

# Characteristics of Postpartum Japanese Mothers Who Were Non-respondents to the Japan Environment and Children's Study

Mika Kigawa<sup>1\*</sup>, Akiko Tsuchida<sup>2,3</sup>, Mika Ito<sup>4</sup>, Tomomi Tanaka<sup>3,5</sup>, Kei Hamazaki<sup>2,3</sup>, Yuichi Adachi<sup>5</sup>, Shigeru Saito<sup>4</sup>, Hidekuni Inadera<sup>2,3</sup>, The Japan Environment and Children's Study Group<sup>6</sup>

<sup>1</sup>Faculty of Health and Social Services, Kanagawa University of Human Services

<sup>2</sup>Department of Public Health, Faculty of Medicine, Graduate School of Medicine and Pharmaceutical Sciences, University of Toyama

<sup>3</sup>Toyama Regional Center for Japan Environment and Children's Study, University of Toyama

<sup>4</sup>Department of Obstetrics & Gynecology, Graduate School of Medicine and Pharmaceutical Sciences, University of Toyama

<sup>5</sup>Department of Pediatrics and Developmental Medicine, Graduate School of Medicine and Pharmaceutical Sciences, University of Toyama

<sup>6</sup>National Institute for Environmental Studies, Tsukuba city, Ibaraki, Japan

## Abstract

**Background:** This study aimed to examine the factors behind survey non-response in mothers who had agreed to participate in a Japanese nationwide birth cohort study- The Japan Environment and Children's Study (JECS)- despite follow-ups. In this longitudinal cohort study, new mothers were asked to answer questionnaires one month after childbirth. Of 104,102 pregnant women who consented to participate, 96,860 were included in the study after excluding those with miscarriages, stillbirths or multiple births.

**Method:** Data from the self-administered questionnaires included participants' socioeconomic status, medical history, details regarding pregnancy and delivery, health status, and health-related behaviors. A JECS participation status for each participant's partner determined whether the partner provided an informed consent form and helped answer the survey. Multivariate logistic regression analysis was used to identify factors that could have potentially affected response/non-response to the survey.

**Results:** Survey non-response was related to the mother's age, socioeconomic status, health-related behavior, history of asthma, and psychological distress during pregnancy, as well as the child's health at birth. Additionally, active participation of partners in the JECS reduced the likelihood of non-response, and lack of partner participation significantly increased the likelihood of non-response.

**Conclusion:** The rate of survey response may be related to the level of the participants' interest and understanding of the study. In the future, approaches to the participant and others responsible for the care of the child should be considered to prevent non-response from ongoing surveys.

**Keywords:** Cohort study, Longitudinal study, Survey response, Survey non-response, Population characteristics, Regression analysis

## Introduction

The Japan Environment and Children's Study (JECS) is a nationwide birth cohort study that began in 2011. The JECS collects data from the gestation period until the child is 13 years old and aims to examine environmental factors related to children's health. Over three years, beginning in 2011, approximately 100,000 pregnant women were recruited to take part in the study. Data were collected via biological and environmental samples as well as survey questionnaires [1]. As of this writing, the data from the samples are available from one month after the child is born [2].

One of the issues when using survey questionnaires is non-response. Response rates for survey research in Japan have seen a decreasing trend, with even surveys conducted by national or local governments receiving only a 70% response rate [3]. This may affect the quality of data, since the statistical power could be affected if the number of data points were less than called for by the study design. More importantly, selection bias may be introduced if non-response is related to certain participant characteristics. In health-related survey research, previous studies have shown that survey responses are affected by participant interest and their understanding of the health problems being studied and relevant health behaviors [4-10]. As a result, in survey research, it is necessary to examine the characteristics of the sample from whom responses could not be obtained.

\***Correspondence to:** Mika Kigawa, Faculty of Health and Social Services, Kanagawa University of Human Services, Japan, Tel: +81-46-828-2752, E-mail:kigawa-c5s@kuhs.ac.jp

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## Materials and Methods

### JECS Study design

The JECS uses self-administered survey questionnaires to collect data about health status and other socioeconomic factors. This research examined two separate baseline surveys, the first conducted during early pregnancy (MT-1)- when pregnant women were recruited- and the second during mid-to-late pregnancy (MT-2). These questionnaires were delivered to women who were enrolled in the JECS and were collected either by the study's Research Coordinators in the obstetrics departments or by mail. At the time of recruitment, partners of the women who consented to participate were also asked to take part. Those who agreed were asked to complete a self-administered questionnaire (FT-1). Delivery and return of the FT-1 took place either directly with the study's Research Coordinators at a participating medical facility or via mail. Medical data of the mothers' pregnancies and deliveries as well as of their children (the population of interest of the JECS), were transcribed by physicians, midwives, nurses, and/or Research Coordinators from the facilities' medical records (Dr-0m). A self-administered survey was provided by the Research Coordinators, who collected responses from participating mothers personally or by mail. The survey was to be completed and returned one month after the child's birth.

### Current study design

The present study was based on two datasets that were distributed only among interested researchers from June 2016 to October 2016. These datasets are scheduled to be made publicly available in the future, but using them will require approval from the research group. The datasets consisted of information on 104,102 fetuses, including cases of miscarriages and stillbirths. Of these, the analysis used data from 96,860 participating mothers. Data from mothers who had joined the study more than once during the study's recruiting period (due to multiple pregnancies) and who withdrew from the study were eliminated. We classified mothers into two groups: respondents, who returned the questionnaire one month after the child's birth, and non-respondents, who did not return it. For analysis, we examined marital status, cohabitation, level of education, household income, medical history, health-related behaviors, and the number of hours worked per week. For health-related quality of life (HRQOL), the data used were from the Japanese version of the Short Form-8 Health Survey (SF-8) [11]. Data on psychological distress were from the Japanese version of the Kessler Psychological Distress Scale (K6) [12]. Data regarding delivery and neonatal health (age of gestation at birth, Apgar scores, physical anomalies, duration of hospital stay, and referral for specialized care) were obtained from medical records.

The mothers' ages at delivery were divided into four categories: under 25 years, 25-29 years, 30-34 years and 35 years and older. As for partner participation in the JECS, we created three categories according to whether a consent form and/or a completed survey questionnaire were received during the mother's pregnancy. The categories were as follows: non-participant (no consent form), inactive participant (agreed to participate in the study but delayed their response or did not return a completed survey), and active participants (agreed to participate and returned a completed survey). The gestational age of the children at birth was divided into three categories: less than 34 weeks, 34-36 weeks, and 37 weeks and more. For the Apgar score, seven was the cutoff point. Two groups appeared: those scoring above and below seven. The cutoff point for the duration of hospital stay for neonates was eight days; the two groups created were those who stayed in the hospital for fewer than eight days and those who stayed for eight days or more. Psychological distress was identified in those whose K6 score was greater than or equal to 13. Two levels of education were identified: those with less than 13 years and those with 13 or more years of schooling. Three household income levels were created: less than ¥4 million, ¥4 million to ¥12 million, and more than ¥12 million. Finally, for hours worked per week during pregnancy, responses were grouped into three categories: not working, working fewer than 40 hours/week (the 50<sup>th</sup> percentile calculated after excluding 'not working' participants), and working 40 or more hours per week.

### Ethical considerations

The JECS protocol was reviewed and approved by the Ministry of Environment's Institutional Review Board on Epidemiological Studies and the ethics committees of all participating institutions. Written informed consent was obtained from all participants.

### Analysis

Pearson's chi-squared and Fisher's exact tests were used to examine the differences in the characteristic of responders and non-responders in the survey as well as the participants' partners' survey participation categories. Binary logistic regression was then performed using the variables found to have a significant relationship with survey response to obtain odds ratios with a confidence interval (CI) of 95%. Additional analysis was performed to confirm the results using multivariate singular value decomposition to impute missing data. For the imputation of missing data, the method of singular value decomposition complement was used. This study used two-tailed tests with a significance level of 0.01. JMP® Pro version 11 (SAS Institute Inc., Cary, NC, USA) was used for the analyses.

## Results

### Participant characteristics

The response rate for the survey at one month after birth was 95.4% (92,464/96,860). Table 1 shows the participant characteristics from the baseline surveys. The participants who belonged to the responder group tended to be older than those who were part of the non-responder group. With regard to medical history, more non-responders had a history of bronchial asthma, depression, dysautonomia or anxiety disorder, while responders tended to have a history of allergic rhinitis, allergic conjunctivitis, or myopia. Based on the K6 scores, more women in the non-responder group were suspected to experience psychological distress during pregnancy. More women in the non-responder group had multiple

**Table 1.** Characteristics of the sample.

		Responder (n = 92,464)	Non-responder (n = 4,396)	
		%	%	p-value
<b>Participants characteristics</b>				
Age at delivery (n = 94,911)		(n = 92,458)	(n = 2,453)	< 0.001
	Less than 25	9.7	21.4	
	25-29	27.5	26.6	
	30-34	35.4	29.2	
	35 and over	27.4	22.8	
Medical history (n = 94,347)		(n = 91,811)	(n = 2,536)	
	Bronchial asthma	10.8	12.9	0.001
	Allergic rhinitis	36.1	32.4	< 0.001
	Allergic conjunctivitis	9.9	8.0	< 0.001
	Depression	3.0	4.6	< 0.001
	Dysautonomia	3.6	5.4	< 0.001
	Anxiety disorder	2.8	4.2	< 0.001
	Myopia	33.2	27.2	< 0.001
Self-Administered mental health (K6) during pregnancy				< 0.001
	Less than 13 points	96.5	94.4	
	13 points and over	3.5	5.6	
Multiple birth		2.4	1.8	0.006
<b>Problems at delivery</b>				
Abnormality of placenta/umbilical cord (n = 91,664)		(n = 89,506)	(n = 2,158)	< 0.001
		23.0	26.2	
Neonatal jaundice (n = 91,758)		(n = 89,667)	(n = 2,081)	< 0.001
		14.6	20.7	
Neonatal referrals for specialized care (n = 91,998)		6.1	20.0	< 0.001
Pregnancy complications (n = 92,826)		(n = 90,581)	(n = 2,245)	< 0.001
		15.1	18.6	
Obstetric complications (n = 92,924)		(n = 90,667)	(n = 2,257)	< 0.001
		46.8	54.1	
Neonatal physical anomalies		6.6	13.9	< 0.001
Apgar Index at 1 minute (less than 7)		2.2	21.0	< 0.001
The gestational age of the children at birth (n = 92,796)		(n = 90,563)	(n = 2,233)	< 0.001
	Less than 34 weeks	0.8	7.7	
	34 – 37 weeks	3.6	7.0	
	37 weeks and more	95.6	85.3	
Date of hospitalization (Child) (n=88,649)		(n = 86,739)	(n = 1,910)	< 0.001
	Less than 8 days	79.9	69.9	
	8 days and more	20.1	30.1	
<b>Health-related behavior and environment</b>				
Smoking habits during early pregnancy (n = 93,637)		(n = 91,153)	(n = 2,484)	< 0.001
	Never smoked	58.5	49.8	
	Smokers who quit before pregnancy	23.4	21.9	
	Smokers who quit after pregnancy	13.5	17.7	
	Smoker	4.6	10.6	
Some family members smokes in same room <sup>a</sup> (n = 93,517)		(n = 91,023)	(n = 2,494)	< 0.001
		53.4	60.0	
Exposed weekly to secondary smoke before pregnancy <sup>b</sup> (n = 93,880)		(n = 91,375)	(n = 2,505)	< 0.001
		18.2	25.2	
Smoking habits during pregnancy (n = 92,541)		(n = 90,933)	(n = 1,608)	< 0.001
	Never smoked	58.1	46.9	
	Smokers who quit before pregnancy	23.6	20.7	
	Smokers who quit after pregnancy	13.9	20.1	
	Smoker	4.4	12.3	
Alcohol consumption during early pregnancy (n = 93,897)		(n = 91,407)	(n = 2,490)	0.001
	Never drink	33.4	34.6	
	Drinkers who quit before pregnancy	55.5	57.5	
	Drinkers	10.1	7.9	
<b>Socioeconomic status</b>				
Education (n = 92,769)		(n = 91,165)	(n = 1,604)	< 0.001

	Less than 13 years	35.9	49.3	
	13 years and more	64.1	50.7	
	Family income per year (n = 86,549)	(n = 85,123)	(n = 1,426)	< 0.001
	Less than 4 million yen	39.9	47.4	
	4 – 12 million yen	57.3	50.6	
	12 million yen and more	1.8	2.0	
	Working time per week during pregnancy (n = 63,391)	(n = 62,362)	(n = 1,029)	< 0.001
	Not working	15.7	28.0	
	Less than 50%ile (40 hours)	33.5	33.6	
	50%ile (40 hours) and over	50.8	38.4	
Relationship with others				
	Participation status of partner <sup>c</sup>			< 0.001
	Non-participant	47.9	71.2	
	Inactive participant	0.9	14.0	
	Active participant	51.2	14.8	

<sup>a</sup>Fisher's exact test, a- "Somebody" is defined as subjects having family members who smoke in the same room. b- "Weekly" is defined as subjects exposed at least once a week. c- Passive participation of a partner refers to those who agreed to participate but did not respond to the questionnaire. Active participation of a partner refers to those who answered the questionnaire.

births than those in the responder group. Based on the SF-8 scores, HRQOL was not significantly different between the non-responder group and responder group.

Significantly more women in the non-responder group had placental or umbilical cord abnormalities and other complications. As for neonatal health, significantly more neonates of non-responders had jaundice, physical anomalies or were referred for specialized care. Additionally, more neonates in the non-responder group versus the responder group had one-minute Apgar scores of less than seven. Significantly more neonates from the non-responder group stayed in the hospital for eight or more days and were referred for specialized care. Babies born to the non-responder group were born at significantly earlier gestational age.

Regarding health-related behaviors, more women in the non-responder group smoked during the early or entire pregnancy period. Furthermore, significantly more women in the non-responder group were often exposed to secondhand smoke before pregnancy. There was no significant difference in alcohol consumption during pregnancy between the response groups.

With regard to socioeconomic factors, more women in the non-responder group had fewer than 13 years of schooling and less than ¥4 million in the household income. The non-responder and responder groups differed in terms of working hours. In the non-responder group, significantly fewer women worked for 40 hours or more per week during pregnancy and some did not work at all.

As for partner participation in the study, significantly more partners of responders took part actively.

### Factors related to survey non-response

Variables that were significantly related to survey response/non-response were analyzed using binary logistic regression. Table 2 shows the likelihood of non-response to the J ECS survey, represented by odds ratios and 95% confidence intervals. The factor that most strongly predicted survey non-response was partner participation status. When compared to participants whose partners did not take part in the J ECS at all, the odds of survey non-response were 0.44 (0.36, 0.52) and 6.92 (4.87, 9.84) when the partners were active participants and inactive participants respectively. Finally, for neonatal health factors, the non-response odds ratios among participants whose children had low one-minute Apgar scores, had referral for specialised care, or had physical anomalies were significantly greater than 1 (2.64, 2.33, and 2.35, respectively).

**Table 2:** Multivariate Logistic Regression Predicting the probability of Survey Non-response.

Variable	Model 1		Model 2	
	AICc = 6124.14		AICc = 16761.5	
	OR (95% CI)	p-value	OR (95% CI)	p-value
Age at delivery				
Less than 25	Reference		Reference	
25 – 29	0.58 (0.45, 0.74)	< 0.001	0.63 (0.54, 0.72)	< 0.001
30 – 34	0.46 (0.36, 0.60)	< 0.001	0.59 (0.51, 0.68)	< 0.001
More than 35	0.44 (0.33, 0.57)	< 0.001	0.59 (0.51, 0.69)	< 0.001
Medical history of bronchial asthma (have/have not)	1.30 (1.04, 1.63)	0.023	2.81 (2.54, 3.11)	< 0.001
K6 during pregnancy (13 and over/less than 13)	1.51 (1.08, 2.11)	0.016	5.26 (4.67, 5.92)	< 0.001
Multiple births (Yes/ No)	1.21 (0.80, 1.83)	0.362	1.44 (1.13, 1.84)	0.003
Neonatal referral at delivery (have/have not)	2.33 (1.82, 2.99)	< 0.001	2.02 (1.74, 2.34)	< 0.001
Neonatal physical anomalies (have/have not)	2.35 (1.89, 2.92)	< 0.001	1.70 (1.47, 1.95)	< 0.001
Apgar Index at 1 min (less than 7/7 and more)	2.64 (1.96, 3.56)	< 0.001	3.70 (3.12, 4.39)	< 0.001
The gestational age of the children at birth				

	37 w and More	Reference		Reference	
	34-36w	1.65 (1.22, 2.23)	0.001	1.41 (1.18, 1.69)	< 0.001
	22-33w	2.82 (1.92, 4.13)	< 0.001	2.32 (1.85, 2.92)	< 0.001
Smoking habits during pregnancy					
	Never smoked	Reference		Reference	
	Smokers who quit before pregnancy	1.17 (0.94, 1.46)	0.147	1.46 (1.31, 1.63)	< 0.001
	Smokers who quit after pregnancy	1.50 (1.20, 1.87)	< 0.001	1.13 (0.99, 1.30)	0.076
	Smoker	2.74 (2.09, 3.57)	< 0.001	1.73 (1.45, 20.6)	< 0.001
Alcohol consumption during pregnancy					
	Never drink	Reference		Reference	
	Drinkers who quit before pregnancy	0.95 (0.79, 1.13)	0.544	0.94 (0.85, 1.03)	0.200
	Drinkers	0.66 (0.47, 0.92)	0.014	0.62 (0.51, 0.75)	< 0.001
	Education status (13 and over/less than 13)	0.78 (0.65, 0.93)	0.007	0.78 (0.70, 0.87)	< 0.001
Family income per year					
	Less than 4 million yen	Reference		Reference	
	4 – 12 million yen	0.98 (0.82, 1.17)	0.843	1.26 (1.13, 1.39)	< 0.001
	12 million yen and more	1.19 (0.65, 2.16)	0.572	0.80 (0.72, 0.88)	< 0.001
Working time during pregnancy					
	Not working	Reference		Reference	
	Less than 40 hours/week	0.69 (0.56, 0.85)	< 0.001	1.28 (1.12, 1.46)	< 0.001
	40 hours/week and over	0.58 (0.47, 0.71)	< 0.001	0.52 (0.44, 0.62)	< 0.001
Participation status of participants' partners					
	Non-participation	Reference		Reference	
	Inactive participation	6.92 (4.87, 9.84)	< 0.001	9.92 (8.46, 11.6)	< 0.001
	Active participation	0.44 (0.36, 0.52)	< 0.001	0.36 (0.32, 0.40)	< 0.001

Model 1: Crude data. Model 2: Model 1 with multivariate singular value decomposition to impute missing values. AIC: Akaike's Information Criterion

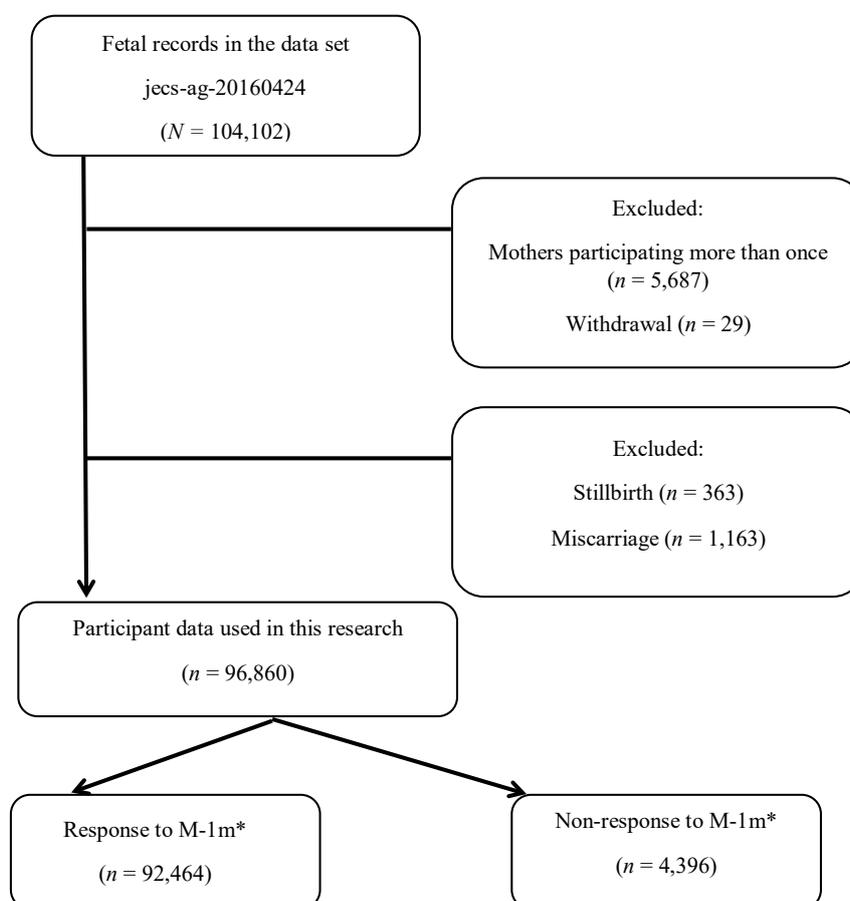


Figure 1: Inclusion of participants. \*M-1m: the questionnaire sheet given one month after birth.

In this study, multivariate singular value decomposition was used to impute missing data. Logistic regression was then used to examine how each factor related to survey non-response. The resulting odds ratios showed similar trends. The odds of non-response were decreased for those who had long working hours during pregnancy, actively participating partners, and transcriptions of multiple prenatal examinations. Conversely, the odds of non-response were significantly increased for those with a history of bronchial asthma, a high score of K6, and a low one-minute Apgar score. However, the odds ratio for working for a short time during pregnancy changed from 0.69 in Model 1 to 1.28 in Model 2.

## Discussion

Mothers (or other guardians) who consented to take part in the study completed the J ECS questionnaire. The current study used data from approximately 100,000 expectant mothers to investigate factors related to whether they responded to a survey that they were to complete one month after giving birth. Factors related to survey non-response were the mother's age at delivery, level of education, hours worked per week, and the child's health at birth. Furthermore, survey non-response differed depending on how actively the mother's partner participated in the J ECS.

Approximately 45% of children in 2013 were part of the J ECS (the number of live births registered in J ECS divided by the number of all live births within the study areas). Nevertheless, the data on the characteristics of the mothers and children we studied showed marked similarity to those obtained from Japan's 2013 Vital Statistics Survey [2].

Previous studies have reported that survey responses may have been affected by participants' health-related behaviors and socioeconomic factors. Several studies have shown that early age, alcohol, smoking, low income, and low educational status were associated with non-response [5,7,13-21]. Nakai et al surveyed health-related behaviors in Japan and showed that there were more elderly people and less current smokers in the early response group [22]. In the non-response group, more individuals were young, currently smoking, exposed to secondary smoke, with low educational levels, and with low income. Alcohol consumption during early pregnancy was associated with a high response rate. One of the objectives of the J ECS was to assess environmental factors that affect children's health (e.g., the mother's health factors). Previous research suggested that the salience of the survey topic was associated with a prompt response [23]. In this survey, participants with an interest in the effect of alcohol consumption on children's health were more likely to respond, and participants with long work hours were more likely to respond to the M1m questionnaire. In the Survey on Time Use and Leisure Activities in 2011 [24], women who worked from home (family workers) spent more time on housework and less time on self-education/training and hobbies/amusement than did those employed outside the home. Associations of time spent answering the questionnaire with responses to the questionnaire were also observed.

This study found that the attitudes of participants' partners toward J ECS participation were an important factor. Having a partner who showed active involvement with the J ECS- by agreeing to participate in the study and sending in their survey- was significantly predictive of participant response. Conversely, having a partner who showed passive involvement in the study by agreeing to take part but not sending in his survey strongly predicted participant non-response. A meta-analysis of 14 articles suggested that partner cooperation during pregnancy and postpartum could have a positive effect on maternal health [25]. A study of prenatal genetic testing found that husbands and wives shared concerns about the test and its results [26]. Additionally, a survey found that pregnant women who stopped smoking after pregnancy but whose husbands smoked frequently were more likely to resume smoking later on. This suggests that partners influence maternal smoking behavior [27]. These results suggest that the response rate is negatively affected if the individuals surrounding participants are familiar with the research but take part inactively.

Previous studies have shown that poor health when completing a survey may be related to survey non-response [28,29]. According to Fukutomi et al, from 2004-2006, the prevalence of bronchial asthma in Japanese women aged 20-44 years was 10.9% (10.1-11.8%). The rate among this study's participants (10.9%) was the same [30]. In a study of women of reproductive age, Fida and colleagues found that increased weight was related to asthma recurrence [31]. Further, when Ronmark et al followed up on the results of a survey on respiratory health, they found significantly higher rates of wheezing, asthma, and chronic cough among non-responders [32]. The current study's results suggest that in participants with a history of bronchial asthma and chronic cough, pregnancy-related weight gain may relate to survey non-response. In a study among participants from the Australian Longitudinal Birth Cohort Study, Ng et al suggested that mothers who have a higher risk of psychological distress were likely to not respond at a one-year follow-up [33]. In this study, there were more non-respondents to M1m among participants with a higher risk of psychological distress during pregnancy.

Survey response is affected not only by the health of the survey participants themselves but also by the health of their neonates. Given that one of the outcomes included among the J ECS hypotheses is the prevalence of physical anomalies in neonates, non-response bias due to neonatal health problems could affect the testing of such hypotheses. However, as the response rate for the survey at one month after childbirth was high, 94.5%, such an effect would be small at the time.

In survey studies, another potential bias may arise when data from returned questionnaires are consistently incomplete. In this study, a second model used multivariate singular value decomposition to impute missing data. Using logistic regression, the values were then examined in relation to survey non-response. The resulting odds ratios showed similar trends. Moreover, the obtained results indicated that we can expect an improvement in the accuracy of estimating significance and measuring odds ratio changes and potentials. The odds ratio of survey non-response changed with family income and time spent working during pregnancy; these factors were unstable across the groups, since those in the response group were much less likely to leave this information out. Both the time spent working and family income had a higher rate of missing data than other factors in both groups. Family income was missing 8.0% and 67.6% of the time for response and non-response groups, respectively. As for time spent working, response and non-response groups did not give this information 32.6% and 76.6% of the time respectively.

## Study Limitations

This study had three important limitations. First, the dataset used contained no information regarding the distribution and collection of the

surveys. Second, in Japan, there is a custom of “*Satogaeribunben*,” wherein pregnant women return to their parents' homes for giving birth; this may have made it difficult to reply to the M1m questionnaire. Third, not all the partners of the participants were asked to take part in the JECS.

Previous studies have indicated that survey response rates differ depending on the type of survey [7,34-38]. In the JECS, Research Coordinators usually gave mothers the survey while they were in the hospital for their deliveries. The surveys were answered one month after childbirth, and the completed surveys were collected by research coordinators Research Coordinators when participants visited participating facilities for their one-month checkup. However, in some cases, the surveys were sent and returned via mail. As this difference was not considered in the dataset used for this analysis, it was not possible to examine how it may have related to non-response. On the other hand, according to the Ministry of Health, Labor, and Welfare (MHLW), about 80% of neonates received a one-month checkup [39], suggesting that most of the survey data used in this study came from surveys that were distributed and collected by the JECS Research Coordinators.

Questions about *Satogaeribunben* were not included in the M1m questionnaire; as such, we were unable to determine who were engaging in the practice and did not assess the effect of *Satogaeribunben* on non-response. However, all M1m questionnaires were delivered to participating women by the Research Coordinators in obstetrics departments participating in the study. As such, the opportunity to return the questionnaires can be considered uniform.

Regarding the final limitation, partners were requested to participate in the JECS by the Research Coordinator when they were interviewed in person. However, when staff members were unable to conduct that interview, those partners were treated as non-participants in the dataset. This would suggest the possibility that partners who may have been interested in JECS were inadvertently included in the non-participant group. This weakens the relationship between active/inactive participation partners and nonparticipation partners. Despite these factors, this study found significant differences in the collected data. Additionally, partners who agreed to participate but had negative thoughts about participation may have been included in the group of inactive participants, which may have delayed the return of questionnaires. The results of this study suggest that such partners (delayed refusals) may have been included in the inactive participation partner group.

## Conclusion

Cross-sectional studies have determined that participant age, socioeconomic status, and interest in the hypothesis are related to survey non-response. This study confirmed that the factors associated with non-response to the JCES as a cohort study were similar to those of cross-sectional studies. Additionally, the research participation status of the participants' partners was related to non-response.

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