

ORIGINAL ARTICLE

Survey of motivation to participate in a birth cohort

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For a longitudinal prospective cohort study to be successful, participants' motivation to provide information must be maintained. Therefore, this study aimed to identify items that effectively promote participants' motivation. Questionnaires were mailed to 4541 mothers and expectant mothers in Chiba Prefecture, Japan who participated in a nationwide birth cohort. A total of 2387 (52.6%) responses were received. The following items were identified as primary motivating factors among our cohort: "benefits to the participants' children", "monetary compensation" and "contribution to a better future environment". More than 30% of the respondents expressed a lack of understanding regarding the study purpose and requirements for participation. About 14% were concerned about the leakage of personal information, and 13% felt burdened by having to make a long-term commitment to the study. Cluster analysis identified four groups, two of which, one with extremely low levels of motivation and the other motivated by only money or goods, lacked an understanding of the study and tended to be concerned about the associated risks and burdens. Participants in these groups were considered to be at a high risk of dropout. Therefore, implementing measures to provide participants with a better understanding of cohort studies could lead to more successful results.

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INTRODUCTION

To address the influence of unfavorable environmental exposure on children's health,^{1,2} large-scale birth cohort studies have been conducted in a number of countries.³ In Japan, the Japan Environment and Children's Study (JECS), a nationwide government-funded birth cohort study, began in January 2011 with the objective of evaluating the impact of environmental factors on children's health and development.^{4,5} The predetermined recruitment goal of 100 000 pregnant women was achieved in March 2014 through the cooperative efforts of 15 medical university-based regional centers. The default follow-up setting was to continue monitoring the participating children until they reach the age of 13. In this birth cohort, mainly healthy pregnant women were recruited, and participants were periodically asked to provide biological samples and information about their living environment, lifestyle and health, as well as their child's development over time.

The success of longitudinal cohort studies largely depends on whether a sufficient number of participants can be recruited and whether their motivation to provide information can be maintained over the course of the study. To that end, it is important to understand the tendencies of participants and the factors that influence their motivation. In several previous studies in hypothetical or actual settings, major incentives to participate in a birth cohort included the following: "contributing to science or the research", "benefit for future mothers and children", "trust in the research institutions" and "learning about their pregnancy or their children's development".^{6–8}

Monetary compensation has also been noted as a possible positive incentive for some women.⁹

However, some participants have a poor understanding of the study purpose and procedure.¹⁰ In a study regarding a hypothetical biobank, which is similar to a birth cohort in terms of biological sample collection, some people showed a willingness to participate even though they had concerns about privacy.¹¹ We hypothesized that those participants might be at risk for dropout from the study. To date, only a few studies have addressed participants' understanding and concerns in a birth cohort, and as far as we know, none have investigated these items in relation to motivation levels.

In the current study, we classified the participants by motivation level and compared their levels of understanding and concerns in order to identify the characteristics of each group and obtain some clues for conducting more successful cohort studies.

MATERIALS AND METHODS

The Japan environment and children's study

The JECS is a birth cohort study conducted to evaluate the impact of various environmental and genetic factors on the health of children up to 13 years of age. A total of 103 106 pregnant women were enrolled in the study. Recruitment was conducted at 15 regional centers nationwide from January 2011 to May 2014. These regional centers conducted recruitment in cooperation with maternity hospitals and local health centers. Biological samples (mother's blood, urine, hair, breast milk and cord blood, and baby's dried blood spots and hair) and medical records continue to be collected, and information about lifestyle, living environment and child development is

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gathered periodically by questionnaire. Portions of the biological samples are stored for genetic analysis, and the plans are being designed to convert the biological sample repository into a biobank for further scientific research.

The Center of Chiba Unit is one of the regional units carrying out the study and it covers a total of 14 cities and towns in Chiba Prefecture, Japan. Recruitment was conducted in consecutive steps as follows: (1) a face-to-face explanation with accompanying briefing materials by trained recruiters; (2) delivery of an explanatory booklet; and (3) receipt of a signed consent form (on the same day or within several days). If prospective participants wanted additional information, it was provided by telephone, e-mail or the study website. As a prerequisite for enrollment, participants were required to have basic literacy so that they could complete the periodic questionnaires.

Questionnaire survey

The questionnaire used in this study was designed to ascertain the following: (1) the participants' initial motivation to participate, (2) the items for which they felt they had insufficient understanding and (3) their concerns. Respondents were classified according to the type and level of their motivation, and then the level of understanding and/or concern was compared between the groups.

For this study, anonymous questionnaires were mailed to 4541 mothers and expectant mothers who were participating in the J ECS at the Center of Chiba Unit on 28 August 2013; 2387 responses had been collected by 30 April 2014 (response rate, 52.6%). The questionnaire items are shown in Supplementary File S1. Categorical data on the age of mothers, month and year of delivery and residential area were also collected, and a query regarding whether their participation was voluntary was included. Motivation for participation (11 items, Figure 1), self-reported understanding of the study (21 items, Figure 2) and concerns about the study (5 items, Figure 3) at the time of enrollment were measured using a 3-point Likert scale. Two motivation items that were found not to be applicable to all the participants were excluded from the analysis.

This study was approved by the Research Ethics Committee of the Graduate School of Medicine, Chiba University.

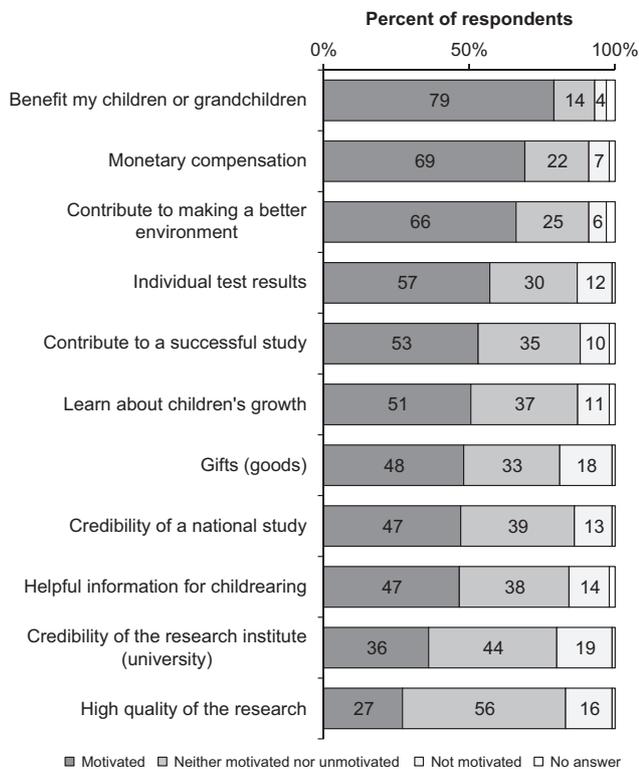


Figure 1 Motivation to participate in the study ($N=2358$).

Data analysis

The relationships between motivation, understanding and concerns were examined using data from 2106 returned questionnaires with complete answers for age, motivation, understanding and concerns. Factor analysis, which is used for data reduction because it can identify a small number of factors that explain most of the variance observed in a much larger number of manifest variables,¹² was conducted on 11 items for motivation, 21 items for understanding and 5 items for concerns. We summarized motivation, understanding and concerns in four, six and two factors, respectively. All factors were named in reference to the items with factor loadings ≥ 0.35 . Factor scores were calculated for each respondent on each factor.

Then, to classify the respondents on their motivation, cluster analysis of standardized factor scores for each motivation factor was performed using Ward's linkage to conceptualize cluster locations and squared Euclidean distance to measure distances between respondents and clusters; this yielded four groups. Group comparisons were conducted for continuous and categorical variables using the Mann-Whitney U -test and χ^2 -test with Bonferroni

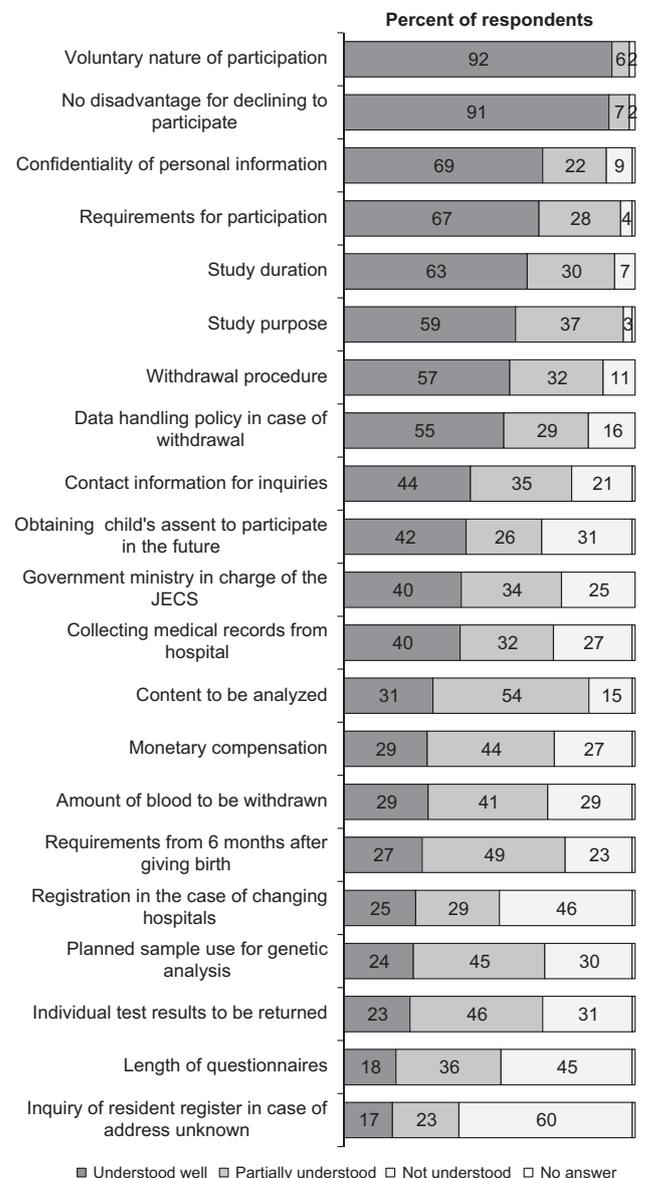


Figure 2 Participants' self-perceived understanding of study aspects at the time of enrollment ($N=2358$).

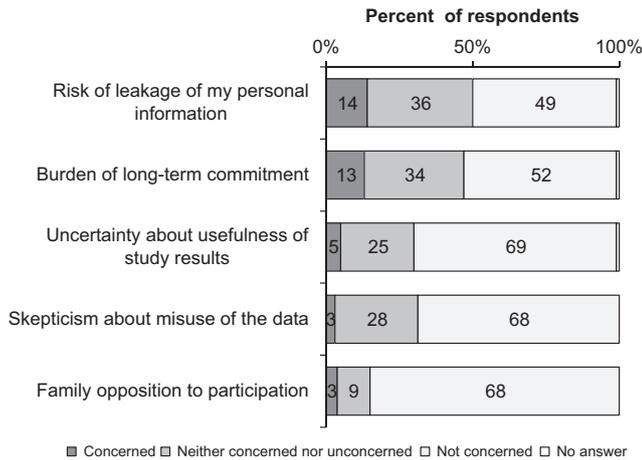


Figure 3 Participants' concerns about the study (N=2358).

Table 1 Comparison of survey respondents to total participants

	Survey respondents (N = 2358)	Total participants (N = 4541)
Age (years)		
<20	5 (0.2%)	32 (0.7%)
20–29	665 (27.9%)	1609 (35.4%)
30–39	1530 (64.1%)	2654 (58.4%)
≥40	176 (7.4%)	246 (5.4%)
No answer	11 (0.5%)	0 (0%)
Birth month of child^a		
July 2011 – June 2012	680 (28.5%)	1396 (30.7%)
July 2012 – June 2013	1086 (45.5%)	2214 (48.8%)
July 2013 – June 2014	600 (25.1%)	923 (20.3%)
No answer	21 (0.9%)	8 (0.2%)
Residence at enrollment		
Chiba (Midori-ku)	382 (16.0%)	713 (15.7%)
Kimitsu	1133 (47.5%)	2199 (48.4%)
Awa	540 (22.6%)	998 (22.0%)
Isumi	286 (12.0%)	527 (11.6%)
Others, no Answer	46 (1.9%)	104 (2.3%)

^aIn the case of multiple participation, the first delivery date was used.

correction, respectively. A *P*-value <0.05 was considered statistically significant. Data analysis was performed using SPSS statistics ver. 22 (IBM corporation, Armonk, NY, USA).

RESULTS

Among the 4541 mothers and expectant mothers who received the questionnaire, responses were received from 2387 (response rate, 52.6%). Respondents were more representative of mothers in their 30 and 40s and those who had recently given birth than other J ECS participant groups (Table 1).

Motivation, understanding and concerns of respondents

Figure 1 shows a list of motivation factors and levels of respondents' motivation to participate in the J ECS at the Chiba Unit. The most frequent motivation factor at the time of enrollment was "benefit my children or grandchildren", followed by "monetary compensation" and "contributing to making a better environment". Less than half of the

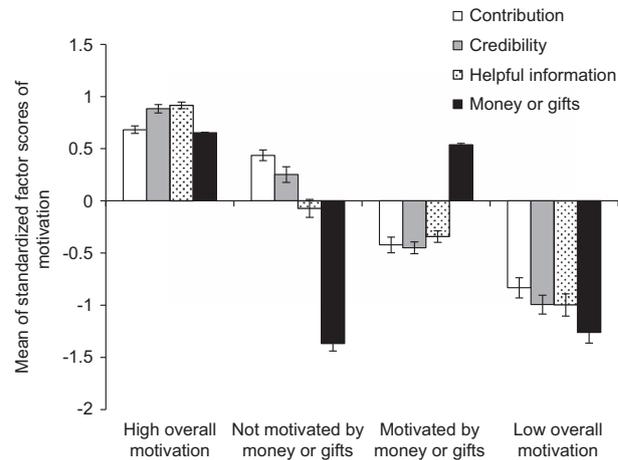


Figure 4 Features of respondent groups classified by motivation factors. Data are expressed as standardized factor scores of motivation. Error bars indicate 95% confidence intervals.

respondents considered the credibility of the research institute or the fact that it was a national research study to be a motivating factor.

Figure 2 shows the level of the respondents' self-rated understanding about information to be disclosed before their decision to participate. Ninety percent of the respondents answered that they understood the voluntary nature of participation. Regarding confidentiality of personal information, requirements for participation, study duration and the purpose, 59–69% of the respondents understood the study well, which rose to >90% when including respondents who partially understood. Concerning study procedures such as obtaining assent from their child in the future, how medical records would be accessed and the use of data collected, less than half of the respondents understood well, which rose to 68–85% when including respondents who partially understood.

Figure 3 shows the level of respondents' concerns about the study at the time of enrollment. About 14 and 13% of the respondents were concerned about leakage of their personal information and the burden of having to make a long-term commitment to the study, respectively. Among the respondents who expressed concern about leakage of their personal information, 14% answered "did not understand well" in response to the item "my name will not be disclosed to anyone other than the institutions involved in the J ECS"; this ratio was significantly higher than the group with less or no concern (8%) (*P*<0.001).

Factor analyses and classification of respondents

Results of factor analyses identified four motivation factors, six understanding factors and two concern factors. The factor loadings are shown in Supplementary Tables S1–S3. From the factor loadings obtained, the motivation factors were named (1) credibility, (2) helpful information, (3) money or gifts and (4) contribution. The understanding factors were named (1) implementation of the study, (2) handling of personal data, (3) voluntary nature of participation, (4) outline of the study, (5) contact information (including withdrawal procedure) and (6) analysis of data. The concern factors were (1) risks and (2) burden. The cumulative contribution ratios before rotation were 54.1% for the four motivation factors, 53.1% for the six understanding factors and 41.8% for the two concern factors.

Respondents were classified into four groups based on cluster analysis using standardized motivation factor scores for each item. A

comparison of standardized motivation factor scores between the four groups is shown in Figure 4. On the basis of results, the groups were named (1) high overall motivation, (2) not motivated by money or gifts, (3) motivated by money or gifts and (4) low overall motivation.

Group comparisons of age, understanding and concerns

A comparison of age, scores for the six understanding factors and scores for the two concern factors between group 1 (high overall motivation) and the other three groups is shown in Table 2. The largest number of respondents were categorized into group 3 (motivated by money or gifts), followed by group 1 (high overall motivation).

Factor scores for understanding, except for "voluntary nature of participation", were highest in group 1 (high overall motivation), followed by group 2 (not motivated by money or gifts), group 3 (motivated by money or gifts) and group 4 (low overall motivation). Meanwhile, factor scores for concern increased in the reverse order. Respondents in group 2 (not motivated by money or gifts) were significantly older than those in group 1 (high overall motivation). More than 90% of the respondents in every group answered that they made the decision to participate in the study on their own.

DISCUSSION

In this study, a substantial number of participants were found to have low overall motivation or to be motivated by money or gifts. These respondents tended to have more concerns and a poorer understanding of the cohort study than the other respondents. This may indicate that these types of participants are at an increased risk of dropping out from this long-term cohort study. If this is in fact the case, appropriate measures need to be taken to maintain their commitment and motivation to continue, such as providing addition

explanations of the study content to deal with their level of understanding and concerns.

Comprehending fully the content of a study and the measures that will be taken to protect personal information through the initial informed consent procedures is almost impossible for participants.^{13–15} In fact, in our cohort, only 59% of the respondents felt that they had sufficient understanding of the purpose of the study, and even fewer felt that they had enough information on data collection methods, the data that would be analyzed, assent from their child in the future and other study procedures. To improve participants' understanding of the study, more information about the study should be provided in an easy-to-understand manner not only at the initial explanation for informed consent, but also periodically throughout the duration of the study.

Although most of the participants were not concerned about the study content or procedures, 14% were concerned about the risk of personal information leakage, and 13% were concerned about the burden of a long-term commitment. A previous study showed that some people showed willingness to participate in a hypothetical biobank while being concerned about privacy.¹¹ In our study, there were significantly more respondents who did not know whether their name would be disclosed to anyone other than participating institutes in the group with concerns about privacy protection compared with those with no or less concern. This result indicates that a limited understanding about privacy protection may increase participants' concerns. To address this issue, more explanations about privacy protection and data management should be provided.

In this study, we identified major motivating factors for participation in a large-scale birth cohort study. We found that many respondents had self-interest motivations (for example, benefit my children or grandchildren, monetary compensation, individual test

Table 2 Comparison of groups classified by motivation with age, self-perceived understanding of study aspects and concerns (N = 2106)

Feature of group	High overall motivation	Not motivated by money or gifts	Motivated by money or gifts		Low overall motivation		
			P-value ^a		P-value ^a	P-value ^a	
Number (%)	674 (32%)	325 (15%)		778 (37%)		329 (16%)	
Age ^b							
<30	29%	19%	<0.001	32%	NS	26%	NS
30–39	64%	69%		62%		70%	
≥40	7%	13%		7%		4%	
Factor scores ^c							
Understanding							
Implementation of the study	0.293 (–0.358, 1.003)	–0.084 (–0.765, 0.577)	<0.001	–0.174 (–0.758, 0.460)	<0.001	–0.312 (–0.927, 0.388)	<0.001
Handling of personal data	0.331 (–0.435, 0.984)	0.037 (–0.518, 0.693)	<0.01	–0.141 (–0.677, 0.526)	<0.001	–0.261 (–0.737, 0.505)	<0.001
Voluntary nature	0.296 (0.230, 0.334)	0.287 (0.228, 0.335)	NS	0.290 (0.206, 0.338)	NS	0.281 (0.185, 0.329)	<0.01
Outline of the study	0.472 (–0.275, 0.789)	0.188 (–0.402, 0.673)	<0.01	0.159 (–0.553, 0.668)	<0.001	0.050 (–0.792, 0.617)	<0.001
Contact information	0.564 (–0.503, 0.892)	0.368 (–0.602, 0.769)	<0.01	0.204 (–0.686, 0.718)	<0.001	–0.034 (–0.790, 0.657)	<0.001
Data analysis	–0.006 (–0.297, 1.147)	–0.148 (–0.475, 0.823)	<0.001	–0.212 (–0.509, 0.753)	<0.001	–0.225 (–0.511, 0.748)	<0.001
Concerns							
Risks	–0.651 (–0.725, –0.313)	–0.482 (–0.725, 0.814)	<0.001	–0.408 (–0.651, 0.983)	<0.001	–0.239 (–0.627, 1.057)	<0.001
Burden	–0.358 (–0.768, 0.101)	–0.130 (–0.768, 0.512)	<0.001	0.010 (–0.449, 0.561)	<0.001	0.193 (–0.449, 0.561)	<0.001

NS, not significant.

^aP values for comparison with the "high overall motivation" group were calculated by χ^2 -test for age and Mann–Whitney *U*-test for standardized factor scores with Bonferroni correction.

^bValues represent percentage of respondents in groups.

^cValues represent median, 25th percentile and 75th percentile.

results and helpful information about childrearing), as well as altruistic motivation factors (for example, contributing to making a better environment). Previous studies also reported that most participants of birth cohort surveys had altruistic motivation.^{6,7} Although altruism is a principal factor in participation, it would be difficult to maintain participants' willingness to take part in a long-term study based on altruistic motivation alone. A review reported a decreased tendency of volunteerism as a factor in recent uncooperativeness in epidemiologic studies with little immediate benefit to participants themselves.¹⁶ However, the perception of making contributions to others may lead to personal benefit in terms of self-satisfaction.¹⁷

Offering incentives to serve participants' self-interests may improve their disposition. Given that studies found that participants expected their individual data to be returned and that this was one of their key motivating factors for participating in a study, highlighting the return of individual test results could be an effective strategy for encouraging long-term participation.^{18,19} Furthermore, the overall results from the cohort can provide some personal benefit for the participants.^{7,19} The majority of respondents in this study were motivated by benefits to their children, learning about their child's growth and gaining helpful information for childrearing. Periodical newsletters or website could also fulfill participants' expectations by providing helpful information. The effect of monetary compensation on encouraging participation has been inconsistent among studies.^{8,11,19–21} In our study, although monetary compensation seemed to be a major motivating factor, it is difficult to employ due to financial limitations and potential ethical concerns.²²

Although 78% of the participants in an Italian hospital-based birth cohort cited the credibility of the research institute as a reason to participate, we did not identify this as a major reason. Differences regarding the type of cohort (hospital-based vs population-based) and/or cultural backgrounds might have influenced this phenomenon.

Our study did have several limitations. First, to avoid false descriptions due to participants worrying that their answers would be identifiable to researchers, this survey was conducted anonymously. Therefore, it was not possible to investigate whether the group with weak overall motivation had a tendency to dropout. Further study is required to address this issue. Second, information about social and educational situations was not available in this survey. Younger J ECS participants tend to have lower household income and lower educational attainment. In this study, the group that was not motivated by monetary compensation had a larger number of older participants than younger ones, which may be associated with their higher household income and educational attainment. Finally, this survey was conducted with participants who enrolled in the Center of Chiba Unit and it is unclear whether the results would apply to all participants of the J ECS. The participants in the Chiba Unit are estimated to cover about 40% of all pregnant women in the research area, which is comparable to estimates in the other J ECS units (47%).

In conclusion, we found for the first time that participants with low overall motivation and participants motivated by money or gifts alone had limited understanding and numerous concerns about the study. These results suggest a need to address these issues by providing further explanations about the study not only at the time of enrollment, but also periodically throughout the duration of the study.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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- Landrigan, P. J., Kimmel, C. A., Correa, A. & Eskenazi, B. Children's health and the environment: public health issues and challenges for risk assessment. *Environ. Health Perspect.* **112**, 257–265 (2004).
- Prüss-Ustün, A. & Corvalán, C. *Preventing Disease through Healthy Environments: Towards an Estimate of the Environmental Burden of Disease*. (Geneva, WHO (World Health Organization) Library Cataloguing-in-Publication Data) http://www.who.int/quantifying_ehimpacts/publications/preventingdisease.pdf. Accessed 22 November 2015.
- Lawlor, D. A., Andersen, A. M. & Batty, G. D. Birth cohort studies: past, present and future. *Int. J. Epidemiol.* **38**, 897–902 (2009).
- Kawamoto, T., Nitta, H., Murata, K., Toda, E., Tsukamoto, N. & Hasegawa, M. Rationale and study design of the Japan environment and children's study (J ECS). *BMC Public Health* **14**, 25–32 (2014).
- Michikawa, T., Nitta, H., Nakayama, S. F., Ono, M., Yonemoto, J., Tamura, K. *et al.* The Japan Environment and Children's Study (J ECS): a preliminary report on selected characteristics of approximately 10 000 pregnant women recruited during the first year of the study. *J. Epidemiol.* **25**, 452–458 (2015).
- Daniels, J. L., Savitz, D. A., Bradley, C., Dole, N., Evenson, K. R., Eucker, B. *et al.* Attitudes toward participation in a pregnancy and child cohort study. *Paediatr. Perinat. Epidemiol.* **20**, 260–266 (2006).
- Brumatti, L. V., Montico, M., Russian, S., Tognin, V., Bin, M., Barbone, F. *et al.* Analysis of motivations that lead women to participate (or not) in a newborn cohort study. *BMC Pediatrics* **13**, 53 (2013).
- Qiu, X., He, J., Qiu, L., Larson, C. P., Xia, H. & Lam, K. B. Willingness of pregnant women to participate in a birth cohort study in China. *Int. J. Gynaecol. Obstet.* **122**, 216–218 (2013).
- Nechuta, S., Mudd, L. M., Elliott, M. R., Lepkowski, J. M. & Paneth, N. Michigan Alliance for the National Children's Study. Attitudes of pregnant women towards collection of biological specimens during pregnancy and at birth. *Paediatr. Perinat. Epidemiol.* **26**, 272–275 (2012).
- Stolt, U. G., Helgesson, G., Liss, P. E., Svensson, T. & Ludvigsson, J. Information and informed consent in a longitudinal screening involving children: a questionnaire survey. *Eur. J. Hum. Genet.* **13**, 376–383 (2005).
- Kaufman, D. J., Murphy-Bollinger, J., Scott, J. & Hudson, K. L. Public opinion about the importance of privacy in biobank research. *Am. J. Hum. Genet.* **85**, 643–654 (2009).
- IBM Corporation IBM SPSS Statistics Base 22 (2013). http://www.sussex.ac.uk/its/pdfs/SPSS_Statistics_Base_22.pdf, Accessed 20 February 2016.
- Helgesson, G., Ludvigsson, J. & Stolt, U. G. How to handle informed consent in longitudinal studies when participants have a limited understanding of the study. *J. Med. Ethics* **31**, 670–673 (2005).
- O'Neill, O. *Autonomy and Trust in Bioethics* (Cambridge University Press, New York, NY, USA, 2002).
- Allen, J. & McNamara, B. Reconsidering the value of consent in biobank research. *Bioethics* **25**, 155–166 (2011).
- Galea, S. & Tracy, M. Participation rates in epidemiologic studies. *Ann. Epidemiol.* **17**, 643–653 (2007).
- Harcombe, H., Derrett, S., Herbison, P. & McBride, D. 'Do I really want to do this?' Longitudinal cohort study participants' perspectives on postal survey design: a qualitative study. *BMC Med. Res. Methodol.* **11**, 8 (2011).
- Harris, E. D., Zinief, S. I., Amatruda, J. G., Clinton, C. M., Savage, S. K., Taylor, P. L. *et al.* The beliefs, motivations, and expectations of parents who have enrolled their children in a genetic biorepository. *Genet. Med.* **14**, 330–337 (2012).
- Kaufman, D., Murphy, J., Scott, J. & Hudson, K. Subjects matter: a survey of public opinions about a large genetic cohort study. *Genet. Med.* **10**, 831–839 (2008).
- Jenkins, M. M., Reed-Gross, E., Barfield, W. D., Prue, C. E., Gallagher, M. L., Rasmussen, S. A. *et al.* Qualitative assessment of study materials and communication strategies used in studies that include DNA collection. *Am. J. Med. Genet. A* **155A**, 2721–2731 (2011).
- Glidewell, J., Reefhuis, J., Rasmussen, S. A., Woomert, A., Hobbs, C., Romitti, P. A. *et al.* Factors affecting maternal participation in the genetic component of the National Birth Defects Prevention Study-United States, 1997–2007. *Genet. Med.* **16**, 329–337 (2014).
- The National Commission for the Protection of Human Subjects of Biomedical and Behavioral Research The Belmont Report: Ethical Principles and Guidelines for the Protection of Human Subjects of Research (1979). <http://www.hhs.gov/ohrp/human-subjects/guidance/belmont.html>, Accessed 22 November 2015.