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ORIGINAL ARTICLE

The effects of an online decision aid to support the reproductive decision-making process of genetically at risk couples—A pilot study

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Abstract

Couples at risk of transmitting a genetic disease to their offspring may experience doubts about their reproductive options. This study examines the effects of an online decision aid (DA) on the (joint) reproductive decision-making process of couples (not pregnant at time of inclusion) at risk of transmitting a genetic disease to their offspring. The primary outcome is decisional conflict, and secondary outcomes are knowledge, realistic expectations, deliberation, joint informed decision-making, and decisional self-efficacy. These outcomes were measured with a pretest–posttest design: before use (T0), after use (T1), and 2 weeks after use (T2) of the decision aid (DA). Usability of the DA was assessed at T1. Paired sample t-tests were used to compute differences between baseline and subsequent measurements. The comparisons of T0-T1 and T0-T2 indicate a significant reduction in mean decisional conflict scores with stronger effects for participants with high baseline decisional conflict scores. Furthermore, use of the DA led to increased knowledge, improved realistic expectations, and increased levels of deliberation, with higher increase in participants with low baseline scores. Decision self-efficacy only improved for participants with lower baseline scores. Participants indicated that the information in the DA was comprehensible and clearly

organized. These first results indicate that this online DA is an appropriate tool to support couples at risk of transmitting a genetic disease and a desire to have (a) child(ren) in their reproductive decision-making process.

KEYWORDS
(joint informed) decision-making, decision aid, decisional conflict, genetic counseling, preimplantation genetic testing (PGT), reproductive genetics

1 | INTRODUCTION

Deciding if, and when to have children, is one of life's most important decisions. Reproductive decisions in couples are usually made together and relate to questions such as whether and when to have children (Alvarez, 2018). Couples with an increased risk of offspring affected with a genetic disorder may face an additional question, namely which reproductive option to choose (De Die-Smulders et al., 2013). Most couples prefer to have a genetically related child (Severijns, de Die-Smulders, et al., 2021). Options for them are a natural conception with prenatal diagnosis (PND), or preimplantation genetic testing (PGT) or no genetic testing, thereby accepting the risk of transmitting the genetic disease to their child(ren). PND pertains to genetic testing of the fetus during pregnancy and possible termination of the pregnancy if the fetus is affected. PGT involves genetic testing of in vitro fertilized (IVF) embryos, after which only embryos unaffected by the familiar genetic condition will be transferred to the uterus (van Dijke et al., 2020). Alternatives for couples with a genetic risk for offspring include adoption, foster parenting, and refraining from having children. Lastly, the use of donor gametes (sperm donation or oocyte retrieval) can be considered. When choosing for this latter option, only one of the parents will be genetically related to the child.

Choosing between these options may be challenging, and couples may face difficulties in their reproductive decision-making process (Derks-Smeets et al., 2014; Myring et al., 2011). The complexity of decision-making may be influenced by several factors, such as the severity and inheritance pattern of the disease, personal experiences with the disease, attitudes toward pregnancy termination, the desire to have genetically related children, preference toward a natural pregnancy, and costs of procedures (e.g., adoption costs) (Derks-Smeets et al., 2014; Genoff Garzon et al., 2018; Hershberger et al., 2012). Generally, two decisional partners (i.e., the prospective parents) are involved in reproductive decision-making and each of these factors may carry different weights for each partner. As reproductive decision-making involves risk and uncertainty of outcomes, and the decision is highly value-laden, decisional conflict may be experienced by the partners individually and as a couple. Decisional conflict pertains to the uncertainty about the course of action to take (O'Connor, 1995). Even after making a reproductive decision and taking actions, couples may experience doubt regarding the decision they made (Decruyenaere et al., 2007).

What is known about this topic

Couples who are at risk of transmitting a genetic disease to their offspring may face challenges regarding reproductive decision-making. Deciding if, and how to pursue their desire to have (a) child(ren), can be a demanding process, and many couples indicate a need for support during decision-making.

What this paper adds to the topic

This paper provides more insight into the joint decision-making process of couples. Additionally, it shows that the online decision aid can be an appropriate addition to genetic counseling in the reproductive decision-making process of couples at risk of transmitting a genetic disease to their offspring.

Informed decision-making (IDM) can mitigate decisional conflict (Gietel-Habets et al., 2018). Different aspects play a role in informed decision-making such as knowledge, deliberation, and value-consistency. In the case of reproductive decisions, ideally, all decisional partners should be involved in the decision-making process. To date, however, literature on decision-making in the preconception and prenatal setting primarily focuses on the perspective of the pregnant woman and relatively little attention has been paid to the involvement of the partner in the joint decision-making process. Previous research has shown that women perceive their partner as an important decisional partner, but that they often do not feel sufficiently supported by them (Underwood et al., 2020; van der Wulp et al., 2013). Furthermore, there are indications that jointly deliberating and settling together, on a decision that affects both partners, may yield better health outcomes (Osamor & Grady, 2018). When reviewing the extent of partner involvement in decision support tools related to prenatal screening and diagnosis, we found that many decision aids (DAs) do not focus on partner involvement (Severijns, van der Linden, et al., 2021). Making a joint informed decision entails the concepts of informed decision-making; meaning that both partners should have sufficient knowledge, that partners should deliberate separately and together and that their decision is value consistent.

Previous research has shown that there is a need for additional support regarding reproductive decision-making in the context of

hereditary cancer (Gietel-Habets et al., 2018). Different strategies are available to support the decision-making process and to promote IDM (Stacey et al., 2014). One of these strategies, which is especially useful in choices that are preference-sensitive and value-laden, includes the use of DAs (Stacey et al., 2017). DAs are tools designed to assist individuals to make a choice and deliberate on the risks, benefits, and consequences of the available options. Furthermore, DAs have been proven to be effective in improving users' knowledge regarding options and to reduce decisional conflict (Nagle et al., 2008; Stacey et al., 2014).

To support couples at risk of transmitting a genetic disease to their offspring who have a desire to have (a) child(ren), we developed an online DA, detailing all reproductive options offered to couples. The DA is not meant to replace reproductive counseling in the clinical genetic setting but to support the counseling process. To promote joint informed decision-making (JIDM), the DA places emphasis on conveying information regarding the reproductive options and on optimizing the decisional process between partners (Nagle et al., 2008). With joint decision-making, we refer to the process in which both partners (mostly romantic partners), who are affected by the decision, contribute to the decision-making process. The DA aims to encourage interaction between partners and to stimulate communication about their values and preferences regarding reproductive options.

The primary aim of this study is to investigate the effectiveness of the DA in reducing decisional conflict. The secondary aim of this study was to explore the effect of the DA on the joint decision-making process between partners. Other secondary outcome measures are knowledge, realistic expectations, deliberation, and decisional self-efficacy. We report the results of the pilot study.

2 | METHODS

2.1 | Study design

A nationwide pilot study was conducted in the Netherlands to investigate the effects of the online DA. The pilot study was conducted in collaboration with all University Medical Centres of the Netherlands (Maastricht University Medical Centre [MUMC+], University Medical Centre Utrecht, University Medical Centre Groningen, Amsterdam University Medical Centre, Erasmus Medical Centre, Radboud University Medical Centre, and Leiden University Medical Centre). A pretest–posttest design was used to evaluate the short-term effects of the DA. The study protocol was approved by the Medical Ethics Committee of MUMC+ (METC 2019-1278).

2.2 | The decision aid

The DA was developed in line with the International Patient Decision Aid Standards (Holmes-Rovner, 2007) and consists of several components:

1. *An introduction page* -
   This page explains the aim and usage of the decision aid. Participants are asked to indicate the inheritance pattern of their condition after which they receive tailored information about the risk of transmitting the disease to their offspring.
2. *Information on the reproductive options* -
   The DA was designed in a way that users can choose about which option they want to know more about. On the first page, basic information on all reproductive options was provided, thereby ensuring users know at minimum the key points of each option. Apart from the basic information on all available options that was provided at the start of the decision aid, participants were free to use the decision aid at their discretion. They were at liberty to read the information about all reproductive options, or only select a few, and to choose the information formats (textual, graphical, and video material) that suited them best.
   Detailed information on the seven different reproductive options is provided: 1. natural conception without genetic testing, 2. PND, 3. PGT, 4. use of donor gametes (i.e., sperm donation or oocyte retrieval), 5. adoption, 6. foster parenting, and 7. refraining from having (further) children. The information includes a description of the medical procedures, risks, costs, and eligibility criteria, is tailored to the inheritance pattern relevant to the couple, and is provided in both text and video.
3. *Option grid* -
   An interactive option grid provides an overview of important characteristics of the different reproductive options, such as medical procedures, risks, and costs. Participants can select and compare the different reproductive options.
4. *Value Clarification Method (VCM)* -
   A VCM that presents users with 17 statements representing important values and motives related to reproductive decision-making and the different reproductive options is included. The content of the VCM is based on motives and considerations derived from previous studies (Derks-Smeets et al., 2014; Severijns, de Die-Smulders, et al., 2021; van Rij et al., 2011). An example is: "*For me it is important to be able to conceive naturally, even if there is a risk that my child will inherit the disease*." Users are asked to answer all statements on a 6-point scale ranging from completely disagree [1] to completely agree [6] or not applicable. When they have answered all the statements, they can generate an overview of all their answers on the statements.
5. *Joint decision-making exercise* -
   This exercise aims to stimulate deliberation and communication about values and the decision-making process between the partners. When both partners have answered all the statements, they receive a link to an overview of their answers. This enables partners to compare their answers on the VCM. After this, partners are encouraged to communicate about their preferences and preferred reproductive options. They can write down the values important to both of them and which reproductive options they consider together.

6. *Question prompt sheet* -
   Participants can note the remaining questions in the question prompt sheet and discuss these with their genetic counselor or other health care professional.
7. *A glossary* -
   Important terms that may be perceived as difficult by patients are explained in a glossary.
8. *Resources* -
   Information regarding resources used to develop the DA, funding resource and information about the developers and their contact details are provided.

2.3 | Participants and recruitment

Participants were recruited during or after consultation by clinical geneticists or genetic counselors of all Clinical Genetics Departments in the Netherlands from September 2020 to December 2020, during the COVID-19 pandemic. Couples were eligible for participation if they [1] had an increased risk of transmitting a genetic disease to their offspring, [2] had not yet implemented a reproductive decision, [3] wished to fulfill their desire to have (a) child(ren) within 5 years, [4] were 18 years or older and, if female, were of reproductive age (18–41) and [5] had sufficient knowledge of the Dutch language and ample experience with the use of computers and Internet. Both partners were asked to complete the questionnaires independently.

2.4 | Procedures and instrumentation

Genetic consultations were held face-to-face or online due to COVID-19 during the recruitment period. If couples indicated that they would like to participate, they received an e-mail including a patient information letter to introduce the study and a link to the online registration page. After registration and providing online informed consent, couples received a login code for the DA and were directed to the baseline questionnaire (T0). After completing T0, they were asked to use the online DA to their preferences. After use of the DA, they were immediately directed to T1. When both partners had filled in the questionnaire, they received an extra e-mail with a link to compare their answers on the VCM. Two weeks after completion of the baseline questionnaire, participants were asked to complete the third questionnaire (T2). Reminders via e-mail were sent 2 and 5 days after registration for the baseline questionnaire T0, 1 day after completion of the decision aid (T1), and 2 and 5 ays after participants received the last questionnaire (T2). After completion of all three questionnaires, participants received an online gift card of 15 euro.

The questionnaires were divided into several topics. Demographic characteristics, assessed at T0, included age, gender, and educational level. Additionally, at T0, several questions were asked regarding their medical and reproductive history and the consultation with the clinical geneticist. Participants were asked to fill in which partner has (the predisposition for) the hereditary disorder and which disease it concerns. Additionally, participants were asked at which University Medical Centre the consultation with the clinical geneticist took place and what the main topic of this consultation was. Couples were also asked to elaborate on their reproductive history and to indicate whether they have an indication for IVF or ICSI because of reduced fertility. Lastly, couples were asked within which time frame they aimed to fulfill their desire to have (a) child(ren) (currently, within 2 years, and within 5 years) (full questionnaire, Appendix S1).

The primary outcome measure was decisional conflict. The secondary outcome measures were the concepts of knowledge, deliberation, realistic expectations, decision self-efficacy, and joint informed decision-making. All primary and secondary outcome measures were assessed at T0, T1, and T2. At T1, the usability of the DA was assessed and at T2 the impact of COVID-19 on participants' reproductive decision-making was assessed.

Decisional conflict was assessed by the 16 items (Cronbach's $\alpha = 0.96$) of the Traditional Decisional Conflict Scale—Statement Format (O'Connor, 1995). This questionnaire has been validated for application in Dutch (Koedoot et al., 2001) and contains five subscales namely effective decision (Cronbach's $\alpha = 0.85$), uncertainty (Cronbach's $\alpha = 0.81$), informed (Cronbach's $\alpha = 0.92$), values clarity (Cronbach's $\alpha = 0.91$), and support (Cronbach's $\alpha = 0.80$). Each item is scored on a 5-point Likert scale ranging from strongly agree [0] to strongly disagree [4] resulting in a total score from no decisional conflict [0] to extremely high decisional conflict [100]. The total score was calculated by dividing the total sum by 16, and this was multiplied by 25. Due to the fact that some couples had not yet made a final reproductive decision, a combined score was also calculated excluding the subscale of "effective decision." The scores for the remaining 12 items were divided by 12 and multiplied by 25.

Knowledge was assessed with 14 questions (true/false/not sure), and one point was provided for each correct answer. Not sure was counted as an incorrect answer.

The level of *deliberation* was assessed by the deliberation scale of van den Berg (van den Berg et al., 2006). This scale consists of 6 questions (Cronbach's $\alpha = 0.89$) on a 5-point Likert scale ranging from strongly disagree [0] to strongly agree [5]. Based on median scores, deliberation scores of ≥23 (total 30) were considered to be high (Reumkens et al., 2021).

The extent to which participants had *realistic expectations* regarding all reproductive options was assessed by an adapted version of the questionnaire used by Reumkens, Tummers, et al. (2019). There were three questions, for example, "*What is the risk that women will develop complications (such as overstimulation of the ovaries) with IVF treatment for PGT.*" The questions contained eight to eleven response options, and one point was scored for each correctly answered question.

Decision self-efficacy was assessed by an adapted version of the validated Decision Self-Efficacy Scale (Bunn & O'Connor, 1996). This version contained four items (Cronbach's $\alpha = 0.70$), with a 5-point Likert scale ranging from not at all confident [0] to very confident [4]. The total score was calculated by dividing the total sum by 4 and this was multiplied by 25, resulting in a score from not at all confident [0]

to very confident [100]. Two items were added separately, that is, (1) "*I feel confident that I am able to make the choice regarding my desire to have children that best suits me*" (0 = totally disagree to 4 = totally agree) and (2) "*I find the reproductive decision-making process*" (0 = very difficult to 4 = very easy). These questions are based on previous research on this topic in a similar study population (Gietel-Habets et al., 2017).

Joint informed decision-making (JIDM) was measured with 12 self-developed questions (Table 4). This questionnaire was developed in co-creation with several experts, for example, psychologists, clinical geneticists, and health communication specialists and covered five themes believed to relate to the dyadic decision-making process, that is, deliberation between partners, social support, dyadic communication, satisfaction with the decision-making process, and decision outcome. The items were scored on a 5-point Likert scale ranging from strongly disagree [1] to strongly agree [5]. An exploratory factor analysis was performed to check for underlying factors in the JIDM questionnaire. Based on the Kaiser criterion (1960), the scale was captured best by a one-factor solution. The internal reliability analysis of the 12 items on JIDM showed a Cronbach's alpha of 0.98. Based on the results of these analyses, it was decided to regard the 12 JIDM-related items as one scale and use the combined score for further analyses. Additionally, based on results from a previous study, we were interested in the assessment of the relative weight of both partners' opinions in the decision-making process (Reumkens, Tummers, et al., 2019). Therefore, couples were asked to respond to the following single statement: "*The woman's opinion is more important in reproductive decision*." Answer categories were yes or no, and an explanation for their answer was asked for.

Usability of the DA was assessed at T1 by 8 items regarding user perceptions (Table 5). These items are related to efficiency, effectiveness, enjoyment, and active trust (Crutzen et al., 2014). Related to this, the effect of the DA on decision-making, as perceived by the participants, was assessed by 6 items of The Preparation for Decision Making (PrepDM) scale. Both scales were assessed by a 5-point Likert scale ranging from totally disagree [1] to totally agree [5] (Crutzen et al., 2014). Additionally, participants were asked to give an appreciation score for the DA [1–10]. Lastly, there were two open-ended questions in which they could provide feedback on positive and negative features of the DA.

Finally, as this study was conducted during the *COVID-19* pandemic, at T2, four closed-ended questions assessed the impact on participants' reproductive decision-making, for example, "*The COVID-19 crisis made me think differently about the possible reproductive options regarding our desire to have (a) child(ren).*" These questions were answered with a 5-point Likert scale ranging from totally agree [1] to totally disagree [5]. Additionally, participants could elaborate on their answers.

2.5 | Data analysis

Data on baseline characteristics were summarized by means of descriptive statistics. Selectivity of dropout (i.e., non-random loss of participants) at T1 and T2 was tested using binary logistic regression. The paired sample t-tests were used to evaluate differences between the baseline and subsequent measurements (T0–T1 and T0–T2) with complete cases, that is, respondents who filled out T0 and T1 and/or T0 and T2. To indicate effect sizes (ES), Cohen's *d* was reported.

To assess the effects of the DA for subgroups (low or high baseline), the sample was split into two groups based on median baseline scores for all main outcome variables before conducting a paired sample t-test, respectively, one for each group. These analyses were explorative to see whether especially those with "decision difficulties" (lower or higher scores) at baseline benefited from the DA (Reumkens, Tummers, et al., 2019). Statistical analyses were performed using IBM SPSS software version 25. *p*-values of <0.05 were considered statistically significant.

3 | RESULTS

3.1 | Participants' characteristics at baseline

The baseline questionnaire (T0) was completed by 48 participants who used the DA for an average duration of 18 minutes, in which they read 13 pages. In total, 36 participants completed T1 (25% dropout) and 39 participants completed T2 (19% dropout). All questionnaires were completed by 35 participants (dropout rate 27%). Some couples only completed T0 and T2. Nineteen couples participated together ($n = 38$), and 10 participants took part without their partner. Two-third of the participants were highly educated ($n = 36$). Participants with different hereditary diseases participated. The most frequent hereditary condition was Huntington's disease (25%). Table 1 shows an overview of the demographic characteristics of the participants. One-third of the participants indicated at T0 that they were familiar with all of the reproductive options. All participants expressed a preference toward an option in which both partners would be genetically related to the child. At T2, the most preferred reproductive option was PGT (67%). Other participants indicated that their preferred option would be natural conception without genetic testing (13%) and PND (21%).

3.2 | Effects of the decision aid

3.2.1 | Primary outcome measure: Decisional conflict

Total mean decisional conflict scores (0–100) decreased from 24.94 at baseline to 21.88 at T1 ($p = 0.068$, ES = 0.37). From baseline to T2, the total mean decisional conflict score significantly decreased to 19.88 ($p = 0.032$, ES = 0.42) (Table 2).

The total mean decisional conflict scores for participants who did not make a decision (excluding the effective decision subscale) significantly decreased from 31.30 at baseline, to 25.64 at T1 ($p = 0.021$, ES = 0.40) to 24.20 at T2 ($p = 0.007$, ES = 0.46). The subcategories of feeling informed and values clarity showed a significant

TABLE 1 Baseline characteristics of participants ($n = 48$)

	N	Percentage
Gender		
Male	21	43.8
Female	27	56.3
Age (years)	31.8[a] (SD 4.4)	-
Male	33.3[a] (SD 5.2)	-
Female	30.7[a] (SD 3.5)	-
Education[b]		
Low	1	2.1
Middle	15	31.3
High	32	66.7
Mode of inheritance		
Autosomal dominant	28	58.3
Autosomal recessive	8	16.7
X-linked	3	6.3
Chromosomal	8	16.7
Unknown	1	2.1
Main topic counseling		
Desire to have children	27	56.3
Genetic condition	16	33.3
Both	2	4.2
Children (with current partner)		
Yes	9	18.8
No	39	81.3
Planning to have children		
Trying to conceive now	10	20.8
Within 2 years	24	50.0
Within 5 years	8	16.7
Not sure yet	6	12.5
IVF/ICSI because of reduced fertility		
Yes	12	25.0
No	19	39.6
Not sure	17	35.4
Preferred reproductive option		
Natural conception without genetic testing	7	14.6
PND	9	18.8
PGT	32	66.7
Donor gametes	0	0
Adoption	0	0
Foster parenting	0	0
Refraining from having children	0	0

[a]Mean age, sd = standard deviation.

[b]Low = less than primary, primary and lower secondary education, middle = upper or post-secondary non-tertiary education, high = tertiary education (Reumkens, Tummers, et al., 2019).

decrease in both T1 and T2, indicating that the decision aid seems to have had the most effect on the aspects of feeling informed and feeling clear about personal values. The other subcategories showed a decrease in both T1 and T2, although not significant.

The analyses based on median subgroup scores indicated that participants with high baseline decisional conflict scores (>33.3) significantly decreased in total scores from baseline (M = 44.79) to T1 (M = 33.68, $p = <0.001$, ES = 1.04, $n = 18$) and T2 (M = 32.08, $p = 0.002$, ES = 0.81, $n = 20$), while participants with low baseline decisional conflict sores (≤33.3) showed no significant reduction at T1 ($n = 18$) or T2 ($n = 19$) (Table 3).

Differences in sex

Analyses split by sex revealed that among males only scores on the informed subscale significantly decreased from 28.57 at baseline to 12.82 at T1 ($p = 0.048$, ES = 0.61). Among females, the total decisional conflict score (excluding effective decision) significantly decreased from 33.08 at T0 to 25.67 at T2 ($p = 0.037$, ES = 0.44). Scores on the informed subscale also significantly decreased from 29.67 at T0 to 24.64 at T1 ($p = 0.043$, ES = 0.45), and scores on the values clarity subscale significantly decreased from 32.00 at T0 to 20.00 at T2 ($p = 0.005$, ES = 0.61).

3.2.2 | Secondary outcome measures

Knowledge

Most participants (81%) knew that opting for natural conception implies the acceptance of the risk of transmitting the genetic disease to offspring and that the woman has to become naturally pregnant if a couple opts for PND (88%). Regarding PGT, 85% of the participants knew that a couple needs to undergo an IVF treatment for PGT and 92% knew that women need to take hormones during an IVF treatment and that PGT takes place before pregnancy (92%). Specific knowledge of other options, such as gamete donation and adoption, was lower, with only 10% of the participants knowing at baseline that anonymous gamete donation is not allowed in the Netherlands and that it is not allowed to adopt a child if one of the adoptive parents is (or will become) seriously ill (19%). The mean level of knowledge significantly increased (Table 2) from 8.38 at baseline to 9.33 at T1 ($p = 0.013$, ES = −0.44) and 9.26 at T2 ($p = 0.021$, ES = −0.39).

Deliberation

The mean level of deliberation (Table 2) significantly increased from 22.18 at baseline, to 23.39 at T1 ($p = 0.044$, ES = -0.35) and 24.13 at T2 ($p = 0.001$, ES = -0.56). However, for participants with low baseline scores on deliberation and with follow-up data at T1 ($n = 15$) or T2 ($n = 17$), the level increased from 18.47 at T0 to 20.67 at T1 ($p = 0.039$, ES = −0.59) to 22.06 at T2 ($p = 0.002$, ES = −0.89) (Table 3),

Realistic expectations

Realistic expectations (Table 2) increased from a score of 0.72 at baseline to 1.19 at T1 ($p = 0.020$, ES = −0.41) and 1.10 at T2 ($p = 0.012$, ES = −0.42). For participants with a low baseline score on realistic expectations (Table 3), scores significantly increased from 0.50 at T0 to 1.23 at T1 ($p = <0.001$, ES = −0.77, $n = 31$) and 1.06 at T2 ($p = <0.001$, ES = −0.68, $n = 34$).

TABLE 2 Short-term effects of the decision aid on main outcome measures

	T0	T1	T2	T0-T1 ($n = 36$)			T0-T2 ($n = 39$)		
	Mean (SD)	Mean (SD)	Mean (SD)	T	p	ES	T	p	ES
Decisional conflict									
Total score (0–100)	24.94 (13.23)	21.88 (17.53)	19.88 (11.89)	1.901	0.068	0.37	2.259	**0.032**	0.42
Total score (excl. effective decision; 0-100)[a]	31.30 (18.43)	25.64 (16.35)	24.20 (12.13)	2.423	**0.021***	0.40	2.863	**0.007**	0.46
Uncertainty	40.17 (24.99)	37.04 (22.93)	35.47 (20.65)	0.728	0.427	0.12	1.620	0.113	0.26
Informed	29.27 (21.53)	20.37 (18.20)	21.58 (12.20)	3.095	**0.004**	0.52	2.349	**0.024**	0.38
Values clarity	29.70 (21.27)	21.53 (16.47)	19.23 (11.34)	2.691	**0.011**	0.49	3.552	**0.001**	0.57
Support	26.07 (21.04)	23.61 (18.31)	20.51 (12.66)	0.851	0.401	0.14	1.641	0.109	0.26
Effective decision	22.19 (14.43)	20.83 (19.76)	16.81 (14.67)	1.383	0.179	0.27	1.970	0.059	0.37
Knowledge									
Total score (0–14)	8.38 (2.22)	9.33 (2.03)	9.26 (2.02)	−2.618	**0.013**	−0.44	−2.407	**0.021**	−0.39
PND (0–5)	2.83 (1.34)	3.36 (1.22)	3.05 (1.23)	−1.836	0.075	−0.31	−1.000	0.324	−0.16
PGT (0–4)	3.38 (0.81)	3.55 (0.65)	3.62 (0.67)	−1.063	0.295	−0.18	−1.462	0.152	−0.23
Other[b] (0–5)	2.17 (1.00)	2.42 (1.30)	2.59 (1.14)	−1.723	0.094	−0.29	−2.515	**0.016**	−0.40
Realistic expectations (0–3)	0.72 (0.76)	1.19 (0.82)	1.10 (0.68)	−2.446	**0.020**	−0.41	−2.649	**0.012**	−0.42
Deliberation (6–30)	22.18 (4.36)	23.39 (4.19)	24.13 (3.97)	−2.092	**0.044**	−0.35	−3.491	**0.001**	−0.56
Decision self-efficacy									
Total score (0–100)	80.77 (14.22)	84.03 (12.80)	83.17 (12.43)	−1.053	0.300	−0.18	−1.212	0.233	−0.19
item 5[c] (1–5)	4.00 (0.76)	4.22 (0.87)	4.15 (0.67)	−1.643	0.109	−0.27	−1.356	0.183	−0.22
item 6[d] (1–5)	2.67 (1.13)	2.64 (0.90)	2.67 (0.93)	0.751	0.457	0.13	0.000	1.000	0.000

Bold values are *p*-values which were considered statistically significant (<0.05).

[a]These 12 items were summed, divided by 12, and multiplied by 25, T1 ($n = 27$), T0/T2 ($n = 29$).

[b]Other consisted of five questions on natural conception (1), use of donor gametes (2), adoption (1), and foster parenting (1).

[c]"I feel confident that I am able to make the choice regarding my desire to have children that best suits me (0 = totally disagree to 4 = totally agree).

[d]I find the reproductive decision-making process…" (0 = very easy to 4 = very difficult).

Decisional self-efficacy

There was no overall significant increase in participants' decision self-efficacy scores. However, for participants with low baseline scores of decisional self-efficacy (Table 3), scores only significantly increased from 69.38 at T0 to 77.57 at T1 ($p = 0.031$, ES = −0.57, $n = 17$).

Joint informed decision-making

The overall mean scores on all JIDM items were high at baseline and did not significantly increase at T1 and T2. Most participants indicated that they agreed or totally agreed with the statements (scale ranging from strongly disagree [1] to strongly agree [5]; Table 4). Many participants indicated that they discussed the pros and cons of the options together (4.08), that they felt satisfied with the way they discussed the various options with their partner (4.10), and that they know which option their partner prefers (4.28). At T2, the item with the highest score indicated that they were satisfied with the way they make the decision together (4.38). Even though the scores did not significantly increase, participants indicated that the joint decision-making exercise helped them to talk with each other and gain more insight into their preferences. At baseline, in total, 11 men and 11 women (46%) agreed that the woman's opinion is more important in the reproductive decision and 5 men and 7 women (25%) disagreed. Five men and 9 women (29%) neither agreed nor disagreed with this statement. Different reasons were mentioned for why participants perceived the woman's opinion as more important, and these were primarily related to the fact that the woman experiences the physical burden (carrying the child, possible medical treatments) and psychological burden (pregnancy termination) of the choice.

Usability of the DA

As shown in Table 5, the mean scores on the Preparation for Decision Making Scale (range 1–5) ranged between 3.19 and 3.61, indicating that participants perceived the DA as useful in preparing oneself to communicate with their counselor and make reproductive decisions. Regarding the usability of the DA, scores ranged from 3.86 to 4.19 indicating that the DA provided relevant information (mean 4.14), that it increased their awareness on options (3.94), that they could use the information from the DA to make a decision about their desire to have (a) child(ren) (3.86), and that they would recommend the DA to other couples at risk of

TABLE 3 Analyses for main outcome measures based on median split baseline scores

	T0 (baseline)	T1 (immediately after completing the decision aid)	T2 (2 weeks after baseline)	T0-T1 ($n = 36$)				T0-T2 ($n = 39$)			
	Mean (SD)	Mean (SD)	Mean (SD)	T	*p*	ES	*n*	T	*p*	ES	*n*
Decisional conflict (0-100)[a]											
Low baseline (≤33.3)	17.10 (8.63)	17.59 (11.90)	15.90 (11.03)	−0.073	0.942	−0.02	18	0.398	0.696	0.9	19
High baseline (>33.3)	44.79 (14.69)	33.68 (16.49)	32.08 (6.67)	4.429	**<0.001**	1.04	18	3.615	**0.002**	0.81	20
Knowledge (0–14)											
Low baseline (≤8)	6.65 (1.53)	8.17 (1.69)	8.40 (1.50)	−3.848	**0.001**	−0.91	18	−4.413	**<0.001**	−0.99	20
High baseline (>8)	10.21 (1.03)	10.50 (1.65)	10.16 (2.14)	−0.449	0.659	−0.11	18	0.096	0.925	0.02	19
Realistic expectations (0–3)											
Low baseline (≤1)	0.50 (0.51)	1.23 (0.84)	1.06 (0.69)	−4.281	**<0.001**	−0.77	31	−3.957	**<0.001**	−0.68	34
High baseline (≥2)	2.20 (0.45)	1.00 (0.71)	1.40 (0.55)	3.207	**0.033**	1.43	5	4.000	**0.016**	1.79	5
Deliberation (6–30)											
Low baseline (<23)	18.47 (3.54)	20.67 (3.85)	22.06 (4.18)	−2.277	**0.039**	−0.59	15	−3.669	**0.002**	−0.89	17
High baseline (≥23)	25.04 (2.48)	25.33 (3.29)	25.73 (3.03)	−0.696	0.494	−0.15	21	−1.334	0.197	−0.28	22
Decision self-efficacy (0–100)											
Low baseline (<84.4)	69.38 (10.12)	77.57 (11.49)	75.63 (11.27)	−2.364	**0.031**	−0.57	17	−2.011	0.059	−0.45	20
High baseline (≥84.4)	92.76 (4.78)	89.80 (11.26)	91.12 (7.88)	1.407	0.176	0.32	19	0.773	0.450	0.18	19
Joint Informed Decision-Making (12–60)											
Low baseline (≤50)	42.85 (9.33)	42.79 (11.37)	48.00 (5.53)	−0.041	0.968	−0.01	19	−1.804	0.087	−0.40	20
High baseline (>50)	57.31 (3.33)	57.06 (4.39)	53.79 (11.11)	0.575	0.573	0.14	17	1.351	0.193	0.31	19

[a]Decisional conflict scale excluding effective decision subscale.

TABLE 4 Statements on joint informed decision-making

	T0 ($n = 39$) Mean (SD)	T1 ($n = 36$) Mean (SD)	T2 ($n = 39$) Mean (SD)
Total score (12-60)[a,b]	49.90 (10.12)	49.53 (11.29)	50.82 (9.07)
My partner and I discussed the pros and cons of the options together	4.08 (0.90)	4.06 (0.89)	4.00 (0.89)
I am confident that I can make the right decision together with my partner	4.15 (1.01)	4.08 (1.05)	4.28 (0.86)
I let my partner know that I appreciate his/her support and advice during the decision-making process	4.21 (0.92)	4.19 (1.04)	4.26 (0.85)
My partner lets me know that he/she appreciates my support and advice during the decision-making process	4.10 (1.0)	4.11 (1.06)	4.21 (0.86)
I think my partner listens carefully to me during the decision-making process	4.15 (1.01)	4.11 (1.09)	4.15 (0.93)
I think I listen carefully to my partner during the decision-making process	4.21 (0.92)	4.17 (0.97)	4.23 (0.81)
My partner has asked me which option I prefer	4.13 (1.03)	4.06 (1.12)	4.13 (0.98)
I know which option my partner prefers	4.28 (0.89)	4.14 (1.05)	4.31 (0.83)
I know how my partner feels about the advantages and disadvantages of the different options and what is important to him/her	4.21 (0.83)	4.14 (0.96)	4.23 (0.87)
I am satisfied with the way my partner and I discuss the various options	4.10 (0.88)	4.11 (1.01)	4.33 (0.80)
I am satisfied with the way we make the decision together	4.15 (0.96)	4.14 (0.99)	4.38 (0.78)
I am convinced that this decision is best made by us together, rather than having one of us decide	4.13 (1.11)	4.22 (1.07)	4.31 (0.95)

[a]All scores did not change significantly.

[b]The items were scored on a 5-point Likert scale ranging from strongly disagree [1] to strongly agree [5].

transmitting a genetic disease to their offspring if they were looking for information about reproductive options (4.19). Participants graded the DA with a mean score of 8.0 (SD = 0.84) on a scale of 1–10.

Participants indicated that the information was comprehensible and clearly structured. They appreciated that the advantages and disadvantages were mentioned for each option and that different reproductive options were explained. The option grid and the VCM were particularly appreciated. Participants indicated that the VCM helped them to communicate with their partner and that it provided insight into the perspective of their partner. Frequently mentioned points of improvement were that the DA could be better adapted to be used on mobile devices and that it would be better to use more bullet points to structure information.

Influence of COVID-19 on the decision-making process

Some participants indicated that the COVID-19 crisis influenced their perspective on the different reproductive options. Particularly, participants indicated that medical procedures (e.g., IVF or PGT procedures) would be postponed or delayed ($n = 10$), with several participants describing insecurities about the start of medical trajectories (e.g., start of a PGT trajectory) ($n = 6$). Other factors that were mentioned pertained to the quality of hospital care during COVID-19 ($n = 1$), the influence of COVID-19 on their own health ($n = 1$), the safety and influence of the COVID-19 vaccines on a pregnancy ($n = 1$) and experiencing more stress due to COVID-19 ($n = 1$). None of the participants were afraid to visit a medical center during the COVID-19 crisis. Only one participant indicated that their preferred reproductive option changed during the COVID-19 crisis as the waiting lists for a PGT trajectory had increased and, therefore, they decided to opt for a natural pregnancy. With regard to changes in the provision of care due to COVID-19, some participants indicated that they experienced the online consultations as less pleasant ($n = 8$) and that the waiting lists for medical procedures had increased ($n = 14$). Eleven participants found the online consultations more pleasant compared with face-to-face consultations. Some participants could not start with a medical trajectory, for example, IVF/PGT ($n = 2$). All participants indicated that the COVID-19 crisis had not influenced the communication between them and their partner regarding decision-making.

4 | DISCUSSION

In this study, we investigated the short-term effects of an online decision aid on reproductive options for couples at risk of transmitting a genetic disease to their offspring. Immediate (T0-T1 before and after viewing the DA) and short-term effects (T0-T2 before and 2 weeks after viewing the DA) were found for decisional conflict, knowledge, realistic expectations, and deliberation. Regarding the main outcome measure, decisional conflict, scores significantly decreased from T0-T1 and T0-T2 for participants who did not make a decision. The effect sizes of 0.40 (T0) and 0.46 (T2) are in line with studies of decision support interventions (O'Connor, 1995). No overall effects were found for decisional self-efficacy. A possible

TABLE 5 Evaluation of the DA: user perceptions

	T1 ($n = 36$) Mean (SD)
Preparation for Decision Making Scale (0 not at all–5 a lot)Did this decision aid…	
Prepare you to make a better decision	3.39 (1.13)
Help you think about the pros and cons of each option	3.50 (1.11)
Help you think about which pros and cons are most important	3.61 (1.05)
Help you organize your own thoughts about the decision	3.33 (1.12)
Help you identify questions you want to ask your doctor?	3.19 (1.26)
Prepare you to talk to your health care professional about what matters most to you?	3.28 (1.23)
Usability of the DA (0 totally agree–5 totally disagree)	
I could easily search for information in the DA	3.94 (0.83)
The DA increased my awareness about the different reproductive options	3.94 (0.75)
The website provided relevant information	4.14 (0.80)
I could trust the information provided by the DA	4.11 (0.82)
I thought my visit to the DA was pleasant	3.97 (0.70)
I could use the information from the DA to make a decision about my desire to have children	3.86 (0.83)
I would return to this DA if I was looking for information about reproductive options	3.89 (0.82)
I would recommend this website to others if they were looking for information about reproductive options for people with an increased risk of transmitting a genetic disease to their offspring	4.19 (0.82)

explanation might be that couples felt that they were not ready to make an actual decision because of all the uncertainties regarding medical procedures (e.g., long waiting lists), since two-thirds of the participants preferred PGT. This might be due to the uncertainties caused by the COVID-19 pandemic.

The subgroup analyses, however, showed sustained effects in increasing decision self-efficacy among participants with lower baseline decision self-efficacy (T0-T1 and T0-T2). Moreover, these analyses showed significant effects for participants with lower baseline levels of knowledge, realistic expectations, and deliberation while significant effects were also found for participants with a higher baseline level of decisional conflict (T0-T1 and T0-T2). The significant effects suggest that the DA especially supports couples with a higher need for reproductive decision support, which is in line with the findings of Reumkens, Tummers, et al. (2019) who evaluated the short-term effects of a DA for persons with a genetic predisposition to cancer and their partners (Reumkens, Tummers, et al., 2019). Reumkens, Tummers, et al. (2019) also found effects in increasing self-efficacy and deliberation among participants with low baseline levels in self-efficacy and deliberation. Additionally, this study showed a stronger decrease in decisional conflict among participants with high baseline decisional conflict levels (Reumkens, Tummers, et al., 2019).

Regarding JIDM, baseline scores were already high. The majority of the participants discussed the pros and cons of the different reproductive options with their partner, felt satisfied with the way they discussed the various options and knew which reproductive option their partner prefers. Additionally, they were satisfied with the way they make the decision together. The JIDM scale was new and self-developed and was tested within a small usability test with the clinical population and was, therefore, only used for exploratory purposes. Even though no significant effects were found on JIDM, the questions provided more insight in this process. For instance, almost half of the participants indicated that they believe that the opinion and preferences of the woman are more important in the decision-making process because she will be more affected by the psychological and physical burden. This is in line with previous studies in which women appeared to have a more important vote regarding reproductive decisions than men (Hollander et al., 2020; Lindgren et al., 2017; Severijns, de Die-Smulders, et al., 2021).

4.1 | Strengths and limitations

This study was the first to address the JIDM process and has provided insight in the JIDM process among a heterogeneous group of couples at risk of transmitting a genetic disease to their offspring. Even though no statistically significant effects were found on JIDM, participants indicated that they found the JIDM exercise pleasant and indicated that it encouraged them to talk to each other about the options. When partners both participate in a decision, it is more likely that they discuss and explore more options by sharing their perspectives and this may result in better outcomes (Osamor & Grady, 2018). A possible explanation for the lack of significant effects on JIDM relates to the high baseline scores on JIDM as these may be indicative of a ceiling effect caused by selective participation, but also the modest sample size of this pilot study. Since many partners participated as a couple, as was encouraged during recruitment, our study group might represent a group of couples in which both partners are committed to the reproductive decision-making process. Even though we asked participants to participate separately and independently, it might be that they participated together. Due to the small sample size, testing for intra-couple correlation was not feasible.

Further limitations of this study pertained to the fact that not all items of the questionnaires used have been validated in the Dutch language or for our specific clinical population. All concepts have however been measured in previous studies in similar clinical populations and/or the Dutch language (Gietel-Habets et al., 2017; Reumkens, Tummers, et al., 2019; Reumkens et al., 2021). The JIDM scale was however self-developed and has only been tested within a small usability test among our study population. Therefore, the

findings with regard to JIDM are to be considered of exploratory nature. It was also not possible to calculate a response rate as we were unable to reliably register the number of study invitations extended by all counselors involved in recruitment in all participating Clinical Genetics Departments.

Overall, the results show that the DA improved several outcomes related to decision-making. However, caution is warranted when interpreting these findings due to the application of a non-experimental design and a relatively small sample size. Some analyses were exploratory in nature, and the related findings should be interpreted with caution. Therefore, in future studies, that is, a planned randomized controlled trial (RCT), a larger sample size will be applied and prolonged effects of the DA will be examined. Additionally, within the RCT, attention will be paid to further confounders and effect modifiers since the current study was not suitable for this.

Lastly, one limitation pertains to the generalizability of the results. The sample included many participants with a high level of education, included only opposite-sex couples and many participants with Huntington disease. Furthermore, although our recruitment efforts aimed at including both same- and opposite-sex couples, only opposite-sex couples participated in this study. Therefore, the effects cannot be generalized to all couples at risk of transmitting a genetic disease to their offspring. In future research, these factors should be taken into account.

4.2 | Conclusion and practice implications

The findings of this pilot study suggest that the DA is effective in supporting couples at risk of transmitting a genetic disease to their offspring in making a reproductive decision. In particular, those who have the highest need for decision support may benefit. The DA lowered participants' decisional conflict and had a positive effect on indicators of informed decision-making (e.g., knowledge). The DA is meant as an additional support tool and is no replacement of (online) consultations with a genetic counselor, but a relevant and appropriate addition to reproductive genetic counseling (Gietel-Habets et al., 2018; Stacey et al., 2014). Preferably, a referral to the tool could be included in the standard report couples receive after consultation (Reumkens, de Die-Smulders, & van Osch, 2019). In this way, couples receive information from the consultations and from the DA. Additionally, in our opinion, supporting partner involvement and optimizing joint informed decision-making in decisions related to reproduction should form an integral part of counseling and decisional support efforts. We believe it is important to focus on both partners because in previous studies, men expressed the desire to be treated as a couple by health care professional and that they want to be involved in the decision-making process regarding reproductive decisions (Hollander et al., 2020; Williams et al., 2011). Currently, many DAs do not focus or involve the partner in decision support (Severijns et al., 2021). Therefore, we aim to involve both partners in our DA and explore the effects of this involvement.

AUTHOR CONTRIBUTIONS

Yil Severijns: Conceptualization; project administration; writing – original draft; writing – review and editing. **Maartje W. F. Heijmans:** Conceptualization; writing – original draft; writing – review and editing. **Christine E. M. de Die-Smulders:** Conceptualization; supervision; writing – original draft; writing – review and editing. **Emilia K. Bijlsma:** Conceptualization; writing – review and editing. **Nicole Corsten-Janssen:** Conceptualization; writing – review and editing. **Sara J. R. Joosten:** Writing – review and editing. **Sander M. J. van Kuijk:** Formal analysis; writing – original draft. **Klaske D. Lichtenbelt:** Writing – review and editing. **Cecile P. E. Ottenheim:** Writing – review and editing. **Kyra E. Stuurman:** Writing – review and editing. **Gita M. B. Tan-Sindhunata:** Writing – review and editing. **Hein de Vries:** Supervision; writing – original draft; writing – review and editing. **Liesbeth A. D. M. van Osch:** Supervision; writing – original draft; writing – review and editing. Authors Y. Severijns, L. van Osch en C. E. M. de Die-Smulders confirm that they had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. All of the authors gave final approval of this version to be published.

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COMPLIANCE WITH ETHICAL STANDARDS

CONFLICT OF INTEREST

Y. Severijns, M.W.F. Heijmans, L. van Osch, C.E.M. de Die-Smulders, H. de Vries, S.M.J. van Kuijk, E.K. Bijlsma, N. Corsten-Janssen, S.J.R. Joosten, K.D. Lichtenbelt, C.P.E. Ottenheim, K.E. Stuurman, and M.B. Tan-Sindhunata declare that they have no conflict of interest.

HUMAN STUDIES AND INFORMED CONSENT

This study was approved by the medical ethics committee of Maastricht University Medical Centre (METC 2019-1278). All procedures performed in this study were in accordance with the ethical standards of the medical ethics committee of Maastricht University Medical Centre and have been performed in accordance with the ethical standards as laid down in the 1964 Declaration of Helsinki and its later amendments.

DATA SHARING AND DATA ACCESSIBILITY

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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