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A Comparison of the Prevalence of Urinary Incontinence in Girls With Cystic Fibrosis, Asthma, and Healthy Controls

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Summary. Urinary incontinence (UI) is recognized as a significant problem in adult females with cystic fibrosis and can often have a marked impact on day-to-day activities. The prevalence and severity of UI in the pediatric cystic fibrosis (CF) female population is less clear and there are no comparative data with healthy children or children with other respiratory disorders. An anonymous self-completed semi-structured questionnaire was used to study the prevalence rates of UI in girls with CF aged between 11 and 17 and compared it to age-matched asthmatic and healthy girls. The prevalence of UI in girls with CF was significantly higher (17/51, 33%) than the asthmatic (4/25, 16%) and healthy girls (2/27, 7%) ($P = 0.02$). It may manifest as early as 11 years of age and is associated with increasing lung disease. Surprisingly it is perceived as a relatively minor problem in terms of the distress it causes. Pediatric CF clinics should be routinely addressing UI as a potential problem in all girls from the age of 11 years. **Pediatr Pulmonol. 2006; 41:1065–1068.** © 2006 Wiley-Liss, Inc.

Key words: cystic fibrosis; asthma; urinary incontinence; female; children.

INTRODUCTION

Cystic fibrosis (CF) is a multi-system disorder. Several studies have reported urinary incontinence (UI) to be a significant problem for many women with CF, with a reported prevalence of 30–68%.[1–5] This compares to 13% in normal young adult females and 35% in those over 35 years of age.[6,7] The increase in UI has commonly been attributed to the stress placed on the pelvic floor by repeated coughing and forced expiratory maneuvers.[2–4] The occurrence and degree of UI is reported to increase with worsening lung disease.[2,4] As coughing is reported to be more frequent and intense in CF than other lung disease,[8] it is likely that this association may in part be caused by more frequent and longer bouts of coughing. Other potential factors, which may contribute, include poorer nutritional status, impaired muscle function, reduced physical fitness, and long-term use of oral steroids.

Urinary incontinence may have a significant impact on quality of life in terms of day-to-day activities and may also reduce adherence to chest physiotherapy and exercise, further exacerbating pulmonary symptoms and potentially increasing the frequency of UI. In the normal childhood population, UI is not uncommon and the prevalence of daytime UI varies between 8 and 15%.[9–11] The cause of UI in the younger population can be attributed to several factors including maturation, behavior, infection, and neurological problems.[12]

Few studies have examined the prevalence of UI in a younger female population with chronic lung disease. Previous unpublished studies of UI in girls with CF all report an increase in prevalence.[13,14] Whether this increase in prevalence is associated with CF specifically is unclear, as no data are currently available on the prevalence of UI in children with other respiratory disorders, such as asthma, where coughing can also be a chronic problem. The aim of this study was to identify the prevalence and severity of UI in girls with CF and compare it to those with asthma and normal healthy controls.

MATERIALS AND METHODS

Girls between the ages of 11.0–17.0 years were invited to complete an anonymous questionnaire, designed to

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identify the presence and impact of UI. Subjects with CF and asthma were recruited from two specialist centers, whilst the healthy controls were either siblings of CF children or girls attending non-respiratory outpatient clinics (general pediatric or hemoglobinopathy clinics, and those attending for urological/neurological complaints were excluded). All eligible girls with CF (n = 68) were asked to participate by a CF nurse specialist in clinic, and those not attending in the time frame were invited by post. Asthmatic girls and normal control children were recruited from successive clinics. Ethical approval was obtained from each of the three participating centers and parents and girls gave written consent/assent to participate in the study.

The questionnaire was adapted from that used by Orr et al.[2] Subjects were asked questions related to the occurrence, severity and impact of UI. The questions included demographic data; height and weight from which body mass index (BMI) was calculated; age of onset of menarche; the use of oral steroids; and frequency and severity of cough. They were also asked about the presence of urinary leakage, when it happened, what triggered it and how it affected daily activities. A visual analog scale (VAS) was used to indicate the degree of distress that UI caused on daily life. This consists of a 10 cm horizontal line with an anchor point at each extreme. To the left (zero) ''I do not feel upset by it at all'' and to the right (10) ''leaking urine makes me extremely upset.'' Although the questionnaire was anonymous, all girls were given the opportunity to provide their names if they wished further referral. In addition, girls with CF and asthma were asked to provide their latest lung function data (forced expiratory volume in 1 sec, FEV_1 % predicted).

Statistical Analysis

Data on incontinence in adult patients with and without CF varies,[2,9] a mid point of the quoted data was taken for each group (49% for CF and 11.5% for non-CF). Assuming a difference of 30% to be clinically significant, 48 in the CF group and 48 in the non-CF group would give a 90% power to detect a difference at $P = 0.025$.

Tests for normality showed age, age at menarche and FEV_1 to be normally distributed so they are expressed as means with standard error of means, and analysis was by one-way ANOVA. Height, weight, and BMI were not normally distributed (in the non-CF groups) so these parameters are expressed as medians with interquartile ranges and analysis was by non-parametric Kruskal Wallis test (for all groups). FEV_1 is expressed as percent predicted for height, gender, and age.[15] Chi-square test was used to analyze differences in UI between the different subject groups. Analysis of variables within the CF group (comparing UI vs. non-UI) was by independent *t*-test as all data were normally distributed. Statistical analyses were performed using Statistical Package for Social Sciences (SPSS version 11.0).

RESULTS

Questionnaires were analyzed from 51 girls with CF, 25 with asthma, and 27 controls. Seventeen CF girls declined to participate (75% participation rate) whilst all asthmatic and normal children agreed to take part. No significant differences in baseline characteristics for age or height were seen in the three groups (Table 1). There was no significant difference in weight and BMI between the asthma and control groups but the CF girls had significantly lower weight and BMI than the other groups. FEV_1 was similar in the asthma and CF groups (Table 1) and not measured in the control group.

There was a significant difference in the prevalence of UI between the groups (Chi^2 $P = 0.02$), with 17/51 (33%) girls with CF reporting UI, compared to 4/25 (16%) in the asthma group and 2/27 (7%) in the control group (Fig. 1). Of the 17 girls reporting UI in the CF group, 10 reported that leakage could happen at any time, whether their chest was ''good'' or ''bad.'' The majority of those with UI (14/17, 82%) reported small volume leakage (defined as a few

TABLE 1— Baseline Characteristics of All Subjects

		CF (n = 51)	Asthma (n = 25)	Normal (n = 27)	
Age (years)	Mean (range); SEM	13.8 (11–16.9); 0.23	14.2 (11.1–16.9); 0.38	13.9 (11.2–16.9); 0.31	ns
Height (cm)	Median (IQR)	156.3 (149.7–161.7)	154.0 (150.8–161.2)	160.0 (151.9–164.2)	ns
Weight (kg)	Median (IQR)	43.4 (37.7–47.1)	51.6 (46.7–61.0)	49.0 (41.0–56.1)	$P = 0.001$
BMI (kg/m^2)	Median (IQR)	17.8 (16.4–19.8)	22.4 (18.8–23.7)	18.9 (16.8–21.7)	$P < 0.001$
FEV_1 (% predicted)	Mean (range); SEM	74 (35–115); 2.8	80 (42–100); 3.5	—	ns
Age onset menarche (years)	Mean (range)	12.6 (11–15)	12.1 (10–14)	11.5 (10–14)	$P = 0.006$
	SEM	0.20 (n = 27)	0.31 (n = 16)	0.21 (n = 19)	

Statistical comparison is made between CF children vs. asthma and controls. Normally distributed data are expressed as mean and standard error of mean (SEM) and analyzed using ANOVA, whilst non-parametric data are expressed as median and interquartile ranges (IQR) with analysis by Kruskal Wallis test (ns, non-significant).

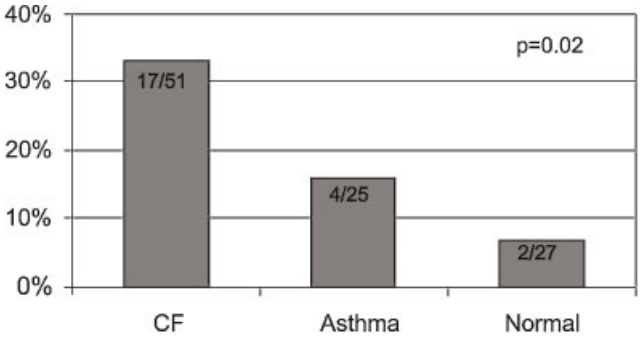

Fig. 1. Prevalence of urinary incontinence. Statistical comparison is made between CF children vs. asthma and controls.

drops only). Three girls reported larger volume leakage, with two reporting complete emptying of the bladder. Two girls in the study reported large volume leakage and requested help. In both cases a referral was made to a specialist uro-gynecology team. In one case, following initial consultation with a female gynecologist with a special interest in pediatrics, further urodynamic investigation was declined but referral was made to a specialist physiotherapist with some success.

The most common everyday triggering factors were lifting (17/17), sneezing (11/17), laughing (10/17), and running (3/17). Seven girls reported UI to interfere with physical education classes when their chests were bad, and one reported problems even when her chest was good. Six girls said that UI interfered with airway clearance regimens and four also reported difficulty performing lung function tests. Twelve of the seventeen CF girls and all four asthmatic girls reported UI with cough. There was no association between amount of coughing and having UI or UI severity. Only one CF girl was on oral steroids.

Within the group of CF girls, those reporting UI had a significantly lower BMI (mean 16.6 vs. 18.6 kg/m^2, $P = 0.005$) and FEV_1 (62% vs. 80%, $P = 0.001$), but were not different in age. The average age of onset of menarche in the CF group (12.6 years) was significantly later than in the other groups (12.1 in asthmatic group and 11.5 years in healthy girls, $P = 0.006$). UI did not seem to be related to menarche within the CF group (Table 2).

The mean score (measured on the 0–10 VAS) of the perceived degree of distress caused by their UI in the 17 girls with CF was 3.5 (standard deviation 2.2) whilst that for the 6 girls in the combined control groups was 3.8 (SD 1.7), which was not statistically different. Only five of the girls reporting UI had previously reported this as a problem.

DISCUSSION

This study has provided evidence that the prevalence of UI is significantly higher in young girls with CF when compared with a similar cohort of girls with asthma and normal healthy controls. This study used a questionnaire based closely on that used by Orr et al.[2] in a previously reported survey of UI in adult females with CF. Only one other study to date has included a normal control group or another respiratory disease group.[5] Whilst no-one was excluded from taking part, the response rate of 75% in girls with CF (it was 100% in other subjects) means we cannot be certain how representative the results are for all girls of this age range in our clinic. We cannot know, for example, whether failure to respond was due to embarrassment from someone with UI or indifference from someone without it.

This study has highlighted the fact that UI can be a problem in girls as young as 11 years of age. Although some reports of an increased prevalence of UI in CF children have included those as young as 5 or 6 years of age,[12,13] this study used an older cohort (girls between the ages of 11 years and 17 years) in order to reduce the risk of reporting pathology more common in a younger pediatric population (e.g., ''giggle'' incontinence, urinary tract infection and behavioral problems).[12]

In contrast to a previous study, an association was found between disease severity and occurrence of UI.[16] The reasons for the discrepancy between these studies are unclear and most adult studies also report a similar association. With deteriorating respiratory status, prolonged coughing becomes an increasing problem and the

TABLE 2—Comparison of Baseline Characteristics Within CF Group of Those With and Without Urinary Incontinence (UI)

	CF UI	CF No UI	Mean difference	95% CI of difference	
Age (years)	13.5	13.9	−0.5	−1.5 to 0.5	ns
BMI (kg/m^2)	16.6	18.6	−2.0	−3.4 to −0.6	$P = 0.005$
FEV_1 (% predicted)	61.9	80.4	−18.4	−29.4 to −7.4	$P = 0.001$
Age at menarche (years)	12.6	12.6	0	−1.0 to 0.9	ns

Data are expressed as means, (ns, non-significant).

requirement of chest physiotherapy (which usually involves forced expiratory maneuvers) also increases. Although the precise mechanism of UI in females with CF is not known, we would speculate that stress incontinence may be due to frequent, excessive, and forceful expiratory maneuvers. The CF group reporting UI in this study had significantly lower BMI scores suggesting that nutritional factors may also be a contributory factor to muscle development and strength. The use of long-term oral steroids in children with CF is relatively uncommon and their use is therefore unlikely to be relevant to the occurrence of UI.

Seven of the girls with UI reported that it interfered with activities such as physical education classes when their chests were bad. Four of the seventeen also said it interfered with the performance of lung function tests and it is possible that this may lead to ''underperformance'' at times. One would expect UI to be perceived as a very distressing problem, which has a significant impact on day-to-day physical and social functioning, yet girls with CF rated the impact of UI to be relatively low. There is no doubt that UI is an embarrassing complication and many feel reluctant to report or discuss it, either with their own family or with their medical team, therefore, the problem may be underreported. All girls experience physiological, hormonal, developmental, and social changes during adolescent years. In CF, along with these changes girls at this age also begin to understand and acknowledge the impact of CF on their lives. Therefore, a possible explanation for the relatively low scores could be that in terms of CF as a whole, the impact of UI is perceived as a minor inconvenience.

Only 2/17 girls wished for referral to a specialist. Investigation of UI can include complex urodynamic studies that are invasive and embarrassing. Orr reported that adult females with CF are generally more likely to live with the problem, particularly if it is relatively mild.[2] A small study of 19 female adults with CF, referred for investigation and treatment of UI, reported a 30% rate of either withdrawal or reluctance to undergo further assessment (despite having requested help in the first instance).[17] Referral of children for urodynamic investigation should only be made after careful discussion with the child and her parents. Addressing this issue sensitively and routinely from an early age may help reduce embarrassment, and also provide an opportunity to discuss concerns before they become a problem.

Girls with CF have a significantly greater problem with UI compared to girls in a similar age group with asthma and healthy controls. The problem may manifest in girls as young as 11 years. This study indicates that the presence and degree of UI is associated with disease severity. Although perceived as a relatively minor problem by girls with CF, in a small number it has a much greater impact both physically and emotionally. UI is of increasing concern in adult females with CF. It is therefore essential that pediatric CF teams should be routinely and sensitively addressing UI as a potential problem in girls of all ages.

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