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Prevalence of wheeze during childhood: retrospective and prospective assessment

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ABSTRACT: The question "Has your child ever had wheezing or whistling in the chest at any time in the past?" is a simple and widely used proxy measure for the lifetime prevalence of asthma. Our aim was to test its validity in a longitudinal survey, comparing retrospective recall with prospective assessment of lifetime prevalence.

A population-based cohort of 1,422 children, surveyed twice previously, was studied again at age 8–13 yrs by postal questionnaire using standardized questions from the International Study of Asthma and Allergies in Childhood (ISAAC).

Of those traced (1,190) questionnaires were returned by 89%. The prevalence of current wheeze was higher than in the previous surveys (20.5% *versus* 12.4% and 12.5%). Reported "wheeze ever" increased significantly from survey 1 (15.6%) to survey 2 (22.4%) and survey 3 (39.2%) and was very similar to the cumulative lifetime prevalence assessed prospectively over three surveys (42.8%). The retrospective question had a good negative predictive value (97%) and a reasonable positive predictive value (65%) compared to prospective assessment. Children reporting "wheeze ever" (but not current wheeze) in surveys 1 and 2 had at survey 3 an asthma prevalence higher than never-wheezers but lower than current-wheezers.

It is concluded that retrospective recall of wheeze at age 8–13 yrs is a valid proxy measure for the lifetime prevalence of wheeze.

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Numerous surveys of wheeze in children have been conducted during recent years. Most of these studies tried to assess the lifetime prevalence of wheeze with questions of the type "Has your child ever had attacks of wheezing?" [1–3], or "Has your child ever had wheezing or whistling in the chest at any time in the past?" [4–6], as well as its current prevalence.

The validity of such a retrospective assessment of the lifetime prevalence of wheeze has been questioned because some surveys of children have failed to show a significant increase in "wheeze ever" with increasing age, and this has been explained by poor parental recall [1, 7, 8]. Most longitudinal surveys of childhood wheeze have been conducted in late childhood [9, 10], or even adults [11], so that poor recall seems likely. Parental recall may be more reliable when children are relatively young. If this is true, the question "wheeze ever" could still be used as a simple and reasonably valid proxy measure for the lifetime prevalence of wheeze in pre-adolescent children.

The Leicester longitudinal cohort is a population-based random sample of children born January 1985–January 1990 who have been followed up thrice between 1990–1998 at median ages of 3, 6 and 11 yrs [1, 2, 12, 13]. The aim of this study was to assess the validity of the question "Has your child ever had wheezing or whistling in the chest at any time in the past?", asked at the age of 8–13, as a proxy measure for the cumulative lifetime prevalence of wheezing illness up to this age.

Methods

Subjects and study design

An age-stratified random population sample of 1,650 Caucasian children born between January 1985 and January 1990 was identified in 1990 using the Leicestershire Health Authority child health database as the sampling frame. The methodology has been published in detail elsewhere [1, 12, 13]. Briefly, in April 1990, the parents of these children received a postal questionnaire on respiratory symptoms, family history of atopy, environmental and social conditions (survey 1 (s1)). A second survey (s2) of the same cohort was performed between March 1992 and April 1994 and included an interview of all children who had originally reported "attacks of wheeze ever" or "cough without a cold", together with a random sample of previously asymptomatic children [2].

In February 1998, when the children were aged 8–13 yrs, another postal questionnaire was sent to the parents of all 1,650 children who were originally selected (survey 3, s3). Nonresponders were mailed the questionnaire twice more at four-weekly intervals. The study was approved by the Leicestershire Research Ethics Committee, and parents were permitted to return the questionnaire uncompleted.

Questionnaire

As there was no suitable and well-validated questionnaire available for use in the preschool age group, the

questionnaire applied in the first two surveys was specifically designed for the study [1]. On the questionnaire, wheeze was defined as "a whistling sound coming from the chest, not the throat" and parents were asked to answer the questions "Has your child ever had attacks of wheezing?" and "During the past 12 months, how many attacks of wheezing has he/she had?". Children who answered positively to the first question and had at least 1 attack in the past 12 months were classified as "current-wheezers".

The questionnaire used in 1998 was significantly shorter than the original and questions on respiratory symptoms were replaced by standardized questions appropriate for school children from the International Study of Asthma and Allergies in Childhood (ISAAC) [5]. Wheeze was assessed with the questions "Has your child ever had wheezing or whistling in the chest at any time in the past?" and "Has your child had wheezing or whistling in the chest in the last 12 months".

Statistical analysis

All questionnaires were double-entered onto a personal computer using Epi Info Software, Version 6.04b (Center for Disease Control and Prevention, Atlanta, Georgia, USA). Subsequent analyses were performed using STATA, version 5.0 for Windows (STATA Corporation, Texas, USA). For the analysis of data from s2 survey estimation methods were used (svyprop, svymean, svylogit), which adjusted for stratification (wheezers, coughers and asymptomatic children) and unequal sampling fractions. Chi square tests and McNemar's Chi square test for paired data were used to test for statistical significance when comparing proportions, and likelihood ratio tests in logistic regressions.

Definitions

Retrospective lifetime prevalence of wheeze at s2 or s3 was defined as a positive answer to either question: "Has your child ever had attacks of wheezing?" (s2) or "Has your child ever had any wheezing or whistling in his chest at any time in the past?" (s3).

Cumulative prospective lifetime prevalence of current wheeze by s2 or s3 was defined as the cumulative prevalence of "wheeze in the last 12 months" up until (and including) the current survey. Cumulative prospective lifetime prevalence of wheeze ever by s2 or s3, was, accordingly, the cumulative prevalence of "wheeze ever" up until (and including) the current survey (s2 or s3).

Validity is the degree to which a measurement measures what it purports to measure [14]. Our data allowed us to assess construct validity, concurrent criterion validity and predictive validity. To assess construct validity (whether the measurement corresponds to theoretical concepts concerning the phenomenon under study [14]), the following were tested: a) whether the reported prevalence of "wheeze ever" increased with increasing age of the children; and b) whether the gap between the prevalence of "wheeze ever" and "current wheeze" increased with age (as we would expect with a periodic disease such as asthma). Concurrent criterion validity (comparison of the measurement with a criterion or "gold standard" at the same point in time) was assessed by comparing the retrospective lifetime prevalence of wheeze at s3 with the corresponding prospective cumulative current prevalence of wheeze (our "gold standard") by s3. Predictive validity (ability of the measurement to predict the criterion) was assessed by comparing the prognosis of children who, at s1 or s2, were said to have "wheezed ever" (but not currently) with the prognosis of "current-wheezers" and "never-wheezers". As prognostic outcomes "current wheeze", "current asthma", "use of a bronchodilator in the past 12 months" and "use of an inhaled steroid in the past 12 months" as reported in s3 were chosen.

Results

Response rate and sample representitiveness

Of the 1,650 children originally selected, two had died, 74 emigrated, 48 had no updated address, and 6 refused to participate. In total, 1,305 children (86% of those traced) returned the questionnaire (table 1). This included 1,190/1,422 of the children of s1 (89% of those traced) and 442/488 of the children who had taken part in both earlier surveys (94% of those traced). There was no significant difference in symptom prevalence by type of respondent (in 87% the mother, in 10% the father, in 4% both) nor by mailing (82% answered the first, 15% the second and 3% the third mailing). Data quality was excellent with few missing data (0–2% per question) and very few inconsistencies or logical errors (<1%). Data from s1 were available for 138/215 nonresponders and for 91/122 children lost to follow-up. Responders did not differ from these two groups of nonparticipants in terms of age, sex, symptom prevalence, severity of wheeze, family history of

Table 1. – The Leicestershire longitudinal cohort: characteristics of the children followed up in each sweep (survey 1, 2 and 3)

	Survey*		
	1	2	3
Date of survey	April 1990	March 92–April 94	February 1998
Time interval from survey 1 yrs	-	3.1 (1.8–4.1)	7.8 (7.4–8.3)
Time interval from survey 2 yrs	-	-	4.7 (3.8–6.9)
Completed questionnaires	1422	488*	1305
Response rate**	86	61	86
Age of children at survey yrs	2.9 (0.2–5.4)	5.8 (3.9–8.8)	10.7 (8.1–13.2)
Number of females n%	698 (49%)	242 (50%)	645 (49%)

For continuous data (time intervals, age), median and range is given. *: survey 2 included all children reporting "wheeze ever" (222) or chronic cough without wheeze (226) in the first survey and a random sample of asymptomatic children (347). Response rates were 65% for wheezers (145), 58% for coughers (130) and 61% for asymptomatic children (213). **: completed questionnaires/traced children.

atopy, diagnosis of asthma and eczema, hospitalizations, medical follow-up, treatment, and social class. However, responders were less likely to be exposed to a smoking father (33% *versus* 43%) or mother (28% *versus* 38%) and to smoke during pregnancy (26% *versus* 36%, all p values <0.01).

Because of the relatively low response rate in the second survey (table 1), it was essential to check if the 442 children with a linked dataset from all three surveys were representative of the whole cohort. In a logistic regression adjusting for the stratified sampling in s2, children with a linked dataset were not significantly different from the other children for: prevalence of any respiratory symptom, asthma severity, diagnosis of asthma, hayfever or eczema, or treatment for asthma either at s1 or at s3. They were, however, less likely to belong to a low social class (odds ratio (OR) 0.7, p=0.002) or to have a smoking father or mother (both OR 0.7, p<0.02) and more likely to keep pets (OR 1.4, p=0.04) compared to the other children (in both surveys).

Prevalence of current wheeze

The prevalence of current wheeze did not change with age within each survey, but was significantly greater in s3 than in s1 and s2 (p<0.0001, table 2). It is unlikely, that this is due to a heightened awareness of wheeze after repeated participation in questionnaire surveys, because there was rather a trend towards a higher prevalence of current wheeze and wheeze ever in the 130 children who participated for the first time in s3 (25.7% and 46%) compared to the children who had taken part in all three surveys (18.8% and 39.2%).

Validity of the retrospective assessment of lifetime prevalence of wheeze

Construct validity. When the cross-sectional data were analysed by comparing prevalence of wheeze in different age groups within each survey, there was no significant increase in the prevalence of "wheeze ever" by age in boys within any survey. In girls, there was a trend for a rising prevalence of "wheeze ever" with age within surveys (significant for s1; p=0.03).

When data were compared longitudinally (between surveys), a highly significant increase of wheeze ever with increasing age (s1 *versus* s3, paired test p<0.0001, table 2 was found). The proportion of children with current wheeze among those who "wheezed ever" decreased with age from 79% (CI 74–85%) in s1 to 48% (40–56%) in s2 and 44% (37–51%) in s3.

Concurrent criterion validity. The retrospective lifetime prevalence of wheeze ("wheeze ever" at s2 or s3) was only insignificantly lower than the corresponding prospective (cumulative) prevalence of "wheeze ever" using data from all earlier surveys and considerably higher than the cumulative prevalence of current wheeze at any survey (table 2). The validity of the retrospective assessment ("wheeze ever" at s3) was determined by comparison with the prospectively assessed cumulative prevalence of current wheeze (table 3). Compared to this standard, the question "wheeze ever" had a very good negative predictive value (97%). This suggests that if parents of a child aged 8–13 yrs report, that their child has never wheezed, it is very likely to be true. The positive predictive value was lower (65%). This means, that one third of the children said to have "wheezed ever" at s3 did not report current wheeze in any of the three surveys. However, as the intervals between the surveys were much longer than 12 months, these children may have wheezed for a short time between the surveys only.

Predictive validity. Table 4 shows the prognosis at s3 of children, who, at survey 1, reported to have wheezed ever (but not currently), in comparison with never-wheezers and current-wheezers. Prognosis for "ever wheezers" was in between the prognosis of current wheezers and never-wheezers and significantly different from both groups after adjusting for age. The same results were found comparing the outcome at s3 of children who, at s2, had been current-wheezers, ever-wheezers or never wheezers (data not shown).

Discussion

The third follow-up of the Leicestershire longitudinal cohort, a representative population-based sample of children who were first surveyed when aged 0–5 yrs and followed up twice since then [1, 12, 13] is reported. The original sample was representative of the Caucasian population of Leicestershire and eight years later, 89% of

Table 2. – Cross sectional prevalence of wheeze as reported in each survey and cumulative lifetime prevalence of wheeze (calculated using data from all earlier surveys)

	Survey		
	1	2	3
Cross-sectional prevalence of wheeze			
Current wheeze (last 12 months):			
All children	12.4 (10.7–14.1)	12.5 (9.7–15.2)	20.5 (18.3–22.7)
Linked data*	12.0 (10.9–13.1)	11.0 (8.3–13.8)	18.8 (15.1–22.5)
Wheeze ever:			
All children	15.6 (13.7–17.5)	22.4 (19.2–25.6)	39.2 (36.5–41.8)
Linked data*		20.8 (17.6–24.0)	39.2 (34.5–43.9)
Cumulative lifetime prevalence of:			
Current wheeze**		18.6 (16.0–21.3)	26.9 (23.1–30.8)
Wheeze ever***		25.6 (22.5–28.6)	42.8 (38.1–47.5)

Data are displayed as proportions with (95% confidence intervals). *: children who took part in all three surveys (n=422); **: current wheeze at any survey (1, 2 or 3); ***: "wheeze ever" at any survey (1, 2 or 3).

Table 3. – Validity of the question "wheeze ever" asked at survey 3 compared to the cumulative lifetime prevalence of current wheeze

"Wheeze ever"	Cumulative prevalence of current wheeze* (prospective) % (n)	
	Yes	No
Yes	26 (152)**	14 (61)
No	2 (11)	59 (216)

*: Cumulative prevalence of current wheeze is defined as a report of wheeze in the previous 12 months at any survey (1, 2 or 3); **: weights were used for calculating the proportions (to adjust for unequal sampling fractions in survey 2). Sensitivity, pint estimate (95% confidence intervals) 0.94 (0.90–0.98); Specificity, 0.81 (0.76–0.86); Positive predictive value, 0.65 (0.58–0.73); Negative predictive value, 0.97 (0.96–0.99).

children who could be traced replied to the questionnaire (84% of the original cohort). This compares favourably with other cohort studies [9, 15]. Over 20% of children aged 8–13 reported current wheeze and 39% reported "wheeze ever". This is similar to results from recent parent-completed questionnaire surveys in the UK [16, 17].

The aim of this paper was to assess the validity of the question "Has your child ever had wheezing or whistling in the chest at any time in the past?" asked at the age of 8–13 yrs, as a measure of lifetime prevalence of wheezing. That question is part of most standardized asthma questionnaires and has been used in hundreds of cross-sectional surveys including the recent ISAAC studies [5].

Data on lifetime prevalence are potentially very interesting, because period prevalence of wheeze gives only an incomplete picture of the burden of illness of a remitting disease such as asthma [18]. Surveys in older children and adults, however, suggest that a substantial proportion of childhood symptoms is forgotten later in life [9–11, 19]. In the large British National cohort, first surveyed at age 7 yrs, the prospectively assessed cumulative prevalence by age 11 yrs was twice as high as the retrospective prevalence by age 11 (22 *versus* 12%) respectively, and the difference persisted at age 16 yrs (25% *versus* 12%) and 33 yrs (43% *versus* 28%) [9, 19]. In Tasmania, 1,494 children recruited at age 7 were questioned when aged 30. Of 741 subjects with parent-reported childhood asthma by the age of 7, 327 (44%) did not recall that they had ever wheezed [11]. Parental recall of preschool wheeze in schoolchildren may be more reliable, but the validity of this widely used question has not yet been assessed for this age-group.

In our cohort the question had good construct validity, with the prevalence of a report of "wheeze ever" increasing significantly with increasing age in the longitudinal data set (15.6%, 22.4%, 39.2%). The proportion of children with current-wheeze among children reported ever to have wheezed decreased with age, as would be expected from a disease with a variable natural history including periods of remission. The retrospective lifetime prevalence in our cohort (39.2%) was almost identical to the prospectively assessed cumulative lifetime prevalence over the previous surveys (42.8%). A negative predictive value of 97% suggests that preschool symptoms had rarely been forgotten by the parents of 8–13 yr olds. The relatively low positive predictive value (only 65% of children reporting "wheeze ever" at s3 had reported current wheeze in one of the surveys), may at least partly be explained by the long intervals between the surveys, since children with transient wheezing between the surveys would have been missed. This incorrectly labels some answers as false positives. Ideally, "current wheeze" should have been assessed at yearly intervals. Finally, good predictive validity for the question "wheeze ever" was found. Children reporting "wheeze ever" (but not current-wheeze) at s1 or s2 had a worse prognosis at s3 for any outcome (current wheeze, current diagnosis of asthma, use of bronchodilators or topical steroids) compared to never-wheezers. This may have implications for future public health interventions. If a preschool population is screened to identify children at risk of suffering from asthma later, simple preventive interventions should be considered for "ever wheezers", who constitute an intermediate risk group, as well as for active wheezers.

Why was the validity of the retrospective assessment of lifetime prevalence of wheeze better in our cohort than in most other longitudinal surveys? The main reason may be the age range of the observation period: recall by parents may be better for events in early childhood, when the child/parent contact is closer, than in the older age-groups (7–23 yrs) studied in the British National cohort [9, 10]. In Tasmania, the answers of grown-up offspring was compared with previous questionnaires answered by their parents [11]. It is not surprising that adults cannot reliably recall transient symptoms from their own childhood. Another reason for our high rate of recall might be the higher proportion of wheezing children who are nowadays given the diagnosis and treatment for asthma. A symptom, which is labelled and treated with a special device (an inhaler), is more likely to be remembered.

The present results might have been influenced by a change in wording between surveys. If the question about "wheezing or whistling in the chest" (s3) were more likely to elicit a positive response than questions about "attacks of wheeze" (s1 and s2) this might falsely suggest good recall. Our data do not support a large effect caused by this

Table 4. – Comparison of prognosis of children who have never wheezed, wheezed "ever" but not currently and current wheeze

Outcome at Survey 3	Wheeze as reported at Survey 1			Odds ratio*	
	Never	Ever	Current	Ever *versus* never	Current *versus* ever
Subjects n	1006	43	141		
Current wheeze %	15 (13–18)	33 (18–47)	48 (40–57)	2.6 (1.3–5.0)	2.4 (1.1–5.2)
Current asthma diagnosis %	12 (10–14)	28 (13–42)	44 (35–52)	3.0 (1.5–6.3)	2.4 (1.1–5.1)
Bronchodilator %	14 (12–17)	38 (23–53)	50 (42–59)	3.5 (1.8–6.7)	2.1 (1.0–4.3)
Steroid inhaler %	8 (6–10)	16 (5–28)	34 (26–43)	2.3 (1.0–5.4)	3.6 (1.3–9.9)

Data is represented as % (range). *: adjusted for age at Survey 1.

change of wording: the number of children who had at least 1 "attack" of wheeze in the past 12 months was identical to the number of children reporting "wheezing or whistling in the chest" (20.1% compared to 20.5%). In the 1998 repeat cross-sectional survey of preschool children [20] current "wheezing or whistling in the chest" was reported in 26% (95% CI 24%–28%) of the Caucasian children, and "attacks of wheezing" in the past 12 months in 24% (23–26%).

Lastly, the design of our study is not perfect for assessing cumulative prevalence of wheezing with precision. As the three surveys were separated by intervals of several years, periods of wheeze of short duration may have been missed. Ideally, the cumulative prevalence of wheeze would need to be assessed by continuous measurement of symptoms or by very frequent surveys (not more than one year apart).

In conclusion, this is, to the authors′ knowledge the first study validating the retrospective recall of preschool wheeze in parents of school-age children. In our longitudinal cohort, the question had reasonable validity as a proxy measure for cumulative lifetime prevalence up to age 8–13 yrs. The results, however, may not apply to other age groups or to regions with a much lower prevalence of or treatment for wheeze.

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