Predictors of non-response to successive waves of surveys in the Japan Environment and Children’s Study during the 3-year postpartum period: a longitudinal cohort study

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ABSTRACT

Objectives We examined changes in factors related to non-response to successive waves of the nationwide birth cohort study, the Japan Environment and Children’s Study (JECS), during the first 3 years after childbirth.

Design Longitudinal cohort study.

Setting As the baseline survey, mothers completed self-administered questionnaires distributed by hand during pregnancy or 1 month after delivery. The self-administered questionnaires that we used in this study were then distributed by mail every 6 months until the children were 3 years old, for a total of six times.

Participants Of 103,060 mothers who consented to participate in the JECS during pregnancy, 88,489 mothers were included in the study after excluding those with multiple births, miscarriages or stillbirths and those who withdrew from the study within 3 years after providing informed consent.

Primary and secondary outcome measures Data were collected at the baseline survey on participants’ socioeconomic status, medical history, health status, health-related behaviours and their children’s health conditions and living situations. The strength of the impact of related factors and the prediction of response status were examined and compared using binomial logistic regression analysis.

Results For all six follow-up questionnaire surveys, higher maternal age was strongly associated with providing a response. Factors that were strongly associated with mothers not providing a response were smoking after childbirth and having more children. The concordance rate of response status based on the presented model was about 70%, suggesting that the response status for the first 3 years after birth can be predicted from the information collected in the baseline survey.

Conclusion By identifying predictors of non-response from information obtained in baseline surveys, researchers may be able to reduce non-response to successive survey waves by issuing reminders, reviewing data collection methods and providing appropriate financial and/or non-financial incentives.

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ The Japan Environment and Children’s Study (JECS) is a large nationwide longitudinal birth cohort study that includes 103,060 mothers with confirmed obstetric outcomes recruited between 2011 and 2014.
⇒ JECS covered 45% of live births that occurred within the study areas in 2013.
⇒ The data on the characteristics of the mothers and children who participated in JECS show similarities with the data from Japan’s 2013 Vital Statistics Survey.
⇒ The sample size of this study was sufficient to examine the risk factors of non-response.
⇒ The study has three main limitations: it had a short follow-up period, the data were analysed from continuing participants only and the mothers’ social environment or sudden life events were not examined.

BACKGROUND

A common issue in conducting longitudinal cohort studies is that some participants are lost to follow-up. This lack of response reduces the sample size set during the planning stage of the study, which negatively impacts the analytical results and statistical power of the study. Also, certain characteristics related to questionnaire response rates may cause selection bias that affects the results. Earlier research has confirmed that participants’ interest in and understanding of the diseases or health-related behaviours being studied affect questionnaire response rates, especially in health-related studies. In cross-sectional studies, participants’ response or non-response was associated with their socioeconomic status, and participants interested in the aim of the study were more responsive.1-4 On the other hand, a longitudinal birth cohort study found that higher socioeconomic status and higher maternal age were associated with a
Figure 1  Flow chart of the participant selection. *Reasons for censoring included death of mother, death of child, loss of custody and inability to follow-up with unknown address. JECS, Japan Environment and Children’s Study.

The first year after delivery. In these studies, we found that children reach 13 years of age. The characteristics of the children from the time of pregnancy until the end of the follow-up period is ongoing, and data on the children’s health status and living situation of their children. The questionnaire about the children that we analysed in this study were distributed by mail to their mothers every 6 months until the children were 3 years of age. The questionnaire was designed to be answered in about 30 min and mothers were given 1 month in which to return it. A reminder about the study was sent by post one or more times until the next questionnaire was sent. Participants received financial and non-financial incentives for each questionnaire returned. Change of address was identified from the participants’ self-reporting of a new address.

METHODS
Study design
The study design has been reported elsewhere. This study used JECS dataset (jecs-ta-20190930), released in October 2019 for stakeholders (The dataset supporting the conclusions of this article will be available after the steering committee of the JECS permits accessibility.). This dataset included 103,060 pregnant women who consented to participate in JECS. After excluding those with multiple births, miscarriages or stillbirths and those who withdrew from the study within the first 3 years, data from 88,489 mothers were analysed in this study (figure 1). The initial baseline survey, which was conducted during pregnancy or at 1 month after delivery, involved the distribution and collection by hand of a self-administered questionnaire to mothers. The data collected were the socioeconomic status, medical history, health status and health-related behaviours of the mothers and the health status and living situation of their children. The questionnaire about the children that we analysed in this study was distributed by mail to their mothers every 6 months until the children were 3 years of age. The questionnaire was designed to be answered in about 30 min and mothers were given 1 month in which to return it. A reminder about the study was sent by post one or more times until the next questionnaire was sent. Participants received financial and non-financial incentives for each questionnaire returned. Change of address was identified from the participants’ self-reporting of a new address.

Definitions
Children diagnosed as having physical anomalies or suspected of having physical anomalies by an obstetrician were classified as ‘having physical anomalies’. Psychological distress during pregnancy, as measured by the Kessler 6 scale (Japanese version), was defined as a K6 score ≥13, and postpartum depression, as measured by the Edinburgh Postnatal Depression Scale (EPDS), was defined as an EPDS score ≥9.

The postpartum health status of the participants at 1 month after delivery was categorised into the following categories according to the participants’ subjective assessment of their health status: ‘poor’, ‘fair’, ‘good’ and ‘very good’.

The primary caregivers of the JECS participants’ children at 1 month of age were categorised into two groups: ‘mother’ and ‘other than mother’. The number of siblings living with the child was categorised into three groups: 0, 1 and ≥2. Participants’ education status was categorised into two groups: <13 years and ≥13 years.

Annual household income was categorised into three groups: <4 million yen, ≥4 million and <12 million yen and ≥12 million yen.

The status of partner participation in JECS was categorised as follows: ‘non-participation’, ‘inactive participation’ (ie, the partner consented to participate in JECS but never returned the questionnaire for
partners) and ‘active participation’ (ie, the partner consented to participate in JECS and returned the questionnaire for partners).

**Patient and public involvement**
There was no patient or public involvement in this study.

**Statistical analysis**
To calculate ORs and 95% CIs, binomial logistic regression analysis was performed using participant characteristics (ie, age at delivery, years of education, smoking, alcohol consumption, psychological distress during pregnancy and postpartum depression), the health status of the child (ie, presence/absence of physical anomalies), partner participation status in JECS, primary caregiver and the number of siblings living with the child. A significance level of 0.01 (two-tailed) was used for all statistical analyses. ORs were considered statistically significant when 1.0 was not included within the 95% CI. SAS V.9.4 (SAS Institute, Cary, North Carolina, USA) was used for all statistical analyses.

Eighty per cent of the subjects were randomly sampled and used to create the prediction formula, and the remaining 20% were used to verify the prediction formula.

**RESULTS**

**Participant characteristics**
Response rates to the six successive follow-up questionnaires, calculated as the number of returned questionnaires divided by the number of participants, were 94.7%, 92.1%, 90.2%, 88.4%, 86.7%, 85.1%, respectively.

The characteristics of the participants are presented in table 1. The age at delivery of non-respondents was lower than that of respondents in all survey periods. Both postpartum smoking and alcohol consumption were higher in the non-respondent group than in the respondent group in all survey periods. In terms of self-reported maternal health status, the rate of ‘very good’ was lower and that of ‘poor’ was higher among non-respondents compared with respondents in all survey periods. Non-respondents had fewer years of education than respondents in all survey periods. Psychological distress during pregnancy and/or depression after delivery was more severe among non-respondents in all survey periods.

The proportion of children diagnosed as having physical anomalies was similar among respondents and non-respondents in all survey periods. The number of siblings living with the child was higher among non-respondents than respondents in all survey periods.

The proportion of mothers who were the primary caregiver of their child was lower among non-respondents in all survey periods. The proportion of partners who did not participate or inactively participated was higher among the non-respondents in all survey periods.

**Factors related to non-response**
Factors related to non-response in each research period were investigated and the results were used to create a prediction formula. The results of the logistic regression analysis of the response status to each questionnaire item (ORs and 95% CI) and accuracy of the prediction formula are shown in table 2.

In all survey periods, age at delivery, psychological distress during pregnancy, education status, postpartum smoking status, number of siblings living with the child and partner’s participation status in JECS were strongly related to response status.

Higher maternal age at delivery and longer years of education were associated with providing a response to follow-up questionnaires in all survey periods. In contrast, having psychological distress during pregnancy, postpartum smoking, having a greater number of children and having another person other than the mother as the child’s primary caregiver were associated with increased non-response to the follow-up questionnaires in all survey periods. Moreover, inactive participation of partners in JECS was associated with increased non-response to questionnaires, while partner’s active participation was associated with providing a response.

Weaker health status after delivery was associated with increased non-response to questionnaires in all survey periods. Postpartum depression was associated with increased non-response to the questionnaires, but the effect was statistically significant in only the survey conducted at 3 years. Drinking alcohol after childbirth was significantly associated with non-response to questionnaires. Higher family income was associated with higher response, but the highest family income bracket showed an increase in non-response to the questionnaires in all survey periods, although the effect was statistically significant in only the survey at 6 months.

The Area Under the Curve (AUC) of Receiver Operating Characteristic curve (ROC) using the results of binomial logistic regression analysis was less than 0.71.

**DISCUSSION**

JECS collects information on the developmental environment and the growth and development of children based on the survey responses of participants, most of whom are mothers. We previously examined the factors related to non-response to questionnaires distributed during the first year after childbirth; these factors included postpartum physical condition, number of siblings living with the child, the child’s caregiver at 1 month of age and partner participation. In the present study, we attempted to predict the questionnaire response status by using related factors collected in the baseline survey and examined the differences in the impact of these factors by survey period. We used data from approximately 100 000 mothers participating in JECS.

According to a number of earlier studies, health-related behaviours, socioeconomic status and poor health at the time of the questionnaire can affect the questionnaire response rate. 2 4 16–28 In addition to these factors, we found higher age at delivery and more years of education were strongly associated with providing a response over all 3 years after childbirth in this study. Higher
Table 1  Response and non-response rates in successive survey waves according to participant characteristics in the baseline survey

<table>
<thead>
<tr>
<th>Survey period (response rate)</th>
<th>6 months (97.4%)</th>
<th>1 year (92.1%)</th>
<th>1.5 years (90.2%)</th>
<th>2 years (88.4%)</th>
<th>2.5 years (86.7%)</th>
<th>3 years (85.1%)</th>
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</thead>
<tbody>
<tr>
<td></td>
<td>Non-respondents (%)</td>
<td>Respondents (%)</td>
<td>Non-respondents (%)</td>
<td>Respondents (%)</td>
<td>Non-respondents (%)</td>
<td>Respondents (%)</td>
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<tr>
<td>Age at delivery (n=88,488)</td>
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<tr>
<td>&lt;25</td>
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<td>30–34</td>
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<td>≥35</td>
<td>21.3</td>
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<td>21.4</td>
<td>27.9</td>
<td>21.0</td>
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<td>46.0</td>
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<td>Good</td>
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<tr>
<td>Never smoked</td>
<td>42.9</td>
<td>59.4</td>
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<td>59.9</td>
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<td>Quit before pregnancy</td>
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<td>20.6</td>
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<td>Never drank</td>
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<td>Quit before pregnancy</td>
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<td>Number of siblings living with the child (n=88,489)</td>
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<td>0</td>
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<td>≥2</td>
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<td>Having child</td>
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<td>Non-respondents (%)</td>
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Table 1 Continued

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<tr>
<th>Survey period (response rate)</th>
<th>6 months (97.4%)</th>
<th>1 year (92.1%)</th>
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<td>Respondents (%)</td>
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<td>Child’s primary caregiver (n=88,489)</td>
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<td>49.4</td>
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<td>Family income per year (n=81,200)</td>
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<td>&lt;4 million yen</td>
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<td>39.3</td>
<td>49.2</td>
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<td>39.0</td>
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<td>4-12 million yen</td>
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<td>49.2</td>
<td>59.0</td>
<td>50.0</td>
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<td>≥12 million yen</td>
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<td>1.8</td>
<td>1.6</td>
<td>1.9</td>
<td>1.7</td>
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<td>Partner’s participation status* (n=88,489)</td>
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<td>56.6</td>
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<td>0.8</td>
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<td>0.8</td>
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<td>Active participation</td>
<td>34.6</td>
<td>51.9</td>
<td>38.2</td>
<td>52.0</td>
<td>38.5</td>
<td>52.3</td>
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</table>

*Partners who did not agree to participate or could not be reached to request their participation in JECS were classified under ‘non-participation’. Partners who agreed to participate but did not return the questionnaire were classified under ‘inactive participation’. Partners who returned the questionnaire were classified under ‘active participation’.

EPDS, Edinburgh Postnatal Depression Scale; JECS, Japan Environment and Children’s Study; K6, Kessler 6 scale.
family income was associated with response, although the highest income households showed some association with non-response. The time needed to answer the questionnaire and the financial incentives offered for responding may have been related to the reduced responses by participants in these high-income households. Thus, to help improve response rates, consideration should be given to the relationship between financial incentives and the time needed to answer the questionnaires, as well as any additional non-financial incentives offered.

Depression during pregnancy has been reported to be a predictor of postpartum depression. This may be
related to our results that psychological distress during pregnancy affected response status more strongly than postpartum depression. Although depression during pregnancy can be difficult to alleviate, postpartum depression often resolves at around 6 months after delivery and then relapses. The difference in the clinical course of depression during pregnancy and after childbirth may affect the response rate to follow-up questionnaires.

Previous studies have shown that health status, number of children and the primary caregiver affect questionnaire response rates, as we found in this study also. Having someone other than the mother as the child’s primary caregiver was associated with non-response to the follow-up questionnaires in all survey periods, although with lower ORs in the later survey periods. In growing families, the primary caregiver needs more time to care for the child’s siblings, which may have strengthened the association with non-response to questionnaires over time.

The effect of the child having physical anomalies on the response status was small and not significant, though a stronger association was seen for non-response to the survey at 6 months compared with the surveys at 1 year and later. The weaker associations seen in the later survey waves may be because mothers come to better understand the importance of providing data to JECS about their children with physical anomalies over time, given that one of the key purposes of JECS is to examine children’s health. To help to reduce the number of non-respondents among participants whose child has physical anomalies, greater efforts may be needed to sufficiently explain the purpose and importance of data collection in JECS during the first 6 months postpartum.

In this study, the partner’s participation status in JECS was associated with the mother’s questionnaire response status. We could find only one related study investigating the relationship between the presence of others and participation in a study, and this involved general practitioners (GPs) and paediatric cancer patients. In our study, there was an association between partners classified as active participants, who had agreed to participate in JECS and had returned their questionnaire(s), and response, and these partners were thus considered to be familiar with the aims of JECS. As such, they might have played a role similar to that of the GP in the other study, and the effect lasted for 3 years after childbirth. In contrast, non-response was associated with having inactive participants, who had agreed to participate but did not return their questionnaire(s), although this effect decreased over the 3 years. It is not clear from this study if the nature of the partner’s support influenced the mother’s response because we did not investigate this factor in detail. It is possible that the presence of partners who are supportive in parenting or housework may improve mothers’ response rates, and future studies should investigate the relationship between nature of their support and response rates.

Prediction using the information from the baseline survey revealed AUC values in the range of 0.66–0.7. This finding suggests that it would be possible to identify predictors of non-response using the participants’ information collected in a baseline survey. By identifying the predictors of non-response in this way, researchers may be able to reduce non-response to successive survey waves by issuing reminders, reviewing data collection methods and providing appropriate financial and/or non-financial incentives, such as offering chances to be involved in activities that the participants express interest in.

This study has several limitations. First, the follow-up period was only 3 years after childbirth. This study used the data of JECS participants who continued participation until the dataset was finalised. The data used in this study were from continuing participants only, so the factors influencing withdrawal from longitudinal birth cohort studies are not clear. Given that we examined the response status for only the first 3 years of follow-up questionnaires, it is not clear what factors may affect response over a longer period. Second, we excluded mothers with multiple births. This was done because, although multiple children would be included as participants in the JECS, the same mother would answer the questionnaire for each child. We considered that responses from the same mother would show the same tendency, resulting in bias. Third, not all partners of the JECS participants were contacted for participation in this study. Whenever possible, the participant’s partner was contacted in person by the investigator during the participant’s pregnancy in order to obtain informed consent. However, when the partner could not be reached, their participation status was classified as ‘participation refusal’ in the dataset due to the lack of informed consent. Finally, this study did not examine the relationship between questionnaire response status and the mother’s social environment or sudden life events or examine the cumulative effect to non-response. These aspects are left to future study. The strengths of this study are its large sample size and the wide range of participant characteristics collected. The total number of participants was 88,489, which is sufficient for examining response-related factors. JECS covered nearly half (45%) of the live births that occurred within the study areas in 2013, and the characteristics of mothers and children who participated in the JECS were similar to those who participated in the 2015 Vital Statistics Survey. Therefore, the results of this study have sufficient power for our analysis.

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Acknowledgements We thank the children and their families who participated in and cooperated with this study. We also thank Think SCIENCE for English language editing of this manuscript. The findings and conclusions of this article are solely the responsibility of the authors and do not represent the official views of the above-mentioned government.

Collaborators We thank all members of the JECs Group as of 2020: Michihiro Kamijima (principal investigator, Nagoya City University, Nagoya, Japan), Shin Yamazaki (National Institute for Environmental Studies, Tsukuba, Japan), Yukhiro Ohya (National Center for Child Health and Development, Tokyo, Japan), Reiko Kishi (Hokkaido University, Sapporo, Japan), Nobuo Yagashi (Tohoku University, Sendai, Japan), Koichi Hashimoto (Fukushima Medical University, Fukushima, Japan), Chisato Mori (Chiba University, Chiba, Japan), Shuichi Ito (Yokohama City University, Yokohama, Japan), Zentaro Nakayama (University of Yamanashi, Chuo, Japan), Takeo Nakayama (Kyoto University, Kyoto, Japan), Tomotaka Sobue (Osaka University, Suita, Japan), Masayuki Shima (Hyogo Medical University, Nishinomiya, Japan), Hiroshi Nakamura (Tottori University, Yonago, Japan), Narufumi Suganuma (Kochi University, Kochi, Japan), Koichi Kusuhara (University of Occupational and Environmental Health, Kitakyushu, Japan) and Takahiko Kato (Kumamoto University, Kumamoto, Japan).

Contributors MK designed and carried out this study and was the principal investigator. HK and MK collected and analyzed the data. All authors have read and approved the final manuscript. MK is responsible for the overall content as the guarantor.

Funding The Japan Environment and Children’s Study was funded by the Ministry of the Environment, Japan.

Competing interests None declared.

Patient and public involvement Patients and/or the public were not involved in the design, conduct, or reporting, or dissemination plans of this research.

Patient consent for publication Not applicable.

Ethics approval This study involves human participants and was approved by The JECs protocol was reviewed and approved by the Ethics Committee of all participating institutions. Written informed consent was obtained from all participants in this study. The protocol of this study was reviewed and approved by the Ethics Committee, University of Toyama (#20180669). Participants gave informed consent to participate in the study before taking part.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement Data are available upon reasonable request. Data are unsuitable for public deposition due to ethical restrictions and legal framework of Japan. It is prohibited by the Act on the Protection of Personal Information (Act No. 57 of 30 May 2003, amendment on 9 September 2015) to publicly deposit the data containing personal information. Ethical Guidelines for Medical and Health Research Involving Human Subjects enforced by the Japan Ministry of Education, Culture, Sports, Science and Technology and the Ministry of Health, Labour and Welfare also restrict the open sharing of the epidemiologic data. All inquiries about access to data should be sent to: jecs-enquiries@ja.jp. The person responsible for handling enquiries sent to this email address is Dr Shoji F Nakayama, JECs Programme Office, National Institute for Environmental Studies.

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