Received: 31 May 2019 | Revised: 5 August 2019 | Accepted: 8 August 2019
DOI: 10.1002/jgc4.1162

National Society of Genetic Counselors WILEY

ORIGINAL ARTICLE

Attitudes of Israeli parents of children with Down syndrome toward non-invasive prenatal screening and the scope of prenatal testing

Tamar Nov-Klaiman | **Aviad E. Raz** | **Yael Hashiloni-Dolev**

Department of Sociology & Anthropology, Ben-Gurion University of the Negev, Beer-Sheva, Israel

Correspondence
Tamar Nov-Klaiman, Department of Sociology & Anthropology, Ben-Gurion University of the Negev, Beer-Sheva, Israel.
Email: tamarnk@gmail.com

Funding information
German Research Foundation

Abstract

Due to its early utilization and increasing ability to provide genetic information, non-invasive prenatal screening (NIPS) has reinforced social and bioethical quandaries concerning prenatal genetics. This paper presents exploratory findings based on 20 semi-structured interviews conducted in 2017–2019 with Israeli parents of children with Down syndrome (DS), four of whom also serving as representatives of DS organizations. Their views are presented regarding the pros and cons of NIPS; the social context of decision-making about NIPS; and views on life with DS and termination of pregnancies on that ground. While illustrating the large heterogeneity of views concerning NIPS and prenatal diagnosis (PND) amongst parents of children with DS, our respondents commonly criticized the imbalanced information provided by professionals regarding DS, seen as sending a discriminating message in line with the public ignorance surrounding DS. These views are further discussed in the multi-cultural, ableist and pro-natal context of Israeli society. We conclude by offering practical implications concerning NIPS, parental autonomy, and informed choice.

KEYWORDS
Down syndrome, Israel, NIPS, parents, prenatal genetic screening

1 | INTRODUCTION

Non-invasive prenatal screening (NIPS) is a new genetic test targeting placenta-derived cell-free DNA (cfDNA) circulating in maternal blood. At the moment it is used mainly for the detection of chromosomal abnormalities, especially targeting Down Syndrome (DS) in some marketing campaigns. The test is effective as early as 9–10 weeks of gestation and being non-invasive it does not entail a risk for miscarriage which is associated with invasive diagnostic testing, such as amniocentesis and chorionic villus sampling (CVS). Cell-free DNA is rapidly cleared from the maternal circulation within hours after delivery, making it pregnancy specific. However, as NIPS analyzes cfDNA originating from the placenta, and since in a small proportion of cases the chromosomal make-up of the placenta is not identical to that of the fetus, the result of the test might not correctly represent the fetal status. Moreover, cfDNA originating from a vanished twin or maternal karyotype anomalies may also lead to false results (Leonard, 2017). Therefore, NIPS is considered as a screening test. As such it requires verification by other means of approved genetic diagnosis, such as CVS or amniocentesis, when abnormal findings are detected (Devers et al., 2013). However, the new developments of non-invasiveness and abundance of genetic information in an early stage of the pregnancy have the potential to revolutionize prenatal genetics (Dondorp et al., 2015; van Schendel et al., 2014). The expansion of NIPS is also fueled by its commercialization (Minear, Lewis, Pradhan, & Chandrasekharan, 2015; Ravitsky, 2017).

Because of its unique features, NIPS has reinforced conflicting social and bioethical perspectives, including support of "procreative autonomy" (Savulescu, 2001), criticism of a new form of eugenics undermining moral equality (Habermas, 2003; Sandel, 2007; Schües, 2014; Thomas & Rothman, 2016), commodification of "babies by design" (Congregation, 2008), or promotion of parental desires for a particular form of family (Franklin & Roberts, 2006). This study focuses on the Israeli public, whose "search for the perfect baby" (Remennick, 2006) arguably takes on unique cultural meanings considering the effects of the Holocaust and the challenges of national survival in the face of on-going military conflicts (Chemke Juan & Steinberg, 1989).

Israeli pregnant women are routinely provided with risk assessments for DS through biochemical blood tests and nuchal translucency ultrasound. For women with screening results indicating a high risk for DS, and for women older than 35, follow-up diagnostic services are offered, including genetic counseling and amniocentesis, whose uptake is voluntary. In Israel, NIPS is marketed by private companies at the cost of a few thousands NIS, depending on the panel of conditions being tested (around 900$ for the "basic panel" which includes trisomies 13, 18, 21 and the sex chromosomes). In some cases, patients get partial reimbursement from their health insurance. A direct consequence of this is inequality in access. Including NIPS in the routine prenatal testing is currently being considered, probably as a first-tier test replacing the existing first and second trimester screens. If and when this happens, it will most likely lead to a dramatic increase in the consumption of NIPS.

1.1 | Attitudes of parents of children with DS toward prenatal screening

Since NIPS and prenatal diagnosis (PND) have the potential to influence the way society views DS and other genetic conditions, an important group of stakeholders are parents of children with DS. It is true for screening programs based on the traditional biochemical tests, and even more so for NIPS, as this test is more accurate in the detection of DS. The test was found to have a sensitivity of 99.4 and specificity of 99.9 for trisomy 21 (Mackie, Hemming, Allen, Morris, & Kilby, 2017). In recent years, various disability rights groups have protested against the routinization of NIPS, for example in the recent campaign titled "Don't Screen Us Out" (Ravitsky, 2017; Thomas & Rothman, 2016). However, little research has investigated whether the approaches of parents of children with DS toward prenatal screening in general and NIPS in particular are indeed in line with those of disability rights organizations that have launched such protests.

Whereas some advocacy groups clearly oppose prenatal testing for DS, parents of children with this condition cannot be regarded as a homogenous group (Inglis, Hippman, & Austin, 2012). Kellogg, Slattery, Hudgins, and Ormond (2014) found that while North American mothers said that NIPS could lead to an increase in pregnancy termination, increased social stigma and decreased availability of services for individuals with DS, over half of the participants agreed that they would consider using NIPS in the future. Dutch parents of children with DS perceived positively NIPS's accuracy and safety, and considered it to lead to less false reassurance and less unnecessary invasive procedures and to enable preparation for a child with special needs. Early testing was perceived as positive in terms of enabling easier coping with termination due to less maternal–fetal bonding while others said that this might lead to less thoughtful terminations that women could later regret (van Schendel et al., 2017). These mixed opinions illustrate the wide spectrum of attitudes toward prenatal screening among family members of children with DS (Bryant, Hewison, & Green, 2005; Skotko, Levine, & Goldstein, 2011). A common critique among these parents is what they describe as imbalanced and inaccurate information provided by medical professionals regarding DS (Kellogg et al., 2014; van Schendel et al., 2017). Balanced information is crucial for autonomous choice in both the decision to take the test and the decision concerning how to act based on its results (Asch & Wasserman, 2009; Kellogg et al., 2014; Skotko et al., 2011).

In some Western European countries, the uptake by the general population of screening tests for DS is high (74% in England, 84% in France and ≥90% in Denmark). However, a low uptake (<30%) is reported in the Netherlands (Crombag et al., 2016). Uptake is influenced by a variety of factors including religiosity, ethnicity, education/knowledge, and maternal age (Gitsels-van der Wal et al., 2014). Some women fail to understand that the test is optional (Al-Jader, Parry-Langdon, & Smith, 2000; Asch & Wasserman, 2009) while others are not informed of its existence or cannot afford it (Rowe, Puddicombe, Hockley, & Redshaw, 2008). Therefore, uptake rates, which are also often limited in terms of sample size and representativeness, do not necessarily reflect attitudes.

Prenatal diagnosis is generally embraced by Israeli women under the pretext of "genetic responsibility" (Raz & Schicktanz, 2009a, 2009b; Remennick, 2006). It is widely supported by professionals, the public and the legal system as preventing suffering rather than jeopardizing human rights (Hashiloni-Dolev, 2006, 2007). Israeli leaders of organizations for disability rights and support groups for people with genetic conditions were found to express support of PND as well as selective abortion, while at the same time stressing their commitment for already-born disabled individuals (Raz, 2004). In terms of actual uptake, Sher, Romano-Zelekha, Green, and Shohat (2003) found that 60.9% of Israeli Jewish women in their sample performed the triple test. The main reason provided by women for declining the test was religious or moral. Indeed, 96% of the secular women compared to only 6.7% of the ultra-religious women in that study performed the triple test. According to Zlotogora, Haklai, and Leventhal (2007), in four towns where almost all residents are ultraorthodox, 95.5% of the pregnancies diagnosed with DS were born alive. Among the Muslim population 81.3% of the pregnancies diagnosed with DS were live born. There is still no research regarding the uptake of, and attitudes toward, prenatal screening among family members of children with DS in Israel. The impetus for this research was the expanding use of the relatively new NIPS test in Israel. NIPS also became the trigger for re-viewing, through the eyes of our respondents, other forms of prenatal testing – diagnostic as well as screening tests – and their perceived implications.

2 | METHODS

Qualitative methodology was chosen as best suited for the study's emphasis on subjective meaning-making in social perspective (Flick, 2014).

2.1 | Participants

Semi-structured interviews were conducted during 2017–2019 following IRB approval. Four representatives of Israeli DS organizations (who are parents of children with DS themselves) were contacted by the first author. They agreed to participate and to aid in recruiting (through the social network of their organization) additional respondents. After interviewing these additional respondents, we used the snowballing method to recruit others. As the recruitment originated from several sources, i.e., four different DS organizations with varied socio-demographic and religious constituencies, bias was minimized. The inclusion criterion was being a parent of a child with DS. This included parents of children with DS whose pregnancies took place when NIPS was available for use as well as parents of older children with DS, who were born before the availability of the test. Some interviewees had subsequent pregnancies, when NIPS was already available. Respondents received a disclosure statement describing the study and signed an informed consent.

2.2 | Instrumentation and procedures

Data were collected through semi-structured interviews conducted by the first author with 20 parents of children with DS (see Table 1). Interviews were conducted in Hebrew over the telephone, lasted 70 min on average and were fully transcribed verbatim. The interview guide was constructed in order to learn how the respondents view the use and aims of NIPS, and how they relate to their own and their society's use (or rejection) of these technologies. Representatives of DS organizations were interviewed about their official position as well as their perspective as parents of children with DS. All respondents were categorized as users/non-users of NIPS, depending on whether in any of their pregnancies they have used NIPS. As a recruitment strategy for ensuring a variety of opinions, we tried to direct our snowballing so that we recruit both users and non-users of NIPS. The respondents were asked about using or not using NIPS – for some these were concrete questions and for others hypothetical, depending on age and family status. This study is part of a larger project for which the first author also interviewed eight Israeli health professionals specializing in Ob/Gyn and/or genetics concerning NIPS, as well as 24 Israeli women (without family history of DS) who were users or non-users of NIPS. This study focuses on the attitudes of parents of children with DS.

2.3 | Data analysis

In the analysis phase interview transcripts were organized and coded using grounded theory approach (Strauss & Corbin, 1990) and analyzed thematically. Interview collection was finalized when the research team agreed that thematic saturation was reached, meaning that no new topics were raised in subsequent interviews. Interview transcripts were translated by the authors from Hebrew to English and pseudonyms were used with quotes. The transcripts were analyzed thematically to uncover discursive themes and categories of themes recurring within and across groups of respondents, for example groups of users and non-users, or religious and secular respondents (Denzin & Lincoln, 1994). Following a review of the relevant literature, preliminary codes included arguments pro and con NIPS as well as views on life with DS. Additional themes, especially concerning the concrete social context of decision-making about NIPS and PND, were inductively gleaned from the transcripts. The research team did the coding together on the first few interview transcripts, discussing the relevance of the themes and agreeing on needed modifications and reclassifications. The first author then continued with the coding, discussing new findings as they appeared and their relationships to the codes in team meetings, where agreements were reached to prevent the potential bias of a single rater and using inter-rater reliability to increase the validity of the results. The quotes used were selected to represent the range of opinions among the respondents.

TABLE 1 Characteristics of participants

Characteristic	*N*, % (*n* = 20)
Female	19 [95%]
Male	1 [5%]
Mean age, years (range)	41.9 (24–74)
Religiosity	
Secular	7 [35%]
Modern religious	2 [10%]
Ultra-Orthodox	11 [55%]
Mean number of children (range)	4.75 (1–13)
Mean age of child with DS, years (range)	11.5 (2–37)
NIPS users	2 (in pregnancy with DS, false negative result) 1 (in later pregnancy, normal result)
Representatives of DS organizations	4

Note: Since most participants have multiple children and reported varied information regarding testing for each of their pregnancies, summarizing other data in the table was not possible.
Abbreviations: DS, Down syndrome; NIPS, non-invasive prenatal screening.

3 | FINDINGS

All respondents were Caucasians and Jews, probably as a result of our snowballing method. The interviewees presented a wide spectrum of attitudes toward screening for DS and the rationale behind the utilization or rejection of such screening, as well as its potential outcomes. Their views are presented under the following categories: (a) arguments pro and con NIPS; (b) the social context of decision-making about NIPS and PND; and (c) views on life with DS and termination of pregnancies on that ground.

3.1 | Arguments pro and con NIPS

The main arguments pro-NIPS emphasized the notions of parental autonomy as well as safe and early testing that allows for early termination of affected pregnancies or to prepare for a child with special needs. The central arguments against NIPS stressed it as being harmful for bonding, stressful, costly and providing false reassurance.

3.2 | Arguments in support of NIPS

The following quote demonstrates the importance of parental autonomy for participants:

> People should go privately to their doctor or geneticist, consult and hear all about the risks, see if it fits them or not. [Bosmat, age >50, secular, chairwoman of DS advocacy organization, mother of 3. Eldest with DS]

A main point raised by parents, both users and non-users of the test, can be presented as NIPS being "only a blood test, without risks", especially when compared to what many saw as "risky" amniocentesis. This was described by users as a reason for choosing NIPS over other forms of invasive tests:

> We didn't want to face the unnecessary risks of amniocentesis and we contemplated what to do. [...] We realized this was a blood test without risks, so we decided to do this blood test and spare ourselves this issue of amniocentesis. [Efrat, age 38, mother of 2. Youngest with DS. Had False-Negative result in NIPS in this pregnancy]

Non-users also described this as a likely reason for others (or even themselves in future pregnancies) to use NIPS. Feasibility of early testing was another characteristic mentioned by participants. This was considered a positive feature of the test, when termination is a valid option for the pregnant woman:

> If it is done early in pregnancy and has no risk for the embryo, for an abortion or for contaminations or anything, then why not do something... both in an early week and without the dangers? [Noa, age 43, secular, mother of 3. Eldest with DS]

Even some ultra-orthodox respondents saw early testing as valuable:

> Those blood tests are done very early, so it could be that in that stage, if you take this test and there is an abnormal finding with high reliability, [...] it could be that we would go to the Rabbi... It could be that the Rabbi says yes. But it is because of the early stage of pregnancy that rabbis would allow terminations. But not for sure and for any condition. [Tzipi, age 35, Ultraorthodox, mother of 4. Youngest with DS]

However, the majority of ultraorthodox participants did not perceive early testing as an advantage, since they would not consider termination regardless of timing:

> Time plays a role if you are thinking of doing something about it. But not for someone who won't do anything with it [with the results]. [Hanna, age 55, ultraorthodox, mother of 13, youngest with DS]

Some parents, including from the ultraorthodox community, pointed out that testing enables to prepare for a child with special needs:

> I think there is great benefit in testing during pregnancy, in order to know... For instance, a blood test that tells you if there is a genetic problem with the fetus, even if it isn't in order to terminate but simply to know and prepare accordingly. [Alona, age 33, secular, mother of 2, youngest with DS. Pregnant when interviewed]

3.3 | Arguments against NIPS

Some of our respondents spoke of testing as a great source of stress:

> ...I am happy I didn't know during pregnancy. Very happy. Because I think it would have made it a difficult pregnancy. You don't know what's going to happen, what is going to come out. [...] It seems to me like a very unpleasant experience. Neither to you nor to the fetus and the fetus feels it. It feels if it is wanted or not. I think the fact that my child felt all through the pregnancy that it was wanted, it is meaningful for the rest of his life. [Adva, age 34, ultraorthodox, mother of 4. Third with DS]

> We think that a mother who knows during pregnancy that there is a high risk for DS – many times it can bring to a state of great emotional stress during the entire pregnancy, because the imagination works, and the emotional stress works extra time. It harms both the mother and the fetus. And we know of mothers

> who remained in this emotional crisis after giving birth because of this thing. [Shmuel, ultraorthodox, head of a DS organization, father of 7. The second with DS]

Some women further pointed out that knowing the diagnosis in advance is harmful for bonding and were grateful for not knowing prior to giving birth:

> If I had known this was a child with DS I would probably break down much more. I am among these women who think it was better not to know [during pregnancy]. When it happened, we dealt with it. If I were carrying it the whole time, it would have looked much bigger and much more frightening. Burdening much more. [When you don't know] you already bond with your baby and then later they can say whatever they say. [Hodaya, age 24, ultraorthodox, mother of 2. Eldest with DS]

Some respondents argued against the false reassurance they associated with NIPS. They described, in retrospect, how they failed to fully acknowledge the limitations of the test, for instance the fact it was a screening test rather than diagnostic, and the scope of conditions being tested in comparison to amniocentesis. Consequently, the test was described as providing false reassurance:

> …The illusion that NIPS gives is that even though you should know it is a screening test, since it is supposedly so precise, I think people look at it as if it is a diagnostic test. [Efrat, age 38, mother of 2. Youngest with DS. Had FN result in NIPS in this pregnancy]

> The whole time I was thinking this was a replacement for amniocentesis, that if you are doing the test and the result is normal, you don't need to go through amniocentesis. [Noga, age 40, secular, mother of 2. Youngest with DS. Had FN result in NIPS in this pregnancy]

Another argument against taking the test was its financial cost. Yet this argument was not against NIPS per se but against lack of access.

3.4 | The social context of decision-making about NIPS

The pros and cons did not exist in an individual vacuum. Rather, decision-making was described as a process influenced by external factors that included societal support and tolerance (or lack of it):

> In the very non-ideal world that we constantly fight against, genetic testing isn't a simple yes or no question. There is social pressure in Israel not to give birth to children with problems. Of course. There is social pressure: "This isn't going to happen to me… I am going to have a perfect baby." [Bosmat, age >50, secular, chairwoman of DS advocacy organization, mother of 3. Eldest with DS]

> I feel that Israeli society is much less accepting of disabilities. There is no legitimacy for disability. […] Doctors, caregivers, everybody who sees a secular woman with a child with DS, they all wonder how come it happened. [Efrat, age 38, mother of 2. Youngest with DS. Had FN result in NIPS in this pregnancy]

Respondents also spoke about NIPS and PND in the broader context of disability and social responsibility:

> I don't support publishing articles that say 'There are no more births of children with DS in Iceland, they solved the problem. Only in Israel children are still born with this severe syndrome.' This is very bad. It is based on ignorance. And it doesn't provide the real data. […] I oppose what the doctors say. I oppose the policy of the ministry of education to separate these children." [Bosmat, age >50, secular, chairwoman of DS advocacy organization, mother of 3. Eldest with DS]

The following quote demonstrates how, according to another DS advocacy organization activist, parental autonomy is undermined by imbalanced information regarding DS:

> I always tell women who consult with me that I don't decide regarding their womb. I want to lower the fear to the minimum and explain what a child with DS is like. I can understand it because they are the product of society, of an ill social-medical discourse. They make us fear these children. We are the product of our society. I can understand those who abandon [the babies] in the hospitals. It is not their fault. They are the product of this sick medical and social discourse to the point that they abandon their babies." [Sivan, age 48, secular, DS advocacy organization, mother of 4. Youngest with DS]

Some respondents spoke of the tension between religious and medical expectations (cf. Ivry, Teman, & Frumkin, 2011):

> Let's say the only ones that tried a bit to convince me to terminate were the doctors. The doctors who pushed me kind of hard. But from my own surroundings I had a strong support. From my family and friends. I still do. […] I can tell you that each time I came to see my doctor, he would simply tell me 'the light is red, and you continue driving'. [Ilanit, age 35, modern religious, mother of 3. Eldest with DS. Abnormal findings during pregnancy pointed to high risk of DS]

> There are doctors who pressured more and doctors who pressured less [into conducting prenatal tests]. There were doctors who have been working with the ultraorthodox sector for some time and gave up on this and understood that there is no point in pressuring. It is the newer doctors who pressured and said 'Why not? It is very important' and tried to convince in that direction. But the senior doctors gave up on this. They know this public so they don't pressure anymore. [Miri, ultraorthodox, mother of 7. 4th with DS]

On the other hand, some religious women described the professionals they encountered as attentive and respectful toward their decisions:

> These doctors know the ultraorthodox sector. They are familiar with the avoidance of testing. My doctor told me "You don't want that? Then no". No pressure at all. She offered me everything that existed at the time and I told her the necessary things I wanted and that's it. She didn't pressure me at all. [...] There are respectful doctors. [Tzipi, age 35, Ultraorthodox, mother of 4. Youngest with DS]

Some respondents claimed that PND continues to carry a discriminating message to those with a disability:

> It sends a message. And the message I get from it is a painful one. It's like we are trying to purify the world and build it only with people who are healthy and successful. And in fact, our society is composed of many, many different kinds of people, who deserve a place just like us. [Hanna, age 55, ultraorthodox, mother of 13, youngest with DS]

> These tests create, to begin with, a discourse that when these children are born it is: 'Oh my god, why were they born?' [Sivan, age 48, secular, DS advocacy organization, mother of 4. Youngest with DS]

In addition to social pressures, some parents described the uptake of tests during pregnancy as a routine – something that is done automatically, regardless of the actual reason for which these tests are offered. That is, they performed tests without thinking of the possibility that these tests might detect an abnormality and without considering the social and ethical aspects of life with disability as a basis for the decision-making process regarding testing.

> Interviewer: Were social and financial aspects of life with a disabled child among your considerations when deciding about testing in pregnancy?
>
> Interviewee: No. Not at all. I didn't even think about that. It wasn't on my mind in any way. I didn't know any baby or child with special needs and it really didn't occupy my thoughts whatsoever. [Alona, age 33, secular, mother of 2, youngest with DS. Pregnant when interviewed]

> Disability? No, we didn't think about that at all. From our perspective, during pregnancy you do tests and if everything is normal – the pregnancy is normal. We didn't think that something might happen. No, we didn't think about it. Absolutely not." [Noa, age 43, secular, mother of 3. Eldest with DS]

In contrast with those who spoke about an automatic routine, others said that not only was testing enhancing autonomy, it was also perceived (by themselves and by society) as a form of responsibility, as the following quote demonstrates:

> The message I receive both verbally and non-verbally is 'how could you be so irresponsible and not do amniocentesis and how did it happen that you have a child with DS?' As if he has no right to exist in this world." [Efrat, age 38, mother of 2. Youngest with DS. Had FN result in NIPS in this pregnancy]

The notion of testing as displaying responsibility was related by respondents to realizing that for many, testing was interpreted as a means to minimize suffering – be it the suffering of the child or that of the parents.

> Of course, people should test. Do whatever you can in order to have a healthy child. Because later, the child suffers and so do the parents. [Noa, age 43, secular, mother of 3. Eldest with DS]

Positive experiences of disability were noted especially amongst religious respondents:

> Whenever I'm walking with my son I think that I'm walking with a king. [...] I never experienced inappropriate behavior or that he was rejected or people distancing themselves from him. Not at all." [Hanna, age 55, ultraorthodox, mother of 13, youngest with DS]

For many parents, considerations of PND were therefore part of a dynamic social experience which was part of their actual course of life. Family history was another important source of impact. Some stated that although they manage life with DS, they perceive testing as crucial since they would not be able to deal with *another* child with special needs:

> This time it was clear to us that we will perform amniocentesis and CMA [chromosomal microarray analysis]. We even considered exome testing. It will be

> very difficult for us to deal with another child, not with DS but with special needs. And I saw and heard of so many cases of severe syndromes and difficult cases that I don't want to deal with. My sweet boy is enough with all his issues that I need to deal with. [...] I cannot settle for screening tests now. [Alona, age 33, secular, mother of 2, youngest with DS. Pregnant when interviewed]

> ...Eliminating the existence of another fetus is like eliminating the existence of our living child. It is very complicated. [...] I believe we will think it over very deeply, according to our life today, which is very different from our life then [before having a child with special needs]. [Ilanit, age 35, orthodox, mother of 3. Eldest with DS. Abnormal findings during pregnancy pointed to high risk of DS]

3.5 | Views on life with DS and termination of pregnancies on that ground

In the broader context for decision-making regarding NIPS, participants expressed their own views regarding DS, as well as the views they perceived to be held by society on this matter. Some parents spoke about the personal change they went through regarding their perception of life with disability. A strong and common theme was that life with DS is a constant fight against authorities and social stigmas:

> If they made it easier on us… I mean, if it were easy to raise such a child… Because in this country it isn't only a problem to know you deserve something. Now that you know you deserve it, go fight for it. It isn't easy. And not many parents have the energy to fight all the time, and you do need to fight all the time. [Dvora, age >60, secular, chairwoman of DS advocacy organization, Youngest child with DS]

> A mother who gives birth to a child with DS turns into a warrior. We must beg for everything we need. Nothing is taken for granted. Many, many fights, even for things that should obviously be given. [Hanna, age 55, ultraorthodox, mother of 13, youngest with DS]

This perception of fight regarding many aspects of life with disability is central to understanding how parents of children with DS refer with open-mindedness to the decision of others to terminate a pregnancy of a fetus diagnosed with DS. It also reflects on their own hypothetical decision, had they known during pregnancy about the diagnosis. However, views regarding termination ranged from full support to outright rejection. On the one end were those who would not consider this for themselves and viewed termination as an atrocity. Such opposition to termination of affected pregnancies was strongly related to religious views, with respondents expressing the view that the fetus has a soul and there is a reason why God gave this child to certain parents:

> The first thing I think about is that it should be considered on a case by case basis and you never know why a person would do that. I try to judge them favorably, but I think it is a crime. Clear cut. Killing a soul is unthinkable. [Tova, age 28, ultraorthodox, mother of 3. Youngest with DS]

> God chose us as parents to take care of him [our child]. Probably only we can help him and nobody else can, and this is our duty as parents [...] I was chosen to raise this child who has a higher soul. [Miri, ultraorthodox, mother of 7. 4th with DS]

On the other end were those who understood the decision to abort, including a few who claimed that they would have decided the same:

> I have no agenda saying that one should not terminate [a pregnancy diagnosed with DS]. Not every family must have this challenge in life and it is absolutely legitimate to say, 'Thank you very much, I'd rather try and get pregnant again later, and I give up on this pregnancy'. [...] I don't have a principle saying 'there must be people with DS in this world'. [Noga, age 40, secular, mother of 2. Youngest with DS. Had FN result in NIPS in this pregnancy]

Some claimed that at the time of being pregnant, while still having no personal knowledge of life with DS, they would have terminated, whereas now they think differently:

> I don't regret for one moment not having done it [amniocentesis], because… It is hard to say if we would have terminated or not, but my husband says we surely would have. …. I know there are 99% chance we would have terminated, so I'm glad we didn't. [Alona, age 33, secular, mother of 2, youngest with DS. Pregnant when interviewed]

Many parents narrated a personal change concerning their acceptance of having a child with DS. For some it was a gradual process, while for others a quick one:

> I told two neighbors in our village: "Listen, I know it is DS. We chose this. We wanted this. This was our decision. I want you to spread this message after I give birth". It was very important for me that people don't pity us. [Tirza, age 61, modern religious, had a

> diagnosis of DS through amniocentesis following abnormal findings on U.S, mother of 12. Youngest with DS]

> Slowly I started feeling a change in my emotions, in my acceptance of DS. [...] If once I said 'I don't want this in my life at all', today I don't say this anymore. [...] Today I think that when he grows up he will be able to do everything like any other person. I'm very optimistic about his future, which is something I wasn't a year ago. [Efrat, age 38, mother of 2. Youngest with DS. Had FN result in NIPS in his pregnancy]

For some parents such acceptance brought about a realization that disability can add value to society:

> They have a meaningful, joyful life. I cannot say their life isn't challenging, but they challenge their environment more than themselves. And perhaps our environment deserves to be challenged and not monotonous. [Tzipi, age 35, Ultraorthodox, mother of 4. Youngest with DS]

> The minute such tests do not exist, people will understand that these children should come into the world and we, as a society that wants to be humane, need to know them and understand that they came to heal it. Period. [Sivan, age 48, secular, DS advocacy organization, mother of 4. Youngest with DS]

Some parents pointed out the ignorance surrounding DS. They claimed that the public does not know what DS is (just as they did not, before having such a child themselves). They speculated that public ignorance regarding DS leads parents to decide to terminate diagnosed pregnancies:

> I just know that most people who receive the results will opt for termination, whether or not they can deal with it, without thinking about it thoroughly. Without really knowing what it means. If I want to tell you where the problem is – it is that people don't really know what DS is and they live by stereotypes and prejudice. [Ilanit, age 35, orthodox, mother of 3. Eldest with DS. Abnormal findings during pregnancy pointed to high risk of DS]

In contrast, other parents claimed that even with their current experience, they believe such pregnancies should not be brought to term:

> I think life isn't easy and preventive medicine is the right medicine. Whatever you can prevent – go ahead and prevent. So do all the tests you can in order to prevent any trouble in the future. [Dvora, age >60, secular, chairwoman of DS advocacy organization, Youngest child with DS]

Heterogenous views were also expressed regarding prevention/support. While supporting children with DS and their families was seen as essential, some parents also stated that prevention is a goal too, given the advanced technologies of PND:

> I don't think that nowadays, when technology is so advanced, and you can detect so many things during pregnancy – I don't think children with disabilities, including Down syndrome, should be born. If they are already born, if it was not found during pregnancy – of course they should be given everything possible. But why… Why let these kids be born if… Why? Why do people do these tests in the first place? [Noa, age 43, secular, mother of 3. Eldest with DS]

> Once a person with DS is born, this person must be taken care of. If the question is whether to invest in them or invest in amniocentesis or the blood tests - then definitely in them! [Bosmat, age >50, secular, chairwoman of DS advocacy organization, mother of 3. Eldest with DS]

4 | DISCUSSION

The interviewees presented a wide spectrum of attitudes toward screening for DS in general and NIPS in particular. The main arguments pro-NIPS emphasized are the notions of parental autonomy as well as safe and early testing that allows preparing for a child with special needs or experiencing a less traumatizing termination due to an earlier pregnancy. Central arguments against NIPS stressed that it was harmful for bonding, stressful, costly, providing false reassurance and carrying a discriminating message. The combination of pro and con arguments expressed by our respondents resembles to a large extent the mixed attitudes concerning NIPS of pregnant women from the general public in other countries (Lewis, Silcock, & Chitty, 2013; van Schendel et al., 2014). In a similar manner to our respondents, pregnant women elsewhere regarded NIPS favorably compared to other PND methods, because of its safety, higher accuracy compared to other screening tests and early stage of usage. This was perceived as an advantage both for those who want to prepare for DS and those who seek termination. Previous studies also report fears from routinization of the test that would compromise true procreative autonomy as well as the test's potential impact on the disabled community (Lewis et al., 2013; van Schendel et al., 2014).

Despite overall similarities between arguments raised by pregnant women in general and the arguments raised by our respondents, the personal experience of raising a child with DS has a strong yet contingent effect on parents' views regarding prenatal testing and life with disability. When 'zooming in,' different consequences of such life experiences can be observed. For some, this personal experience strengthened previous thoughts on PND and disability. For others, their life circumstances changed their views. Some respondents described how their personal experience taught them the value of PND in preventing suffering. For others, the experience of having a child with DS made them more aware of the importance of preparing for a challenging life. Others reported a change in perceiving life with disability – a shift from rejection and fear to acceptance and optimism and even to the realization of the contribution of such a condition to society. Having a child with DS had a paramount effect which was nevertheless different for each respondent. While most of our respondents were non-users of NIPS, this did not reflect a single attitude but rather a plurality of (sometimes conflicting) reasons, from religious ideologies to rejecting NIPS in favor of more thorough PND technologies such as amniocentesis and chromosomal microarray analysis (CMA).

In a similar manner, parents of children with DS in other countries also had a wide spectrum of views concerning NIPS and screening for DS (Bryant et al., 2005; Inglis et al., 2012; van Schendel et al., 2017; Skotko et al., 2011). While some recognized the advantages of NIPS, it was seen by others as having potentially negative implications like increased social stigma and decreased availability of services for individuals with DS, as well as increasing the pressure on women to test. Notwithstanding their awareness of the negative potential of the test, many reported that they would personally consider using NIPS in the future (Kellogg et al., 2014).

While illustrating the large heterogeneity of views concerning NIPS and PND amongst parents of children with DS, our findings also point to a strong critique of the imbalanced information provided by professionals regarding DS (Kellogg et al., 2014; van Schendel et al., 2017). NIPS and PND in general were seen by some of our respondents as sending a discriminating message, in line with the public ignorance surrounding DS. As parents of children with DS, our respondents stressed the importance of such knowledge for true informed choice and decision-making. To respect the variety in values and preferences of parents, as illustrated by our respondents too, balanced information is essential for enabling informed choice (Asch & Wasserman, 2009; Kellogg et al., 2014; Skotko et al., 2011).

4.1 | Parents' views in cultural context

Previous studies promoted the popular view of the Israeli public as characterized by a very high uptake and support of PND, considered as a medical priority and a moral duty by health professionals and the public (Hashiloni-Dolev, 2007; Raz & Schicktanz, 2009a, 2009b; Remennick, 2006; Zlotogora, Grotto, Kaliner, & Gamzu, 2015). However, empirical studies demonstrate PND uptake of about 60% in the overall Israeli population, an uptake which increases considerably amongst the secular Israeli public (Gofin, Adler, & Palti, 2004; Grinshpun-Cohen, Miron-Shatz, Berkenstet, & Pras, 2015; Sher et al., 2003). Yet even with the availability and accessibility of a national program for "the detection of Down Syndrome" (which includes first and second trimester screening, and free amniocentesis for women older than 35 years), more than 50% of DS cases in Israel are brought to term—with most of the affected babies born in Orthodox Jewish and Muslim Arab communities where termination of pregnancy is banned by religion (Zlotogora et al., 2007).

It is intriguing to consider our respondents' views in the multicultural context of Israeli society, and how they reflect as well as problematize broader cultural views found in previous studies about PND in Israel. At first glance it may seem that the personal decision-making of our respondents reflects a polarity of religiosity versus secularism. This is true to some extent. However, variety of views were observed in both groups. While many religious respondents indeed expressed opposition to testing and termination of affected pregnancies, their views were heterogenous, also reflecting different streams of religious lifestyle. Whereas most of the secular parents emphasized the need for parental autonomy and saw NIPS as enhancing such autonomy, some also pointed out that autonomy is compromised due to social pressures coming from medical professionals as well as from family members and society at large. Indeed, far less children with DS are born in the secular population in Israel compared to the religious one, making them and their families more exceptional in their environment. Therefore, the decision to test during pregnancy or to bring a pregnancy of a fetus with DS to term falls beyond the considerations of the physical/cognitive disability of the child and the financial implications of raising them. This decision is closely linked to the degree of exceptionality of such a child in their surrounding, which is far more extreme in the secular Israeli environment.

Some parents described testing during pregnancy as a routine ("pregnancy is checkups (*bdikot*, in Hebrew)"), done automatically in accordance with the recommendations of the medical system, without considering the meaning of life with the disability that is being tested. This non-reflective uptake matches the concern raised by parents in other studies about a possible 'routinization' of prenatal screening. The ease and safety of NIPS thus could lead to feeling a reduced need to reflect on the pros and cons of prenatal screening (Van Schendel et al., 2014). We found that for parents of children with DS, the justification underlying both routinization and responsibilization was linked to the perception of a constant fight and lack of societal support regarding many aspects of life with DS. All parents acknowledged that support for individuals with DS is essential, and some stated that prevention (through termination) is a goal too. Our respondents related this seemingly contradicting embracing of both prevention and care to the lack of societal support and their view of suffering. These findings portray the difference between Israeli DS advocacy and DS advocacy organizations in Europe that protest the use of PND and termination of affected pregnancies. The emphasis on suffering and lack of societal support may explain why representatives of DS organizations support NIPS rather than protest it (Raz, 2004).

4.2 | Study limitations

Although the size of our group of respondents was small, thus lacking the ability to generalize from it, this is a common limitation in qualitative studies that instead allow for in-depth exploratory findings and insights. To the best of our knowledge, this is the first study concerning attitudes of Israeli parents of children with DS toward NIPS. A limitation of the study is that while we did manage to recruit both secular and religious respondents stratified by level of education and number of children, other Israeli ethnic groups (such as Israeli-Arabs) are not represented. In addition, most of our respondents were women. Future studies should include larger samples of males and other ethnic groups.

5 | CONCLUSIONS AND PRACTICE IMPLICATIONS

Israeli parents of children with DS have a wide spectrum of attitudes toward NIPS. Realizing the various concerns and needs of this heterogeneous group of clients, as depicted in this study, is beneficial for genetic counselors in providing more comprehensive and culturally tailored service. A strong practice implication emanating from our findings is that in order to ensure true parental autonomy, there is a need for clear pre-test counseling, without which testing might be done automatically implying false reassurance for some. The same goes for post-test counselling, in case of abnormal test results. This requires educating all professionals who encounter such parents, to be able to present unbiased, up-to-date information that is free of pressures. Given the heterogeneity of views presented among both secular and religious participants, it is important that health professionals do not base their counseling on pre-assumptions since such 'cultural profiling' may be misleading. There are too many personal and social contingencies creating a variety of attitudes beyond religiosity or secularism. Counseling should hence start with listening to the client. Moreover, knowledge regarding the status of the fetus (in terms of having certain genetic disorders) is not beneficial to all parents. It is therefore also important to explain to patients about the option of opting out and implementing it in guidelines and laws.

AUTHORS CONTRIBUTIONS

All authors have substantial contributions to the conception and design of the work, with the first author having carried the acquisition of data for the work as part of her doctoral studies.

ACKNOWLEDGMENTS

We thank all the respondents who participated in this study for sharing their experiences and insights. We are grateful for the funding provided by the German Research Foundation (DFG) as part of the comparative research project "Meanings and Practices of Prenatal Genetics in Germany and Israel" (PreGGI) #RE 2951/3-1. We thank our project partners, Christoph Rehmann-Sutter, Christina Schües, Stefan Reinsch and Hannes Foth, as well as the two anonymous reviewers and the Editor, for their helpful suggestions concerning this study.

COMPLIANCE WITH ETHICAL STANDARDS

Conflict of interests

The authors declare no conflict of interests.

Human studies and informed consent

Informed consent was obtained from all participants for being included in the study.

Animal studies

No animal studies were carried out by the authors for this article.

REFERENCES

Al-Jader, L., Parry-Langdon, N., & Smith, R. (2000). Survey of attitudes of pregnant women towards Down syndrome screening. *Prenatal Diagnosis*, *20*, 23–29. https://doi.org/10.1002/(SICI)1097-0223(200001)20:1<23:AID-PD746>3.0.CO;2-T

Asch, A., & Wasserman, D. (2009). Informed consent and prenatal testing: The Kennedy-Brownback act. *American Medical Association Journal of Ethics*, *11*(9), 721–724. https://doi.org/10.1001/virtualmentor.2009.11.9.oped1-0909

Bryant, L., Hewison, J., & Green, J. (2005). Attitudes towards prenatal diagnosis and termination in women who have a sibling with Down's syndrome. *Journal of Reproductive and Infant Psychology*, *23*, 181–198. https://doi.org/10.1080/02646830500129214

Chemke Juan, J., & Steinberg, A. (1989). Ethics and medical genetics in Israel. In D. C. Wertz, & J. C. Fletcher (Eds.), *Ethics and human genetics: A cross cultural perspective* (pp. 271–284). Berlin, UK: Springer-Verlag.

Congregation for the Doctrine of the Faith: Instruction Dignitas Personae. On Certain Bioethical Questions. (2008). Vatican

Crombag, N., Boeije, H., Iedema-Kuiper, R., Schielen, P., Visser, G., & Bensing, J. (2016). Reasons for accepting or declining Down syndrome screening in Dutch prospective mothers within the context of national policy and healthcare system characteristics: A qualitative study. *BMC Pregnancy and Childbirth*, *16*(1), 121. https://doi.org/10.1186/s12884-016-0910-3

Denzin, N. K., & Lincoln, Y. (1994). *Handbook of qualitative research*. Thousand Oaks, CA: Sage.

Devers, P., Cronister, A., Ormond, K., Facio, F., Brasington, C., & Flodman, P. (2013). Noninvasive prenatal testing/noninvasive prenatal diagnosis: The position of the National Society of Genetic Counselors. *Journal of Genetic Counseling*, *22*(3), 291–295. https://doi.org/10.1007/s10897-012-9564-0

Dondorp, W., De Wert, G., Bombard, Y., Bianchi, D., Bergmann, C., Borry, P., … Cornel, M. (2015). Non-invasive prenatal testing for aneuploidy and beyond: Challenges of responsible innovation in prenatal screening. *European Journal of Human Genetics*, *23*, 1592.

Flick, U. (2014). *An introduction to qualitative research*. London, UK: SAGE.

Franklin, S., & Roberts, C. (2006). *Born and made, an ethnography of preimplantation genetic diagnosis*. Princeton, NJ: Princeton University Press.

Gitsels-Van Der Wal, J., Verhoeven, P., Manniën, J., Martin, L., Reinders, H., Spelten, E., … Nl, J. (2014). Factors affecting the uptake of prenatal screening tests for congenital anomalies; a multicentre prospective cohort study. *BMC Pregnancy and Childbirth*, *14*, 264. https://doi.org/10.1186/1471-2393-14-264

Gofin, R., Adler, B., & Palti, H. (2004). Screening tests in prenatal care: A national study in Israel. *Israel Medical Association Journal*, *6*, 535–539.

Grinshpun-Cohen, J., Miron-Shatz, T., Berkenstet, M., & Pras, E. (2015). The limited effect of information on Israeli pregnant women at advanced maternal age who decide to undergo amniocentesis. *Israel Journal of Health Policy Research*, *4*(1), 23. https://doi.org/10.1186/s13584-015-0019-6

Habermas, J. (2003). *The future of human nature*. Cambridge, UK: Polity Press.

Hashiloni-Dolev, Y. (2006). Between mothers, fetuses and society: Reproductive genetics in the Israeli-Jewish context. *Nashim: A Journal of Jewish Women's Studies & Gender Issues*, *12*, 129–150.

Hashiloni-Dolev, Y. (2007). *A Life (Un)Worthy of Living. Reproductive Genetics in Israel and Germany*. Dordrecht, the Netherlands: Springer.

Inglis, A., Hippman, C., & Austin, J. (2012). Prenatal testing for Down syndrome: The perspectives of parents of individuals with Down syndrome. *American Journal of Medical Genetics Part A*, *158A*, 743–750. https://doi.org/10.1002/ajmg.a.35238

Ivry, T., Teman, E., & Frumkin, A. (2011). God-sent ordeals and their discontents: Ultra-orthodox Jewish women negotiate prenatal testing. *Social Science & Medicine*, *72*(9), 1527–1533. https://doi.org/10.1016/j.socscimed.2011.03.007

Kellogg, G., Slattery, L., Hudgins, L., & Ormond, K. (2014). Attitudes of mothers of children with down syndrome towards noninvasive prenatal testing. *Journal of Genetic Counseling*, *23*(5), 805–813. https://doi.org/10.1007/s10897-014-9694-7

Leonard, S. (2017). Current concepts in noninvasive prenatal screening (NIPS). *Journal of Fetal Medicine*, *4*(3), 125–130. https://doi.org/10.1007/s40556-017-0122-6

Lewis, C., Silcock, C., & Chitty, L. S. (2013). Non-invasive prenatal testing for Down's syndrome: Pregnant women's views and likely uptake. *Public Health Genomics*, *16*(5), 223–232. https://doi.org/10.1159/000353523

Mackie, F. L., Hemming, K., Allen, S., Morris, R. K., & Kilby, M. D. (2017). The accuracy of cell-free fetal DNA-based non-invasive prenatal testing in singleton pregnancies: A systematic review and bivariate meta-analysis. *BJOG: An International Journal of Obstetrics & Gynaecology*, *124*(1), 32–46. https://doi.org/10.1111/1471-0528.14050

Minear, M. A., Lewis, C., Pradhan, S., & Chandrasekharan, S. (2015). Global perspectives on clinical adoption of NIPT. *Prenatal Diagnosis*, *35*(10), 959–967. https://doi.org/10.1002/pd.4637

Ravitsky, V. (2017). The shifting landscape of prenatal testing: Between reproductive autonomy and public health. *Hastings Center Report*, *47*, S34–S40. https://doi.org/10.1002/hast.793

Raz, A. (2004). Important to test, important to support: Attitudes toward disability rights and prenatal diagnosis among leaders of support groups for genetic disorders in Israel. *Social Science and Medicine*, *59*(9), 1857–1866. https://doi.org/10.1016/j.socscimed.2004.02.016

Raz, A., & Schicktanz, S. (2009a). Diversity and uniformity in genetic responsibility: Moral attitudes of patients, relatives and lay people in Germany and Israel. *Medicine, Health Care, and Philosophy*, *12*(4), 433–442. https://doi.org/10.1007/s11019-009-9215-x

Raz, A., & Schicktanz, S. (2009b). Lay perceptions of genetic testing in Germany and Israel: The interplay of national culture and individual experience. *New Genetics and Society*, *28*(4), 401–414. https://doi.org/10.1080/14636770903314533

Remennick, L. (2006). The quest for the perfect baby: Why do Israeli women seek prenatal genetic testing? *Sociology of Health and Illness*, *28*, 21–53. https://doi.org/10.1111/j.1467-9566.2006.00481.x

Rowe, R., Puddicombe, D., Hockley, C., & Redshaw, M. (2008). Offer and uptake of prenatal screening for Down syndrome in women from different social and ethnic backgrounds. *Prenatal Diagnosis*, *28*, 1245–1250. https://doi.org/10.1002/pd.2125

Sandel, M. (2007). *The case against perfection: Ethics in the age of genetic engineering*. Cambridge, MA: Harvard University Press.

Savulescu, J. (2001). Procreative beneficence: Why we should select the best children. *Bioethics*, *15*(5–6), 413–426. https://doi.org/10.1111/1467-8519.00251

Schües, C. (2014). Improving deficiencies? Historical, anthropological, and ethical aspects of the human condition. In M. Eilers, K. Grüber, & C. Rehmann-Sutter (Eds.), *The human enhancement debate and disability. New bodies for a better life* (pp. 38–63). Hampshire, UK: Palgrave.

Sher, C., Romano-Zelekha, O., Green, M., & Shohat, T. (2003). Factors affecting performance of prenatal genetic testing by Israeli Jewish women. *American Journal of Medical Genetics*, *120A*(3), 418–422. https://doi.org/10.1002/ajmg.a.20047

Skotko, B. G., Levine, S. P., & Goldstein, R. (2011). Having a son or daughter with Down syndrome: Perspectives from mothers and fathers. *American Journal of Medical Genetics Part A*, *155*(10), 2335–2347. https://doi.org/10.1002/ajmg.a.34293

Strauss, A. C., & Corbin, J. M. (1990). *Basics of qualitative research. Grounded theory procedures and techniques*. London, UK: SAGE.

Thomas, G., & Rothman, B. (2016). Keeping the backdoor to eugenics ajar?: Disability and the future of prenatal screening. *American Medical Association Journal of Ethics*, *18*(4), 406–415. https://doi.org/10.1001/journalofethics.2016.18.4.stas1-1604

van Schendel, R., Kater-Kuipers, A., van Vliet-Lachotzki, E., Dondorp, W., Cornel, M., & Henneman, L. (2017). What do parents of children with Down syndrome think about non-invasive prenatal testing (NIPT)? *Journal of Genetic Counseling*, *26*, 529–531. https://doi.org/10.1007/s10897-016-0012-4

van Schendel, R. V., Kleinveld, J. H., Dondorp, W. J., Pajkrt, E., Timmermans, D. R. M., Holtkamp, K. C. A., … Henneman, L. (2014). Attitudes of pregnant women and male partners towards non-invasive prenatal testing and widening the scope of prenatal screening. *European Journal of Human Genetics*, *22*(12), 1345. https://doi.org/10.1038/ejhg.2014.32

Zlotogora, J., Grotto, I., Kaliner, E., & Gamzu, R. (2015). The Israeli national population program of genetic carrier screening for reproductive purposes. *Genetics in Medicine*, *18*(2), 203. https://doi.org/10.1038/gim.2015.55

Zlotogora, J., Haklai, Z., & Leventhal, A. (2007). Utilization of prenatal diagnosis and termination of pregnancies for the prevention of Down syndrome in Israel. *Israel Medical Association Journal*, *9*, 600–602.

How to cite this article: Nov-Klaiman T, Raz AE, Hashiloni-Dolev Y. Attitudes of Israeli parents of children with Down syndrome toward non-invasive prenatal screening and the scope of prenatal testing. *J Genet Couns*. 2019;28:1119–1129. https://doi.org/10.1002/jgc4.1162