Pregnant women’s opinions toward prenatal pretest genetic counseling in Japan

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Abstract
In-person models of genetic counseling (GC) have been the common method in Japan for pregnant women to receive GC. However, recent increases in the number of pregnant women considering undergoing prenatal testing have made it challenging to retain individualized in-person care. To explore pregnant women’s opinions toward pretest GC models and the ideal time duration, a self-administered questionnaire survey was conducted for women at their first prenatal visit. A total of 114 valid respondents (93.4%) were included in the analyses. Of these, 80.7% of women preferred in-person GC, followed by classroom (9.6%), group (3.5%), and telegenetic-based GC (2.6%). Women with experience in undergoing prenatal testing significantly did not prefer in-person GC ($p = 0.05$). Sixty-two women (54.4%) preferred a duration of 15–29 min for pretest GC sessions, followed by 30–59 min (28.9%) and <15 min (14.9%). Women’s preference of ≥30 min in length was significantly associated with anhedonia, singleton pregnancies, acquaintance with people with trisomy 21, and awareness of prenatal testing. Women who were unaware of the need for agreement with the partner for prenatal testing and who did not know the average life expectancy of a trisomy 21 patient significantly preferred <15 min in length over other durations. While the majority of women preferred in-person GC for <30 min, their preferences varied by their background characteristics, experiences, attitudes, and knowledge. These findings will help establish a prenatal GC system offering a choice of GC models in Japan; however, further large-scale studies are needed to confirm these findings.

Introduction
Prenatal testing for aneuploidy is mostly used for advanced maternal age in Japan [1]. Given the increasing trend in the proportion of pregnant women over 35 years old in Japan [2], the introduction of noninvasive prenatal testing (NIPT) for the detection of aneuploidy in a nationwide trial in April 2013 has influenced an increased women’s awareness of prenatal testing [3]. The introduction of NIPT has also helped to increase the number of prenatal testing options; however, this situation may have resulted in the phenomenon that pregnant women may feel overwhelmed by the number of choices available [4]. Therefore, prenatal pretest GC has become increasingly important, as GC helps pregnant women sufficiently consider prenatal testing, giving them an appropriate understanding of the test and effectively facilitating their informed choice after adequate consideration [5].

The National Society of Genetic Counselors proposed four service delivery models (SDMs): in-person (referred to as face-to-face traditional), telephone, group, and telegenetic-based (provided remotely using videoconferencing) GC [6]. In-person models of GC have been the common method in Japan. However, this model can be time-intensive and is not practical for serving a large population [7]. Although the implementation rate of prenatal testing in Japan where prenatal screening policies have not been adopted was reported to be <10% [8, 9], a recent study in Japan suggested that one-third of pregnant women would choose to undergo prenatal testing after being provided information on prenatal testing at their first prenatal visit [10].
Given the limited number and involvement of Japanese qualifications for genetic healthcare professionals, such as clinical geneticists and certified genetic counselors, in the clinical practice of prenatal GC [11, 12], the current and emerging demand for prenatal testing may overburden presently available genetic healthcare professionals. Therefore, in Japan, it is necessary to explore appropriate prenatal GC SDMs that do not compromise the quality of care and improve the efficiency and access to GC, with the goal of serving the growing number of pregnant women seeking prenatal testing.

While researchers in other countries have explored alternative GC SDMs in the prenatal setting [13–21], pregnant women’s preferences toward models and durations of pretest GC have been poorly studied. In addition, pregnant women’s preferences may vary by country and culture. Thus, the present study investigated Japanese pregnant women’s opinions toward different models and durations of prenatal pretest GC.

Methods

Participants

We conducted a prospective observational study to assess preferences regarding prenatal GC among Japanese pregnant women at the National Center for Child Health and Development (NCCHD) in Tokyo, Japan. As a pilot study to explore pregnant women’s preferences concerning prenatal pretest GC models, we set the sample size at over 100 pregnant women in the research plan. Pregnant women attending a general outpatient clinic were recruited at their first prenatal visit before 17 weeks’ gestations from June to August 2019.

Participants were asked to complete a self-administered anonymous questionnaire on their preferences toward GC models and prenatal pretest GC duration for chromosomal disorders, their socioeconomic status, attitudes, knowledge and experience with prenatal genetic testing, and knowledge and experience with people affected with trisomy 21. Women’s demographics, their receipt of prenatal testing in this pregnancy, and employment status were retrieved from medical records. In addition to the questionnaire, the participants’ maternal mental state was assessed at the women’s first prenatal visit using a self-report two-question instrument that asked about depressed mood and anhedonia [22] at the clinical practice in the NCCHD. Participants with missing data were excluded from the data analysis.

All participants provided their written informed consent prior to the questionnaire survey. This study protocol was approved by Institutional Review Board at the NCCHD on May 14, 2019 (project number 2193).

Instrumentation

A survey instrument on preferences regarding prenatal GC was originally developed with input from maternal fetal medicine physicians, medical geneticists, and genetic counselors at NCCHD. The questionnaire consisted of 28 questions focused on experience and attitudes of prenatal testing (ten questions), preferences toward pretest GC models (five questions), knowledge of congenital disorders including paternal or maternal age effect disorders (five questions), knowledge and experience with people affected with trisomy 21 (four questions), and demographic data, including the age of the father and socioeconomic status (four questions). These items were presented in a multiple-choice format ranging from two to seven potential choices with women allowed to select one of the choices.

Our primary outcome of interest was pregnant women’s preferences toward SDMs and the ideal duration of prenatal pretest GC. On the question of GC SDMs, pregnant women were asked, “What model of pretest GC do you prefer?” Possible answers were “in-person GC,” “classroom GC,” “group GC,” “tele genetic-based GC,” and “don’t know.” In the options of “classroom GC” and “group GC,” the following description was shown on the questionnaire: “an individual meeting with a genetic counselor one-on-one is available after classroom or group GC sessions.”

Regarding the preferences for pretest GC SDMs, we divided the respondents into two groups: in-person GC and other three SDMs (classroom, group, or tele genetic-based), as pretest GC at NCCHD has been delivered as in-person GC models. In addition, we divided the respondents into another two groups (classroom GC or group GC, and in-person or tele genetic-based GC models) to identify factors influencing a preference for non-individualized care at pretest GC.

Regarding the preferences for prenatal pretest GC duration, possible answers were “<15,” “15–29,” “30–59,” “≥60 min,” and “don’t know.” We divided these respondents into groups of <30 and ≥30 min, as pretest GC at NCCHD has been designed to take <30 min in clinical practice. In addition, we divided these respondents into another two groups (those who preferred <15 and ≥15 min) to identify factors influencing a preference for short-duration pretest GC.

Questionnaire

The first section focused on respondents’ experiences and attitudes concerning prenatal testing and contained questions about whether or not they had received a brochure about prenatal testing at their first visit (Q1), when they became aware of prenatal testing (before or after pregnancy) (Q2), their awareness of prenatal testing options (Q3), their
experience with prenatal testing in a prior pregnancy (Q4), opinions concerning the brochures about prenatal testing at the first visit (Q5), opinions concerning the appropriate indications for prenatal testing (Q6), their understanding of the frequency with which prenatal testing is performed among Japanese pregnant women (Q7), experiences discussing prenatal testing with their partners (Q8), whether or not their partner agrees with them about prenatal testing (at the point of the questionnaire) (Q9), and opinions concerning the need for agreement from their partner about prenatal testing (Q10).

The next section focused on opinions concerning pretest GC models and contained questions about the awareness of genetic counseling (Q11), preferences for the duration of pretest GC (Q12), GC models (Q13), people receiving GC together (Q14), and their present plan for prenatal testing when answering the questionnaire (Q15).

The next section assessed respondents’ knowledge of and experiences with people affected by trisomy 21 and contained questions about their awareness of trisomy 21 (Q21), understanding of the mechanisms involved in trisomy 21 (Q22), knowledge of the average life expectancy of someone with trisomy 21 (Q23), and the growth of trisomy 21 (Q24).

The final section contained questions asking about the age of the respondents’ partner (Q25), method of conception (Q26), own educational background (Q27), and annual household income (Q28).

Data analyses

Co-variables, including maternal age, parity (nulliparous, parous), history of spontaneous abortion (yes, no), method of conception (natural conception, assisted reproductive techniques, other infertility treatment), fetal number (singleton, multiple gestation), gestational age, receipt of prenatal testing in this pregnancy, and employment status (full-time, part-time, unemployed), were retrieved from the medical records.

We assessed pregnant women’s preferences regarding prenatal GC descriptively. The pretest GC session at the NCCHD was conducted using an in-person model and designed to be <30 min in duration. To compare maternal demographics among pregnant women who preferred in-person GC, classroom or group GC, and among those who preferred sessions ≥30 and <15 min in length, the chi-square test was used. The influence of pregnant women’s attitudes, knowledge and experience with prenatal testing and trisomy 21 on their preferences for in-person GC, and a length of ≥30 and <15 min was also evaluated using a chi-square test. The participants who responded “don’t know” to the two questions about preferences for pretest GC SDMs and time duration were excluded from the statistical analyses for GC SDMs and time duration.

All descriptive and statistical analyses were performed using the statistical software package Stata SE 15 (STATA Corp., College Station, TX, USA). Statistical significance was set at ≤0.05, and all statistical tests were two-tailed.

Results

Of the 122 pregnant women who consented to participate in our study at the prenatal visit, we excluded 8 with missing data on the following variables: annual household income \((n = 4)\), planned prenatal testing at questionnaire \((n = 2)\), preference for GC SDMs \((n = 1)\), and preference for brochures of prenatal testing \((n = 1)\). We therefore conducted our analysis in the remaining 114 women, except for an analysis of the preference for GC SDM based on 110 women (4 were excluded for answering “do not know”) and time duration based on 112 women (2 were excluded for answering “do not know”).

Participants’ background characteristics

The demographic and background characteristics of the participants are shown in Table 1. Sixty-eight women (59.6%) were ≥35 years old. Twenty-six (22.8%) and 19 (16.7%) women answered “yes” to the questions about depression and anhedonia during the past month, respectively. Approximately 25% of women had a history of spontaneous abortion. Approximately half of pregnancies were achieved by natural conception. Multiple pregnancies accounted for 11.4% in this study population.

Attitudes and understanding about prenatal testing and people with trisomy 21

As shown in Table 2, seven (6.1%) women had experience with undergoing prenatal testing in a prior pregnancy. Participants who knew someone with trisomy 21 or had spoken with people with trisomy 21 accounted for 35.1%. Ultimately, 57.0% of women did not undergo prenatal testing in this pregnancy. At the questionnaire, 47 women (41.2%) decided to undergo prenatal testing in this pregnancy, and 33 women (28.9%) were undecided about whether or not to undergo prenatal testing. Eighty-nine women (78.1%) indicated the need agreement with the father to undergo prenatal testing. Over 80% of women had been aware of prenatal testing before this pregnancy. Approximately half of women correctly understood that trisomy 21 is not an inherited disease. Forty-eight women (42.1%) correctly understood that the life expectancy of an individual with trisomy 21 was ≥40 years.
Table 1 Demographic and background characteristics of study participants

|                                       | n = 114 | Age at questionnaire (years) |   |   |
|---------------------------------------|---------|-------------------------------|---|---|
|                                       |         | Number                        | % of all |   |
| Age at questionnaire (years)          |         |                               |   |   |
| <35                                   |         | 46                            | 40.4% |   |
| 35–39                                 |         | 42                            | 36.8% |   |
| ≥40                                   |         | 26                            | 22.8% |   |
| Age of husband at questionnaire (years)|         |                               |   |   |
| <35                                   |         | 38                            | 33.3% |   |
| 35–39                                 |         | 40                            | 35.1% |   |
| ≥40                                   |         | 36                            | 31.6% |   |
| Age difference among couples          |         |                               |   |   |
| Pregnant women > husbands             |         | 36                            | 31.6% |   |
| Husbands > pregnant women             |         | 61                            | 53.5% |   |
| No age difference                     |         | 17                            | 14.9% |   |
| Education                             |         |                               |   |   |
| Middle school                         |         | 1                             | 0.9%  |   |
| High school                           |         | 8                             | 7.0%  |   |
| Junior college                        |         | 24                            | 21.1% |   |
| University                            |         | 74                            | 64.9% |   |
| Post-graduate                         |         | 7                             | 6.1%  |   |
| Annual household income (×10,000 yen) |         |                               |   |   |
| <500                                  |         | 15                            | 13.2% |   |
| 500–699                               |         | 22                            | 19.3% |   |
| 700–999                               |         | 31                            | 27.2% |   |
| 1000–1499                             |         | 27                            | 23.7% |   |
| ≥1500                                 |         | 19                            | 16.7% |   |
| Employment                            |         |                               |   |   |
| Full-time employment                  |         | 71                            | 62.3% |   |
| Part-time                             |         | 12                            | 10.5% |   |
| Unemployed                            |         | 29                            | 25.4% |   |
| Others                                |         | 2                             | 1.8%  |   |
| Self-reported depressed mood during the past month |         |                               |   |   |
| Yes                                   |         | 26                            | 22.8% |   |
| No                                    |         | 88                            | 77.2% |   |
| Self-reported anhedonia during the past month |         |                               |   |   |
| Yes                                   |         | 19                            | 16.7% |   |
| No                                    |         | 95                            | 83.3% |   |
| Gestational age at questionnaire (weeks) |         |                               |   |   |
| <10                                   |         | 46                            | 40.4% |   |
| ≥10                                   |         | 68                            | 59.6% |   |
| Parity                                |         |                               |   |   |
| Nulliparous                           |         | 77                            | 67.5% |   |
| Parous                                |         | 37                            | 32.5% |   |
| History of spontaneous abortion       |         |                               |   |   |
| Yes                                   |         | 30                            | 26.3% |   |
| No                                    |         | 84                            | 73.7% |   |
| Method of conception                  |         |                               |   |   |
| Natural conception                    |         | 58                            | 50.9% |   |
| Assisted reproductive techniques      |         | 38                            | 33.3% |   |
| Other infertility treatment           |         | 18                            | 15.8% |   |
| Fetal number                          |         |                               |   |   |
| Singleton                             |         | 101                           | 88.6% |   |
| Multiple                              |         | 13                            | 11.4% |   |

Table 2 Participants’ attitudes and knowledge about prenatal testing and people with trisomy 21

|                                       | n = 114 | Experience |   |
|---------------------------------------|---------|------------|---|
|                                       |         | Prenatal testing in a prior pregnancy |   |
|                                       |         | Yes        | 7 |
|                                       |         | No         | 107 |
| Had someone in their vicinity with T21 or talked with people of T21 |         | Yes        | 40 |
|                                       |         | No         | 74 |
| Prenatal testing in this pregnancy    |         | None       | 65 |
|                                       |         | NIPT       | 26 |
|                                       |         | MSS        | 22 |
|                                       |         | AC/CVS     | 1 |
| Attitudes                             |         | Preference on brochures of prenatal testing at the first visit |   |
|                                       |         | Wanted to receive | 77 |
|                                       |         | Neither    | 28 |
|                                       |         | Did not want to receive | 1 |
|                                       |         | Don’t know | 8 |
| Planned prenatal testing at questionnaire |         | No testing | 34 |
|                                       |         | Undecided  | 33 |
|                                       |         | Will have testing | 47 |
| Needed agreement with the partner for prenatal testing |         | Yes        | 89 |
|                                       |         | Neither    | 9 |
|                                       |         | No         | 14 |
|                                       |         | Don’t know | 2 |
| Want to have GC with husband          |         | Yes        | 104 |
|                                       |         | No         | 10 |
| Knowledge                             |         | Aware of prenatal testing before this pregnancy |   |
|                                       |         | Yes        | 97 |
|                                       |         | No         | 17 |
| Aware of GC                           |         | Yes        | 27 |
|                                       |         | No         | 87 |
| Understanding of the mechanism of T21 |         | Not an inherited disease | 58 |
|                                       |         | Inherited disease or don’t know | 56 |
| Understanding of the average of life expectancy of T21 |         | ≥40 years old | 48 |
|                                       |         | 40 years old or don’t know | 66 |
| Recognition of MAE-related disorders  |         | Yes        | 105 |
|                                       |         | No         | 9 |

AC amniocentesis, CVS chorionic villus sampling, GC genetic counseling, NIPT noninvasive prenatal testing, MAE maternal age effect, MSS maternal serum screening, T21 trisomy 21
The participant characteristics by preference for in-person GC, classroom or group GC, or a duration ≥30 or <15 min for pretest GC.

### Influence of participant’s experience, attitudes, and knowledge on preferences regarding prenatal GC

The influence of participant’s experience, attitudes, and knowledge on their preferences for in-person GC, classroom or group GC, or session durations of ≥30 or <15 min for pretest GC is shown in Table 5. Women who underwent prenatal testing in a prior pregnancy did not prefer in-person GC significantly (p = 0.05). In-person GC was preferred by 31/33 (93.9%) and 25/32 (78.1%) of those who were undecided and who had decided to undergo prenatal testing at the time of the questionnaire, respectively (p = 0.157). Among the 24 women who did not need agreement with the partner to undergo prenatal testing, 22 (91.7%) preferred in-person GC. Thus, among such women, none of them preferred classroom or group GC, while those who felt that they needed agreement with their partner to undergo testing significantly preferred classroom or group GC to other options (p = 0.028). No other variables were significantly associated with the preference for in-person GC or classroom or group GC.

With regard to the pretest GC duration, women who were acquainted with someone with trisomy 21 or had talked to people with trisomy 21 as well as those who were aware of prenatal testing before this pregnancy significantly preferred ≥30 min sessions for pretest GC (p = 0.017 and p = 0.037, respectively). Over 20% of women who did not need agreement with the partner for prenatal testing and those who were unaware of the average life expectancy of a trisomy 21 individual significantly preferred <15-min sessions for pretest GC (p = 0.043 and p = 0.027, respectively). Among women who were unaware of GC and those who lacked an understanding of the mechanism of trisomy 21, the frequency of preference for <15-min sessions for pretest GC was ~20%, although the difference was not significant (p = 0.056 and p = 0.054, respectively).

### Discussion

In this study population, 80.7% of pregnant women preferred traditional in-person GC for pretest GC. Pregnant women who had undergone prenatal testing in a prior pregnancy did not prefer in-person GC significantly. Pregnant women with singleton pregnancy, anhedonia during the past month, recognition of prenatal testing, and personal experience with people with trisomy 21 significantly preferred ≥30-min sessions for pretest GC to other durations. Women who did not need agreement with the partner for prenatal testing and were unaware of the average life

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**Table 3 Participants’ preferences for prenatal pretest GC**

| Preferred GC models                          | Number | % of all |
|----------------------------------------------|--------|----------|
| In-person (face-to-face)                     | 92     | 80.7%    |
| Classroom                                    | 11     | 9.6%     |
| Group                                        | 4      | 3.5%     |
| Telegenetics (provided remotely using videoconferencing) | 3      | 2.6%     |
| Don’t know                                   | 4      | 3.5%     |

**Preferred duration of pretest GC**

| Duration  | Number | % of all |
|-----------|--------|----------|
| <15 min   | 17     | 14.9%    |
| 15–29 min | 62     | 54.4%    |
| 30–59 min | 33     | 28.9%    |
| ≥60 min   | 0      | 0.0%     |
| Don’t know| 2      | 1.8%     |

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Table 4 Demographic and background characteristics of study participants with a preference for pretest GC model and duration

| Preferences toward pretest GC model | Preferences toward pretest GC duration (minutes in length) |
|-------------------------------------|----------------------------------------------------------|
| In-person GC (n = 92) | Classroom or group GC (n = 15) | ≥30 (n = 33) | <15 (n = 17) |
| Number | Number (%) | Number (%) | Number | Number (%) | Number (%) | Number (%) | Number (%) |
| Age at questionnaire (years) | 0.611 | 0.461 | 0.120 | 0.071 |
| <35 | 49 | 40/49 (81.6) | 8/49 (16.3) | 50 | 11/50 (22.0) | 11/50 (22.0) |
| ≥35 | 61 | 52/61 (85.2) | 7/61 (11.5) | 62 | 22/62 (35.5) | 6/62 (9.7) |
| Age of husband at questionnaire (years) | 0.906 | 0.915 | 0.402 | 0.182 |
| <35 | 38 | 32/38 (84.2) | 5/38 (13.2) | 37 | 9/37 (24.3) | 8/37 (21.6) |
| ≥35 | 72 | 60/72 (83.3) | 10/72 (13.9) | 75 | 24/75 (32.0) | 9/75 (12.0) |
| Age difference among couples | 0.306 | 0.079 | 0.515 | 0.898 |
| Pregnant women > husbands | 35 | 32/35 (91.4) | 1/35 (2.9) | 36 | 10/36 (27.8) | 6/36 (16.7) |
| Husbands > pregnant women | 58 | 46/58 (79.3) | 11/58 (19.0) | 59 | 16/59 (27.1) | 9/59 (15.3) |
| No age difference | 17 | 14/17 (82.4) | 3/17 (17.7) | 17 | 7/17 (41.2) | 2/17 (11.8) |
| Education, | | | | |
| University or higher level degree | 0.539 | 0.888 | 0.844 | 0.617 |
| Others | 31 | 27/31 (87.1) | 4/31 (12.9) | 32 | 9/32 (28.1) | 4/32 (12.5) |
| Annual household income (x10,000 yen) | 0.167 | 0.292 | 0.462 | 0.089 |
| <1000 | 65 | 57/65 (87.7) | 7/65 (10.8) | 67 | 18/67 (26.9) | 7/67 (10.5) |
| ≥1000 | 45 | 35/45 (77.8) | 8/45 (17.8) | 45 | 15/45 (33.3) | 10/45 (22.2) |
| Employment | 0.320 | 0.677 | 0.477 | 0.408 |
| Full-time employment | 68 | 55/68 (80.9) | 5/68 (11.9) | 69 | 22/69 (31.9) | 12/69 (17.4) |
| Others | 42 | 37/42 (88.1) | 10/42 (23.7) | 43 | 11/43 (25.6) | 5/43 (11.6) |
| Self-reported depressed mood during the past month | 0.502 | 0.786 | 0.190 | 0.077 |
| Yes | 25 | 22/25 (88.0) | 3/25 (12.0) | 25 | 10/25 (40.0) | 1/25 (4.0) |
| No | 85 | 70/85 (82.4) | 12/85 (14.1) | 87 | 23/87 (26.4) | 16/87 (18.4) |
| Self-reported anhedonia during the past month | 0.150 | 0.242 | 0.015 | 0.186 |
| Yes | 19 | 18/19 (94.7) | 1/19 (5.3) | 19 | 10/19 (52.6) | 1/19 (5.3) |
| No | 91 | 74/91 (81.3) | 14/91 (15.4) | 93 | 23/93 (24.7) | 16/93 (17.2) |
| Gestational age at questionnaire (weeks) | 0.739 | 0.626 | 0.513 | 0.599 |
| <10 | 45 | 37/45 (82.2) | 7/45 (15.5) | 46 | 12/46 (26.1) | 6/46 (13.0) |
| ≥10 | 65 | 55/65 (84.6) | 8/65 (12.3) | 66 | 21/66 (31.8) | 11/66 (16.7) |
| Parity | 0.542 | 0.957 | 0.691 | 0.730 |
| Nulliparous | 74 | 63/74 (85.1) | 10/74 (13.5) | 75 | 23/75 (30.7) | 12/75 (16.0) |
| Parous | 36 | 29/36 (80.6) | 5/36 (13.9) | 37 | 10/37 (27.0) | 5/37 (13.5) |
| History of spontaneous abortion | 0.074 | 0.571 | 0.139 | 0.742 |
| Yes | 30 | 22/30 (73.3) | 5/30 (16.7) | 30 | 12/30 (40.0) | 4/30 (13.3) |
| No | 80 | 70/80 (87.5) | 10/80 (12.5) | 82 | 21/82 (25.6) | 13/82 (15.9) |
expectancy of trisomy 21 individuals significantly preferred <15-min sessions to other durations. While the majority of women preferred in-person GC with ≥30-min sessions for pretest GC, their preferences varied by background characteristics, experience, attitudes, and knowledge. To our knowledge, this is the first study on pregnant women’s preferences concerning different models and durations of prenatal pretest GC.

**A strong preference for in-person GC**

In our study population, the proportion of pregnant women who preferred traditional in-person GC was over 80%, which was higher than that in previously published studies from Latin and Italian populations, which reported that roughly half of pregnant women indicated a preference for in-person GC [17, 23]. Such differences may reflect cultural and personal preferences in the study populations. One-third of pregnant Latina women stated that advantages of group GC included the value of hearing the perspectives of other individuals in a similar situation to their own and the opportunity to learn from others [23]. In this study, 3.5% and 9.6% of women stated preferences for group GC and classroom GC, respectively. One possible explanation for this difference is that Japanese pregnant women may be uncomfortable sharing GC information in front of others and wish for privacy and the focused attention of a genetic counselor, which they can receive in individual sessions. In this study, a history of having undergone prenatal testing in a prior pregnancy was the only significant factor associated with non-preference for in-person GC. This finding suggests that these women thought that they already understood the information of prenatal testing and the process of GC.

**Table 4 (continued)**

| Method of conception                  | Preferences toward pretest GC model | Preferences toward pretest GC duration (minutes in length) |
|---------------------------------------|-------------------------------------|----------------------------------------------------------|
|                                       | Number (n = 110)                     | Number (n = 112)                                         |
|                                       | Number (%)  | p value  | Number (%)  | p value  | Number (%)  | p value  | Number (%)  | p value  |
| Natural conception                    | 0.666      | 0.449    | 0.457       | 0.216    | 0.643       | 0.879    |
| Pregnancy after infertility treatment | 0.492      | 0.634    |             |          |             |          |
| Fetal number                          |            |          |             |          |             |          |
| Singleton                             | 56         | 46/56 (82.1) | 9/56 (16.1) | 57       | 15/57 (26.3) | 11/57 (19.3) |
| Multiple                              | 54         | 46/54 (85.2) | 6/54 (11.1) | 55       | 18/55 (32.7) | 6/55 (10.9) |
|                                     |            |          |             |          |             |          |
|                                     | 99         | 82/99 (82.8) | 14/99 (14.1) | 100       | 33/100 (33.0) | 0.018 |
|                                     | 11         | 10/11 (90.9) | 1/11 (9.1)   | 12       | 0/12 (0.0)   | 2/12 (16.7) |

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*a*Four participants who answered “don’t know” to the questions about preference for GC models were excluded from this analysis

*b*Two participants who answered “don’t know” to the questions about preference for duration of pretest GC time were excluded from this analysis

*c*Includes participants who preferred 30–59 min only, as no participants preferred ≥60 min

**Alternative GC models for the Japanese population**

While it is important to improve pregnant women’s access to prenatal GC, it is paramount that their reported outcomes be considered when implementing alternative GC SDMs. Previous studies have demonstrated the utility of providing digital information on prenatal testing before in-person GC, which was confirmed to help improve patients’ knowledge [13, 21], shorten the time required [21], and reduce decision conflict [13, 16, 20]. Considering our finding that Japanese women tend to prefer in-person GC, such an approach would likely be acceptable in a Japanese population. Although telegenic-based GC, described as web-based or videoconferencing GC, also has advantages of interactive visual communication and satisfying pregnant women’s preference for individual counseling [14], this approach was preferred by the smallest proportion of pregnant women (2.6%) in this study population. Telegenic GC was less acceptable to pregnant women in Japan. To understand why in-person (face-to-face) GC was preferred among Japanese women, further studies are warranted.

**Influence of women’s experiential knowledge of trisomy 21**

While pregnant women’s understanding of the mechanism and average life expectancy of trisomy 21 were not significantly associated with their preferences for prenatal pretest GC models and durations, women who had an acquaintance with trisomy 21 or had talked with people with trisomy 21 significantly preferred ≥30-min sessions for pretest GC to other durations. This finding suggests that their experiential knowledge acquired from personal experience with trisomy 21 may play a role in prenatal...
Table 5: Influence of participant’s attitudes, knowledge, and experience on preferences toward in-person GC model and duration

| Experience                                                                 | Preferences toward pretest GC model | Preferences toward pretest GC duration (minutes in length) |
|----------------------------------------------------------------------------|------------------------------------|----------------------------------------------------------|
|                                                                            | Number (n = 110)                   | Number (n = 112)                                          |
|                                                                            | p value                            | Number (%)                                               |
|                                                                            | p value                            | Number (%)                                               |
|                                                                            | p value                            | Number (%)                                               |
|                                                                            | p value                            | Number (%)                                               |
| **Prenatal testing in a prior pregnancy**                                  |                                    |                                                          |
| Yes                                                                        | 7                                  | 6/7 (57.1)                                               |
|                                                                            |                                    | 1/7 (14.3)                                               |
|                                                                            |                                    | 1/7 (14.3)                                               |
|                                                                             | 0.050                              | 0.959                                                    |
| No                                                                         | 103                                | 88/103 (85.4)                                            |
|                                                                            |                                    | 14/103 (13.6)                                            |
|                                                                            |                                    | 105                                                       |
|                                                                            |                                    | 32/105 (30.5)                                            |
|                                                                            |                                    | 16/105 (15.2)                                            |
|                                                                             | 0.837                              | 0.853                                                    |
|                                                                             | 0.017                              | 0.289                                                    |
| **Had someone in their vicinity with T21 or talked with people of T21**   |                                    |                                                          |
| Yes                                                                        | 39                                 | 33/39 (84.6)                                             |
|                                                                            |                                    | 5/39 (12.8)                                              |
|                                                                            |                                    | 39                                                         |
|                                                                            |                                    | 17/39 (43.6)                                             |
|                                                                            |                                    | 4/39 (10.3)                                              |
| No                                                                         | 71                                 | 59/71 (83.1)                                             |
|                                                                            |                                    | 10/71 (14.1)                                             |
|                                                                            |                                    | 73                                                         |
|                                                                            |                                    | 16/73 (21.9)                                             |
|                                                                            |                                    | 13/73 (17.8)                                             |
|                                                                             | 0.587                              | 0.417                                                    |
| **Prenatal testing in this pregnancy**                                     |                                    |                                                          |
| None                                                                       | 62                                 | 50/62 (80.7)                                             |
|                                                                            |                                    | 10/62 (16.1)                                             |
|                                                                            |                                    | 64                                                         |
|                                                                            |                                    | 21/64 (32.8)                                             |
|                                                                            |                                    | 8/64 (12.5)                                              |
| NIPT                                                                       | 25                                 | 23/25 (92.0)                                             |
|                                                                            |                                    | 1/25 (4.0)                                               |
|                                                                            |                                    | 25                                                         |
|                                                                            |                                    | 4/25 (16.0)                                              |
|                                                                            |                                    | 2/25 (8.0)                                               |
| MSS                                                                        | 22                                 | 18/22 (81.8)                                             |
|                                                                            |                                    | 4/22 (18.2)                                              |
|                                                                            |                                    | 22                                                         |
|                                                                            |                                    | 8/22 (36.4)                                              |
|                                                                            |                                    | 7/22 (31.8)                                              |
| AC/CVS                                                                     | 1                                  | 1/1 (100.0)                                              |
|                                                                            |                                    | 0/4 (0.0)                                                |
|                                                                            |                                    | 1                                                          |
|                                                                            |                                    | 0/1 (0.0)                                                |
|                                                                            |                                    | 0/1 (0.0)                                                |
| **Attitudes**                                                              |                                    |                                                          |
| Preference on brochures of prenatal testing at the first visit             | 0.687                              | 0.645                                                    |
|                                                                             |                                    | 0.476                                                    |
|                                                                             |                                    | 0.763                                                    |
| Wanted to receive                                                          | 75                                 | 62/75 (85.7)                                             |
|                                                                            |                                    | 11/75 (14.7)                                             |
|                                                                            |                                    | 76                                                         |
|                                                                            |                                    | 24/76 (31.6)                                             |
|                                                                            |                                    | 11/76 (14.5)                                             |
| No or neither or don’t know                                                | 35                                 | 30/35 (82.7)                                             |
|                                                                            |                                    | 4/35 (11.4)                                              |
|                                                                            |                                    | 36                                                         |
|                                                                            |                                    | 9/36 (25.0)                                              |
|                                                                            |                                    | 6/36 (16.7)                                              |
| Planned prenatal testing at questionnaire                                  | 0.157                              | 0.292                                                    |
|                                                                             |                                    | 0.561                                                    |
|                                                                             |                                    | 0.790                                                    |
| No testing                                                                  | 32                                 | 25/32 (78.1)                                             |
|                                                                            |                                    | 6/32 (18.8)                                              |
|                                                                            |                                    | 33                                                         |
|                                                                            |                                    | 11/33 (33.3)                                             |
|                                                                            |                                    | 6/33 (18.2)                                              |
| Undecided                                                                  | 33                                 | 31/33 (93.9)                                             |
|                                                                            |                                    | 2/33 (6.1)                                               |
|                                                                            |                                    | 33                                                         |
|                                                                            |                                    | 11/33 (33.3)                                             |
|                                                                            |                                    | 4/33 (12.1)                                              |
| Will have testing                                                          | 45                                 | 36/45 (80.0)                                             |
|                                                                            |                                    | 7/45 (15.6)                                              |
|                                                                            |                                    | 46                                                         |
|                                                                            |                                    | 11/46 (23.9)                                             |
|                                                                            |                                    | 7/46 (15.2)                                              |
| Needed agreement with the partner for prenatal testing                     | 0.229                              | 0.028                                                    |
|                                                                             |                                    | 0.855                                                    |
|                                                                             |                                    | 0.043                                                    |
| Yes                                                                        | 86                                 | 70/86 (81.4)                                             |
|                                                                            |                                    | 15/86 (17.4)                                             |
|                                                                            |                                    | 87                                                         |
|                                                                            |                                    | 26/87 (29.9)                                             |
|                                                                            |                                    | 10/87 (11.5)                                             |
| No or neither or don’t know                                                | 24                                 | 22/24 (91.7)                                             |
|                                                                            |                                    | 0/24 (0.0)                                               |
|                                                                            |                                    | 25                                                         |
|                                                                            |                                    | 7/25 (28.0)                                              |
|                                                                            |                                    | 7/25 (28.0)                                              |
| Want to have GC with husband                                               | 0.744                              | 0.725                                                    |
|                                                                             |                                    | 0.157                                                    |
|                                                                             |                                    | 0.171                                                    |
| Yes                                                                        | 100                                | 84/100 (84.0)                                            |
|                                                                            |                                    | 14/100 (14.0)                                            |
|                                                                            |                                    | 102                                                       |
|                                                                            |                                    | 32/102 (31.4)                                            |
|                                                                            |                                    | 14/102 (13.7)                                            |
| No                                                                         | 10                                 | 8/10 (80.0)                                              |
|                                                                            |                                    | 1/10 (10.0)                                              |
|                                                                            |                                    | 10                                                         |
|                                                                            |                                    | 1/10 (10.0)                                              |
|                                                                            |                                    | 3/10 (30.0)                                              |
| **Knowledge**                                                              |                                    |                                                          |
| Aware of prenatal testing before this pregnancy                           | 0.275                              | 0.397                                                    |
|                                                                             |                                    | 0.037                                                    |
|                                                                             |                                    | 0.183                                                    |
| Yes                                                                        | 95                                 | 78/95 (82.1)                                             |
|                                                                            |                                    | 14/95 (14.7)                                             |
|                                                                            |                                    | 97                                                         |
|                                                                            |                                    | 32/97 (33.0)                                             |
|                                                                            |                                    | 13/97 (13.4)                                             |
| No                                                                         | 15                                 | 14/15 (93.3)                                             |
|                                                                            |                                    | 1/15 (6.7)                                               |
|                                                                            |                                    | 15                                                         |
|                                                                            |                                    | 1/15 (6.7)                                               |
|                                                                            |                                    | 4/15 (26.7)                                              |
| **Aware of GC**                                                            | 0.877                              | 0.312                                                    |
|                                                                             |                                    | 0.140                                                    |
|                                                                             |                                    | 0.056                                                    |
| Yes                                                                        | 26                                 | 22/26 (84.6)                                             |
|                                                                            |                                    | 2/26 (7.7)                                               |
|                                                                            |                                    | 27                                                         |
|                                                                            |                                    | 11/27 (40.7)                                             |
|                                                                            |                                    | 1/27 (3.7)                                               |
| No                                                                         | 84                                 | 70/84 (83.3)                                             |
|                                                                            |                                    | 13/84 (15.5)                                             |
|                                                                            |                                    | 85                                                         |
|                                                                            |                                    | 22/85 (25.9)                                             |
|                                                                            |                                    | 16/85 (18.8)                                             |
| **Understanding of mechanism of T21**                                       | 0.933                              | 0.724                                                    |
|                                                                             |                                    | 0.617                                                    |
|                                                                             |                                    | 0.054                                                    |
| Not an inherited disease                                                   | 56                                 | 47/56 (83.9)                                             |
|                                                                            |                                    | 7/56 (12.5)                                              |
|                                                                            |                                    | 57                                                         |
|                                                                            |                                    | 18/57 (31.6)                                             |
|                                                                            |                                    | 5/57 (8.8)                                               |
| Inherited disease or don’t know                                            | 54                                 | 45/54 (83.3)                                             |
|                                                                            |                                    | 8/54 (14.8)                                              |
|                                                                            |                                    | 55                                                         |
|                                                                            |                                    | 15/55 (27.3)                                             |
|                                                                            |                                    | 12/55 (21.8)                                             |
decision-making, as shown in a previous report among Canadian women [24]. For instance, women declining prenatal testing tended to know someone with trisomy 21 [25]. Those findings suggest that our study participants with experiential knowledge of trisomy 21 need time at pretest GC to carefully consider the value of prenatal testing and whether or not to undergo prenatal testing based on their experiential knowledge of trisomy 21.

Maternal mental stress as a predictor of an appropriate GC method

In this study, the prevalence of a depressive mental status, such as depression or anhedonia, was over 20%, which was higher than the previously reported prevalence of depression at the first trimester, where 11% of pregnant women had been diagnosed with depression according to a diagnostic interview [26]. This difference may be due to the fact that the prevalence of depression among this study population relied solely on a self-reported two-question instrument to identify depression. Some patients who test positive for depression with such an instrument were reportedly not found to have depression in a diagnostic interview [22]. The prevalence of a depressive mental status using a self-report two-question instrument may therefore be overestimated. Approximately 95% of pregnant women with anhedonia during the past month preferred in-person GC, with a significant preference for a longer duration of prenatal GC. This finding suggests that anhedonia during the past month is a predictor of women’s preference for prenatal GC method. Although there have been no studies assessing the association between maternal mental stress and women’s preferences for prenatal GC method, difficulty making decisions is a core symptom of depressive disorders. Our findings also suggest that depressed women require more time to decide whether or not to undergo prenatal testing. At in-person GC, there is more time for an exploration of women’s values, concerns and emotions, so in-person GC allows for more personalized psychosocial support.

Influence of women’s awareness of prenatal testing

Among women who had been aware of prenatal testing before this pregnancy, the proportion who preferred ≥30-min sessions for prenatal pretest GC was significantly higher than those of >30 min. This significant association may be due to the knowledge acquired before they become pregnant leading them to be more aware of the importance of pretest GC, although this study did not assess the level of their understanding. No studies have examined the relationship between pregnant women’s awareness of prenatal testing and their preference for prenatal GC methods. Our findings suggest that providing sufficient information about

| Preferences toward pretest GC model | Preferences toward pretest GC duration (minutes in length) | Number | (%) | Number | (%) | Number | (%) | Number | (%) | Number | (%) |
|-----------------------------------|-----------------------------------------------------------|--------|-----|--------|-----|--------|-----|--------|-----|--------|-----|
| In-person GC (n = 92) | Classroom or group GC (n = 15) | | | | | | | | | | |
| Understanding of average of life expectancy of T21 | Yes | No | | | | | | | | | |
| ≥40 years old | 46 | 49/46 (106.3) | 20/46 (42.6) | | | | | | | | |
| <40 years old or don’t know | 64 | 52/64 (81.3) | 12/64 (18.8) | | | | | | | | |
| Recognition of MAE-related disorders | Yes | No | | | | | | | | | |
| Includes participants who preferred ≥30-60 min only, as no participants preferred ≥60 min | 46 | 49/46 (106.3) | 20/46 (42.6) | | | | | | | | |

Table 5 (continued)
prenatal testing to women before pregnancy might influence the decision-making process concerning prenatal testing once they become pregnant.

Pregnant women who preferred short-duration pretest GC

Approximately 15% of women preferred <15-min sessions for pretest GC. Factors that were significantly associated with a preference for <15-min sessions were not knowing whether or not agreement with the partner was necessary for prenatal testing and a misunderstanding of the average life expectancy of trisomy 21 individuals. These findings suggest that women without the partner’s agreement and lacking basic education on trisomy 21 might more easily agree to prenatal testing than others. If prenatal testing is conducted in these pregnant women, the test results might create anxiety and conflict concerning the decision. While the present study did not address these issues, the involvement of the partner in the prenatal testing decision-making process and understanding of trisomy 21 might affect pregnant women’s awareness of the importance of pretest GC.

Limitations of the study

Several limitations associated with the present study warrant mention. First, our study was conducted in a population of a fairly high socioeconomic status: over 70% of the pregnant women in the present study were university graduates or better, and ~70% of the participants had an annual household income of ≥7 million yen. In addition, our study included a high prevalence of subjects with an advanced maternal age, pregnancies achieved by infertility treatment, and multiple pregnancies. Thus, to confirm our findings, further replicative studies on other population are encouraged. Second, in the absence of a validated instrument specific to our research questions, our study used one developed originally for this study. However, the current study focused on pregnant women’s preferences toward the models and duration of pretest GC, which will enable us to explore Japanese pregnant women’s preferences regarding prenatal GC and establish valuable pretest GC SDMs in this country. At present, various GC methods are being considered due to the ongoing COVID-19 outbreak. This study was conducted prior to the COVID-19 outbreak.

Conclusion

We reported that over 80% of Japanese pregnant women preferred traditional in-person GC for pretest GC. While most women preferred in-person GC for <30 min in length for prenatal pretest GC SDMs, women’s preferences may vary by their background characteristics, experience, and knowledge. These findings will help establish a prenatal GC system offering a choice of GC SDMs in Japan, along with further large-scale studies of pregnant women’s preferences and the reported outcomes of prenatal GC SDMs.

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Compliance with ethical standards

Conflict of interest

The authors declare that they have no conflict of interest.

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