitating factors in pollakiuric children with tics were not as high as previous reports on those without mention of co-existing TD (33% vs. > 40%). This could be due to our study being retrospective, or to different cultural backgrounds. Older children and adolescents may be shy of talking about voiding problems. The reasons for this difference could have a direct relationship to the underlying pathogenesis of TD or the special personalities of children with TD, rather than psychological factors alone.

Could pollakiuria be a clinical manifestation of basal ganglia dysfunction? We know that movement disorders are not the only problems caused by basal ganglia dysfunctions.⁽¹²⁾ The characteristic clinical features of a developing basal ganglia syndrome in children include the following four categories: (1) involuntary movements and other motor disturbances (i.e. delays in attaining age-appropriate gross and fine motor skills, general motor clumsiness, poor athletic abilities, and speech articulation disorders

including stuttering); (2) OCB; (3) ADHD; and (4) aggressive behavior. Eight TD children with pollakiuria in our study had co-morbidities with usual frequency, except OCB, which had a higher occurrence rate than in previous studies.^(13,14) Pollakiuria could be a compulsive symptom as it is easily triggered by suggestion. However, our number of cases was too small to conclude this. In their paper about PANDAS, Murphy and Pichichero used the term “‘compulsive’ daytime urinary urgency and frequency”, perhaps because of the significant association with other symptoms of acute and explosive onset OCD.⁽²⁾ There were so many other articles on PANDAS that did not mention pollakiuria, that it might be just neglected.

Male dominance in pollakiuria could be due to either male dominance in TD or externalization of male genitalia. TD occurs in boys five to ten times more frequently than in girls,⁽¹²⁻¹⁴⁾ and in our series of children there were eight boys and one girl. The male genitalia, as such a significant externalized symbol, may have some contribution too, if we believe Freud's hypothesis. However, the young age factor in children with tics and pollakiuria is significant. Enuresis has been mentioned in children with TD but not pollakiuria. Some medications, such as fluoxetine and milnacipran, may result in pollakiuria.^(15,16) Our patients were not exposed to these medications. Some children in the control group did receive some anti-tic or psychotropic agents without developing pollakiuria at any time. Spontaneous resolution of pollakiuria occurred in all 48 patients, aged 3.9 to 7.8 years, within 12 days to five months after diagnosis, in the series of Chan and Huang,⁽¹¹⁾ and also in other reports.⁽¹⁷⁻¹⁹⁾ Most of our patients had a good outcome too. However, psychotherapy did not cause an immediate cessation of pollakiuria in our patients, which has been described in a previous article.⁽²⁰⁾ It indicated that pollakiuria in children with TD might have a different pathogenesis from that in children without tics. Those reporting previously might have neglected associations with tics and related disorders: TS is frequently not just a TD alone.⁽²¹⁾

Children with pollakiuria urinate in a similar manner to tic manifestations, repetitively, involuntarily and consciously. The frequent co-morbid obsession and/or compulsion have a similar character to uncontrolled repetition. It is difficult for us to determine whether the pollakiuria in our children with TD

is a tic itself, a compulsion alone, or just an anxiety. Further studies, especially prospective ones, are needed in the future to evaluate the true etiology of pollakiuria in children with tics.

REFERENCES

- Schmitt BD. Daytime wetting (diurnal enuresis). *Pediatr Clin North Am* 1982;29:9-20.
- Murphy ML, Pichichero ME. Prospective identification and treatment of children with pediatric autoimmune neuropsychiatric disorders associated with group A streptococcal infection (PANDAS). *Arch Pediatr Adolesc Med* 2002;156:356-61.
- American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders*. 4th ed. Washington DC: American Psychiatric Association, 1994.
- Anonymous. Definitions and classification of tic disorders. The Tourette Syndrome Classification Study Group. *Arch Neurol* 1993;50:1013-6.
- Basar H, Aydoganli L, Yuksel M, Basar M, Akdemir G, Kaptanoglu E. The outcome of urological findings in operated tethered cord patients. *Int Urol Nephrol* 1997;29:167-71.
- Garcia Nieto V, Monge Zamorano M, Sanchez Almeida E, Garcia Garcia M, Castro Diaz D. Pollakiuria and idiopathic hypercalciuria in childhood. *Arch Esp Urol* 1995;48:1017-21.(Spanish)
- Zoubek J, Bloom DA, Sedman AB. Extraordinary urinary frequency. *Pediatrics* 1990;85:1112-4.
- Asnes RS, Mones RL. Pollakiuria. *Pediatrics* 1973;52:615-7.
- Stephens FD, Whitaker J, Hewstone AS. True, false and sham urinary tract infection in children. *Med J Aust* 1966;2:840-2.
- Koff SA, Byard MA. The daytime urinary frequency syndrome of childhood. *J Urol* 1988;140:1280-1.
- Chen WF, Huang SC. Pollakiuria: Extraordinary daytime urinary frequency a common problem in pediatric practice. *Chang Gung Med J* 1995;18:115-9.
- Cohen DJ, Leckman JF, Pauls D. Neuropsychiatric disorders of childhood: Tourette's syndrome as a model. *Acta Paediatr Suppl* 1997;422:106-11.
- Leckman JF, Cohen DJ, eds. *Tourette's Syndrome - Tics, Obsessions, Compulsions: Development Psychopathology and Clinical Care*. New York (NY): John Wiley & Sons, Inc. 1999.
- Wang HS. Clinical Diagnosis and Management of Tourette syndrome. *Acta Neurol Taiwan* 2001;10:219-27.
- Sancho A, Martinez-Mir I, Palop V. Pollakiuria and micturition syndrome related to fluoxetine. *Med Clin (Barc)* 1995;105:598-9.(Spanish)
- Regina W, Vandel P, Vandel S, Sechter D, Bizouard P. Clinical tolerance of a new antidepressant - milnacipran. *Encephale* 1999;25:252-8.(French)

17. Cohen HA, Nussinovitch M, Kauschansky A, Straussberg R, Ashkenasi A, Frydman M, Varsano I. Extraordinary daytime urinary frequency in children. *J Fam Pract* 1993;37:28-9.
18. Watemberg N, Shalev H. Daytime urinary frequency in children. *Clin Pediatr (Phila)* 1994;33:50-3.
19. Brock JW 3rd. The frequency and frequency dysuria syndromes of childhood: hypercalciuria as a possible etiology. *Urology* 1994;44:411-2.
20. Glazier DB, Ankem MK, Ferlise V, Gazi M, Barone JG. Utility of biofeedback for the daytime syndrome of urinary frequency and urgency of childhood. *Urology* 2001;57:791-4.
21. Wang HS. Tourette syndrome: Not just a tic disorder. *Chang Gung Med J* 2005;28:527-9.

抽動異常兒童的日間頻尿現象

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- 背景：**過去，抽動異常兒童出現日間頻尿的現象很少被文獻提及；本文旨在報告我們的經驗以便引起適當的討論與關切。
- 方法：**以一年為期，所有到我們妥瑞症門診求診並確定有抽動異常者，皆詳細研究他們有無日間頻尿的現象。
- 結果：**全部 467（男 407，女 60；平均年齡 8.7 歲）位抽動異常者中，9 位有日間頻尿的現象，發生率為 1.9%。這 9 位有 5 位屬一過性的抽動異常，1 位慢性抽動異常，3 位屬妥瑞症。其中 7 位的日間頻尿現象在 2 至 6 個月內一次就消失了，其餘 2 位（6 歲及 19 歲的男生）則拖了一年以上還不好。可能導致焦慮的誘因只有 3 位問得到。強迫症的傾向則有 8 位滿明顯的。
- 結論：**日間頻尿的現象在抽動異常者並不常出現，雖然與過去無抽動異常者之日間頻尿文獻一樣多數自然消失，少數卻好好壞壞拖了好多年造成個案異常地困擾。我們懷疑抽動異常者的日間頻尿並非焦慮現象而已，也可能是一種抽動或強迫行為，值得進一步更大規模前瞻性地探討。
(長庚醫誌 2005;28:773-8)

關鍵字：日間頻尿，抽動異常，妥瑞症。

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受文日期：民國94年2月25日；接受刊載：民國94年9月23日

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