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Incidence of Enuresis and Encopresis Among Children with Attention Deficit Hyperactivity Disorder in a Population-Based Birth Cohort

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Abstract

OBJECTIVE—This study reports the incidence of enuresis and encopresis among children with attention-deficit/hyperactivity disorder (AD/HD) versus those without AD/HD.

METHOD—Subjects included 358 (74.5% male) children with research-identified AD/HD from a 1976-1982 population-based birth cohort (N = 5718) and 729 (75.2% male) non-AD/HD control subjects from the same birth cohort, matched by gender and age. All subjects were retrospectively followed from birth until a diagnosis of enuresis or encopresis was made or last follow-up prior to 18 years of age. The complete medical record for each subject was reviewed to obtain information on age of initial diagnosis of an elimination disorder, frequency and duration of symptoms, identification of exclusionary criteria specified by DSM-IV, with confirmation of the diagnosis by expert consensus.

RESULTS—Children with AD/HD were 2.1 (95% CI, 1.3-3.4; p = 0.002) times more likely to meet DSM-IV criteria for enuresis than non-AD/HD controls; they were 1.8 (95% CI, 1.2 – 2.7; p = 0.006) times more likely to do so than non-AD/HD controls when less stringent criteria for a diagnosis of enuresis were employed. Though not significant, children with AD/HD were 1.8 (95% CI, 0.7-4.6; p = 0.23) times more likely to meet criteria for encopresis than non-AD/HD controls. The relative risk was 2.0 (95% CI, 1.0-4.1; p = 0.05) when a less stringent definition for encopresis was utilized.

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CONCLUSIONS—The results of this population-based study demonstrate that children with AD/HD are more likely than their peers without AD/HD to develop enuresis with a similar trend for encopresis.

Keywords

enuresis; encopresis; AD/HD; birth-cohort; incidence

Introduction

Of the numerous behavior disorders studied in children, attention-deficit/hyperactivity disorder (AD/HD) may be the most common, with estimates of incidence as high as 7.4% to 16% in the school-aged population.¹ Similarly, idiopathic enuresis is reported to occur in as many as 12% to 16% of 5 year-old, 7% to 11% of 9 year-old, and 1% to 3% of 16 year-old children.² Although typically reported to be a less common childhood disorder, encopresis has historically been reported to occur in 1% to 3% of primary school aged children.³⁻⁴ A more recent population-based study in more than 13,000 children reported the prevalence of encopresis to be 4.1% among 5 to 6 year-old and 1.6% among 11 to 12 year-old children.⁵

Attention Deficit/Hyperactivity Disorder, enuresis and encopresis have been shown to have a wide range of adverse effects on psychosocial functioning. Barbaresi et al.⁶ report disturbing school outcomes in children with AD/HD including significantly lower reading achievement and increased rates of school absenteeism, grade retention and school dropout. The psychosocial impact of nocturnal enuresis is reported to be minimal in pre-pubescent children but having a greater impact in older children, with emotional symptoms being more likely.⁷⁻⁸ Cox et al.⁹ compared children with encopresis to a matched control group and found significantly more anxiety/depressive symptoms, attention difficulties, social and disruptive problems, poorer school performance, and 20% of the encopretic children exceeding thresholds for clinically significant behavior problems on standardized questionnaires of psychosocial functioning.

Researchers have employed multiple methods to better understand the complex relationship between AD/HD, enuresis and encopresis. Baeyens et al.,¹⁰ reviewed all epidemiological and clinical studies examining the AD/HD and enuresis relationship and found nearly all studies reporting an increased prevalence of behavioral problems in general, and AD/HD in particular in children with enuresis. In a birth cohort of 1,265 children from New Zealand followed for 15 years, children who met criteria for enuresis after 10 years of age showed a slightly increased risk of attention deficit behaviors and conduct problems.¹¹ Baeyens et al.¹² reported that of children diagnosed with nocturnal enuresis, 15% met the diagnostic criteria for AD/HD, and 22.5% were diagnosed with the inattentive subtype of AD/HD. In a retrospective review of medical records, Friman et al.⁷ reported that children with nocturnal enuresis, compared with clinical and nonclinical samples without enuresis, did not present with clinically significant behavioral problems. However, AD/HD symptoms were not specifically assessed. More recently, in a nationally representative study of the prevalence of enuresis in 1,136 children between the ages of 8 and 11, the 12 month prevalence of enuresis was 4.45% (6.21% in boys, 2.51 % in girls), with AD/HD being strongly associated with enuresis (odds ratio of 2.88).¹³

To our knowledge, there is only one clinical study of the association between AD/HD and enuresis which found that age 6 years children with AD/HD were 2.7 times more likely than controls to have nocturnal enuresis and 4.5 times more likely to have daytime enuresis.¹¹ Only one population-based study has investigated the relationship between encopresis and behavior problems. Van der Wall, Benninga and Hirasing⁵ reported on a population-based

study of encopresis in 18,456 5- to 6-year-old children and 16,293 11- to 12-year-old children. Regardless of age, children with encopresis were 1.71 times more likely to exhibit behavioral problems and 2.2 times more likely to exhibit emotional problems compared to children without encopresis. Unfortunately, standardized behavior questionnaires were not employed, nor were specific behavioral diagnoses made in this study.

One study of 20 children with both urinary incontinence and encopresis found that they were more likely to exhibit hyperkinetic syndromes, and emotional and conduct problems compared to children who only had urinary incontinence.¹⁴ Of the children with comorbid enuresis/encopresis, 65% fell within the clinical range on a standardized behavior rating scale.

With one exception [Sheeram],¹³ the epidemiological and clinical studies published thus far to explore the frequency of occurrence of these childhood disorders begin with samples of children with enuresis and encopresis, and attempt to estimate the occurrence of AD/HD among these children. A thorough review of these studies reveals many methodological limitations that complicate the efforts to generalize these findings. For example, the confounding effect of variables such as age, gender, SES, and clinic referred samples with enuresis creates a problem in understanding the relationship between AD/HD and enuresis. A similar review of the association between encopresis and AD/HD has yet to be published, although many of the same concerns would be relevant.

To date, no study has investigated the incidence of enuresis or encopresis in a carefully defined population-based cohort of children with AD/HD. Therefore, the cumulative incidence of developing enuresis or encopresis among children with AD/HD cannot yet be accurately determined. The current study utilizes cumulative incidence in order to measure the probability of occurrence for enuresis and encopresis in a clearly defined population of children with research-identified AD/HD. To our knowledge, this retrospective study is the first population-based investigation of a research-identified group of children with AD/HD, utilizing clinical definitions of enuresis and encopresis in order to more accurately determine the rate of occurrence of these elimination disorders among children with AD/HD.

Method

Study Setting and Subjects

We have previously published descriptions of the identification of the 5,718 members of the 1976-1982 birth cohort in Rochester, MN who remained in the community at least until 5 years of age, and who were included as subjects for this study.^{1, 15, 16} Briefly, the two main resources used in this study included school and medical records. Through a contractual research agreement, permission was obtained to access the cumulative school records for every child born in Rochester between January 1, 1976 and December 31, 1982 to mothers residing in Independent School District (ISD) #535 (Rochester, Minnesota). We included all children who were ever enrolled at any of the public, private or parochial school in ISD #535, as well as children who were home-schooled; all school records were accessible for review and abstracting. Through the Rochester Epidemiology Project (REP), we also had access to the complete, dossier medical record for all members of the birth cohort.^{15, 17} These medical records are available from the Mayo Clinic, Olmsted Medical Center and the few private medical practitioners in the community, encompassing essentially all of the providers of primary and specialty medical care for residents of Rochester. The medical records contain a detailed history of all medical encounters in the community including ambulatory medical and social services, hospitals, emergency department and home visits, laboratory, psychiatry and psychology reports and test results, from birth until the patient no longer resides in the community. Through the Rochester Epidemiology Project, all

diagnoses and surgical procedures recorded at these medical facilities, including their affiliated hospitals, are indexed for computerized retrieval (Medical Diagnostic Index). Authorization to review the medical records of birth cohort subjects was obtained in accordance with Minnesota statutes and approved by the Mayo Clinic institutional review board.

Identification of AD/HD Incident Cases and Non-AD/HD Controls

As previously described,^(1,15) we employed a five-step approach, beginning with a review of school records of all 5,718 subjects for evidence of any concern about learning or behavior, regardless of whether or not the subject had any specific, clinical diagnosis, including AD/HD. Second, for all subjects with any type of learning or behavioral concern identified in the school records (n=1961), we searched for additional information from school and medical records, including DSM-IV defined symptoms of AD/HD, results from teacher and/or parent AD/HD questionnaires, clinical diagnoses of AD/HD, and documentation of treatment with medication commonly prescribed for children with AD/HD.¹⁸ Third, for the subjects whose school records did *not* include any suggestion of learning or behavioral problems (n=3757), we searched the computerized index of medical diagnoses, which captures diagnoses on all patients seen at the Mayo Clinic and Olmsted Medical Center, for diagnoses related to or including AD/HD. Fourth, we reviewed the records of birth cohort members who had received care at the only private community provider of psychiatric care other than the mentioned institutions and identified four additional potential AD/HD cases. For the fifth and final step, the following, explicit, *research criteria* were applied to identify AD/HD incident cases among the 1,344 possible cases identified in the preceding four steps: 1) *symptoms of AD/HD as specified in DSM-IV* positive results on AD/HD questionnaires, and 3) *documented clinical diagnoses of AD/HD*. The exclusion criteria specified in DSM-IV were also applied (i.e., subjects could not be considered as AD/HD incident cases if they had a diagnosis of pervasive developmental disorder, severe mental retardation, schizophrenia, or other psychotic disorder).¹⁸ **Table 1** describes the 379 subjects who fulfilled various combinations of these criteria, and were designated as research-identified AD/HD incident cases. All subtypes of AD/HD were included as cases. Subjects fulfilled research criteria for AD/HD at a mean age of 10.4 years, with over 50% fulfilling criteria before age 10 years.

Of the 379 subjects who fulfilled various criteria for AD/HD, 21 were excluded from the analyses in the current study because they denied ongoing access to their medical records for research purposes. For each of the original 379 AD/HD cases, two controls were previously selected from the birth cohort. Controls were matched on gender and date of birth (± 6 months) using an optimal matching algorithm applied to the values of each matching factor. However, 29 controls were excluded because they denied access to their records for research purposes. The study sample therefore consisted of 358 research-identified AD/HD incident cases and 729 matched controls without AD/HD. All of the cases and controls were followed through their medical records from birth until their last medical visit prior to 18 years of age.

Identification of Enuresis and Encopresis Cases

All cases of enuresis and encopresis were identified using the specific criteria listed in the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision (DSM-IV-TR), including age of onset of symptoms, frequency and duration of symptoms, and exclusionary criteria as summarized in **Table 2**. We first employed the Medical Diagnostic Index to identify all potential cases of enuresis or encopresis based on the presence of at least one of the following diagnoses: enuresis-psychogenic, encopresis-psychogenic, enuresis-nos, encopresis-nos, incontinence-anal, incontinenceurinary, incontinence of feces, incontinence of urine, enuresis, and encopresis. A data abstraction

protocol was created from the research literature regarding typical diagnostic methods, types of treatment, and clearly defined treatment outcome measures. Abstracted information included demographic details abstracted from the birth certificate for children and their parents, symptomatology such as voiding and soiling type, frequency and duration of symptoms, length of episodes of continence, specific recommended treatments and outcomes.

In order for a child to be classified as an incident case of enuresis or encopresis, all abstracted data were reviewed by an experienced pediatric psychologist and developmental pediatrician, with consensus between the two reviewers required for classification. Cases were defined as “definite” if they met all DSM-IV criteria, and as “borderline” if the medical record included one of the previously mentioned diagnoses but did not provide information regarding frequency or duration of symptoms criteria. As indicated in the DSM-IV, exclusion criteria for enuresis or encopresis included incontinence symptoms that were directly related to a substance (e.g., diuretic, laxative) or medical condition (e.g., spina bifida, diabetes).

Statistical Analyses

Characteristics from the birth certificate were compared between AD/HD cases and non-AD/HD controls based on the two-sample t-test for birth weight and maternal age and the chi-square test for race and maternal education. Subjects were followed from their date of birth to their date of last medical follow-up prior to 18 years of age. Subjects who met criteria for either enuresis or encopresis were followed until the date of their first elimination disorder diagnosis of enuresis or encopresis; otherwise they were censored at the date of their last follow-up. The cumulative incidence by age 15 was calculated using the Kaplan-Meier method; age 15 was chosen since all of the diagnoses of enuresis or encopresis in the study sample were prior to 15 years of age. Cox proportional hazards models were fit to estimate the relative risk (RR) and corresponding ninety-five percent confidence interval (95% CI) to compare the risk of enuresis (or encopresis) between subjects with and without AD/HD. All calculated p-values were two-sided and p-values less than 0.05 were considered statistically significant. Statistical analyses were performed using the SAS version 9.2 software package (SAS Institute, Inc.; Cary, NC).

Results

The study sample consisted of 358 research-identified AD/HD incident cases and 729 matched controls without AD/HD. Seventy-five percent of the subjects were male (270 AD/HD cases and 548 controls). Based on characteristics from the birth certificates, subjects with ADHD tended to have lower birth weight (mean, 3465 vs. 3525 grams; $p=0.08$) and were more likely to be Caucasian (99.2% vs. 97.3%; $p=0.04$), have mothers with less education (47.7% (156/327) vs. 37.1% (241/649) with no education past high school, $p=0.002$), and have younger mothers at birth (mean, 26 years vs. 26.8 years; $p=0.005$), compared with controls.

Enuresis

Among the 358 AD/HD cases, 35 cases met DSM-IV criteria for enuresis at a mean (SD) age of 6.7 (2.1) years (range, 4.4 - 14.8 years). An additional 8 cases were classified as having “borderline” enuresis at a mean (SD) age of 6.1 (1.9) years (range, 4.1 - 9.5 years).

Among the 729 non-AD/HD controls, 34 subjects met the DSM-IV criteria for enuresis at a mean (SD) age of 7.0 (2.5) years (range, 4.0 - 13.8 years). An additional 16 subjects were classified as having “borderline” enuresis at a mean (SD) age of 6.1 (2.2) years (range, 3.5 -

11.5 years). Among the AD/HD cases and controls who did not meet either criterion for enuresis, 96.5% and 89.0%, respectively, had medical visits up through 15 years of age.

As summarized in **Table 3**, the cumulative incidence of meeting DSM-IV criteria for enuresis by 15 years of age was 9.9% and 4.8%, respectively, among AD/HD cases and controls. Subjects with AD/HD were 2.1 (95% CI, 1.3-3.4; $p = 0.002$) times more likely to have met the DSM-IV criteria for enuresis than non-AD/HD controls. Further, subjects with AD/HD were 1.8 (95% CI, 1.2-2.7, $p = 0.006$) times more likely than controls to have met either the DSM-IV criteria for enuresis or have “borderline” enuresis. The above results were unchanged after adjusting for race, birth weight, maternal education, and maternal age in each of the regression models (2.1; 95% CI, 1.3-3.4; $p = 0.002$ and 1.8; 95% CI, 1.2-2.6; $p = 0.007$; respectively).

Encopresis

Among the 358 AD/HD cases, 8 cases met DSM-IV criteria for encopresis at a mean (SD) age of 8.0 (3.1) years (range, 4.0 - 12.5 years). An additional 7 cases were classified as having “borderline” encopresis at a mean (SD) age of 7.3 (1.3) years (range, 5.7 - 9.3 years).

Among the 729 non-AD/HD controls, 9 subjects met the DSM-IV criteria for encopresis at a mean (SD) age of 6.7 (2.1) years (range, 4.1 - 9.9 years). An additional 6 subjects were classified as having “borderline” encopresis at a mean (SD) age of 5.9 (1.9) years (range, 3.7 - 8.2 years). Among the AD/HD cases and controls who did not meet either criterion for encopresis, 92.7% and 89.0%, respectively, had medical visits up through 15 years of age.

As summarized in **Table 4**, the cumulative incidence of meeting the DSM-IV criteria for encopresis by 15 years of age was 2.3% and 1.3%, respectively, among AD/HD cases and controls. Subjects with AD/HD had a non-significant trend toward being more likely to have met the DSM-IV criteria for encopresis than non-AD/HD controls (RR, 1.8; 95% CI, 0.7-4.6; $p = 0.23$). However, subjects with AD/HD were 2.0 (95% CI, 1.0-4.1, $p = 0.05$) times more likely than controls to have met either the DSM-IV criteria for encopresis or have “borderline” encopresis. The above results were unchanged after adjusting for race, birth weight, maternal education, and maternal age in each of the regression models (1.8; 95% CI, 0.7-4.7; $p = 0.23$ and 2.0; 95% CI, 1.0-4.1; $p = 0.06$; respectively).

Comorbid Enuresis and Encopresis

Among the 358 AD/HD cases, 5 met the DSM-IV criteria for both enuresis and encopresis, 1 case met the criteria for enuresis and borderline encopresis, and 1 case met the criteria for borderline enuresis and encopresis. Among the 729 non-AD/HD controls, 2 met the DSM-IV criteria for both enuresis and encopresis and 1 met the criteria for enuresis and borderline encopresis. The small number of combined cases precluded statistical analysis. Encopresis preceded enuresis for two cases and both conditions were diagnosed at the same date for two other cases.

Discussion

This study is part of our ongoing effort to better understand co-morbidities among members of a population-based cohort of children with research-identified AD/HD. It also represents the first report of the relative risk of being diagnosed with enuresis or encopresis for children with AD/HD, compared to children without AD/HD. We found that AD/HD is associated with a significantly greater risk of developing a co-morbid elimination disorder, specifically, enuresis. We were unable to demonstrate a statistically significant association between AD/HD and increased risk for encopresis. However, it should be noted that there were relatively

few cases of encopresis overall among study subjects, limiting the statistical power for our assessment of this association.

Methodological strengths of this study include ascertainment of cases in a population-based birth cohort, and use of a non-referred cohort of research-identified cases of AD/HD of ample size with two controls matched on pertinent characteristics. Furthermore, this is the first study to attempt to obtain a more precise estimate of the association between AD/HD and encopresis in a population-based sample of children with research-identified AD/HD and encopresis; however, the small number of cases of encopresis among our subjects potentially limited our ability to accurately assess this association. Although we do have access to information about stimulant medication treatment provided to our research-identified childhood AD/HD cases, the current study did not include an assessment of the potential for stimulant treatment or other factors that might increase or decrease the likelihood of an elimination disorder. A future study of factors that affect the rate of elimination disorder among children with AD/HD would be an important contribution to our understanding of these commonly co-morbid conditions.

Notwithstanding the previously described methodological strengths of this study, several potential limitations should be noted. First, emigration: 43% of the original birth cohort of 8,548 left the community before age 5. However, this potential source of bias was previously addressed and found unlikely to impact the findings reported here.¹⁶ Second, we did not directly assess the study subjects for AD/HD nor for an elimination disorder at the time of the study. However, multiple sources of information (school, medical) were utilized, combined with expert consensus (i.e., pediatric psychologist, behavioral pediatrician) to confirm diagnoses. All records for each of the 5,718 subjects were reviewed and abstracted; thus, it is unlikely that a significant number of AD/HD cases were missed. Similarly, every record that was electronically identified with a possible elimination disorder was thoroughly reviewed by the investigators. Since our method involved a retrospective review of medical and school records, caution is warranted in suggesting that AD/HD causes an elimination disorder or visa versa. Finally, Rochester is primarily a white, middle class community so inferences to other populations or settings may be limited. Despite these potential limitations, the ability to employ a population-based, non-referred, precisely defined cohort of both boys and girls with AD/HD and access to the complete, detailed medical records of all subjects offers a unique opportunity to understand the relationship between AD/HD and enuresis and encopresis.

Given the strong association between AD/HD status and having an elimination disorder, primary care clinicians should routinely inquire about a child's mastery of urinary and bowel continence in all children with AD/HD. This information would be helpful in directing the child and family to effective interventions (i.e., urine-alarm-conditioning treatment for nocturnal enuresis; behavior therapy combined with bowel management for encopresis). It is also important for clinicians to be aware of the challenging behaviors associated with AD/HD that may impact the effectiveness of treatment for enuresis and encopresis, as these behaviors (e.g., oppositional responses, distractibility, task-avoidance) are associated with poorer outcomes.^{19, 20} Limited research has been done to elucidate the mechanism of action that links enuresis with AD/HD. Ornitz et al.²¹ have suggested a deficit in the prepulse inhibition of the startle response in children with AD/HD and comorbid enuresis implying an immaturity in the developing nervous system of children who have this combination of problems. Studies aimed at understanding the relationship between AD/HD and elimination disorders may identify other common, underlying neurological differences that may lead to more effective treatment for both disorders.

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What's New: Based on stringent diagnostic criteria for AD/HD and enuresis and encopresis, a more accurate estimate of a positive significant association between AD/HD and elimination disorders has been demonstrated in a retrospective, population-based birth cohort study.

Table 1

Research criteria for AD/HD case definition

AD/HD cases	Meets * DSM-IV research criteria for AD/HD	AD/HD questionnaire results	Clinical diagnosis of AD/HD [†]	Number of subjects		
Research identified AD/HD cases	+	+	+	170		
	+	+	-	41	228	
	+	-	+	17		
	-	+	+	122		379
	-	-	+	29	151 [‡]	

AD/HD indicates attention deficit/hyperactivity disorder.

Pluses and minuses indicate the presence or absence, respectively, of a given criterion.

* All DSM-IV criteria were met, only age criterion was not used.

[†] Clinical diagnosis of AD/HD was already made, recorded, and abstracted as stated in the medical record.

[‡] Because of the retrospective nature of the study; 70% (N = 106) had number of symptoms just below required, 30% (N = 45) either had symptoms noted only by one person, or noted for less than 6 months or both.

Table 2

DSM-IV Diagnostic Criteria for Enuresis and Encopresis

Enuresis	Encopresis
A. Repeated voiding of urine into bed or clothing, either involuntary or intentional.	A. Repeated passage of feces into inappropriate places (i.e., clothing) whether involuntary or intentional.
B. Behavior is clinically significant in frequency (i.e., twice per week for at least 3 consecutive months) or behavior causes significant distress or impairment in psychosocial functioning).	B. At least one such event a month for at least 3 months.
C. Chronological age [*] at least 5 years	C. Chronological age [*] of at least 4 years.
D. Behavior not due to medication side-effect or medical condition.	D. Behavior not due to medication side-effect or other medical condition except constipation.

* If diagnosis made prior to earliest age for the disorder and symptoms persisted until the earliest age for the disorder, subjects were included in this study.

Table 3

Cumulative incidence of enuresis among AD/HD cases and non-AD/HD controls.

	No. with enuresis	Cumulative incidence of enuresis by age 15 years	RR (95% CI)	P-value
Definite [†] enuresis				
AD/HD cases (N=358)	35	9.9%	2.1 (1.3 - 3.4)	0.002
Non-AD/HD controls (N=729)	34	4.8%	referent	
Definite or borderline [‡] enuresis				
AD/HD cases (N=358)	35 + 8	12.1%	1.8 (1.2 - 2.7)	0.006
Non-AD/HD controls (N=729)	34 + 16	7.0%	referent	

[†] Subjects were defined as having “definite” enuresis if they met all DSM-IV criteria.

[‡] Subjects were classified as having “borderline” enuresis if their medical record did not provide information regarding either the frequency of symptoms or duration of symptom criteria.

Table 4

Cumulative incidence of encopresis among AD/HD cases and non-AD/HD controls.

	No. with encopresis	Cumulative incidence of encopresis by age 15 years	RR (95% CI)	P-value
Definite [†] encopresis				
AD/HD cases (N=358)	8	2.3%	1.8 (0.7 - 4.6)	0.23
Non-AD/HD controls (N=729)	9	1.3%	referent	
Definite or borderline [‡] encopresis				
AD/HD cases (N=358)	8 + 7	4.3%	2.0 (1.0 - 4.1)	0.05
Non-AD/HD controls (N=729)	9 + 6	2.1%	referent	

[†]Subjects were defined as having “definite” encopresis if they met all DSM-IV criteria.

[‡]Subjects were classified as having “borderline” encopresis if their medical record did not provide information regarding either the frequency of symptoms or duration of symptom criteria.