METHODS

Estimating non-response bias in family studies: Application to mental health and lifestyle

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Abstract. Non-response to mailed surveys reduces the effective sample size and may introduce bias. Non-response has been studied by (1) comparison to available data in population based registers, (2) directly contacting non-respondents by telephone or single-item reply cards, and (3) longitudinal repetition of the survey. The goal of this paper was to propose an additional method to study non-response bias: when the variable of interest has a familial component, data from respondents can be used as proxy for the data from their non-responding family members. This approach was used with data on smoking, alcohol consumption, physical activity, coffee- and tea-use, education, body mass index, religion, burnout, life events, personality and mental health in large number of siblings and DZ twins registered with the Netherlands Twin Register. In addition, for smoking behavior, we also used the second strategy by sending a reply card. Results show that scores of members from less cooperative families or incomplete twin pairs tended to be more unfavorable than the scores from highly cooperative families or complete twin pairs. For example, family members from less cooperative families cycled less often and scored higher on anxious depression and neuroticism. For smoking, both the results of the reply card and the results of the additional method suggested a higher percentage smokers among the non-respondents but this was only significant with reply card method. In general, differences between highly/less cooperative families and complete/incomplete DZ twins were small. Results suggest that, even for studies with moderate response rates, data collected on health, personality and lifestyle are relatively unbiased.

Key words: Health and lifestyle, Mailed survey, Mental state, Non-response, Personality, Twin-family design

Introduction

Mailed surveys are widely used to collect data on health and lifestyle in large populations. In Europe, response rates to mailed surveys vary from 52 to 95% [1]. Non-response to mailed questionnaires reduces the effective sample size and therefore the statistical power of the study. Moreover, survey results will be biased by non-participation if refusal to participate is not distributed randomly, and is either directly or indirectly related to the traits under study. Although studies usually recognize the risk of response bias, they are often unable to quantify the degree of bias.

Studies quantifying response bias may use different methods to obtain information on the non-respondents. First, when access is available to population based registers like health insurance databases, utilization databases or population registers, it is possible to compare respondents with non-respondents with regard to the information provided by the registers. In general, studies using this method [2–4] have shown differences between non-respondents and respondents; for example, non-respondents had lower annual incomes, more sickness benefit days and were more often unmarried. A drawback of this method is that the response bias can only be examined with regard to the available – often rather general – characteristics in population registers and cannot indicate the degree of response bias regarding the more specific characteristics of interest in comprehensive survey studies.

A second method to quantify response bias is obtaining this specific information by contacting the non-respondents themselves either by telephonic interview or by sending a reply card. A study that used a telephonic interview to obtain information on the non-respondents showed statistically significant differences between respondents and non-respondents for smoking status, hazardous alcohol consumption and lack of vigorous activity [5, 6]. Although such a telephonic interview provides valuable information on non-response, there will always remain a group of non-respondents who either cannot be reached by phone or will be unwilling to participate.

Longitudinal studies provide a third source of information on non-response by allowing the
comparison of respondents and non-respondents at later follow-up, using the information obtained at the start of the study. Most of those studies have found small or no differences between respondents and non-respondents [7–10]. Subjects who repeatedly returned a questionnaire tended to be married, non-smokers and more physically active than those who returned it only once. However, a possible problem with these studies is that they are not based on random samples; the original study population at the first measurement itself may already have been a selected sample.

Here we propose an additional method to obtain information on non-respondents which is based on family and/or twin designs. We will concentrate on general demographic (education), lifestyle variables (smoking, alcohol use, physical activity, coffee- and tea-consumption, religious practice) and personality/mental health (body mass index, burnout, problem behavior, neuroticism). These variables are familial, that is family members resemble each other for those characteristics [11–18]. Therefore, data from non-respondents will be correlated with the data from the respondents and data from responding family members thus will offer information on the non-respondents. We illustrate this approach with data of twins and their siblings collected in 2000 in a survey study on health, personality and lifestyle of the Netherlands Twin Register (NTR).

Acting on the idea that health, lifestyle and personality of the non-responding subjects is reflected by the values obtained on the responding family members we first investigate whether the answers on health, lifestyle and personality variables are different for siblings from highly cooperative families (more than 80% of the family members participated) compared to siblings from less cooperative families (less than 80% of the family members participated). With this method the non-response bias is estimated using information of the responding family members. In addition, we compared data from DZ twins from complete pairs (both twins completed a questionnaire) with data from DZ twins from incomplete twin pairs (co-twin did not participate in the survey study). Data of dizygotic (DZ) twins were used because DZ twins share on average 50% of their genes, just like siblings. However, DZ twins are a select group and may have some distinct features in common which are not generalized to a singleton population (e.g. same age).

Methods

Participants

This study is part of an ongoing twin family study on health-related behavior of the NTR that assesses families with adolescent and (young) adult twins every two/three years since 1991 [19]. For the present study, the data from the 2000 survey were used.

In May 2000, questionnaires were sent to 13,724 twins/triplets and 2889 siblings. In July 2000 a reminder was send to the non-respondents. Because smoking was an important theme in the 2000 survey, the reminder contained a pre-stamped reply card with a question on their smoking status (smoker/ex-smoker/non-smoker). The reply card was returned by 2676 persons (2138 twins and 538 siblings) who were not willing to complete the questionnaire. The question on smoking behavior on the reply card was answered by 2473 of the 2676 non-respondents.

Twins and siblings who registered after May 2000 also received a questionnaire (n = 564 twins and n = 776 siblings) but not a reminder. Twins registered themselves, while most siblings were recruited by asking their mother for their addresses. In total, questionnaires were send to 14,288 twins/triplets and 3665 siblings from 7223 families. The average family size of the families that were invited to complete a questionnaire was 2.48 (SD 0.99). At the end of the data collection, 4609 twins/triplets and 1474 siblings from 3178 families had completed a questionnaire booklet (Figure 1). For the same sex twins, zygosity was based on questionnaire data or, when available, on DNA typing (zygosity based on DNA was available for 26.1% of the same sex twins). Agreement between zygosity based on questionnaire data and zygosity based on DNA results was 98%. For the opposite sex twin pairs, zygosity is known (DZ) based on their sex. The average family size was 1.91 (SD 0.94). The triplets (41 persons from 22 families), the half-siblings (n = 27) and adoption siblings (n = 5) were excluded from the analyses.

Data analyses

The percentage smokers were compared for respondents and non-respondents using χ² tests.

Familial correlations were calculated for all dependent variables. For the categorical traits tetrachoric correlations were calculated with a threshold model on raw data using MX [20]. The correlations between DZ twins and siblings were constrained to be equal to estimate the familial correlation. For the continuous variables intraclass correlations were calculated from an ANOVA analyses using all DZ twins and sibships [21].

Data from respondents of highly cooperative families were compared with data from respondents of less cooperative families. For each family, the number of respondents was divided by the number of family members who were asked to complete a questionnaire. When less than 80% of the family members participated, the family was marked as ‘less cooperative family’ and when 80–100% of the family members participated the family was marked as ‘highly cooperative family’. The dataset contained