

Decision making on prenatal screening

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Decision making on prenatal screening

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geboren te Dordrecht

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*Mere functional capacities aside,
Test measurements, though accurate, ignore
The essence of the person deep inside,
For science ne'er can grasp the vital core.*

*Truly the test that science fails to see
Is satisfied by all humanity.
Though worn and weary from infirmity,
All people have immeasurable dignity.*

The last two verses of the poem "The overlooked test"
Printed with permission from the author, William P. Cheshire

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Chapter 1

General introduction

Congenital abnormalities

About 2-4% of all newborn babies have a major congenital abnormality apparent at birth (1-4). In the Netherlands, this livebirth prevalence of congenital anomalies is estimated at 2.4% (5). It should be noted that the numerator of this prevalence is the number of liveborn children. If the number of stillbirths is also taken into account this percentage is higher, as the rate of congenital defects among stillbirths is estimated at 10-15% (3;5). It is even estimated that over 80% of the spontaneously aborted embryos and foetuses have structural abnormalities (1;4).

About 15% of the congenital birth defects have a genetic cause, with Down syndrome being the most common chromosomal abnormality; 20% to 30% have multifactorial causes, and up to half of the birth defects have an unknown cause (1;3). Multifactorial disorders have both a genetic component and an environmental component, and one example of such a disorder is neural tube defects (1;6;7).

Neural tube defects

A neural tube defect results from a failure of the neural tube to close during early embryogenesis. The two most frequent neural tube defects are anencephaly and spina bifida. Anencephaly is a lethal condition, and the viability of spina bifida is determined by the type of lesion. The symptoms of spina bifida vary from very mild symptoms to incontinence, paralysis, and hydrocephalus (8;9). Patient care is advancing and improving, although little progress has been made in postnatal surgery of children with spina bifida. Promising results are being achieved in foetal surgery in utero, which is aimed at correcting the structural effect at a point in time when significant neuronal damage has not yet occurred or may be reversible (9).

The birth prevalence of neural tube defects varies according to geographic location, time, and ethnicity (6;10), and the total birth prevalence (i.e. including live births, stillbirths and induced abortions) in Europe is about 1 in 1000 (2). This total birth prevalence can be reduced by periconceptional supplementation of folic acid, and the livebirth prevalence might further be lowered by the availability and utilisation of prenatal diagnosis and termination of pregnancy (6;11-13). Since the policies for prenatal screening and folic acid supplementation vary widely among European countries, the livebirth prevalence of children with neural tube defects also varies greatly (2;14). In the Netherlands, the total birth prevalence has decreased from around 1 in 750 in the 1980s, to around 1 in 950 in the 1990s, to around 1 in 1600 between 2000 and 2003 (2). As both folic acid supplementation and prenatal screening for neural tube defects will continue to increase, it is anti-

pated that the livebirth prevalence of neural tube defects will further decrease in the future.

Down syndrome

The most common chromosomal abnormality in liveborn children is Down syndrome. Down syndrome is caused by trisomy 21. It is characterized by typical physical features, and involves mental retardation, although the severity of the mental retardation varies greatly from child to child. Moreover, Down syndrome entails an increased risk for other birth defects, like heart defects and bowel problems (15). A multifaceted healthcare approach includes having a healthy lifestyle, following certain preventive measures, participating in appropriate health screening, and treating health problems early in their course. Many medical advances, educational techniques, and cultural changes have improved the quality and length of life of individuals with Down syndrome; the life expectancy has increased from 9 years in the 20th century, to 56 years of age currently (16).

In Europe, Down syndrome has a total birth prevalence of approximately 1 in 500 (2). However, since the risk of having a child with Down syndrome increases exponentially with age, the age-related risk runs from around 1 in 1400 at the age of 20, to around 1 in 400 at 35 years of age, to around 1 in 25 for a woman of 45 years of age (17). The livebirth prevalence of Down syndrome is subject to two opposing trends (18). On the one hand, the mean age of pregnant women is increasing (e.g. the Netherlands: Statistics Netherlands, accessed May 22nd 2006), so the mean risk of being pregnant with a child with Down syndrome is increasing. On the other hand, the utilisation of prenatal screening for Down syndrome is also increasing (19), as is the sensitivity of the available tests (20-23). So more cases of Down syndrome are being detected prenatally, and as the vast majority of diagnosed fetuses with Down syndrome are being aborted (24-26), fewer children with Down syndrome will be born. It is expected that the effect of prenatal detection followed by termination of pregnancy will prevail and that livebirth prevalence of Down syndrome will further decrease. For instance, in France the active national policy for prenatal screening has had a major impact on the livebirth prevalence of Down syndrome (27).

Prenatal testing

Prenatal testing is aimed at the antenatal detection of a certain disorder. Prenatal testing can either be done at the request of the expectant parents (e.g. when a disorder runs in the family, or when they are otherwise at increased risk for a disorder).

der), or it can be offered unsolicitedly to pregnant couples. The latter situation is called prenatal screening. The basic difference between screening and testing on request is that screening involves an unsolicited offer of a test to an entire population or subpopulation of apparently healthy people (28;29).

Prenatal testing can either be done through tests that determine the absence or presence of a specific disorder with certainty, or through tests that are only able to provide an individualised estimation of the risk that the foetus has a specific disorder. Amniocentesis is an example of a diagnostic test for the prenatal detection of Down syndrome, while nuchal translucency measurement is an example of a prenatal screening test that determines whether or not there is an increased risk of having a child with Down syndrome.

Prenatal diagnostic testing

Two of the most frequently used invasive prenatal diagnostic tests are chorionic villus sampling and amniocentesis. Chorionic villus sampling is performed in the first trimester of pregnancy (gestational age: 10 to 13 weeks) through either transcervical or transabdominal removal of a sample of chorionic villi (i.e. placental tissue). (30) Amniocentesis is usually done in the second trimester of pregnancy (gestational age: 15 to 18 weeks) through the transabdominal withdrawal of a small amount of amniotic fluid (30). Both tests involve laboratory culturing of the foetal cells that are present in the samples. The test results become available after two weeks, when a foetal karyogram provides the chromosomal diagnosis. Because prenatal diagnostic tests are invasive tests, they carry a risk of an iatrogenic abortion. This risk of a procedure-related abortion depends on the experience of the physician, but is generally estimated at about 0.3 - 0.5% for both tests (31). Besides, neural tube defects can also be detected by detailed ultrasound examination of the spine of the foetus, which is a no-risk diagnostic test.

Prenatal screening

Prenatal screening programmes generally make use of non-invasive, risk-estimating tests, and invasive diagnostic tests are carried out only in high-risk pregnancies (e.g. due to advanced maternal age, or a positive prenatal screening test). The study presented in this thesis focuses on decision making concerning prenatal screening for the two disorders described above: Down syndrome and neural tube defects. During the period in which this study was carried out, the two most commonly used prenatal screening tests were nuchal translucency measurement and the second-trimester maternal serum screening test. Although both tests result in an individualised risk estimation, each has its own characteristics.

Nuchal translucency measurement

It was in the early 1990s that the association between a fluid accumulation behind the foetal neck and Down syndrome became known (32;33). This fluid accumulation appears and then disappears between 11 and 14 weeks of pregnancy in all foetuses, and is observable via ultrasound scanning. In most foetuses with Down syndrome, this so-called nuchal translucency zone is enlarged. The likelihood that the foetus has Down syndrome actually increases with the thickness of the nuchal translucency. So, nuchal translucency measurement involves the determination of gestational age and measurement of the thickness of the nuchal translucency using transabdominal ultrasound examination (34). Based on these parameters, the deviation of the nuchal translucency from what is normal at that gestational age is calculated. This deviation is calculated into a likelihood ratio, which in turn is multiplied by the woman's age-related risk, producing an individualised risk estimation (20;34). Large-scale international review studies on the performance of the nuchal translucency measurement showed generally good test characteristics: the sensitivity of the nuchal translucency measurement is about 70 to 75%, with a specificity of 95% (20;21;35). However, since measuring the thickness of the nuchal translucency is performed by humans, its performance is dependent on the operator. Studies on the repeatability of the nuchal translucency measurement reported high inter-operator and intra-operator correlations (36-38). Thus, it appears that the nuchal translucency measurement is a valid and reliable prenatal screening test.

Maternal serum test

The association between neural tube defects and alpha-fetoprotein in the maternal blood became known in the early 1970s (39;40). The relation between maternal serum alpha-fetoprotein and Down syndrome was shown in the 1980s (41;42). In the late eighties the association between two other serum markers and Down syndrome was discovered, which resulted in the development of the so-called triple test (43). This prenatal screening test involves the laboratory analysis of the levels of alpha-fetoprotein, human chorionic gonadotrophin, and unconjugated oestriol in the maternal serum. On the basis of the levels of these markers, combined with the maternal age, a likelihood ratio is calculated. Multiplying this factor with a woman's age-related risk results in an individualised risk of having a child with Down syndrome. The levels of the triple test markers also indicate the risk that the foetus has neural tube defects. The sensitivity of the triple test is about 60 to 70%, with a specificity of 95% (21;22).

Prenatal screening practice

International situation

Since prenatal screening tests have become available, an increasing number of western countries have started to admit prenatal screening into standard antenatal care. For instance, in the United States, Australia, Taiwan, Israel, and in many countries of Europe (e.g. United Kingdom, France, Finland, Switzerland), pregnant women are routinely offered a prenatal screening test, either in the first or second trimester of pregnancy (44-51). Those with a positive test result (i.e. an increased risk) are being offered diagnostic testing to provide certainty about the presence or absence of Down syndrome or neural tube defects. The increasing utilisation of prenatal testing in general is illustrated by the continuous increase in the proportion of congenital malformations that are diagnosed prenatally (45). For instance, the EUROCAT data indicate an increasing proportion of the total prevalence of Down syndrome that is diagnosed prenatally and subsequently aborted (from around 5% in 1980, and 30% in 1990, to around 55% in 2003) in Europe (2). The same trend is observed for neural tube defects (from less than 20% in 1980, and 45% in 1990, to over 70% in 2003). However, the strength of these trends in the separate European countries varies according to the different national policies for prenatal screening.

Dutch situation

Population Screening Act

While prenatal screening for Down syndrome and neural tube defects has become a standard part of antenatal care in many western countries, at the time our study was performed, this was not the case in the Netherlands. In 1996 the Population Screening Act (PSA) became law in the Netherlands (52). This Act is aimed at protecting the population against screening programmes that could be a threat to either physical or psychological health. The PSA states that screening for serious disorders that can neither be treated nor prevented is such a threat, and is therefore prohibited without ministerial approval. As termination of pregnancy is considered as neither treatment nor prevention, and as there is no approval for this type of population screening, prenatal screening for Down syndrome and neural tube defects was prohibited during our study. Prenatal diagnostic screening was only offered to pregnant women older than 35 years of age. Younger women could only have prenatal testing done if they requested it themselves. However, recently the policy in the Netherlands has changed. In 2004 it was determined that every pregnant woman should be informed about prenatal screening, whereas it would be proactively offered only to pregnant women over 35 years of age (53). Since the

study described in this thesis involves offering prenatal screening to pregnant women of all ages, the minister of health had to grant permission for the study. After the Health Council of the Netherlands had advised the minister to allow the study (54), the permit was granted.

Antenatal care

The Netherlands has a decentralised antenatal care system. The vast majority of pregnant women are counselled by midwives who are fully qualified, autonomous practitioners (55-58). Community midwives take care of the physiological pregnancies and only high-risk pregnancies are supervised by gynaecologists. In the Netherlands, the natural character of pregnancy is generally highly valued, and pregnancy and delivery is generally considered as a natural event rather than a medical event. This appears, for example, from the high number of home deliveries (over 30%, although a decreasing trend has been observed during the last decades (59)), and the low use of medication during delivery (55).

The Dutch debate

In the Netherlands, although prenatal screening has been available for many years, the question of whether it should be generally introduced in antenatal care has been a matter of debate for over two decades (60;61). The participants of the societal debate in the Netherlands can roughly be divided into two coalitions of opponents and proponents of a nationwide prenatal screening programme. The coalition of proponents consisted of the Health Council of the Netherlands, the Dutch society of obstetrics and gynaecology, and patient communities, while that of the opponents included the government, the majority in the Dutch parliament, the Dutch society of midwives, and a few individual gynaecologists (60). The proponents of a prenatal screening programme were of the opinion that it would provide decision options for pregnant couples, and they stated that prevention of suffering and grief would justify termination of pregnancy. The opponents claimed that there was no reliable prenatal screening test (low sensitivity and low specificity). Moreover, they questioned whether there was sufficient societal acceptance for termination of pregnancy following a diagnosis of Down syndrome or neural tube defects. The government repeatedly rejected the start of a nationwide prenatal screening programme. Thus, societal arguments (termination of pregnancy, medicalisation of pregnancy, stigmatisation of disabled persons, social pressure) seem to overrule individual arguments (freedom of choice, reassurance). Indeed, the history of prenatal screening in the Netherlands shows a tension between promotion of the development and use of prenatal screening, mainly by the medical commu-

nity, on the one hand, and control and regulation of prenatal screening by the political decision makers on the other hand (61).

Decision making on prenatal screening

Notwithstanding the many beliefs and suppositions in relation to prenatal screening, much is unknown about the actual advantages and disadvantages of prenatal screening, about pregnant women's reasons for accepting or declining the offer of a prenatal test, about their decision making processes, and about the extent to which pregnant women actually make informed decisions. The study presented in this thesis was aimed at investigating these issues. In this section, the three central themes of this thesis will be introduced:

1. Uptake of prenatal screening and women's motivations for decision making
2. Factors influencing prenatal screening decisions
3. Informed decision making about prenatal screening

Uptake of prenatal screening and women's motivations for decision making

In the debate about prenatal screening, many assumptions are being made regarding how many pregnant women would accept or decline the offer of a prenatal screening test, and what motivates them to choose for or against prenatal screening. For instance, it is argued that prenatal screening entails an offer one can not refuse, or that pregnant women will feel pressurised to accept prenatal screening (62;63). International studies report that pregnant women generally have positive attitudes toward prenatal screening, and that the vast majority of pregnant women who were offered a prenatal screening, accept the test (64). It is, however, not known how many pregnant women actually would accept an unsolicited offer of a screening test in the Netherlands, and what motives they would have for their decisions. Moreover, although pregnant women generally prefer first-trimester screening as opposed to second-trimester screening (65;66), evidence is lacking concerning whether a screening programme offering a first-trimester test would result in a higher percentage of utilisation as compared to second-trimester screening.

Factors influencing prenatal screening decisions

There are many theories that describe factors influencing human behaviour and decision making (67). Most of these theories consider decision making as a rational

process weighing the pros and cons of the different options. Several theories also take into account the social context in which decisions are made, and include social influences on people's decision making (68). More recently, theories that recognise the role of emotions in decision making have been developed (69). Below, these three facets of decision making are described and their relevance for prenatal screening decisions is discussed.

Rational decision theory

A decision on prenatal screening is a decision with uncertain outcomes. In decision making literature, such decision situations are called "decisions under uncertainty" (70;71). Rational decision theory is concerned with making a trade-off between several options with different probabilities and utilities (i.e. the subjective valuation of that outcome) (70). More specifically, subjective expected utility (SEU) theory poses that people who face a decision between different alternatives with uncertain outcomes choose the option that has the highest SEU (70-72). For instance, the choice of a patient about whether to live with a disturbing health problem, or to risk surgery that will probably help but that could also make the condition worse (e.g. serious complication, or death) depends on the utility that is assigned to the possible outcomes and on their subjective probabilities (70). The SEU of each option is calculated by multiplying the utilities of the possible outcomes by their subjective probability of occurrence. According to SEU theory, the option that maximises the decision maker's subjective expected utility is considered the rational choice (71).

In the case of prenatal screening, this rational approach supposes that a pregnant woman would accept a prenatal test if its subjective expected utility exceeds that of declining the test. This implies that her perception of the risk of having a child with, for example, Down syndrome (i.e. its subjective probability) together with her perception of the seriousness of having Down syndrome (i.e. its utility), is important for the choice to accept or decline prenatal screening. For instance, Marteau et al. applied SEU theory to prenatal screening decisions, and found that those undergoing prenatal screening perceived the subjective expected utility attached to testing as greater than those declining it (73). This corresponds with Van der Meulen et al.'s decision analysis, which concluded that the value of a prenatal test depends on the risk of Down syndrome and iatrogenic abortion due to invasive diagnostic testing, and on the value that a pregnant woman assigns to these outcomes (74). However, that study made use of the objective risks of having a child with Down syndrome, while an SEU approach uses people's perceptions of that risk. Since perceived risk and severity of Down syndrome are considered to play an important role in the decision making on prenatal screening, prenatal

screening information booklets generally include information about the risk and severity of having a child with Down syndrome (75;76). As it has been shown that people generally do not make decisions in such a rational way, and frequently deviate from the rational model (70;77), the question that needs to be asked is what role perceptions of risk and severity really play in pregnant women's considerations and decision making.

Social influences

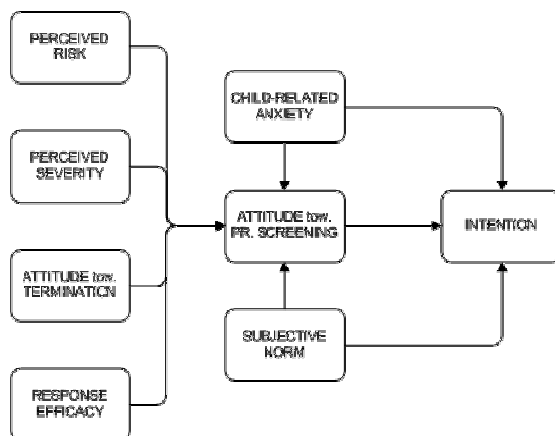
Many health behaviour theories have their roots in SEU theory (78). Like SEU theory, these models emphasize the rationality of human behaviour by assuming that decision making is based upon elaborate cost-benefit analysis of the likely outcomes of the different options. Besides the assessment of probability and utility, these so-called social cognition models incorporate other factors in their model (67). The theory of planned behaviour outlines how the influences upon an individual determine that individual's decision making (78). This theory includes attitude, subjective norm, and perceived behavioural control as determinants of a decision maker's behavioural intention (68;79). According to the theory of planned behaviour, the decision maker's attitude toward a behaviour is based upon beliefs about the probability that a behaviour leads to a specific outcome and the evaluation of this outcome (79). Evidently, this expectancy-value model of attitude is in line with SEU theory (80). Subjective norm is defined as the social pressure an individual feels to perform or not perform a behaviour (68;79). The constructs of the theory of planned behaviour appear to explain health behaviours in general (68). Moreover, one's attitude toward prenatal screening was found to be associated with prenatal screening utilisation (64), and perceived pressure to accept or decline prenatal screening was repeatedly suggested as being an important factor in pregnant women's screening choices (81-83). It was expected that both attitude and subjective norm would be important additions to the model, and would explain substantial proportions of the variance in pregnant women's intentions concerning undergoing prenatal screening.

Emotional influences

The above-mentioned theories are cognitive models, and do not take into account emotional factors (77;84;85). More recently, hypotheses that recognised the role of emotions in people's decision making processes have been developed (69;85;86). For instance, Loewenstein's risks-as-feelings hypothesis poses that responses to risky situations result in part from emotional influences, including feelings such as worry, fear and anxiety (69). In other words, it is assumed that people's behavioural intentions are not only based on a cognitive evaluation of the anticipated

outcomes and their subjective probabilities, but also on an emotional response. Since such an emotional reaction might diverge from the cognitive evaluation, it is relevant to include emotions in the analysis of pregnant women's decision making processes on prenatal screening. It was anticipated that feelings such as being anxious about the health of the unborn baby influence pregnant women's intentions to undergo a prenatal test that would provide more information about the health of this child. Therefore, the different analyses of the decision making process about prenatal screening also address the role of emotions in relation to the risks of having a handicapped child.

Figure 1. Prenatal screening model



Prenatal screening model

Based on these theories and on earlier findings (e.g. 64), a comprehensive prenatal screening model was constructed (Figure 1). In chapter 3 the whole model is described in detail and tested using path analysis, while in chapters 4 and 5 two individual constructs of the model are examined in greater depth (risk perception and subjective norm, respectively).

Informed decision making about prenatal screening

A central topic in the debate about prenatal screening has concerned whether pregnant women who are offered a prenatal screening test will be able to make autonomous, informed decisions regarding whether to accept or decline the test (25;87). It is generally recognised that informing and counselling individuals about screening is aimed at facilitating informed decision making, rather than at achiev-

ing as high uptake rates as possible (87-90). This emphasis on informed decision making implies a shift from the traditional public health perspective on screening to the perspective of individual choice, which might result in a tension between the two perspectives (88;90-93). From a traditional public health perspective, the cost-effectiveness of a specific screening programme is dependent on a high uptake of the screening, while promoting informed choice might compromise this objective (93). Nevertheless, patient autonomy and informed choice are increasingly considered as being most important.

Informed decision making applies in particular to health decisions for which the choice is dependent on individual values (94-96). These so-called preference-sensitive decisions include situations that involve high risks, and situations where there is little medical justification for one option over the other, and where the selected option is dependent on the values of the decision maker (94;97;98). Examples of such preference-sensitive decisions are the decision to choose lumpectomy or mastectomy for treating early-stage breast cancer, or the decision in case of prostate cancer between surgery (lower morality, but risk of sexual dysfunction) and watchful waiting (higher mortality, but no additional risks) (94;99).

Prenatal screening involves preference-sensitive decisions, as the information about the screened disorders and the available test has to be evaluated in accordance with the pregnant women's preferences and values. Prenatal screening can lead to invasive diagnostic testing, which entails a risk for the foetus, and to the option of termination of pregnancy (100). So, decision making is expected to be associated with values concerning pregnancy, disability, and termination of pregnancy. Therefore, counselling and informing about prenatal screening is not aimed at achieving as high uptake rates as possible, or to prevent as many cases of Down syndrome and neural tube defects as possible, but to enable people to make their own, informed choice about prenatal screening (101-103). This is justified by two arguments. In the first place, by enabling people to make informed decisions the ethical principle of autonomy is respected (91;101;104). Respect for autonomy is one of the most important ethical principles in health care (104). An autonomous action is defined as one which is performed intentionally, with understanding, and without controlling influences (91;104). In the second place, informed decisions lead to better psychological outcomes (97;105). For instance, an individual who has made an informed choice to accept a certain screening test, will be less frightened and worried if she receives a positive test result, as compared to those who made an uninformed choice. An informed test acceptor probably knows the meaning of such a test result and has anticipated on this possibility, while an uninformed acceptor possibly has not thought about this option. In general, it is assumed that in-

formed decision making leads to more satisfaction, less decisional conflict, and less anxiety.

International studies provide ample evidence that pregnant women are currently not making fully informed choices about prenatal testing (64;97;106;107). A recent review on the psychosocial aspect of prenatal screening concluded that “most women are not evaluating complete decision-relevant information in accord with their beliefs before making their screening choices, that is most are not making informed choices about screening.” (64). However, as there are no studies that assessed the level of informed decision making on prenatal screening in the Netherlands, it is currently not known to what extent this international status quo refers to the Dutch situation. The aim of the third part of this study was to address the issue of informed decision making on prenatal screening in the Netherlands.

There is, however, much debate about how to evaluate informed decision making. Should the evaluation be based on the final choice (outcome), or on the way the choice was made (process) (97)? The former approach implies that the actual choice is compared to a rational standard. This includes assessment of the extent to which choices conform to the prescriptions of SEU theory (77;108) or the extent to which choices are consistent with certain values (100;102;109). The vast majority of studies assessing informed decision making focus on decision outcomes (110). The latter approach focuses on the evaluation of the decision making process, i.e. the way the decision is made (64;70;97;111). This implies that the measurement of informed decision making includes the assessment of how the decision is reached: is a decision based on the evaluation of all the relevant information, including risks and alternatives? (97;110). These different approaches are reflected by the many different definitions of informed decision making (See Table 1, chapter 7). They all include that an informed decision needs to be based on sufficient knowledge of the relevant information, and should be consistent with the decision maker’s values (64;97;102;110;112-115). However, some of the definitions include an additional prerequisite of an informed decision, namely that it implies a process of deliberation about the alternatives and weighing up their pros and cons (64;110;113;115). According to these two different approaches, this thesis used two different operationalisations of informed decision making (chapters 6 and 7, respectively).

Objectives

In sum, the central objective of this study was to gain more insight into the decision making of pregnant women concerning prenatal screening for congenital de-

fects. This thesis addresses three central themes, and accordingly, the thesis is divided into three parts that deal with the following specific research questions:

Uptake of prenatal screening and women's motivations for decision making

How many pregnant women accept a prenatal screening offer, and what demographic factors influence the uptake rate? (chapter 2)

What are the motivations of pregnant women for accepting or declining prenatal screening? (chapter 2)

Factors influencing prenatal screening decisions

What factors determine pregnant women's decisions regarding prenatal screening? (chapter 3)

Can different components of risk perception be discerned and what is the role of risk perceptions in the decision making process? (chapter 4)

Do prenatal counsellors' attitudes toward prenatal screening influence the decision making of pregnant women? (chapter 5)

Informed decision making about prenatal screening

Do pregnant women make informed choices about prenatal screening, and what are the psychological effects of informed choice? (chapter 6)

Can a measure of informed decision making be constructed that includes the assessment of pregnant women's decision making processes? (chapter 7)

Methods

The study presented in this thesis was part of a larger research project that aimed to study risk perception, decision making and psychological well-being of pregnant women who were offered prenatal screening for congenital defects. As the separate chapters only describe the design and methods of the particular sub-study, the design of the overarching research project is described below.

Design

The research project was a prospective randomised controlled trial among pregnant women in the Netherlands. The recruitment of study participants was done by midwifery and gynaecology practices. Forty-four practices in several parts of the Netherlands consented to be engaged in the study. The practices included both urban and rural areas throughout the Netherlands to ensure a representative sample of pregnant women that would be asked to participate in the study.

In the Netherlands, when a woman has found out that she is pregnant, she usually calls a nearby midwifery practice to make a first appointment with a midwife. The participating practices were instructed to ask pregnant women permission during this first contact to be sent a research information letter. The pregnant women were sent an information letter about the research project and an informed consent form. The inclusion criteria were: command of the Dutch language, and less than 16 weeks of gestation. The inclusion period lasted from May 2001 to May 2003, and the very last follow-up measurement was made in April 2004.

Pregnant women who consented to participate in the study were randomised by the researchers into one of three groups. Women in the first intervention group were offered the nuchal translucency measurement; women in the second intervention group were offered the maternal serum test; women in the control group were not offered prenatal screening, but they received the standard antenatal care. The test offer consisted of an information booklet sent to the pregnant woman's home, and a consultation with the midwife or gynaecologist. The booklet contained information about the disorder or disorders screened for, the particular test, the possible test outcomes, and the options available should there be a positive test result. Moreover, the booklet explicitly paid attention to the decision making process, e.g. by listing advantages and disadvantages of having the test done. In our study, prenatal screening required a separate visit to a hospital or prick point for the maternal serum test, or to a hospital or an ultrasound centre for the nuchal translucency measurement.

Data collection took place through five postal questionnaires at different points in time. The first questionnaire was sent before any information about screening was given and included questions about demographic background, and scales that assess pregnancy experience, emotions, attitudes, and risk perceptions. The second questionnaire was filled in after the test information but before the test result. This questionnaire contained the questions from the first questionnaire, and assessed reasons for accepting or declining prenatal screening, evaluation of the information and of the screening test, knowledge about prenatal screening, decisional conflict, decisional satisfaction, and decision making strategy and style. The third questionnaire was sent to the participants after the test result, and included the scales from the first questionnaire, together with the participants' evaluations of their decision and of the test result. This questionnaire also assessed decision making regarding invasive testing. Pregnant women who were not offered prenatal screening (the control group) or those in the intervention group who declined the screening test were sent the second and third questionnaires at comparable points in time. The fourth questionnaire was sent at 28 weeks of pregnancy and the

fifth was sent after delivery. These latter questionnaires contained several scales that were also included in the previous questionnaires.

Participants

Figure 2 shows the flow of participants through the larger research project. During the recruitment period 4077 pregnant women were approached to participate in the study. Of these, 2986 (73%) gave informed consent and filled in the first questionnaire. Of these participants, 2877 (96,3%) were randomised: 1066 (37%) in the NTM group, 1061 (37%) in the MST group, and 750 (26%) in the control group. The second, third, fourth and fifth questionnaires were filled in by 2277 (79%), 1986 (68%), 1846 (64%), 1779 (62%) participants, respectively (Figure 2).

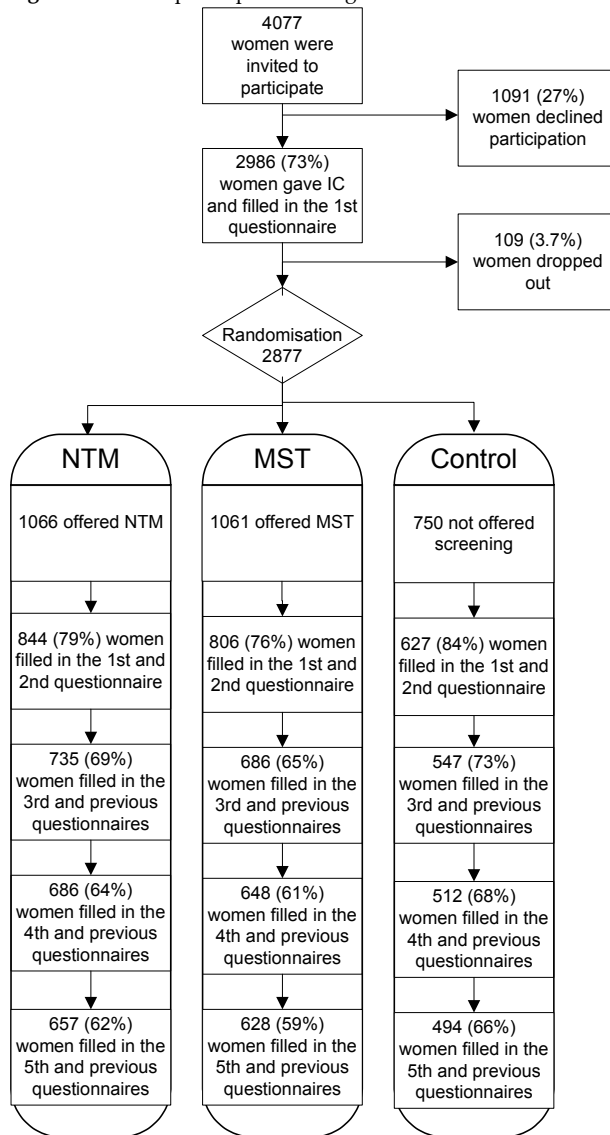
The studies described in this thesis only made use of the first three questionnaires. Moreover, the participants in the control group were not offered prenatal screening, and as this thesis involves decision making on prenatal screening, only pregnant women in the intervention groups were used.

Contents

Chapter 2 describes the uptake of prenatal screening offered to an unselected population of pregnant women. It examines which demographical factors are related to this uptake rate, and whether there are differences between the nuchal translucency measurement and the maternal serum test. This chapter also investigates pregnant women's reasons for accepting or declining prenatal screening.

Chapter 3 describes the development and testing of a prenatal screening decision model that was based on decision theory and health behaviour theory, as well as on previous findings. This model hypothesised that pregnant women's prenatal screening behaviour can be predicted by their attitudes toward prenatal screening, social norm, and worries about the health of the foetus. The attitude toward prenatal screening was hypothesised to be determined by pregnant women's perceptions of risk and severity of Down syndrome, attitude toward termination of pregnancy, and perceived test efficacy. Chapter 4 focuses on how pregnant women perceive the risk of having a child with Down syndrome, and it describes components of risk perception and their role in the decision making concerning prenatal screening. Chapter 5 focuses on the possible influence of the prenatal counsellor on pregnant women's decisions on whether or not to undergo prenatal screening. This potential influence was explored at different stages of the decision making process (pregnant women's test uptake, attitude toward prenatal screening, and their change in attitude).

Figure 2. Flow of participants through the trial



The issue of informed choice or informed decision making in relation to prenatal screening decision making is the focus of chapters 6 and 7. Chapter 6 describes to what extent the participants of the study make informed choices concerning prenatal screening, and it describes the psychological effects of informed choice. For this analysis, an informed choice was defined as one that is based on

the relevant information and consistent with the decision maker's values. Chapter 7 discusses the difference between informed choice and informed decision making, and it describes the construction of a measure of informed decision making, which included a measure of the decision making process (i.e. a measure of deliberation about the alternatives and their pros and cons). This chapter also presents the differences in the level of informed decision making between pregnant women who accepted the prenatal screening test and those who declined the test.

Chapter 8 discusses the findings of the different chapters. Furthermore, it addresses the contribution of this study to the body of knowledge, and its strengths and limitations. This chapter also discusses questions that ensue from the findings of the study, and makes recommendations for further research. Chapter 9 summarises the findings of the study.

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Chapter 2

**Accepting or declining the offer of
prenatal screening for congenital
defects: test uptake and women's
reasons**

Abstract

Prenatal screening for Down syndrome has become standard practice in many western countries. In the Netherlands, however, prenatal screening tests for congenital defects are not offered routinely. The present study aims to assess test uptake in a large, unselected population of pregnant women, and to give more insight into the decision for or against prenatal screening through nuchal translucency measurement or maternal serum screening. The study is part of a randomised controlled trial with two groups each being offered a different prenatal screening test, and a control group. Pregnant women received postal questionnaires at three stages of their pregnancy. Of the women being offered the nuchal translucency measurement or the second trimester maternal serum test, 53% and 38% respectively accepted the test offer. The main reasons for accepting were 'gaining knowledge about the health of the foetus/curiosity' (50%), 'favourable characteristics of the screening test' (18%), 'increased risk of having a child with DS' (15%). The main reasons for declining were 'unfavourable characteristics of the screening test' (42%), 'not applicable/not necessary' (35%), 'anxiety/uncertainty' (36%), 'adverse characteristics of the invasive tests' (32%), and 'being against abortion' (15%). The uptake of prenatal screening was relatively low, and different distributions of reasons were reported, compared to other studies. These differences may be due to the specific Dutch situation in which prenatal screening is not part of standard antenatal care. The question arises as to whether informed decision making would be reduced if prenatal screening became routinized.

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Introduction

Prenatal screening can identify a high-risk subgroup within a population of pregnant women. Prenatal screening for Down syndrome (DS) and neural tube defects (NTD) provides an individualized risk estimation of having a child with one of these disorders. The subgroup of women with an increased risk can be offered prenatal diagnostic testing in order to provide a certain diagnosis.

Two of the available methods of prenatal screening for congenital defects are: the Nuchal Translucency Measurement (NTM) and the Maternal Serum screening Test (MST). Although both screening tests result in a risk estimation, it should be stressed that these tests have different characteristics: NTM screens only for DS in the first trimester of pregnancy, and is performed through ultrasound scanning (1). The MST ('triple-test') is a blood test in the second trimester of pregnancy, and tests for both DS and NTD (2).

Although prenatal screening for DS and NTD has become standard practice in many western countries, this is not the case in the Netherlands. In July 1996, the Population Screening Act (PSA) became law in the Netherlands. Its purpose was to protect the population against screening programmes that could be a threat to the psychological and physical health of the person being screened. One of the categories of the population screening that requires ministerial approval is screening for 'serious disorders that can neither be treated nor prevented'. Prenatal screening for congenital disorders comes under this category, since the legislature considers termination of pregnancy as being neither treatment nor prevention (3). Since there is no approval for this type of screening in the Netherlands, it is forbidden to offer prenatal screening to pregnant women, unless they request it. However, permission is granted to offer prenatal diagnostic tests to pregnant women only if they are over 35 years of age. In contrast to the practice in other western countries, in the Netherlands the antenatal care system is strongly decentralized: midwives operate as autonomous medical practitioners (4). Generally, physiological pregnancies are taken care of by midwives, and it is only in cases where pathology is suspected or identified that the pregnancy is managed by a gynaecologist (5).

In the United States, prenatal screening has been offered as part of customary antenatal care for decades, and having such a screening test done has become common (6;7). Based on interviews and observations Markens et al. and Press and Browner concluded that prenatal screening ceases to be something about which a deliberate decision needs to be made. A recently published review by Green et al. on psychosocial aspects of prenatal- and newborn screening, which included 106 publications from 12 different countries, also concluded that women's understanding of screening is poor and that most women are not making informed choices

about screening (8). Accepting prenatal screening is not a result of a considered, conscious decision, but rather perceived as something self-evident: “Thus, the acceptance of routines because they are routine means that pregnant women do not necessarily make an informed decision [...]” (9). A Scottish report, which presents a technology assessment concerning ultrasound scanning before 24 weeks of pregnancy (this includes NTM), also concluded that many women did not feel that they were being offered a choice about testing, and did not consider their decision making as being informed (10). This lack of informed decision making conflicts with the commonly-held belief that the objective of prenatal screening is to enable people to make their own, informed decision regarding whether or not to have the screening test done, rather than preventing as many affected children as possible from being born (11;12).

Uptake rates of screening tests are generally high in countries where prenatal screening is part of usual care, e.g. in the UK: uptake percentages were 87% for the second trimester MST, and over 95% for NTM combined with first trimester serum biochemistry (13-15). Studies from other countries also reported high uptake rates (16-20). However, uptake rates vary considerably across health care settings, even in the same region (21). Several studies that have been done to assess motivations concerning prenatal screening, focused solely on reasons for declining the screening test (22-26). These studies reported religious and moral reasons as the main reasons for declining a screening test, as well the unreliability of the test. Some other studies examined only reasons for accepting prenatal screening (17;27-30). The most frequently mentioned reason in the majority of these studies was reassurance (8).

Most of the above-mentioned studies were performed in countries where prenatal screening is part of customary antenatal care. Given the questions about informed choice in these countries, our study—which is performed in a situation where this screening is not a routine or implicit decision—offers a unique opportunity to investigate uptake and reasons for accepting or declining the screening offer in an unbiased way. According to the PSA, ministerial approval was needed for this study, since it involves offering prenatal screening. The aim of this study is to assess the uptake of prenatal screening tests offered to an unselected population of pregnant women, and to assess how uptake differs across the background variables. The second aim of the study is to investigate reasons for either accepting or declining prenatal screening for congenital defects.

Patients and methods

Women attending one of 44 participating midwifery and gynaecology practices from May 2001 to May 2003 before 16 weeks' gestation were asked permission to be sent a research information letter and an informed consent form. Women who granted informed consent were randomised into one of two intervention groups or into the control group. Women in the first intervention group were offered the NTM, and women in the other randomisation group were offered the MST. Participants in the control group received the customary care. As the NTM has to be performed in the first trimester of pregnancy, women whose first contact with their midwife or gynaecologist occurred later than ten weeks into the pregnancy could be randomised solely in the MST group or the control group.

The test offer consisted of a sent-home booklet with information about the particular test, and an oral explanation by the woman's midwife or gynaecologist during a consultation. The following topics were covered in the information booklets: characteristics of DS and NTD (information about NTD is covered only in the MST booklet), age-specific risks of DS, population risk of NTD, procedure of the screening test, options available following a positive test result, procedure of the diagnostic tests (amniocentesis and chorionic villus sampling). The last section of the booklet was entitled: "to decide whether or not to undergo the screening test". This section listed some advantages and disadvantages of having the prenatal screening test done. The booklets were pilot-tested for clarity. Women who gave birth in our hospital during the year preceding the start of the study (N=659) were asked to read the booklet and fill in a questionnaire. Of these women, 368 (58%) returned the questionnaire. It appeared that the booklets were comprehensible and suitable to be used as information booklets in our study. After this pilot, the wording of the booklets was adapted in some areas. The oral explanation was standardized by means of a plasticized paper consultation guide, which covered the same topics as the information booklets as well as some counselling guidelines. These guidelines included issues of non-directiveness: "avoid giving your own opinion", "it has to be the pregnant woman's own decision". A separate visit to the hospital was required to have the screening test done.

Data collection took place using five questionnaires. For the present paper, only data of the first three questionnaires were used. The first questionnaire was filled in before the pregnant women received the screening information booklet, and the second questionnaire was filled in after they had read the booklet and decided for or against prenatal screening, but (if applicable) before they received the result of the screening test. The third questionnaire was sent after receiving the result (or at a comparable point in time). The first questionnaire contained questions

about background variables such as age, education, parity, and religion. The second questionnaire contained open-ended questions in which participants were asked for their personal reasons (including a decisive one) for accepting or declining the prenatal screening offer. The third questionnaire asked whether or not participants had had the screening test done. Overall, these answers matched the official result forms. The self-reported test uptake rates were used in the analyses.

During the recruitment period, 4076 women were asked to participate in the study; 2978 (73%) women gave informed consent and returned the first questionnaire. Of these women, 74% (n=2203) also filled in and returned the second questionnaire (after one reminder letter). The third questionnaire was returned by 1968 women. For the present paper, only data of the respondents in the intervention groups were used (n = 1399). For analysis of non-response all women who were asked to participate between September 2002 and January 2003 were sent a short questionnaire. Of these 259 women, 130 (50%) replied. It appeared that the main reason not to participate in the study was lack of time or lack of interest.

Analysis

Categories and subcategories for given reasons for accepting or declining prenatal screening were created by the first author on the basis of a selection of hundred answers, and discussed by others (JHK, EG, DRMT). After some adaptations had been implemented, all answers were coded by three persons (MB, JHK, EG). At random, 20% of the answers were double-coded into the main categories by one of the authors (DRMT). The correspondence rate was 94%. Most non-corresponding codes were a result of a systematically different interpretation by the double-coder of the meaning of three of the 16 main categories. After some discussion, these differences in interpretation were resolved.

To compare the distributions of test uptake with respect to the background variables (age, education, parity and religion), and with respect to the two different screening tests, χ^2 tests were used. Ordinal variables were analysed using a χ^2 test for trend (χ^2_{trend}). Multiple logistic regression was used for multivariate analysis.

Approvals

According to the Dutch Population Screening Act, the Minister of Health, Welfare, and Sports had to grant permission for this study to be performed. After receiving advice from the Health Council, the permission was granted (31). The present study was also approved by the Ethical Committee of the VU University Medical Centre.

Results

To assess the representativeness of the sample, background variables (age, education and parity) were compared to data of the general pregnant population in the Netherlands (32). The only relevant difference was that women in our sample had higher education levels (19% and 43% for the general pregnant population and our study sample, respectively). The mean age of the participants was 31 years of age.

Table 1. Numbers of participating pregnant women accepting or declining the nuchal translucency measurement or the maternal serum test

	Accepted screening N (%)	Declined screening N (%)	Total N
NTM ¹	387 (53)*	342 (47)	729
MST ²	254 (38)	416 (62)	670
Total	641	758	1399

¹ NTM: Nuchal Translucency measurement

² MST: Maternal Serum screening Test

* $\chi^2 = 32.4$, $p < 0.001$

Uptake

The overall test uptake was 46%. Table 1 shows the distribution of acceptors and decliners for the two screening tests. Test uptake was significantly higher among women being offered the NTM compared to those being offered the MST ($\chi^2=32.4$, 1 df, $p<0.001$). Table 2 shows that test uptake was significantly lower for women who reported being more actively religious, and for women who were highly educated. Using a model that started with all four background variables, multiple logistic regression analysis revealed that degree of religiosity and level of education remained significantly and independently associated with test uptake (see Table 3).

Table 2. Background characteristics of the participating pregnant women who accepted or declined prenatal screening

	Total N (%)	Accepted screening N (%)	Declined screening N (%)	
Age*				$\chi^2_{trend} = 0.54$
<26	89 (7)	39 (44)	50 (56)	$p = 0.463$
26-30	499 (37)	215 (43)	284 (57)	
31-35	639 (47)	316 (50)	323 (50)	
>35	123 (9)	49 (40)	74 (60)	
Number of children				$\chi^2_{trend} = 0.15$
0	602 (43)	287 (48)	315 (52)	$p = 0.701$
1	574 (41)	247 (43)	327 (57)	
2	163 (12)	79 (49)	84 (51)	
≥3	60 (4)	28 (47)	32 (53)	
Degree of religiosity*				$\chi^2_{trend} = 6.3$
Actively	58 (4)	12 (21)	46 (79)	$p = 0.012$
Somewhat actively	255 (18)	105 (41)	150 (59)	
Not actively	412 (30)	213 (52)	199 (48)	
Not religious	668 (48)	306 (46)	362 (54)	
Educational level*				$\chi^2_{trend} = 10.1$
Low	165 (12)	95 (58)	70 (42)	$p = 0.001$
Middle	616 (45)	280 (46)	336 (54)	
High	579 (43)	244 (42)	335 (58)	

* The totals of age, degree of religiosity and educational level do not add up to 1399 because of missing values on these questions.

Table 3: Multiple logistic regression analysis: two significant factors associated with test uptake

	OR	95% CI	
Degree of religiosity			$p < 0.001$
Not religious	1.00		
Not actively	1.29	1.00-1.66	
Somewhat actively	0.82	0.61-1.11	
Actively	0.34*	0.17-0.65	
Educational level			$p = 0.005$
Low	1.00		
Middle	0.61*	0.42-0.87	
High	0.55*	0.38-0.78	

* Significant at 0.01 level

Reasons for accepting or declining the screening test

The mean number of reasons for accepting or declining prenatal screening that participants specified was 1.9. The test acceptors gave significantly fewer reasons compared to the test decliners (1.7 versus 2.0, $t = 7.5$, $p < 0.001$). The participants were also asked to give one decisive reason, but 45% of the test acceptors and 33% of the test decliners failed to give a decisive reason. The respondents either did not fill in this question, or they indicated that there was not one decisive reason. So, the numbers of participants giving a decisive reason are: 353 (55% of 641) of the acceptors and 500 (66% of 758) of the decliners. Women's (decisive) reasons for accepting or declining prenatal screening are shown in Table 4 and 5 respectively.

There were no differences in frequencies of decisive reasons between the NTM group and the MST group, except for the category 'research/science' with regard to the acceptors (3% versus 13%, $\chi^2 = 12.4$, 1 df, $p < 0.001$) and the category 'unfavourable test characteristics of the screening test' with regard to the decliners (17% versus 26%, $\chi^2 = 5.9$, 1 df, $p < 0.05$)

Table 4. Reasons to accept the offer of a prenatal screening test

Reasons to accept screening	One of the reasons (N = 641) %	Decisive reason (N = 353) %
Gaining knowledge / curiosity	50	29
Favourable characteristics of screening test	18	15
Increased risk (age, family history)	15	12
Research / science	13	7
Reassurance	8	4
Wanting a healthy child	5	5
Reasons against testing	21	6
Other	36	21

Test acceptors

The most frequently mentioned reason for accepting a screening offer was: 'gaining knowledge about the health of the foetus' (50%, and 39% as decisive reason). This category included sub-categories such as: 'I just want to know whether or not the child has Down syndrome', 'I want more certainty about the health of the foetus', and 'It's just out of curiosity'. Of the women who accepted the screening offer, 18% mentioned that they did so because of the favourable test characteristics (15% as decisive reason). This category consisted of two sub-categories: 'The test does not involve any risk', and 'There's no harm in trying'. Another group (15%, and 12% as decisive reason) decided to have the screening test done because (they

thought) they had an increased risk of having a child with congenital defects, either because of their age or because of family history. Reassurance was mentioned by 8% of the women. The 'Other' category involved reasons such as: 'to be prepared' and 'the ability to make a better choice concerning invasive diagnostic testing'.

Table 5. Reasons to decline the offer of a prenatal screening test

Reasons to decline screening	One of the reasons (N = 758)	Decisive reason (N = 500)
	%	%
Unfavourable characteristics of screening test	42	22
Anxiety / uncertainty	36	13
Not applicable / not necessary	35	12
Adverse characteristics of invasive test	32	11
Against abortion	15	13
Acceptance / destiny	16	10
Reasons in favour of testing	17	12
Other	7	6

Test decliners

The main reasons for declining a screening test were 'unfavourable characteristics of the screening test' (42%), 'anxiety/uncertainty' (36%), 'not applicable/not necessary' (35%), and 'adverse characteristics of the invasive tests' (32%). Almost a quarter of all women mentioned 'unfavourable characteristics of the screening test' as their decisive reason for refusing prenatal screening. This reason contained three sub-categories: 'It is just a calculation of probability', 'The test gives no certainty', and 'The test is not reliable'. Another important reason concerned the feeling in women that screening causes anxiety and uncertainty. The three most frequent sub-categories were: 'I do not want to become unnecessarily anxious', 'The test result will make me uncertain and anxious', and 'I am afraid of a positive test result'. Relevant sub-categories within the group of women who considered screening as 'not applicable' or 'not necessary' were: 'I am not in a risk group' and 'I have already done a prenatal test'. The fourth main reason women gave was coded as 'adverse characteristics of invasive tests'. This category included the following sub-categories: 'I will not do diagnostic testing because of the risk of miscarriage that is involved', and 'I do not want to make a possibly difficult decision'.

Among the reasons for declining the screening test, two ethical reasons could be discerned: women who were opposed to selective abortion (15%), and women who see the health status of the baby as destiny or fate, and would accept the child

anyway (16%). This last reasoning included the following sub-categories: 'The child is welcome, whether it is disabled or not', and 'I want nature to take its course'.

Ambivalence in reasoning

Of the test accepters, 21% also mentioned reasons against prenatal screening, in addition to reasons in favour of prenatal screening (Table 4). The most frequently mentioned reasons were 'adverse characteristics of invasive test', and 'anxiety/uncertainty'. On the other hand, 17% of the women who declined the screening offer also mentioned reasons in favour of screening (Table 5). The contra-reasons among test decliners included 'gaining knowledge/curiosity', 'increased risk of having a child with DS', and 'favourable characteristics of the screening test'.

Discussion

The overall uptake of prenatal screening for congenital defects was 46%. This is considerably lower than most other studies report. Some studies presented uptake rates of 90% and above (15). These differences in uptake are probably related to the different situation in the Netherlands compared with other countries. Generally in the Netherlands, antenatal care is not considered as something 'medical', and the natural character of pregnancy is highly valued (29). This is evident not only from the high rate of home deliveries and the low rate of epidural analgesia during labour in the Netherlands, but also from the fact that prenatal screening is not part of customary care. Consequently, accepting the screening test offer is not a routine decision for Dutch pregnant women. These factors might account for the relatively low uptake rates of prenatal screening. Nevertheless, two Dutch studies did report high uptake rates of around 80% (33;34). The study of Kamerbeek et al. was performed in a particular region of the Netherlands where prenatal screening was offered routinely even though this is not allowed formally. In contrast to our study, in the study of Muller et al. no separate visit was required to have the screening test done. Dormandy et al. have shown that higher uptake of serum screening was seen at hospitals that offered the test as part of a routine visit than at those where screening required a separate visit (21;35). It might be that prenatal screening as part of a routine visit stimulates test uptake without giving too much thought to it (6;36). However, it could also reflect barriers to testing incurred by requiring a separate visit (35).

In our study, uptake rates of 53% for the NTM and 38% for the MST were found. Because there were hardly any differences in given reasons between the

two groups, other factors may be relevant in explaining these different uptake rates. It might be related to the fact that the NTM is performed by ultrasound scanning, resulting in the visualization of the foetus, which is often seen as an advantage of this screening test. In general, pregnant women consider ultrasound examination as a benign procedure allowing them to see their baby, and they attach positive value to being able to see it (9;10;37). Another contributing factor could be women's preferences for screening in the first trimester of pregnancy (NTM) compared to screening in the second trimester (MST) (38;39).

The three most frequently mentioned reasons for accepting the screening test offer (obtaining knowledge, good test characteristics, and increased risk) did not include reassurance. However, other studies (17;28;29) reported reassurance as the most important reason for having a screening test done. This might be due to a difference in information given in the booklet women received, or in the counselling by their midwife or gynaecologist. In our study, participants received well-balanced information about the pros and cons of the screening tests, while this may not always be the case in countries where prenatal screening is part of customary antenatal care. Consequently, it could be that women participating in our study were more aware of the fact that a screening test is not able to give reassurance by itself, since it provides merely a risk estimation.

Two of the main arguments for declining the screening test offer (mentioned by three quarters of the participants) were linked to disadvantageous test characteristics. The unfavourable characteristic of the screening tests is that they provide only a risk estimation, and cannot give certainty. On the other hand, a risk of induced miscarriage of the foetus is inherent in diagnostic testing. Women specifying one of these reasons were not opposed to prenatal screening in general, and did not consider it problematic to know the health status of their baby, but they basically said "the test isn't good enough", or they just did not want to put their baby at risk. These data suggest that many pregnant women will make a different decision when a risk-free screening test providing a certain diagnosis is available. Accordingly, we expect higher uptake rates in the future when better screening tests will have been developed.

Of the women, 23% mentioned reasons against screening that could be interpreted as ethical in nature. This percentage is considerably lower than reported by Sher et al. (25). Cultural and religious differences may be responsible for this difference. Furthermore, it might be due to the fact that at the time Sher et al.'s study was carried out, screening had already become widely accepted. In such a situation, declining prenatal screening may mainly be based on strong moral values. Again, the well-balanced information booklets may also play a part here.

Over 25% of the women also gave reasons in support of the opposite decision option. On the one hand, this could be an indication of ambivalence: the choice of whether or not to screen is not a clear-cut decision, but rather one that is imbued with mixed feelings. In this case, these women need to be given special attention during the counselling process. On the other hand, it may also be an indication of someone balancing the pros and cons of a decision. In this situation, giving opposite reasons might be a sign of a rational, informed decision (40).

To conclude then, offering prenatal screening in a context where it is not part of routine prenatal care resulted in lower uptake rates and different reasons for accepting or declining such a screening test than in a situation in which screening is offered routinely. This raises the question as to what extent a high uptake rate involves a high number of informed choices. Are all test acceptors making considered, deliberate decisions?

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Chapter 3

**Understanding pregnant women's
decision making concerning prena-
tal screening**

Abstract

This study is aimed at enhancing understanding of prenatal screening decision making through testing a hypothesized model based on decision theory and health behaviour theory. 1666 pregnant women who were offered prenatal screening for Down syndrome filled in a questionnaire. Path analysis (using LISREL) resulted in a final model with reasonable model fit, which was verified by split-sample cross-validation. Attitude toward termination of pregnancy, perceived test efficacy, and subjective norm determined a woman's attitude toward having a prenatal test. Anxiety was influenced by perceived risk and perceived severity of having a child with Down syndrome, and by subjective norm, but appeared to be a weak predictor of intention to test. Pregnant women with a positive attitude toward prenatal screening, and who perceived a subjective norm to undergoing prenatal screening, showed a greater intention to have prenatal screening done. These findings may implicate that more attention should be paid toward the values and social context of the pregnant women during the counselling process.

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Introduction

In most western countries, prenatal screening for Down syndrome is routinely offered to pregnant women. Accordingly, all pregnant women are faced with the choice of deciding whether or not to undergo prenatal screening. Reassurance and knowledge about the health of the fetus are found to be the main reasons for accepting prenatal testing; test characteristics and attitude toward abortion are found to be the main reasons for declining (1;2). In a review of the literature on prenatal screening behaviour, it was found that having a prenatal test was associated with negative attitude toward the abnormality tested for, high perceived risk of having a child with the abnormality, strong subjective norm to accept the test, and positive attitude toward termination of pregnancy (1). However, there seems to be a lack of theoretical approaches to decision making regarding prenatal screening, as only a few of these studies referred to a theory for the selection of measures or the interpretation of the results. The present study is aimed at enhancing the understanding of prenatal screening decisions, by developing and testing a hypothesized prenatal screening behaviour model based on decision theory and health behaviour theory.

The decision regarding whether or not to undergo prenatal screening is a complex one, as information about different risks, tests, and outcomes needs to be processed (3;4). The current paradigm of informed decision making implies that pregnant women should make their own choices about prenatal screening, based on correct and relevant information. This information is generally provided through written information booklets in addition to verbal information from gynaecologists or midwives. Such information booklets contain information about the characteristics and risks of the disorder screened for, the characteristics and risks of the available tests, and the meaning and implications of the possible test results (5;6). An important component of the information consists of risk information: the age-related risk of having a child with Down syndrome, the test result –which is an individualized estimation of the risk that the child has Down syndrome– and the risk of an iatrogenic abortion that is involved with diagnostic testing (7). Another significant component of prenatal screening information booklets is information about the characteristics and meaning of Down syndrome (5;8). This indicates that information about the risk, as well as the seriousness of having a child with Down syndrome, are considered essential elements in the decision making process concerning prenatal screening.

The supposition that considerations of both risk and severity play an important role in decision making concerning prenatal screening, is in accordance with expected utility (EU) theory. EU theory prescribes that individuals aim to maximize utility, and thus choose the behaviour or option with the highest expected

utility (9). Expected utility of an alternative is calculated by multiplying the subjective value of an outcome by its probability of occurrence. This expected utility approach corresponds with Van der Meulen et al.'s decision analysis of the maternal serum screening decision, which concluded that the value of a prenatal screening test is determined by the risk of having a child with Down syndrome and the risk of an iatrogenic abortion, and the value that a pregnant woman assigns to these outcomes (10). However, as people interpret these objective probabilities in different ways, subjective probability (i.e. perceived risk) might be a better predictor of prenatal screening decisions. Subjective expected utility (SEU) theory uses the decision maker's subjective evaluations of both probabilities and outcomes (11;12). In the context of prenatal screening, this supposes that pregnant couples accept prenatal screening if they perceive their risk of being pregnant of a child with Down syndrome as high, and if they perceive Down syndrome as serious and severe. In other words, the higher pregnant couples perceive the risk and severity of Down syndrome, the greater their intention to undergo prenatal screening.

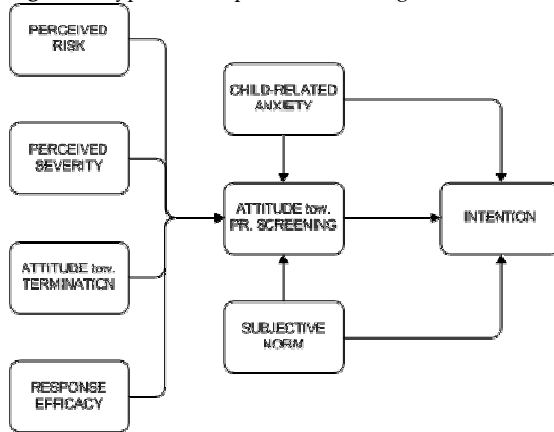
The above-mentioned constructs also play a role in expectancy-value models of attitude (13). According to this model, one's attitude toward a behaviour is based on beliefs about the probability that a behaviour leads to certain outcomes and the evaluations of those outcomes (14;13). In the case of prenatal screening, perceiving the risk of having a child with Down syndrome and the severity of Down syndrome as very low, is likely to result in a negative attitude toward undergoing prenatal screening. Attitude is a central construct of the theory of planned behaviour (TPB) (14;15). The TPB is a widely-applied model in studies attempting to explain health behaviour (including uptake of screening tests), and it postulates that the intention to engage in health behaviour is predicted by three factors: one's attitude toward the behaviour, subjective norm, and perceived behavioural control (15). According to the TPB, attitude toward a behaviour is the degree to which an individual has a favourable or unfavourable appraisal of the behaviour in question. Subjective norm is the perceived social pressure to engage or not to engage in a behaviour. Perceived behavioural control measures an individual's efficacy to perform a certain behaviour regularly or constantly. Since undergoing prenatal screening is a one-time decision, and does not require an individual to be efficacious to perform the behaviour (e.g. the test itself is executed by a practitioner), perceived behavioural control does not play a role in this decision, and is therefore not included in this study.

Although the role of emotions in decision making is increasingly recognized (16;17), the above-mentioned health behaviour models do not take into account emotional factors in decision making (17;18). Loewenstein et al. address this emotional side of decision making, as their risk-as-feelings hypothesis poses that re-

sponses to risky situations result in part from emotional influences, including feelings such as worry, fear and anxiety (16). So, besides the cognitive evaluation, the emotional response (i.e. anxiety about the health of the future child) to a threat also plays an important role in the decision making process. This indicates that, for instance, a pregnant woman may accept prenatal screening because she feels anxious about the health of her baby. So, we hypothesize that prenatal screening intention is also determined by emotions like child-related anxiety.

Furthermore, our study investigating women's reasons for accepting or declining prenatal screening (2), indicated two additional variables that play a role in this decision. Firstly, considerations about the quality of the screening test appeared to be frequently mentioned reasons both for accepting and declining prenatal screening. This corresponds with the construct of response efficacy in protection motivation theory (19). Response efficacy indicates to what extent it is expected that the behaviour (i.e. undergoing prenatal screening) is effective in reducing the threat (i.e. having a child with Down syndrome). Secondly, ethical considerations appeared to be important arguments for declining prenatal screening. If prenatal screening, and subsequently, prenatal diagnostic testing, lead to the detection of Down syndrome, the pregnancy can either be continued or terminated. Thus, attitude toward termination of pregnancy is assumed to play a significant role in decision making concerning prenatal screening.

Based on the above-mentioned theories and findings, a comprehensive prenatal screening model was hypothesized that contained the following constructs: perceived probability of having a child with Down syndrome, perceived severity of having a child with Down syndrome, attitude concerning undergoing prenatal screening, subjective norm concerning undergoing prenatal screening, attitude concerning termination of pregnancy, response efficacy, and child-related anxiety. The model is graphically depicted in Figure 1. It was hypothesized that perceived probability, perceived severity, attitude toward termination, and response efficacy all influence one's attitude toward prenatal screening, and will therefore have an indirect effect on prenatal screening intention. It was also hypothesized that attitude and subjective norm would directly affect prenatal screening intention, and that subjective norm affects attitude. Both a positive attitude and a positive subjective norm will increase prenatal screening intention. Child-related anxiety was assumed to be positively related to both attitude and intention to undergo prenatal screening. Using path analysis, the hypothesized relations and the model as a whole were analyzed, tested, and cross-validated.

Figure 1. Hypothesised prenatal screening behaviour model

Methods

Design

Pregnant women attending one of 44 midwifery and gynaecology practices located in several areas throughout the Netherlands were asked to participate in the study. Exclusion criteria were: a gestational age of more than 16 weeks, and no command of the Dutch language. Pregnant women who gave informed consent were offered a prenatal screening test for Down syndrome (either nuchal translucency measurement, or maternal serum screening). This test offer consisted of a sent-home information booklet about prenatal screening, and an oral explanation by the woman's midwife or gynaecologist. The booklet was pilot-tested for comprehensibility, and covered the following topics: characteristics of Down syndrome, age-related risks of having a child with Down syndrome, test procedure, possible test results and their implications, procedure and risks of diagnostic testing, and a decision support section in which advantages and disadvantages of prenatal screening were listed. The participants were asked to fill in a questionnaire after the test offer, but before the test result (if applicable). A sample of 1666 pregnant women gave informed consent to participate in the study, were offered prenatal screening for Down syndrome, and filled in the questionnaire.

Setting

The present study forms part of a larger project that aims to provide more insight into the risk perception, decision making, and psychological well-being of pregnant women who are offered prenatal screening for congenital defects (2; 20). Dur-

ing the time this study was performed, in the Netherlands it was not allowed to offer prenatal screening for Down syndrome routinely to all pregnant women. Prenatal diagnostic testing was exclusively offered to women with an increased risk of having a child with Down syndrome (including all women over 35 years of age). As this study implies the routine offer of prenatal screening to pregnant women of all ages, it required ministerial approval. Once the permit had been granted, and the study also approved by the Ethical Committee of the VU University Medical Center, the midwifery and gynaecology practices initiated the recruitment process for participants.

Measures

The questionnaire included the following constructs:

- Perceived risk of having a child with Down syndrome was measured using a seven-point item, ranging from 1 ("very low") to 7 ("very high").
- Perceived severity of having a child with Down syndrome was measured using a 10 cm. visual analog scale, ranging from 0 ("the best that can happen to me") to 10 ("the worst that can happen to me").
- Attitude toward termination of pregnancy was measured using a scale that consisted of four five-point semantic differentials: "In my opinion, the possibility of termination of pregnancy if the child appears to be severely handicapped, is... bad – good, frightening – not frightening, not reassuring – reassuring, not self-evident – self-evident". Mean scores were calculated, so the scale ranged from 1 (very negative attitude) to 5 (very positive attitude). The scale was internally consistent, with Cronbach's alpha = 0.78.
- Response efficacy of the screening test was measured using seven five-point questions about different aspects of the perceived efficacy of the test (e.g. perceived informativeness of the test result, perceived effort of undergoing the test). Sum scores were calculated, resulting in a possible range from 7 (low perceived test efficacy) to 35 (high perceived test efficacy).
- Attitude toward having a prenatal screening test was measured by a scale that consisted of four five-point semantic differentials: "In my opinion, testing for congenital defects during my pregnancy, is... bad – good, frightening – not frightening, not reassuring – reassuring, not self-evident – self-evident". Mean scores were calculated, so the scale ranged from 1 (very negative attitude) to 5 (very positive attitude). The scale was internally consistent, with Cronbach's alpha = 0.79.
- Subjective norm was measured by assessing normative beliefs and weighing these for motivation to comply (14). Normative beliefs were assessed for three

referents: the woman's partner, her midwife/gynaecologist, and another significant referent (respondents could nominate someone themselves; most frequently mentioned referents were 'my mother' and 'a friend'), by three five-point items. For instance: "I think my partner wants me to...", with answer categories ranging from "certainly accept the test" to "certainly decline the test". The respondent's motivation to comply with these normative beliefs was measured by three five-point items. For instance: "I find my partner's opinion about accepting or declining the prenatal test...", with answer categories ranging from "very important" to "very unimportant". A final subjective norm scale was constructed by calculating the mean of the three products of normative beliefs with motivation to comply. This scale ranged from 1 (strong subjective norm to decline the screening test) to 5 (strong subjective norm to accept the test). The internal reliability of this scale was 0.59.

- Anxiety was measured using the first subscale of the Pregnancy Related Anxieties Questionnaire (21). This subscale assessed the fear of bearing a physically or mentally handicapped child, and consisted of four five-point items. Mean scores were calculated, so the scale ranged from 1 (low anxiety) to 5 (high anxiety). In our sample, Cronbach's alpha was 0.88.
- Intention to test was measured by a five-point item: "Are you intending to have the prenatal screening test done?". The scale ranged from 1 ("absolutely not") to 5 ("absolutely"). We used the intended choice instead of actual behaviour because LISREL requires a continuous dependent variable. In our study, intention to undergo prenatal screening was strongly correlated with test uptake (Spearman's correlation coefficient: 0.91), so intended choice is considered to be a reliable measure of the actual decision. We therefore presume that the associations with intended choice will also apply to actual prenatal screening behaviour.

Analysis

The path analysis was performed using LISREL 8.54. As the full information maximum likelihood (FIML) procedure was used, missing data were only imputed to calculate the variance-covariance matrix. The analysis consisted of three steps. In the first step, the adequacy of the hypothesized model was assessed by investigating its goodness of fit. As the χ^2 -statistic is sensitive to sample size, other fit indices were included to assess the model fit: the root mean square error of approximation (RMSEA), the standardized root mean square residual (SRMR), the adjusted goodness of fit index (AGFI) and the non-normed fit index (NNFI, is equal to the classic Tucker-Lewis index) (22; 23). For the RMSEA and the SRMR, values close to zero indicate good model fit, and for the AGFI and the NNFI, values close to one (24). In

a following step, modified models were tested. Modifications based on modification indices were only executed when this was theoretically justified. To avoid overestimation of the model's performance, split-sample cross-validation was used to validate the path model. The sample was randomly split into two mutually exclusive sub-samples, both containing 50% of the data. The initial model was evaluated and modified (the first two steps of the analysis) using the first sub-sample (development sample), while the second sub-sample (validation sample) was reserved for final validation (the third step of the analysis).

Table 1. Means (standard deviations) and bivariate correlations

	Means (SD)	1	2	3	4	5	6	7
1. Intention	2.9 (1.8)							
2. Perceived risk	2.5 (1.1)	0.05						
3. Perceived severity	6.7 (2.7)	0.23***	0.07*					
4. Attitude toward termination	2.8 (1.0)	0.28***	-0.03	0.26***				
5. Response efficacy	24.1 (3.1)	0.42***	0.04	0.09**	0.21***			
6. Attitude toward prenatal screening	3.1 (0.9)	0.61***	0.06*	0.25***	0.51***	0.35***		
7. Subjective norm	3.0 (0.9)	0.79***	0.03*	0.17***	0.27***	0.41***	0.54***	
8. Anxiety	2.4 (0.8)	0.20***	0.32***	0.20***	0.04	0.09**	0.11***	0.16***

* $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

Table 2. Goodness of fit indices of the models

Index	Initial model	Modified model	Final model	Cross-validation
χ^2 -statistic (df)	24.1 (4)	56.9 (9)	37.0 (9)	35.6 (9)
RMSEA (90%CI)	0.08 (0.05-0.11)	0.08 (0.06-0.10)	0.06 (0.04-0.08)	0.06 (0.04-0.08)
SRMR	0.02	0.05	0.03	0.03
AGFI	0.92	0.92	0.95	0.95
NNFI	0.91	0.91	0.95	0.95

Results

Characteristics of the participants and test uptake rates are described in detail elsewhere (2). Compared with the general pregnant population in the Netherlands, participants in our study were on average more highly educated. The mean age of the participants was 31 years of age. Means and correlation coefficients of the model variables are shown in Table 1. Intention to undergo prenatal screening was significantly correlated with perceived severity, attitude toward termination of

pregnancy, response efficacy, attitude toward prenatal screening, subjective norm, and child-related anxiety.

Step 1: initial model

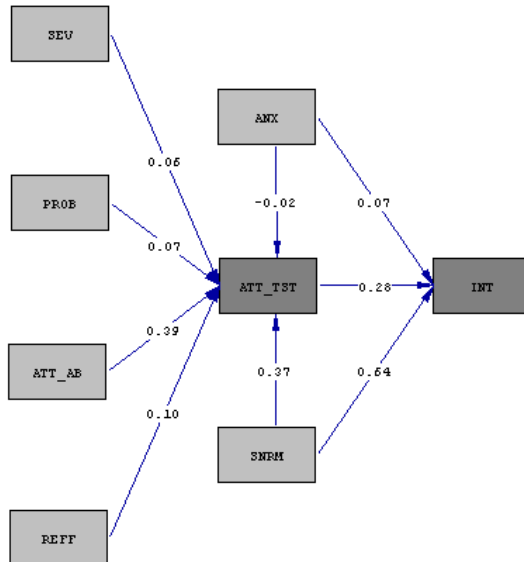
Path analysis of the initial model in the development sample showed that all paths were statistically significant, except for the path between anxiety and attitude toward prenatal screening. Standardized path coefficients are shown in Figure 2. Fit indices of the initial model indicate a moderate fit. (Table 2).

Step 2: model modifications

Three modifications to the initial model were made. First, perceived risk and perceived severity were considered basic ingredients of the decision whether or not to undergo prenatal screening. As they appeared to be only weakly related to attitude toward prenatal screening, we wanted to test the significance of other paths. Several studies found that perceived risk and perceived severity may influence anxiety (25; 26). So, we modified the model by deleting the paths between perceived risk and attitude toward prenatal screening, and between perceived severity and attitude toward prenatal screening, on the one hand; and added the paths from perceived risk to anxiety, and from perceived severity to anxiety, on the other hand (thus shifting those paths from attitude to anxiety) (Figure 3). This modification step led to a small improvement of the fit indices (Table 2).

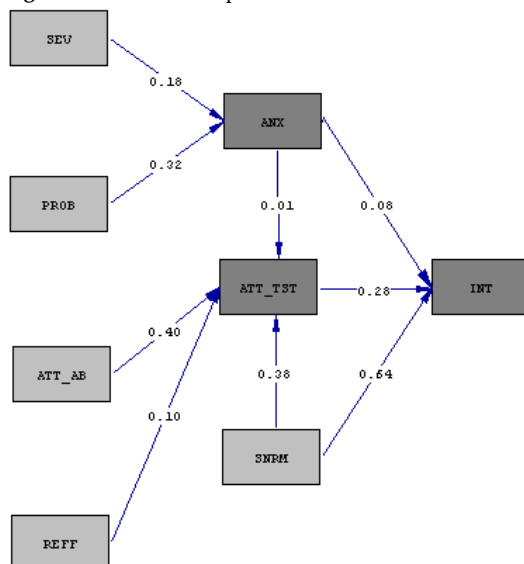
A second and third modification were made by deleting the non-significant path between anxiety and attitude toward prenatal screening, and by adding the direct effect from subjective norm to anxiety (modification index = 54.6), thus indicating that a strong subjective norm to undergo prenatal screening is associated with increased anxiety about the health of the fetus. These modifications led to our final model (Figure 4). This final model showed acceptable model fit, and explained 70% of the variance in the development sample.

Figure 2. Standardized path coefficients of initial model



Abbreviations: SEV: perceived severity; PROB: perceived risk; ATT_AB: attitude toward abortion; REFF: response efficacy; ANX: child-related anxiety; ATT_TST: attitude toward prenatal screening; SNRM: subjective norm; INT: intention to test

Figure 3. Standardized path coefficients of modified model



Abbreviations: see Figure 2

Step 3: cross-validation

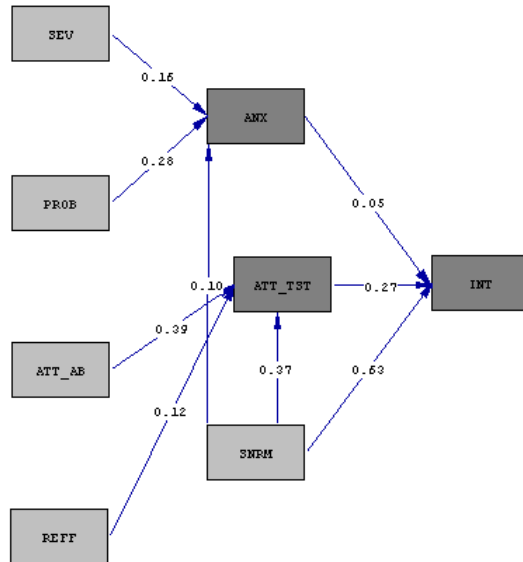
As split-sample cross-validation was used for model validation, the final model that was developed on a random sub-sample of the data was validated on the other mutually exclusive sub-sample (validation sample). Applying the final model to the validation sample showed that the standardized path coefficients were largely in accordance with the development sample (Figure 5). As can be seen from the last two columns of Table 2, cross-validation resulted in a model fit that was almost as good as in the development sample. The model explained 65% of the variance in the validation sample.

Discussion

A hypothesized prenatal screening model was tested. The results of the univariate analyses showed that all variables were significantly associated with intention to test, except for perceived risk of having a child with Down syndrome. Path analysis showed that, after a few modifications, the model had a reasonable fit, implying that the hypothesized model was largely confirmed. After cross-validation, the model still showed acceptable fit. Two variables appeared to play a central role in decision making concerning prenatal screening: attitude toward undergoing prenatal screening, and subjective norm concerning undergoing prenatal screening. Perceived risk and perceived severity appeared to be determinants of anxiety; however, anxiety was only a weak predictor of intention to undergo prenatal screening.

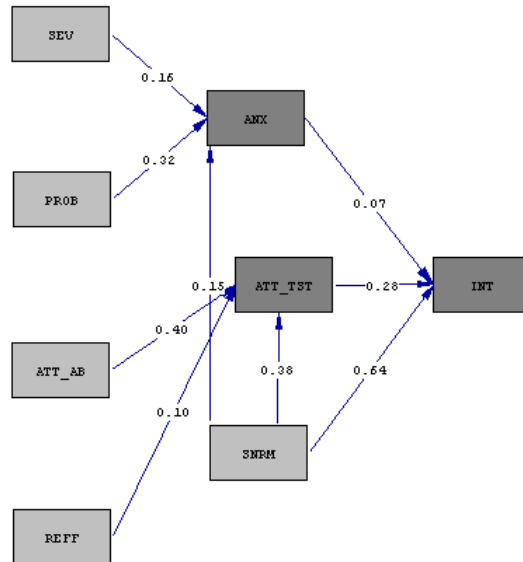
Attitude and subjective norm concerning prenatal screening appeared to significantly influence intention to undergo prenatal screening. These variables, both originating from the TPB, accounted for the major part of the variance that our model explained. These findings were in accordance with a study of Michie, Dormandy, French and Marteau (27) in which prenatal screening uptake was regressed on the TPB variables. As we used multiple path analysis, the effect of subjective norm was adjusted for the other variables in the model. For instance, in addition to a woman's attitude, her decision whether or not to accept the test is mainly determined by the subjective norm she experiences. Women who felt a subjective norm of, for example, their midwife or gynaecologist to accept prenatal screening, had a greater intention of undergoing prenatal screening, and subjective norm to decline prenatal screening led to a lesser intention to test. However, in accordance with the ethical principle of autonomy, a prenatal screening decision should be guided by the decision maker's own beliefs and attitudes, and not by that of the midwife or gynaecologist, for example. This is why, especially in the

Figure 4. Standardized path coefficients of final model



Abbreviations: see Figure 2

Figure 5. Standardized path coefficients of final model applied to validation sample



Abbreviations: see Figure 2

context of prenatal screening, non-directive counselling is advocated. Therefore, our finding that subjective norm is a main determinant of prenatal screening intention is noteworthy. Conversely, the major role of subjective norm could also be interpreted in a way that pregnant women, facing this complex decision, seek support and advice from their circle of family and friends (28). In this train of thought, the effect of subjective norm might not be interpreted as negative (social pressure), but rather as an indication of autonomous decision making using advice from the environment (social support). Further research should examine the potential influence of midwives and gynaecologists on pregnant women's screening decisions.

The variable 'subjective norm' consisted of subjective norms from three persons: the pregnant woman's midwife or gynaecologist, her partner, and another important person. One could argue that the influence of the partner must not be seen as subjective norm, since the partner is part of the pregnant couple and, accordingly, one of the decision makers. On the other hand, the partner's opinion can deviate from the woman's view. From several qualitative interviews we performed with pregnant women (unpublished data), it appeared, for example, that although some women themselves were not intending to undergo prenatal screening, they accepted the test because their partner wanted it. In this way, the partner's influence may be considered as a subjective norm experienced by the pregnant woman, rather than as being part of the decision making process of the pregnant couple.

Perceived risk and perceived severity of having a child with Down syndrome were both very weakly associated with a pregnant woman's attitude toward undergoing prenatal screening (Figure 2). This seems not to correspond with the expectancy-value model of attitude, in which attitude is assumed to be based on beliefs about the probability and evaluation of the outcomes (14). However, in our model we included the beliefs and evaluations of only one outcome (having a child with Down syndrome), while other outcomes may be salient to pregnant women considering prenatal screening. For instance, as prenatal screening provides an individualized risk estimation, a certain diagnosis can only be achieved by amniocentesis or chorionic villus sampling, which involves a risk of iatrogenic termination of pregnancy. Many pregnant women were aware of this disadvantage of prenatal screening, since this risk of prenatal diagnostic testing was a frequently mentioned reason for declining prenatal screening (2). Therefore, attitude toward prenatal screening might also have been influenced by perceived probability and perceived severity of this potential outcome (iatrogenic abortion). Our model for decision making concerning prenatal screening showed that attitude toward prenatal screening was mainly influenced by a woman's attitude toward termination, but also by the perceived efficacy of the screening test. This indicates that a pregnant woman's general attitude toward prenatal screening is based on both ethical con-

siderations (attitude toward termination) and technical considerations (efficacy of the test).

Perceived risk and perceived severity were significant determinants of the level of child-related anxiety the pregnant woman experienced. However, since the standardized path coefficient of anxiety to intention was only 0.06, their (indirect) effect on intention to undergo prenatal screening was very small. This is not in line with Green et al. (1), who concluded that attitude toward the abnormality and perceived risk of the abnormality were major determinants of undergoing prenatal screening. Apparently, in our study, considerations about the severity of Down syndrome or the risk of having a child with this disorder played a limited role in the decision whether or not to undergo prenatal screening, and other factors are more important in the decision making process than considerations about risk and severity of having a child with Down syndrome. This might be caused by a lack of understanding of the risks and consequences of having a child with Down syndrome. Another possible explanation is that the exact levels of perceived risk and perceived severity do not play a part, since decision making concerning prenatal screening might be insensitive to probability or severity. It has been found that affectively-laden decisions are relatively insensitive to changes in probability (29;30). It is suggested that this principle of probability-insensitivity applies to decision making about prenatal screening (31). It may be the possibility that the child has Down syndrome that counts, and not the exact probability. This may explain the minor role of risk perception in our model. An analogous reasoning might be valid for perceived severity: It may be the fact that Down syndrome is a severe disorder that counts, and not the exact magnitude of its disutility. However, since the principle of probability-insensitivity is assumed to be restricted to low probabilities, the marginal role of risk perception in our study might not be translated to decisions that involve high risks (for instance breast cancer screening). This might explain why, for example, van Dijk et al. (32) found that intention for prophylactic mastectomy was influenced by the perceived risk of breast cancer.

Nowadays, there is increasing interest in the role that emotional responses play in decision making processes. Path analysis showed, however, that child-related anxiety was a poor predictor of intention to undergo prenatal screening. This confirms our earlier finding that pregnant women who underwent prenatal screening were only slightly more anxious about the health of their baby (33). This suggests that anxiety plays a marginal role in decision making concerning prenatal screening. This implies that, while prenatal screening is aimed at giving more information about the health of the baby, increased anxiety about the baby's health does not have a high impact on the intended choice to have a prenatal test carried out. A possible explanation of this finding might be that the role of anxiety is

dominated by the effect of other factors. For instance, a pregnant woman can be very anxious about the health of her baby, but she may believe that the test would not give her certainty about it, or she may be opposed to termination of pregnancy. This woman would not accept prenatal screening, despite being very anxious.

A limitation of our study is that path analysis was based on cross-sectional data, implying that the model consists of correlational paths and not of causal relations. Therefore, the causal interpretations of our findings should be considered with care.

In conclusion, this study increases the understanding of decision making concerning prenatal screening. Subjective norm and attitude toward prenatal screening, but not perceptions of risk and severity of having a child with Down syndrome, seem to play a major role in the decision making process. As attitude toward undergoing prenatal screening was mainly determined by attitude toward termination, this attitude may be closely related to one's moral values. So, as social and moral considerations seem to be important for pregnant women in their decision making about prenatal screening, counselling should take into account these aspects of the decision. Therefore, it is suggested that informing and counselling about prenatal testing should not be limited to the communication of (risk) information only, but that it should include discussion about the counselee's moral values and social context.

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Chapter 3

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Chapter 4

Cognitive and emotional components of risk perception and the decision to undergo prenatal screening for Down syndrome

Abstract

This study aimed to investigate whether a cognitive component and an emotional component of risk perception could be identified, using two different risk perception scales. Furthermore, the role of the different risk perception components in the decision to accept or decline prenatal screening was assessed. In a sample of 1650 pregnant women, risk perception (both a numerical and a verbal scale), age-related risk, child-related anxiety, and the intended choice to undergo the prenatal test were assessed. Factor analysis confirmed that the perception of a risk includes both a cognitive and an emotional component. Furthermore, logistic regression analysis showed that cognitive risk perception was significantly associated with the intended choice on prenatal screening, while emotional risk perception was not significantly associated with prenatal screening. However, the role of both risk perceptions in prenatal screening behaviour seemed to be limited. The implications for prenatal screening decision making are discussed.

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Introduction

It is generally recognized that healthcare decisions should be informed decisions (1-3). An important prerequisite for informed decision making is knowledge about and understanding of the risks and benefits of the various options, and their potential consequences (4-8). Therefore, risk communication is a standard part of counselling for treatment or screening decisions (9). Although risk communication is a critical component of the counselling process, research has shown that people find it difficult to adequately understand risks (7;10-13). Within the prenatal testing context, it was found that pregnant women overestimated their personal risk of having a child with Down syndrome, both before and after prenatal counselling (14;15). Similar results were found in studies examining women's perceptions of the risk of having hereditary breast cancer: even after counselling the perception of the risk of getting breast cancer remained inaccurate (16-20).

Risk perception is a central construct in many decision theories and health behaviour theories (21;22). In general, these theories assume that an individual's perception of a risk of, for instance, getting a certain disease, motivates health behaviour, rather than the actual risk.(23;24). Many studies indeed showed that perceived risk is a better predictor of behaviour than actual risk (25-28). Moreover, studies applying the health belief model repeatedly found that risk perception was a significant predictor of health behaviour (29). However, these correlations are generally rather small. For instance, in a review, McCaul et al. found a small but significant effect size ($r = 0.16$) for the relation between perceived risk and breast cancer screening (30). In general, in the cancer screening context it was concluded that risk perception was at best weakly correlated to behaviour (23;31;32). Thus, the role of perceived risk in health decisions is not clear-cut. This is also reflected by different studies in the prenatal testing context. Marteau et al. found that the uptake of a prenatal test was associated with a higher perceived risk of having an affected child (26). Moreover, a review of Green et al. found that the perception of risk of having a baby with an abnormality was an important predictor of prenatal genetic screening behaviour (33). Oppositely, French et al. and Marteau et al. reported that risk perception was not associated with test uptake (34;35).

Risk perception is considered to be a multidimensional concept, comprising both cognitive and emotional components (36-43). The importance of the role of affect and emotion in risk perception has been acknowledged increasingly in recent years (41;44;45). People react to the prospect of risk at two levels: they evaluate the risk cognitively, and they react to it emotionally (41). In other words, risk perception is based on what people think about the risk, but also on how people feel about it (38;40). Windschitl described these risk perception components as:

“beliefs about the objective probability of a hazard” versus “intuitive feelings about one’s vulnerability to the hazard” (38). We refer to these distinct components of risk perception as: cognitive risk perception, and emotional risk perception. This relates to the affect heuristic, which assumes that people use the emotional evaluation of a risk for decision making, rather than the deliberate, cognitive evaluation (40;44;45). People seem to rely even more on the affect heuristic if the decision is complex, time-limited, or emotionally laden (45). This applies to many health care decisions in general, and to decisions about prenatal screening in particular. The decision whether or not to undergo prenatal screening is a complex and emotional choice, and has to be taken in a limited amount of time. Information about different risks, tests, and outcomes needs to be processed, and the decision has to be taken in a specific gestational age frame (e.g. for a first trimester test: 11-14 weeks). As prenatal screening involves values and feelings toward handicap, pregnancy, and termination of pregnancy, decision making on prenatal screening is considered as emotion-laden. Given these specific aspects of the prenatal screening context, it is expected that emotional risk perception would play a more significant role in the decision making than cognitive risk perception.

We have performed several qualitative interviews with pregnant women who were in the process of making a decision whether or not to undergo prenatal screening for Down syndrome, about their thoughts and feelings about prenatal screening, and their risk perceptions (unpublished data). One of the interviewees indeed demonstrated the presence of the different dimensions of risk perception. (see Box 1). In the present study a representative sample of pregnant women were offered prenatal screening for Down syndrome (46). It was aimed to investigate whether a cognitive component and an emotional component of the perception of the risk of having a child with Down syndrome could be discerned using two different risk perception scales: numerical and verbal, respectively. It was assumed that the numerical scale measures the cognitive risk perception component, by asking for one’s belief about the magnitude of the risk, and that the verbal scale measures the emotional component of risk perception, by asking for one’s feeling about the vulnerability to the risk (38;47). Numerical risk perception was assumed to be more related to the objective risk of getting a child with Down syndrome than the verbal risk perception, while the latter would be more related to emotions such as one’s anxiety to get a child with Down syndrome. We hypothesized that factor analysis would reveal a cognitive factor containing numerical risk perception and an objective variable (i.e. age-related risk), and an emotional factor containing verbal risk perception and an emotional variable (i.e. child-related anxiety). The second aim of this study was to assess the role of these risk perception components in the decision whether to accept or decline prenatal screening for Down syndrome.

As this decision is a complex, emotionally laden decision, we hypothesized that the emotional risk perception is a better predictor of prenatal screening behaviour than the cognitive risk perception.

Box 1. An example of how a pregnant woman thinks about her pregnancy-related risks

Interviewer (talking about the risk of an iatrogenic abortion due to prenatal diagnostic testing): "What do you think about that risk?"

Pregnant woman: "It's logical, you puncture into something that ought to be an entirety. So, yes, you're not able to get around this. But, in my feeling, it's simply very large, that chance. *It's a very small chance, I know, but my feeling tells me that it's very large.* That's why I say, I'd rather not."

Subjects and methods

Setting

The study presented in this paper is part of a larger research project, which is designed to give more insight into the risk perception, informed decision making and psychological well-being of pregnant women who are offered prenatal screening for congenital defects (8;46). At the time this study was performed prenatal screening for Down syndrome was not allowed to be offered routinely to pregnant women in the Netherlands, but only to pregnant women over 35 years of age, and to women with an otherwise increased risk. For this reason it was necessary to ask permission from the Minister of Health before starting the study. After a recommendation from the Health Council of the Netherlands, the permit was granted (48). The present study was also approved by the Ethical Committee of the VU University Medical Center

Design

Midwifery and gynaecology practices in various parts of the Netherlands were approached, 44 of which consented to participate. Pregnant women attending one of these practices from May 2001 till May 2003 were asked to participate in the study. Only women with a gestational age of at most 16 weeks and command of the Dutch language were approached. Pregnant women who gave informed consent were offered a prenatal screening test that provides an individualized risk estimation of having a child with Down syndrome. The participants were asked to fill in a questionnaire after the test offer, but before the eventual test result. A sample of 1650 pregnant women were offered the screening test and filled in the questionnaire.

The screening test offer consisted of a sent-home booklet containing information about the particular test, and a standardized oral explanation by the woman's midwife or gynaecologist during a consultation. The following topics were covered in the information booklets: characteristics of Down syndrome, age-specific risks of Down syndrome, procedure of the screening test, meaning of the possible test results, options available after a positive test result (i.e. an increased risk that the child has Down syndrome), procedure and risks of the diagnostic tests. The risk of having a child with Down syndrome increases exponentially from 1 in 1,400 for a woman of 25 years of age, to 1 in 400 for a woman of 35 years of age, to 1 in 30 for a woman of 45 years of age. The booklet paid special attention to the decision making process, and several advantages and disadvantages of prenatal screening were listed. The booklets were pilot-tested for comprehensibility.

Measures

Risk perception of having a child with Down syndrome was measured by two different scales: The numerical risk perception question asks for the participant's estimate of the magnitude of the probability of having a child with Down syndrome. Risk perception was measured on a numerical 10-points scale (ranging from 1 in 50,000 to 1 in 10). The verbal risk perception question asks for the participant's feeling about the risk of having a child with Down syndrome. Risk perception was measured on a verbal 7-points scale (ranging from very small to very large). The actual age-related risk was based on the tables presented by Bray et al. (49). The risk figures were categorized into the categories of the numeric risk perception question. Child-related anxiety was measured using the first subscale of the Pregnancy Related Anxieties Questionnaire Revised (PRAQ-R) (50). This scale assesses the fear of bearing a physically or mentally handicapped child, and consisted of 4 five-point Likert items. The mean scores were used, so the minimum score on this subscale is 1 (low anxiety) and the maximum score is 5 (high anxiety). The scale was internally consistent: Cronbach's alpha was 0.88. The intended choice whether or not to have the prenatal screening test done was measured with a single yes/no question.

Analysis

Factor analysis

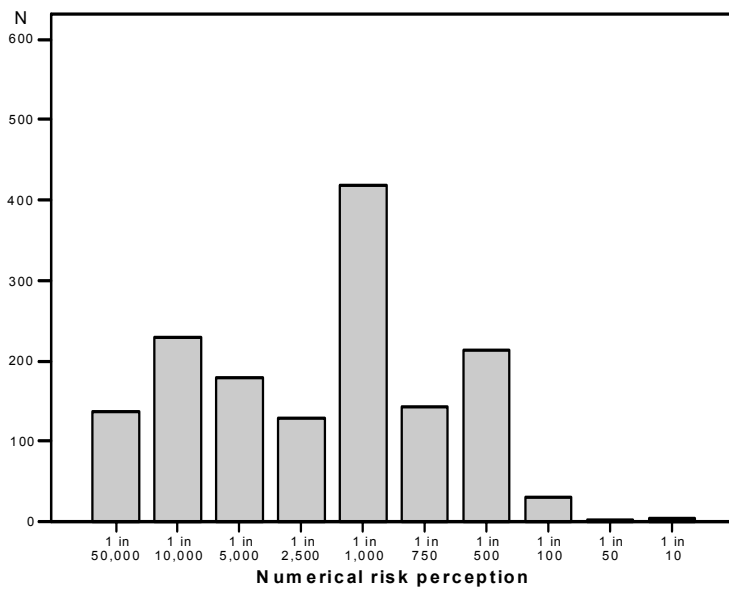
To investigate whether a cognitive component and an affective component can be found in the perceived risk of having a child with Down syndrome, factor analysis was carried out. Four variables were used for this analysis: numerical risk perception, verbal risk perception, age-related risk, and child-related anxiety. If the verbal risk perception question measures the emotional component of risk perception,

then we expect that this question will form one factor with child-related anxiety. If the numerical risk perception question measures the cognitive component of risk perception, then the numerical question should form one factor with the actual age-related risk. To determine the number of extracted factors, the eigenvalues were used (cut-off value of 1), and the scree-plot was analysed. As it was anticipated that the factors would be interrelated, a factor structure that allowed for interdependence was used (promax rotation).

Regression analysis

The role of risk perception in the decision making process was evaluated using logistic regression analysis including the two risk perceptions as independent variables and the intended choice on prenatal screening as dependent variable. If decision making on prenatal screening is primarily based on emotion then we expect that emotional risk perception, rather than cognitive risk perception, will be predictive for the decision to undergo prenatal screening. The analysis was controlled for background variables (age, education, religiosity)

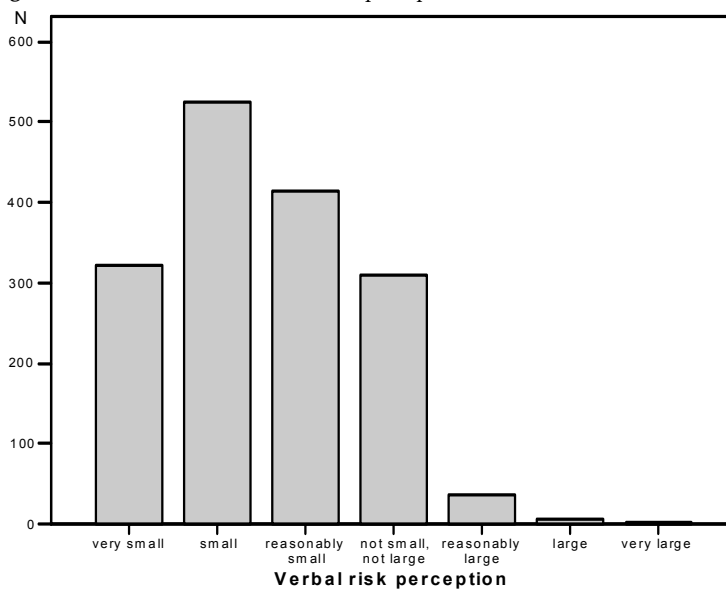
Figure 1: Distribution of the numerical risk perception scores



Results

The mean age of the participants was 31 (SD = 3.6; range: 18-42). The age-specific risk at 31 years of age is 1 in 725 (49). According to the categories of the numerical risk perception scale, this mean age-related risk was 6.2 (SD: 1.0). The mean numerical risk perception was 4.3 (SD: 2.0), and the mean verbal risk perception score was 2.5 (SD: 1.1). Distributions of the numerical and verbal risk perception scores are presented in Figure 1 and Figure 2. The mean level of child-related anxiety was 2.4 (SD: 0.8). Of the pregnant women, 45% were intending to have the prenatal test done.

Figure 2: Distribution of the verbal risk perception scores



The factor analysis extracted two factors with eigenvalues greater than one. These factors explained 42.9 and 25.8% of the variance. After promax rotation, the eigenvalues of these factors were 1.50 and 1.41, respectively. The factor correlation was 0.24. As can be seen from Table 1, the first factor consisted of numerical risk perception and age-related risk was discerned as a cognitive component (rotated factor loadings: 0.73 and 0.89, respectively). The second factor consisted of verbal

risk perception and child-related anxiety, and was identified as the emotional component (rotated factor loadings: 0.67 and 0.90, respectively).

Table 1. Factor analysis: promax rotated pattern matrix

Variables	Component 1	Component 2
Numerical risk perception	0.73	0.21
Verbal risk perception	0.27	0.67
Age	0.89	-0.18
Pregnancy-related anxiety	-0.19	0.90

Multiple regression analysis revealed that, controlling for numerical risk perception and background variables, verbal risk perception was not significantly associated with the decision on prenatal screening (OR: 1.01, 95%CI: 0.90-1.13). Controlling for verbal risk perception and background variables, the numerical risk perception did significantly predict prenatal screening decision making (OR: 1.10, 95%CI: 1.03-1.17). However, as both regression coefficients are very small, and the sample is very large, it is suggested that both risk perceptions play a very modest role in predicting intended choice on prenatal screening.

Table 2. Logistic regression analysis*

	Beta (SE)	Wald	OR	95%CI
Verbal risk perception	0.01 (0.06)	0.03	1.01	0.90-1.13
Numerical risk perception	0.09 (0.03)	7.30	1.10	1.03-1.17

*controlled for age, education, religiosity

Discussion

Risk perception is considered to be a multidimensional concept, with both cognitive and emotional components. Using a numerical and a verbal risk perception scale, the factor analysis confirmed this multidimensionality. Two factors were extracted: The first factor contained numerical risk perception and age-related risk, and was identified as a cognitive factor. The second factor consisted of verbal risk perception and child-related anxiety, and was defined as an emotional factor. The hypothesis that emotional component of risk perception was a better predictor of prenatal screening intention than the cognitive component could not be confirmed, as the verbal risk perception scale (representing the emotional component of risk perception) did not significantly predict the intended choice on prenatal testing.

Oppositely, cognitive risk perception was the better predictor of intended choice. This indicates that the decision whether or not to undergo prenatal screening is less based on emotions than we expected. It may be that prenatal screening is more a value-laden issue, than an emotionally laden issue. It may be that prenatal screening decision making is less motivated by emotional risk perception or emotions like anxiety about the health of the unborn child, and more by a pregnant woman's values toward prenatal screening and termination of pregnancy.

Our findings are not in line with the study of Marteau et al. who concluded that risk perception (measured on a verbal scale) predicted the uptake of prenatal diagnostic testing (26). This might be related to the fact that prenatal diagnostic testing involves a risk of an iatrogenic abortion (due to the procedure). It could be that this makes the decision about prenatal diagnostic testing more a weighing of risks, as compared to the decision about prenatal screening (which is a risk-free test). Another possible explanation might be that this study used a sample of older pregnant women (over 38 years of age), while our study concerned an unselected population of pregnant women of all ages. Young pregnant women have low age-related risks of having a child with Down syndrome, and they might accept prenatal testing primarily for other reasons than perceived risk. These other reasons include 'gaining knowledge', 'curiosity', 'it doesn't hurt to try', and 'reassurance' (46). Furthermore, older pregnant women run higher risks of having a child with Down syndrome, and they were probably more aware of those risks. So, it is likely that for these women perceptions of risk play a larger role in their decision making. This reasoning is supported by another study of Marteau et al. in which they examined several psychological models to predict uptake of prenatal screening. (35). In this study, in which they used a sample of women of all ages, excluding those over 38 years, it was found that verbal risk perception of having a child with a serious abnormality did not differ significantly between test decliners and test acceptors. This latter finding is in line with the results of present study.

As this study shows that risk perception includes both cognitive and emotional components, we propose that counselling about risks and benefits of a treatment or screening, should not focus merely on the correct understanding of the risk figures, but it should also give attention to the emotional aspects of risks. Consideration of both cognitive and emotional aspects of risk perception may give a better insight into the decision making process of patients or counselees. This is particularly important because the cognitive and emotional evaluations of a risk might diverge from each other (41) as was illustrated by the case in Box 1. A counselee may understand, for example, that the risks of a certain treatment or screening are very small, but at the same time the emotional reaction could diverge from

this cognitive evaluation. This individual might be afraid of this risk and its occurrence might feel so real, that she will decline the treatment or screening anyway.

As the different risk perceptions were not, or only weakly, associated with the intended choice on prenatal screening, prenatal screening decision making is probably mainly based on other considerations. It was found that attitude toward the test, subjective norm, and attitude toward abortion were important factors in the decision making about prenatal testing (33). Additionally, the characteristics of the screening test appeared to be both a reason to accept the test ('it doesn't hurt to try'), as a reason to decline the test ('the test gives no certainty') (46). Further research has to be performed to assess which factors play a role in the decision making process about prenatal screening, and how these factors interrelate with each other.

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Chapter 5

Are counsellors' attitudes influencing pregnant women's attitudes and decisions about prenatal screening?

Abstract

It is generally recognised that choices concerning treatment or screening should be people's own, autonomous decisions. However, in the context of genetic counselling many studies found that counsellors deviate from nondirectiveness, or that subjective norm influences behaviour. The present study aimed to investigate whether prenatal counsellors (midwives, gynaecologists) influence pregnant women's decisions and their attitudes regarding prenatal screening. It was hypothesised that uptake rates and attitudes would be associated with the counsellor's attitude toward prenatal screening. Pregnant women attending their midwifery or gynaecology practice were asked to fill out postal questionnaires before and after they were offered prenatal screening for Down syndrome. Their prenatal counsellors also filled in a questionnaire. These questionnaires assessed attitudes toward prenatal screening and background variables. The study sample consisted of 945 pregnant women, being guided by 97 prenatal counsellors. Multilevel regression analyses revealed that neither uptake rates, nor attitude toward prenatal screening were significantly predicted by counsellors' attitudes toward prenatal screening. It is suggested that the advice these pregnant women reported to have received, should rather be interpreted as an indication of shared decision making and social support than of social pressure and undesired influence.

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van den Berg M, Timmermans DRM, Kleinveld JH, van Eijk JThM, Knol DL, van der Wal G, van Vugt JMG. Are counsellors' attitudes influencing pregnant women's attitudes and decisions about prenatal screening?

Introduction

Genetic counselling is aimed at informing counsellees about the risks and characteristics of a specific genetic disorder and the different test options available, and at supporting counsellees in their decision making process (1-6). It is generally recognised that decisions concerning whether or not to have a genetic test done should be the counsellees' own, autonomous decisions. Therefore, especially in the context of genetic counselling, the principle of nondirectiveness and value-neutrality is advocated (7;8). Although counsellors acknowledge that nondirectiveness is important in genetic counselling, and that they generally strive for value-neutrality in their counselling, in the everyday practice of genetic counselling, they frequently depart from nondirectiveness (2;7;9;10). Several studies came to the same conclusion about the prenatal counselling practice specifically, and they raise the question of the extent to which pregnant women's decisions on whether or not to undergo prenatal genetic testing can be considered autonomous and uninfluenced (5;11-15).

As counsellors frequently seem to deviate from nondirectiveness, this may be based on their own attitudes and preferences concerning genetic testing. Possibly, counsellors either implicitly hint at their views, or explicitly impose their own views upon counsellees. Several studies suggest that this kind of influence indeed might occur (16;17). However, Dormandy et al. found that healthcare professionals' attitudes toward prenatal screening were unrelated to pregnant women's uptake of prenatal screening, although they indicate that their correlation study was only powered to detect a large correlation (0.5 and over) (18).

In several social cognitive models of health behaviour, the influence of others on people's decisions is recognized. In for instance the theory of planned behaviour subjective norm is one of the determinants of the intention to perform a certain behaviour (19;20). In the context of prenatal testing, a subjective norm consists of a pregnant woman's belief that other persons (e.g. family members, friends, health professionals) think she should have the test done, and her motivation to comply with these persons. Few studies found subjective norm to be a significant predictor of the decision whether or not to undergo a genetic test (21-23). The conclusion that subjective norm – including the opinion of the counsellor – plays an important role in decision making about genetic testing implies that prenatal counsellors might influence pregnant women in their decision of whether or not to undergo a prenatal test.

It is therefore relevant to investigate to what extent counsellors actually exert influence on pregnant women's decisions concerning genetic testing. We performed a study in which we measured attitudes and background variables of both

pregnant women, and their prenatal counsellors (midwives, gynaecologists). This study aimed to investigate whether prenatal counsellors influence pregnant women's decision making process regarding prenatal screening. The three research questions were: Do prenatal counsellors' attitudes toward prenatal screening predict pregnant women's intended test uptake? Do prenatal counsellors' attitudes predict pregnant women's post-counselling attitudes toward prenatal screening? Do prenatal counsellors' attitudes predict pregnant women's attitude change from pre- to post-counselling? It was hypothesized that the influence of the counsellor can be detected at these three levels of the decision making process. Furthermore, it was hypothesized that pregnant women's own perceptions of the counsellor's advice (to accept or decline the prenatal screening test) would influence their decision making process, and that the point in time at which the pregnant women made their decision (either before, during of after the counselling) would interact with the influence of the counsellor.

Methods

Design

Midwifery and gynaecology practices in various parts of The Netherlands were approached: 44 consented to participate in the study. Pregnant women who enrolled at one of the participating practices were asked permission by their midwife or gynaecologist to be sent a research information letter and an informed consent form. Pregnant women who were more than 16 weeks pregnant, and women who had no command of the Dutch language were not included in the study. Those women who gave informed consent were offered a prenatal screening test (either the nuchal translucency measurement, or the maternal serum test). Prenatal screening was offered and explained to the participants by means of an information booklet and a standardized oral explanation given by the pregnant woman's prenatal counsellor. The information included characteristics of and risks on the screened disorders, characteristics and procedure of the screening test offered, the possible test results, the options available after a positive test, the characteristics and procedure of prenatal diagnostic testing, and a paragraph focussing on advantages and disadvantages of having the test. The screening tests provide an individualized risk estimation of having a child with Down syndrome, and thus can not provide any certainty about it. Pregnant women who were tested positive (i.e. an increased risk) were subsequently offered a prenatal diagnostic test (amniocentesis or chorionic villus sampling), which gives certainty concerning the presence

or absence of Down syndrome. However, prenatal diagnostic testing involves the risk of induced abortion due to the test procedure.

The participating pregnant women were asked to fill in two postal questionnaires. The first questionnaire was filled in before the offer of the test was made (i.e. before they received the information booklet). This questionnaire assessed the women's background characteristics, and their attitudes toward undergoing prenatal screening. The second questionnaire was filled in after the offer of the test, but before they received the test result. Pregnant women who declined the screening test were sent a second questionnaire at a comparable point in time. The second questionnaire measured attitude toward undergoing prenatal screening, the point in time of decision, the perception of the counsellor's advice, and test uptake. So, attitude toward undergoing prenatal screening was assessed both before the screening offer and consultation, and thereafter. These attitudes will be referred to as pre-counselling attitude and post-counselling attitude. The participating prenatal counsellors were also asked to fill in a postal questionnaire. This questionnaire contained questions concerning demographic variables, characteristics of the practice, and their views and preferences regarding prenatal screening.

Setting

The present study is embedded in a larger research project that aimed to give more insight into the decision making and psychological wellbeing of pregnant women who were offered prenatal screening for congenital defects (24;25). At the time this study was performed, prenatal screening was not offered routinely to all pregnant women, as part of standard antenatal care. As the Population Screening Act states that offering prenatal screening is not allowed without ministerial approval, it was necessary for the minister of health to grant permission for the study (26). As a consequence of this situation, prenatal screening could only be offered to participants in a research setting.

Participants

Pregnant women

1650 participants gave informed consent, and filled in and returned the two questionnaires. These pregnant women were counselled by 144 different prenatal counsellors.

Prenatal counsellors

The group of 144 prenatal counsellors worked in 44 different practices or hospitals. Ninety-seven (67%) of the counsellors filled in and returned the questionnaire.

Combined sample

Combining the two samples, we had to exclude some questionnaires of pregnant women from analysis due to two factors. Firstly, for 460 (28%) pregnant women it was not known by which midwife or gynaecologist they were being counselled, leaving a sample with data of 1190 pregnant women. Secondly, since 33 % of the 144 prenatal counsellors did not return the questionnaire, the pregnant women who were counselled by these midwives or gynaecologists also had to be excluded. The final sample of cases that were available for analysis consisted of 945 pregnant women being guided by 97 counsellors.

Measures

Pregnant women

Demographical variables

The following demographical variables were measured: age, level of education, and degree of religiosity.

Attitude toward prenatal screening

Attitude was measured by a scale using four semantic differentials (for details see (25)). The internal reliability of the scale was 0.79. The change in attitude from pre- to post-counselling was calculated by subtracting the two attitude scores so that a positive change indicated that the participant had become more positive toward undergoing prenatal screening.

Time of decision

The point in time at which the decision was made was assessed by a single question asking when the pregnant woman had made her decision concerning prenatal screening (before, during, or after the consultation).

Perceived advice

One question asked whether the counsellor had given an advice to have or not to have prenatal screening done. This was measured using the following scale:

- Yes, to have prenatal screening done
- No, but it was clear to me that she/he was positive about the test
- No advice at all
- No, but it was clear to me that she/he was negative about the test
- Yes, she advised me not to have prenatal screening done

As we did not observe the actual consultations these are the women's interpretations of what actually happened. Therefore, it will be referred to as the pregnant

women's perception of whether or not the counsellor had given or hinted at an advice about prenatal screening.

Intended test uptake

Intended prenatal screening behaviour was measured using a single yes/no question. In 96% of the pregnant women their intended choice matched their actual choice.

Prenatal counsellors

Background variables

The following background characteristics of the prenatal counsellors were measured: age, sex, working field, years of experience, number of counselees, urbanization of working area.

Attitude toward prenatal screening

The counsellors' attitude toward prenatal screening was measured using two self-developed scales, both consisting of 7 semantic differentials (good-bad, useful-useless, etc.). The first scale assessed the attitude toward prenatal screening in general, while the second attitude scale assessed the attitude of the counsellor toward routinely offering prenatal screening to pregnant women. These two scales were internally reliable (Cronbach's alpha's were 0.79 and 0.92 respectively).

Analysis

For the description of the characteristics of both pregnant women and prenatal counsellors, the variables were categorized. However, in the multilevel regression analyses continuous variables were used. To investigate the attitude change of the pregnant women, both pre- and post-counselling attitude scores were recalculated into a positive, neutral, and negative category (as we have described elsewhere (25)), and a Wilcoxon signed ranks test was performed.

Multilevel regression analyses were performed to examine the influence of counsellors' attitudes on the decisions and attitudes of pregnant women. By using multilevel analysis, it was acknowledged that pregnant women were hierarchically nested within prenatal counsellors (27). Multilevel models allow for counsellor effects on the outcome variables. Analyses were done using the MLwiN package, version 2.02.

To test the hypotheses, three multilevel models were tested:

Model 1: prenatal counsellors' influence on intended test uptake

In the first model (logistic regression), intended test uptake was the dependent variable, the first-level independent variables were the pregnant women's pre-counselling attitude toward prenatal screening, their attitude change, their perception of the counsellor's advice, and time of decision, the second-level independent variable was prenatal counsellors' attitude toward prenatal screening.

Model 2. prenatal counsellors' influence on post-counselling attitude toward undergoing prenatal screening

In the second model (linear regression), the dependent variable was post-counselling attitude toward prenatal screening, the first level independent variables were pregnant women's perception of the counsellor's advice, and time of decision, and the second level independent variable was the prenatal counsellors' attitude toward prenatal screening.

Model 3. prenatal counsellors' influence on pre- to post-counselling change in attitude

In the third multilevel model (linear regression), attitude change was the dependent variable, the first level independent variables were pregnant women's perception of the counsellor's advice, and time of decision, the second level independent variable was the prenatal counsellor's attitude toward prenatal screening.

It was expected that time of decision might influence the relation between the counsellor's attitude toward screening and a woman's prenatal screening intention (deciding during or after the consultation might increase the possible influence of the counsellor, as opposed to having decided before the consultation). Therefore, in all models the interaction between time of decision and attitude of the counsellor was also tested. All models were corrected for first-level demographic variables.

Table 1. Pregnant women's pre- and post-counselling attitudes

		Post-counselling attitude			
		Negative	Neutral	Positive	Total
Pre-counselling attitude	Negative	195 (22%)	68 (8%)	19 (2%)	282 (32%)
	Neutral	85 (10%)	95 (11%)	71 (8%)	251 (29%)
	Positive	42 (5%)	75 (8%)	222 (26%)	339 (39%)
	Total	322 (37%)	238 (27%)	312 (36%)	872*

*less than 945 due to missing values

Results

Pregnant women

The demographic characteristics of these pregnant women, and their association with test uptake, have been described earlier (28). With regard to the attitudes of pregnant women, before the test was offered 32% of the pregnant women had a negative attitude, 29% had a neutral attitude, and 39% had a positive attitude toward undergoing prenatal screening. After the test was offered significant changes in attitudes toward prenatal screening occurred (Wilcoxon signed ranks test: $Z = -2.94$, $p = 0.003$) (Table 1). Fifty-nine percent of the participants fell in the same category, while 23% of the participants had a more negative attitude and 18% had a more positive attitude. Seventy percent of the women stated that they had made their decision before the consultation, 10% said that they had decided during the consultation, and 20% indicated that they had made their decision whether or not to have prenatal screening after the consultation. Almost three-quarters of the pregnant women stated that their counsellor did not reveal his/her opinion about prenatal screening, while 9% of them indicated that the counsellor was positive about prenatal screening, and 19% indicated that the counsellor was negative about it.

Prenatal counsellors

The characteristics of the prenatal counsellors are described in Table 2. The sample consisted of 11 (11%) gynaecologists and 86 (89%) midwives. Of all the counsellors, 91% were female and 9% male. Almost all of the midwives (99%) were female, and 3 (27%) of the gynaecologists were female. Of the counsellors, 7% had a negative attitude toward prenatal screening, 58% had a neutral attitude, and 35% had a positive attitude. However, 62% of the counsellors were negative towards offering prenatal screening routinely to all pregnant women, while 24% were neutral, and 14% were positive.

Table 2. Characteristics of the prenatal counsellors

	Mean (SD)	N (%)
Age	39 (10.8)	
Sex*		
Male		9 (9%)
Female		87 (91%)
Working field		
Gynaecologist		11 (11%)
Midwife		86 (89%)
Years of experience	12 (9.5)	
Number of counsellees	288 (155)	
Urbanization of working area*		
Not / little		21 (22%)
Moderate		36 (38%)
Strong / very strong		38 (40%)
Attitude toward prenatal screening*		
Negative		6 (7%)
Neutral		52 (58%)
Positive		32 (35%)
Attitude toward routinely offering prenatal screening*		
Negative		56 (62%)
Neutral		22 (24%)
Positive		13 (14%)

* less than 97 due to missing values

Multilevel analyses

Model 1: intended test uptake

Table 3 shows that the counsellors' attitude did not significantly contribute to the prediction of intended test uptake. The pregnant women's pre-counselling attitude toward undergoing prenatal screening, and their pre- to post counselling change in attitude were important predictors of test uptake (OR = 1.38; $p < 0.001$, and OR = 3.06; $p < 0.001$, respectively). The extent to which the pregnant women reported having been advised by their counsellor was also significantly associated with intended test uptake (OR = 4.48; $p < 0.001$).

Model 2: post-counselling attitude toward prenatal screening

The counsellors' attitude did not predict the pregnant women's attitude toward prenatal screening (Table 3). The pregnant women's perception of the counsellor's advice significantly predicted their attitude toward prenatal screening (Beta = 1.75; $p < 0.001$). Moreover, women who decided after the consultation has less positive post-counselling attitudes as compared to those who decided during the consultation.

Table 3. Regression coefficients of the three multilevel models

Model	Dependent variable	Level	Independent variables	Regression coefficient (SE)	OR	Chi ²	df	p	
1	Intended test uptake	1	Pre-counselling attitude	0.32 (0.03)	1.38	90.7	1	<0.001	
			Attitude change	1.12 (0.14)	3.06	62.3	1	<0.001	
			Advice	1.50 (0.16)	4.48	37.0	1	<0.001	
		2	Counsellors' attitude	Time of decision*			0.38	2	>0.5
				during consultation	0.15 (0.36)	1.16			
				after consultation	0.13 (0.25)	1.14			
					-0.001 (0.19)	1.00	0.00	1	>0.5
2	Post-couns. attitude	1	Advice	1.75 (0.23)		60.7	1	<0.001	
			Time of decision*			6.59	2	<0.05	
			during consultation	0.47 (0.47)					
		after consultation	-0.75 (0.34)						
		2	Counsellors' attitude	-0.03 (0.28)		0.01	1	>0.5	
3	Attitude change	1	Advice	0.70 (0.21)		11.2	1	<0.01	
			Time of decision*			4.16	2	>0.1	
			during consultation	-0.08 (0.43)					
		after consultation	-0.64 (0.32)						
		2	Counsellors' attitude	0.27 (0.26)		1.14	1	>0.1	

* reference category: decision made before consultation

Model 3: pre- to post counselling change in attitude

In the third model, it was analyzed whether the counsellor's attitude did predict pregnant women's change in attitude from pre-counselling to post-counselling. This was not the case (Table 3). Attitude change was significantly associated with the perception of the counsellor's advice (Beta = 0.70; $p < 0.01$).

None of the models showed a significant interaction between time of decision and attitude of the counsellor toward prenatal screening.

Discussion

The majority of the pregnant women made their decision concerning prenatal screening before the consultation with their prenatal counsellor, and most women said that their prenatal counsellor did not reveal his/her opinion about prenatal

screening. This could be interpreted as most counsellors being nondirective in their counselling about prenatal screening. However, substantial minorities of participants reported that their counsellor deviated from nondirectiveness either by hinting at or advising positively or negatively about undergoing prenatal screening (9% and 19%, respectively). Interestingly, although a proportion of the pregnant women perceived that they were being advised by their counsellor to accept or decline the screening test, and that this perception of the advice was strongly related with intended test uptake, multilevel analyses could not confirm that counsellors' attitudes toward prenatal screening influence pregnant women's intended test uptake or attitude (or change in attitude) toward prenatal screening. Moreover, as the counsellors' attitude toward prenatal screening did not interact significantly with the time of decision making in any of the models, it can be concluded that even in the group of women who decided during or after the consultation, no influence of the counsellor could be detected.

Our finding that prenatal counsellors' attitudes did not influence pregnant women's decision making is neither in accordance with the frequently mentioned suggestion that counsellors may be hinting at or imposing their own views on counsellees decision making (17), nor with studies that observed counselling sessions and concluded that counsellors indeed did influence counsellees' decisions (e.g. 9;11;13). This could possibly be explained by the fact that the counsellors were explicitly instructed by the researchers that they should avoid giving their personal opinion to pregnant women, so that accepting or declining prenatal screening would be the pregnant women's own choice. Moreover, the information booklet that pregnant women received did pay special attention to the decision making process, and stressed that they should make their own decision. These factors taken together might have resulted in a practice that differs from that in countries where prenatal screening is part of routine prenatal care.

A possible explanation for the discrepancy between pregnant women's perception of the counsellor's advice on the one hand, and the absence of a significant influence of the counsellor on the other hand, could be that the advice counsellors sometimes give does not reflect their own opinion about prenatal screening. Counsellors might give advice that is in line with the attitude of the pregnant women, instead of influencing women's attitude by imposing their own view on prenatal screening. According to this explanation, the advice given by counsellors should be interpreted as an indication of supporting the decision making process. This would imply that our earlier finding that the important role of subjective norm in the decision making process (23) indeed should rather be interpreted as a sign of social support.

This is in line with the more recent interpretation of optimal counselling, in which counselling is defined as a psychoeducational process, which is primarily aimed at supporting and facilitating the decision making process, and not solely at providing information in an as objective, neutral, and nondirective way (29). This conception of the task of genetic counselling sometimes implies being directive, e.g. by tailoring the information based on the specific needs of a counsellee, by trying to give the counsellee a balanced picture of the situation, or by providing emotional support. This is in accordance with Shiloh's formulation of the aim of genetic counselling, which is to help counsellees reach a decision wisely, rather than to reach a wise decision (30). It may be that some directiveness is needed to help a counsellee in making his/her decision wisely. However, it is important to note that the counsellor's directiveness should be limited to the decision making process, and not aimed at the decision outcome (10).

A limitation of our study is that it relates to self-reported measures. For instance, self-reported advice is found to differ from objective measures of advice (9). Therefore, further research should involve the observation of the actual counselling sessions to gain a more objective picture of the potential influence of counsellors on counsellees' decision making, and of the interaction between counsellor and counsellee. In conclusion, although in our study substantial numbers of counsellees reported having been given advice, either implicitly or explicitly, the regression analyses did not confirm the frequently mentioned influence of genetic counsellors' attitudes on counsellees' attitudes and decisions.

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Chapter 6

Are pregnant women making informed choices about prenatal screening?

Abstract

Prenatal screening should enable pregnant women to make informed choices. An informed decision is defined as being based on sufficient, relevant information and consistent with the decision maker's values. This study aims to assess to what extent pregnant women make informed choices about prenatal screening, and to assess the psychological effects of informed decision making. The study sample consisted of 1159 pregnant women who were offered the nuchal translucency measurement or the maternal serum screening test. Level of knowledge, value consistency, informed choice, decisional conflict, satisfaction with decision, and anxiety were measured using questionnaires. Of the participants, 83 % were classified as having sufficient knowledge about prenatal screening, 82% made a value-consistent decision to accept or decline prenatal screening, and 68% made an informed decision. Informed choice was associated with more satisfaction with the decision, less decisional conflict (this applied only to test acceptors), but was not associated with less anxiety. Although the rate of informed choice is relatively high, substantial percentages of women making uninformed choices due to insufficient knowledge, value inconsistency, or both, were found. Informed choice appeared to be psychologically beneficial. The present study underlines the importance of achieving informed choice in the context of prenatal screening

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Introduction

Screening programs should aim at enabling people to make informed choices, rather than achieving as high as possible uptake rates (1-5). The paradigm of informed choice is based on both ethical and psychological considerations. By means of informed choice, the ethical principle of autonomy is respected (5-7), and better psychological outcomes are assumed to be achieved (8). Accepting or declining prenatal screening in particular should be founded on informed choice, because of the risks and moral values that play a part: prenatal screening may lead to diagnostic testing, which involves the risk of an iatrogenic abortion, and, in case of a positive test result, to the option of termination of pregnancy (9).

Although both practitioners and pregnant women consider informed choice as the main objective of offering prenatal screening (3,10), the decision to accept or decline prenatal screening is often not an informed choice (11). Green et al. found that, although women want to make informed choices about screening, and when questioned directly state that their choices were informed, women do not possess the required understanding of prenatal tests to be able to make an informed choice (11).

Several different definitions of informed choice or informed decision making exist. They all include at least the following two dimensions: firstly, the decision should be based on relevant information, and secondly, it should be consistent with the decision maker's values (1, 12-14).

Whether or not a decision is based on relevant information can be assessed by measuring the decision maker's knowledge of the different aspects of a certain screening. Although there is no gold standard regarding what constitutes sufficient, relevant knowledge, there is consensus that, with regard to screening, several points are essential to know: characteristics of the condition for which screening is being offered, characteristics of the screening test, and implications of the possible test results⁸. Research has shown that women lack sufficient knowledge about the different aspects of prenatal screening, which impedes informed decision making (11,15,16).

The second requisite for an informed choice is value consistency. Values are abstract ideals representing a person's beliefs about ideal modes of conduct (17). As attitudes are tendencies to respond to a concrete object with some degree of favour or disfavour (18,19), they can be considered as a reflection of one's values (1). So, with respect to prenatal screening, in order to assess the decision maker's values, attitudes towards prenatal testing for congenital defects could be measured. Subsequently, value consistency can be assessed by comparing one's attitudes with the actual behaviour. Having the screening test done while having a positive atti-

tude towards screening during pregnancy, and declining the prenatal screening test while having a negative attitude towards screening, are both value-consistent decisions.

Although an informed choice should be based on sufficient knowledge as well as value consistency, most studies on informed decision making did not assess both aspects, but rather focused on measures like the health behaviour, knowledge, values or attitudes, or psychological effects, separately (12). Recently, a measure of informed choice (MMIC) was developed and validated by Marteau and colleagues (1,9). This measure is based on a conceptualization of informed choice that respects its multidimensional character and integrates the basic elements of informed choice (i.e. knowledge, attitude, and actual behaviour).

Furthermore, little is known about the actual psychological effects of informed choice. Studies assessing the relation between informed choice and outcome measures like decisional conflict, decision satisfaction, and anxiety, are scarce (11), and reported mixed results. Bekker et al. found that women who made more informed choices expressed less decisional conflict over time about their decision making, but there was no effect on anxiety (20). This association between informed choice and decisional conflict was also found by Michie et al (9). Green et al., however, suggested that informed decision making is associated with more anxiety and less satisfaction with the decision, as compared with uninformed decision making (11). Therefore, it is unclear what the effects of informed choice are.

The aim of this study was to assess to what extent pregnant women who are offered prenatal screening for congenital defects make informed choices. This was done by measuring its basic dimensions – knowledge and value consistency – and combining these two into a measure of informed choice. In addition, this study aimed to assess the psychological effects of informed decision making.

Subjects and methods

Setting

Up till now in the Netherlands –contrary to many other modern western countries– prenatal screening for Down syndrome (DS) and Neural Tube Defects (NTD) is not offered routinely to pregnant women. Only prenatal diagnostic tests are offered routinely to pregnant women over 35 years of age (among women in this age category, the diagnostic test uptake rate is 34% (21)), and to women with an otherwise increased risk. For this reason, it was necessary to ask permission from the Ministry of Health to carry out this study. Therefore, it is important to

stress that the prenatal screening tests that were offered to pregnant women were exclusively offered to the participants in our study.

The study presented in this paper is part of a larger research project, designed to give more insight into the risk perception, informed decision making and psychological well-being of pregnant women who are offered prenatal screening for congenital defects. The research project is a randomized controlled trial in which participants were randomized into one of two intervention groups or into the control group. The intervention consisted of offering prenatal screening for congenital defects. Women in the first intervention group were offered the Nuchal Translucency Measurement (NTM), and women in the second intervention group the Maternal Serum screening Test (MST). The NTM screens for DS in the first trimester of pregnancy, and is done by ultrasound scanning (22). The MST is a blood test in the second trimester of pregnancy ('triple-test'), and screens for DS and NTD (23).

Both prenatal screening tests provide an individualized risk estimation of having a child with one of these disorders, and thus identify a high-risk subgroup within a population of pregnant women. Subsequently, this subgroup of women with an increased risk is offered prenatal diagnostic testing in order to provide a certain diagnosis.

Information booklet

The test offer consisted of a sent-home booklet containing information about the particular test, and a standardized oral explanation by the woman's midwife or gynaecologist during a consultation. The following topics were covered in the information booklets: characteristics of DS and NTD (information about NTD is covered only in the MST booklet), age-specific risks of DS, population risk of NTD, procedure of the screening test, the meaning of a negative or a positive test result, options available after a positive test result, procedure of the diagnostic tests: amniocentesis and chorionic villus sampling. The booklet paid special attention to the decision making process, and several advantages and disadvantages of prenatal screening were listed. The leaflets were pilot-tested for comprehensibility.

Procedure

Women attending one of 44 participating midwifery and gynaecology practices from May 2001 to May 2003 before 16 weeks' gestation were asked permission to be sent a research information letter and an informed consent form. Practices were allocated in several areas throughout the Netherlands to ensure a representative sample. Women who gave informed consent to participate in the study were asked to fill out postal questionnaires before and after the prenatal screening offer. The first questionnaire was sent before the pregnant women received the screening in-

formation booklet, and this contained questions about background variables such as age, education, parity, and religion. The second questionnaire was sent and filled out after they had read the booklet and decided for or against prenatal screening, but before they had received the test result. It included measures of knowledge, attitude, decisional conflict and anxiety. The third questionnaire was sent after receiving the test result. Among other measures, this questionnaire contained a measure of satisfaction with the decision. The third questionnaire also asked about test uptake. Women in the control group, and women who declined screening received the questionnaires at comparable points in time.

Sample

During the recruitment period, 4076 women were asked to participate in the study; 2986 (73%) women gave informed consent and returned the first questionnaire. Of these women, 76% (n=2277) also filled out and returned the second questionnaire, and 66% (n=1968) also returned the third questionnaire. Analysis of non-response revealed that the main reasons for not participating in the study were lack of time or lack of interest. For the present study, only data of the intervention groups were used (n=1421). Of the women who accepted the prenatal screening test, only 20 turned out to have an increased risk. These cases were excluded. Due to missing values in the different questionnaires, another 242 cases had to be excluded, which resulted in a study sample with data of 1159 women.

Measures

The following socio-demographic variables were assessed: age, parity, educational level, level of religiosity. Whether or not a pregnant women had had a prenatal screening test was asked in the third questionnaire.

Knowledge about prenatal screening for congenital defects was measured by a scale that consisted of seven items for women in the NTM group and ten items for women in the MST group (see appendix). The MST scale corresponded with the NTM scale and had some additional questions about NTD. The items were based on the information that was covered in the prenatal screening information booklets. The scales were composed of yes/no items about three topics: characteristics of DS and NTD (only for the MST group), characteristics of the screening test, and implications of the possible test results. Because no gold standard exists as to what constitutes sufficient knowledge, it was determined that the guess-corrected mid-point would serve as cut-off. Correction for guessing was carried out using Abbott's blind guessing formula (24). This indicated sufficient knowledge when more than five (NTM scale) or seven (MST scale) questions were answered correctly, and insufficient knowledge when fewer questions were answered correctly.

Attitude towards having a prenatal screening test for congenital defects was measured by an attitude scale that consisted of four items (see appendix). The scale ranged from 4 to 20. The scale was internally consistent: Cronbach's alpha was 0.79. The attitude scores were normally distributed with the median score at the midpoint of the scale. The midpoint of the scale equals a neutral attitude. It is theoretically incorrect to classify women with a neutral attitude as positive or negative towards prenatal screening. Consequently, attitude scores were not dichotomized, but they were reclassified into three equal categories: negative attitude, neutral attitude, and positive attitude.

To assess value consistency, attitude scores were combined with test uptake. When positive attitudes involved accepting the prenatal test, or negative attitudes involved declining the test a value-consistent decision was made. Conversely, a value-inconsistent decision was made when positive attitudes were accompanied with declining the test, or negative attitudes were accompanied with accepting the test.

To determine whether a decision was an informed choice or not, dichotomized knowledge scores and value consistency scores were integrated. This resulted in informed decisions that were value-consistent and based on good knowledge, and in uninformed decisions that were value-inconsistent and/or based on poor knowledge.

Decisional conflict was measured by O'Connor's Decisional Conflict Scale (DCS), in which decisional conflict is defined as a state of uncertainty about the courses of action to take (13). Total scores were divided by the number of items, so the scale ranged from 0 to 5. In our sample, Cronbach's alpha for the total scale was 0.84. Satisfaction with the decision was assessed by a scale that consisted of four five-point Likert items about decision satisfaction after the decision was made, with items like 'I think I made a good decision', and 'I am happy with the decision I made'. The total scores were divided by the number of items, thus the scale ranged from 0 to 5. The scale was internally consistent with a Cronbach's alpha of 0.69. Anxiety was measured using the Dutch version of the State Trait Anxiety Inventory version Y (25). In our sample, Cronbach's alpha was 0.93.

Analysis

Group differences were tested using χ^2 tests for categorical variables, and *t* tests for continuous variables.

Approvals

According to the Dutch Population Screening Act, permission for this study had first to be granted by the Minister of Health. After a recommendation from the

Health Council, the permit was granted (26). The present study was also approved by the Ethical Committee of the VU University Medical Center.

Results

The socio-demographic characteristics of the study participants are shown in Table 1. Forty-four percent of the pregnant women decided to accept the offer of the prenatal screening test. Test uptake was significantly higher among participants being offered the NTM (52%) compared to those being offered the MST (35%) ($\chi^2 = 33.3, p < 0.001$).

Table 1. Demographic characteristics of the sample

	N (%)
Age	
<26	69 (6)
26-30	433 (37)
31-35	556 (48)
>35	101 (9)
Parity	
Primiparous	506 (44)
Multiparous	653 (56)
Degree of religiosity	
Actively	47 (4)
Somewhat actively	216 (19)
Not actively	349 (30)
Not religious	547 (47)
Educational level	
Low	128 (11)
Middle	507 (44)
High	524 (45)

Of the participants, 83% (n=957) were classified as having sufficient knowledge about prenatal screening to be able to make an informed choice; 17% (n=202) had insufficient knowledge. There were no significant differences between the test acceptors and the test decliners, nor between the NTM and the MST group. Higher proportions of sufficient knowledge were observed among women with higher levels of education ($\chi^2_{trend} = 55.1, p < 0.001$), and among women in higher age groups ($\chi^2_{trend} = 7.63, p < 0.01$) (Table 2).

Table 2. Percentages of pregnant women who made knowledgeable, value-consistent, and informed choices according to the demographic characteristics of the sample

	Sufficient knowledge %	Value consistency %	Informed choice %
Age			
<26	77 ^a	86 ^a	65 ^a
26-30	79	84	68
31-35	85	84	72
>35	86	59	51
Parity			
Primiparous	80	80	67
Multiparous	84	85	70
Degree of religiosity			
Actively	94	90	86
Somewhat actively	81	81	62
Not actively	83	93	69
Not religious	82	81	69
Educational level			
Low	63 ^a	86	56 ^a
Middle	80	80	64
High	90	83	75

^a statistically significant χ^2 test ($p < 0.05$)

Table 3. Attitudes towards having a prenatal screening test of pregnant women who accepted or declined the test offer

Attitude	Acceptors	Decliners	Total
	N (%)	N (%)	N (%)
Positive	313 (76) ^a	98 (24) ^b	411
Neutral	148 (45)	179 (55)	327
Negative	52 (12) ^b	369 (88) ^a	421
Total	513	646	1159

^a These categories represent value-consistent decisions

^b These categories represent value-inconsistent decisions

The mean attitude score was 12.2 (SD = 3.7), the median was 12. Participants who accepted the prenatal screening test had significantly more positive attitudes compared to women who declined the screening test ($F = 481.4$, $p < 0.001$). As women with neutral attitudes can not be classified as value-consistent or not, these cases are left out of consideration for calculation of value consistency rates ($n: 1159 - 327 = 832$). Numbers of value-consistent and value-inconsistent decisions are shown in Table 3. Of the participants with a positive attitude, 313 women accepted prenatal screening, and 369 participants with a negative attitude declined prenatal screening. This resulted in 682 (82%) participants making value-consistent deci-

sions. Of the acceptors, 52 participants had a negative attitude, and 98 decliners had a positive attitude towards prenatal screening. This totalized to 150 participants (18%) who did not make value-consistent decisions. From Table 2, it can be seen that women over 35 years of age made significantly less value-consistent decisions as compared to younger women ($\chi^2 = 28.4, p < 0.001$).

Of the pregnant women who participated in this study, 68% had made an informed choice, i.e. they made a choice that was based on sufficient knowledge, and consistent with the decision maker's values (Table 4). Consequently, 32% (n=265) of the women made an uninformed choice, 47% (n=123) of which was due to value inconsistency, 43% (n=115) was due to poor knowledge, and 10% (n=27) was due to both value inconsistency and poor knowledge. Participants over 35 years of age made significantly less informed decisions (51%, $\chi^2 = 11.6, p < 0.01$). Furthermore, informed choice appeared to be associated with education; the higher the educational level, the higher the rate of informed choice ($\chi^2_{trend} = 16.0, p < 0.001$) (see Table 2).

Table 4. Value (in-)consistency and knowledge about prenatal screening

	Sufficient knowledge N (%)	Insufficient knowledge N (%)	Total N (%)
Value consistency	567 (68) ^a	115 (14) ^b	682 (82)
Value inconsistency	123 (15) ^b	27 (3) ^b	150 (18)
Total	690 (83)	142 (17)	832

^a This category represent informed choices

^b These categories represent uninformed choices

Table 5. Outcome measures for test acceptors and decliners according to informed choice

	Informed choice Mean (SD)	Uninformed choice Mean (SD)	<i>t</i>	<i>p</i>
Test acceptors				
Decisional conflict	1.9 (0.5)	2.3 (0.6)	5.7	<0.001
Decision satisfaction	4.8 (0.4)	4.5 (0.6)	4.9	<0.001
Anxiety	34.5 (9.5)	36.4 (9.8)	1.8	>0.05
Test decliners				
Decisional conflict	2.0 (0.6)	2.1 (0.5)	1.5	>0.1
Decision satisfaction	4.7 (0.6)	4.5 (0.6)	2.1	<0.05
Anxiety	35.0 (9.1)	35.3 (10.8)	0.3	>0.5

Decisional conflict, satisfaction with the decision, and anxiety

As can be seen from Table 5, informed choice was associated with less decisional conflict; however, this applied only to test acceptors. Both informed acceptors and decliners had significantly higher scores on the satisfaction with the decision scale as compared to uninformed deciders ($t = 4.9, p < 0.001$ and $t = 2.1, p < 0.05$, respectively). Although informed choice was associated with less anxiety for the test acceptors, this difference was not statistically significant ($t = 1.8, p = 0.07$). For the test decliners, no difference in anxiety scores between the women who made an informed choice and those who made an uninformed choice was observed.

Discussion

The vast majority of the pregnant women had sufficient knowledge about the different aspects of prenatal screening for congenital defects, and most women made value-consistent decisions. Assessment of informed choice revealed relatively high levels of informed decision making. Informed choice to accept prenatal screening was associated with less decisional conflict and with more decision satisfaction as compared to uninformed choice. Informed choice to decline prenatal screening was related only with higher levels of satisfaction. Anxiety scores were lower (although not statistically significant) only for informed acceptors.

The conclusion that many women were sufficiently knowledgeable about prenatal screening is not supported by other studies, neither is the finding that most pregnant women made informed decisions (11). Both findings may be related to the specific situation in which the screening was offered. Currently, in the Netherlands, prenatal screening is not offered routinely. Therefore, for most of the participants in our study, receiving the offer of a prenatal screening test was something new. Because of the unfamiliarity with prenatal screening as a standard practice, pregnant women in our study presumably made the decision more consciously. This is supported by research from countries where prenatal screening is part of routine prenatal care, which establishes that prenatal screening is no longer something about which a deliberate decision is made (27,28). This so-called normalization or routinization of prenatal screening leads to less informed choices about having prenatal screening. In the Netherlands, the case is quite the opposite because up till now it has not been allowed to offer prenatal screening routinely. Furthermore, the study participants received extensive, well-balanced information (by means of both a booklet and counselling by the midwife or gynaecologist) that paid special attention to the decision making. This also could be an explanation for the high number of informed choice in our study.

The finding that the rate of informed choices was associated with level of education is due to the association of the level of knowledge with educational level. This last association is not surprising, and in accordance with previous literature (29-31). More uninformed choices were observed among participants over 35 years of age compared to younger women. This finding seems to be illogical since older women have higher levels of knowledge. However, there is a high rate of value inconsistency in this age group, which is caused by test decliners with a positive attitude concerning prenatal screening, rather than by test acceptors with a negative attitude. These test decliners with a positive attitude presumably declined screening because they preferred prenatal diagnostic testing. At the time of the study, in the Netherlands, these women over 35 years of age were routinely offered prenatal diagnostic testing. So, in the model, these older participants were frequently classified as value-inconsistent and, consequently, as uninformed decision makers. This is a shortcoming of the model, because these women did in fact make value-consistent, informed choices to decline prenatal screening.

Although the majority of the decisions were informed choices, substantial percentages of uninformed choices were found. Almost one third of all decisions were categorized as uninformed choices. Forty-seven percent of these decisions were ascribed to value inconsistency, 43% to insufficient knowledge, and 10% to both. Value inconsistency due to declining prenatal screening while having a positive attitude, may be the result of practical barriers like requiring a separate return visit (32,33). Another explanation might be that these participants were of the opinion that the screening test was not good enough, as research has shown that many women give this reason ('unfavourable test characteristics') for declining prenatal screening (34). On the other hand, value inconsistency due to accepting prenatal screening while being negative about it might be attributed to social pressure or normalization of prenatal screening. Further research is needed to investigate the real causes behind value inconsistency .

From the effects on the outcome measures, it can be concluded that informed choice is indeed associated with better psychological outcomes. This underlines the importance of informed choice. However, these positive effects mainly applied to informed choice to accept prenatal screening. That informed choice is associated with less decisional conflict was also found by Michie et al. (9) and Bekker et al. (20), and is contradictory to the suggestion of Green et al. (11) that informed choice is associated with less decision satisfaction and increased anxiety.

A limitation of this study is that the number of women whose prenatal test resulted in an increased risk of having a child with DS or NBD was too small to incorporate them in the analysis, and therefore these cases had to be excluded. This low screen positive rate was caused by the small number of high-risk results in the

NTM group, probably due to the poor quality of the NTMs performed within our study. Of all pregnant women, those with high-risk results will presumably suffer most from uninformed choice, and benefit most from informed choice (9). Studies aiming especially at the high-risk group should be performed to assess the rates of informed choice and its benefits for these women.

Another limitation of the present study is that no measure of the decision making process is involved in the assessment of informed choice. Some definitions of informed choice include a measure of the decision making process in addition to knowledge and value consistency (35). By not incorporating the decision making process it is still unknown how the information was perceived, and whether or not the knowledge was used. Involving the process of decision making in a measure of informed choice would, for instance, make clearer how risk perception influences decision making. Integrating a measure of the decision making process into the assessment of informed choice would probably reveal that some decisions that were formerly classified as informed choices are not the result of a process of deliberation. More research involving the process of decision making in the assessment of informed choice is needed.

This study underlines the importance of achieving informed choice in the context of prenatal screening. Pregnant women will experience less decisional conflict, and will be more satisfied, when having made an informed choice. Therefore, every effort that can be made to increase the number of informed choices, should be carried out. For instance, research has shown that decision aids are able to improve the quality and the level of informedness of prenatal testing decisions (20), and such decision aids should be developed and implemented in the prenatal screening setting. Introducing prenatal screening for congenital defects as part of standard prenatal care should go hand in hand with an adequate system of informing and counselling women about prenatal screening, to ensure informed decision making.

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Appendix

Knowledge measure

	correct	incorrect
By using the NTM/MST, the chance of several congenital defects can be calculated	<input type="checkbox"/>	<input type="checkbox"/>
By using the NTM/MST, it can be determined whether the child is healthy	<input type="checkbox"/>	<input type="checkbox"/>
By using the NTM/MST, Down syndrome can be determined with certainty	<input type="checkbox"/>	<input type="checkbox"/>
By using the MST, a neural tube defect can be determined with certainty	<input type="checkbox"/>	<input type="checkbox"/>
Most children with Down syndrome die at birth	<input type="checkbox"/>	<input type="checkbox"/>
Children with Down syndrome will always have some mental disability	<input type="checkbox"/>	<input type="checkbox"/>
Children with a neural tube defect will always have some physical disability	<input type="checkbox"/>	<input type="checkbox"/>
If the outcome of NTM/MST is increased, an amniocentesis has to be performed	<input type="checkbox"/>	<input type="checkbox"/>
If the outcome of NTM/MST is increased, it means that the child has Down syndrome or a neural tube defect.	<input type="checkbox"/>	<input type="checkbox"/>
Children with a neural tube defect often die at birth	<input type="checkbox"/>	<input type="checkbox"/>

Attitude measure

In my opinion, testing for congenital defects during my pregnancy, is ...		
Bad	<input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/>	Good
frightening	<input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/>	not frightening
not reassuring	<input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/>	reassuring
not self-evident	<input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/> <input type="checkbox"/>	self-evident

Chapter 7

**Informed decision making in the
context of prenatal screening**

Abstract

This study aimed to construct a measure of informed decision making that includes knowledge, deliberation, and value-consistency, and to assess the level of informed decision making about prenatal screening, and differences between test acceptors and test decliners. Women attending one of 44 midwifery and gynaecology practices were asked to fill out postal questionnaires before and after the prenatal screening offer. The principal outcome was the level of informed decision making. For this purpose, knowledge about prenatal screening, deliberation about the pros and cons of the alternatives, test uptake, and attitude towards having a prenatal screening test were measured. 84% of the participants were sufficiently knowledgeable about prenatal screening, 75% of the decisions were deliberate, and 82% were value-consistent. 51% of the participants made an informed decision. Test acceptors made less informed decisions as compared to test decliners. This difference was mainly caused by the lower rate of deliberation in this group. It appears from this study that prenatal screening decisions are often not informed decisions. This is inconsistent with the main objective of offering screening, which is to enable people to make informed decisions. Decision makers should be encouraged during the counselling to deliberate about the various alternatives.

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Introduction

It is generally accepted that the central objective of informing about screening is to enable people to make informed decisions (1-4). There are many different definitions of informed choice or informed decision making (5-13) (see Table 1). All these definitions have two elements in common. Firstly, an informed choice is based on sufficient knowledge about the relevant aspects of the available alternatives. Secondly, an informed choice is consistent with the decision maker's values. A choice refers to the end product of a decision, whereas a decision refers to the process of choosing between alternatives, preceding that choice. In this respect, a definition of informed decision making needs to include an additional element, as compared to the definition of informed choice above. Some of the definitions that are mentioned in Table 1 include such an additional element, namely: an informed decision also implies an evaluation of the alternatives, i.e. a process of deliberation about the alternatives and weighing up their pros and cons(5,6,8,12,13).

Furthermore, most health behaviour theories (e.g. health belief model, protection motivation theory, theory of planned behaviour) consider health decisions to be the end result of a decision making process that is based on deliberative processing of the available information (14-16). Janis and Mann determine the quality of decision making by seven criteria –forming a scale of vigilance– in which deliberation and evaluation take a prominent place (17). Dual-process theories distinguish between systematic, analytic processing, in which different alternatives are evaluated and their pros and cons are weighed, and heuristic, intuitive processing, in which simple decision rules or cognitive heuristics are used (18). Although heuristic decision making requires less cognitive effort, and can be very adequate in e.g. expert decision making, it may produce behavioural inconsistencies and systematic errors (19-21): “One fundamental point (...) is that we often think in automatic ways when making judgements and choices, that these automatic thinking processes can be described by certain psychological rules (e.g. heuristics) and that they can systematically lead us to make poorer judgements and choices than we would by thinking in a more controlled manner about our decisions.” (19)

Although both patients and care providers put a high value on informed decision making, Green et al. argues that most women do not make informed decisions about screening(8). However, two extensive reviews concluded that in most of the reviewed studies, informed decision making was not assessed explicitly or adequately(8,22). In these studies, cognitions that are associated with

Table 1. Definitions of informed decision making

Author	Definition
Summers (11)	Informed choice or decision making generally involves three components: information, comprehension, and voluntary choice.
O'Connor (10)	An effective decision is defined as informed, consistent with personal values, and acted upon.
Marteau <i>et al.</i> (9)	An informed choice is one that is based on relevant knowledge, consistent with the decision maker's values and behaviourally implemented.
Kohut <i>et al.</i> (12)	(...) informed choice is considered a process of decision making which evolves through the evaluation of information and personal values (...).
Bekker (5)	An informed decision is made when an individual evaluates the relevant information about the advantages and disadvantages of all the possible courses of action, in accord with their beliefs, to reach a decision, (...).
Green <i>et al.</i> (8)	A decision is said to be informed when the relevant information about the advantages and disadvantages of all the possible courses of action is evaluated in accord with the decision maker's beliefs, in order to reach a decision and take steps to make a choice.
Briss <i>et al.</i> (7)	Informed decision making is defined as occurring when an individual understands the nature of the disease or condition being addressed; understands the clinical service and its likely consequences, including risks, limitations, benefits, alternatives, and uncertainties; has considered his/her preferences as appropriate; (...) and makes a decision consistent with his/her other preferences and values (...).
Rimer <i>et al.</i> (13)	An informed decision making process includes: understanding the screening test, its risks, benefits, and alternatives, understanding personal values and preferences, weighing the pros and cons of the test (...).
Bekker <i>et al.</i> (6)	The informed model of physician-patient interaction requires (a) professionals to provide accurate, complete and neutral information about the alternatives and consequences about the treatment options, and (b) patients to deliberate this information in accord with their own values and take steps to make a choice.

informed decision making (knowledge, risk perception, attitude), or decision outcomes (decisional conflict, satisfaction, anxiety) were measured only separately.

Marteau and colleagues have developed a multidimensional measure of informed choice (MMIC) based on knowledge and value consistency (9,23). The MMIC characterizes a decision maker as having made an informed choice if this choice was based on sufficient knowledge and was consistent with one's values. The MMIC refers to informed choice, therefore no measure of the decision making process is included. A pregnant woman, with both sufficient knowledge about prenatal screening and a positive attitude towards prenatal screening, who accepts prenatal screening, will be classified by the MMIC as having made an informed choice because this choice was knowledge-based and value-consistent. However, it could be that she perceives prenatal screening as something self-evident because it is part of routine prenatal care. (Research has shown that, in countries where pre-

natal screening is offered routinely, having prenatal screening done has indeed become something self-evident (24.) In this hypothetical case accepting prenatal screening is not the result of a deliberated decision. As informed decision making occurs when the relevant information about the advantages and disadvantages of all the possible courses of action is evaluated value-consistently (5), a (common implicit) assumption of informed decision making is that it concerns systematic, deliberated decision making (25). Thus, this choice should not be classified as an informed decision. This example demonstrates why we incorporated a process-related measure (i.e. a measure of deliberation) in our assessment of informed decision making.

Informed decision making is not only of importance to screening decisions, but also to many treatment decisions, especially preference-sensitive decisions and decisions that involve high risks (26-28). Preference-sensitive decisions refer to situations in which the ratio of risks to benefits is either uncertain, or dependent on patient preferences or values (e.g. the decision to choose lumpectomy or mastectomy for treating early stage breast cancer) (26,27). Because informed decision making is currently seen as the cornerstone of many health care decisions, evaluation of the decision making process is needed to assess whether or not this is achieved(29). To our knowledge, no studies have been performed that assess informed decision making by measuring and integrating the three theoretically-founded dimensions: knowledge, value consistency, and deliberation. The present study aimed to assess these three elements and to construct a measure of informed decision making. Furthermore, this study investigated differences in the level of informed decision making between different choices, i.e. accepting prenatal screening versus declining it.

Methods

Setting

Up till now, in the Netherlands prenatal screening for Down syndrome (DS) and Neural tube defects (NTD) is not offered routinely to pregnant women, which is in contrast with many other modern western countries. Solely prenatal diagnostic tests are offered routinely to pregnant women over 35 years of age, and to women with an otherwise increased risk.

The study presented in this paper is part of a larger research project, aiming to give more insight into the risk perception, informed decision making and psychological well-being of pregnant women who are offered prenatal screening for congenital defects. The research project is a randomized controlled trial in which par-

ticipants were randomized into one of two intervention groups or into the control group. The intervention consisted of offering prenatal screening for DS and NTD, by means of a sent-home booklet containing information about a particular prenatal screening test, and an oral explanation by the woman's midwife or gynaecologist. The last section of the booklet was entitled 'to decide whether or not to undergo the screening test', and listed some advantages and disadvantages of having the test done. Women in the first intervention group were offered the Nuchal Translucency Measurement (NTM), and women in the second randomization group were offered the Maternal Serum screening Test (MST). The NTM tests for DS, in the first trimester of pregnancy, and is done by ultrasound scanning (30). The MST is performed in the second trimester of pregnancy, and tests for DS and NTD (31). Both prenatal screening tests provide an individualized risk estimation of having a child with one of these disorders, and thus select a high-risk subgroup within a population of pregnant women. Subsequently, this subgroup of women with an increased risk is offered prenatal diagnostic testing in order to provide a certain diagnosis.

Procedure

Women attending one of 44 participating midwifery and gynaecology practices from May 2001 to May 2003 before 16 weeks' gestation were asked for their informed consent to participate in the study. These practices were located in several areas throughout the Netherlands to ensure a representative sample. Participants were asked to fill out postal questionnaires before and after the prenatal screening offer. The first questionnaire was sent before the pregnant women received the screening information booklet. The second questionnaire was sent and filled out after they had read the booklet and decided for or against prenatal screening, but before they had received the test result. The third questionnaire was sent after receiving the test result. Women in the control group, and women who declined screening, received the second and third questionnaires at comparable points in time.

Participants

During the recruitment period, 4076 women were asked to participate in the study; 2986 (73%) women gave informed consent and returned the first questionnaire. Of these women, 76% (n=2277) also filled out and returned the second questionnaire, and 66% (n=1968) also returned the third questionnaire. Analysis of non-response revealed that the main reason not to participate in the study was lack of time or lack of interest. For the present study only data from the two intervention groups was used (n=1421). Due to missing values in the different questionnaires, another

91 cases had to be excluded (n=1330). Furthermore, in order to calculate value consistency, participants need to have either a positive or a negative attitude with respect to prenatal screening. Thus, participants with a neutral attitude were excluded; which resulted in a study sample with data of 962 women.

Measures

The questionnaires cover the following measures (see Table 2):

- Knowledge about prenatal screening for congenital defects (second questionnaire).
- Deliberation about the pros and cons of the alternatives (second questionnaire).
- Test uptake measured by a single yes/no question (third questionnaire).
- Attitude towards having a prenatal screening test (second questionnaire).

The variable value consistency was constructed by combining test uptake and attitude category (see Table 2), and the variable informed decision was constructed from knowledge, deliberation, and value consistency (see Table 2). See appendix for a description of the exact items of the deliberation scale. The knowledge and attitude scales are described elsewhere(32).

Analysis

The scales of knowledge and deliberation were dichotomised into sufficient or insufficient knowledge, and deliberation or a lack of deliberation, respectively (see Table 2). Together with the variable of value consistency (either value-consistent or value-inconsistent), this resulted in three dichotomous variables. According to our measure of informed decision making, all three requisites had to be met to reach an informed decision. Group differences were tested using χ^2 tests. To determine which factor(s) account for different levels of informed decision making between two groups, χ^2 statistics were partitioned (33).

Approvals

According to the Dutch Population Screening Act, permission for this study had first to be granted by the Minister of Health. After a recommendation from the Health Council, the permit was granted. The present study was also approved by the Ethical Committee of the VU University Medical Center.

Table 2. Description and characteristics of the measures

Measure	Description	Items	Reliability	Range	Mean (SD)	Cut-off	Outcome
Knowledge score	Knowledge about characteristics of the tested disorder(s) and of the screening test, and meaning of the possible test results.	7 correct / incorrect items	-	0-7	6.4 (0.9)	>5 ^a	Sufficient knowledge: 84%
Deliberation scale	Evaluating the alternatives, thinking about the consequences, weighing up the pros and cons.	6 five-point Likert items	0.83	6-30	21.2 (4.6)	>18 ^b	Deliberated: 75%
Test uptake	Whether the prenatal test was accepted or declined.	1 yes/no question	-	-	-	-	Test uptake: 46%
Attitude scale	Attitude towards having a prenatal test for congenital defects.	4 five-point items	0.79	4-20	12.3 (3.7)	>14 ^c <10 ^c	Positive: 37% Negative: 35%
Value consistency	Consistency between value (attitude) and behaviour (test uptake).	Calculated ^d	-	-	-	-	Value-consistent: 82%
Informed decision	A knowledgeable, deliberate, value-consistent decision.	Calculated ^e	-	-	-	-	Informed choice: 51%

^a Guess-corrected midpoint

^b Midpoint of the scale

^c Attitude scores were classified into three equal categories. The middle category represents neutral attitudes, so only the positive and negative attitudes were used.

^d Value consistency was calculated by classifying test acceptors with positive attitude, and test decliners with a negative attitude as value-consistent. Test acceptors holding a negative attitude, and test decliners holding a positive attitude were classified as value-inconsistent.

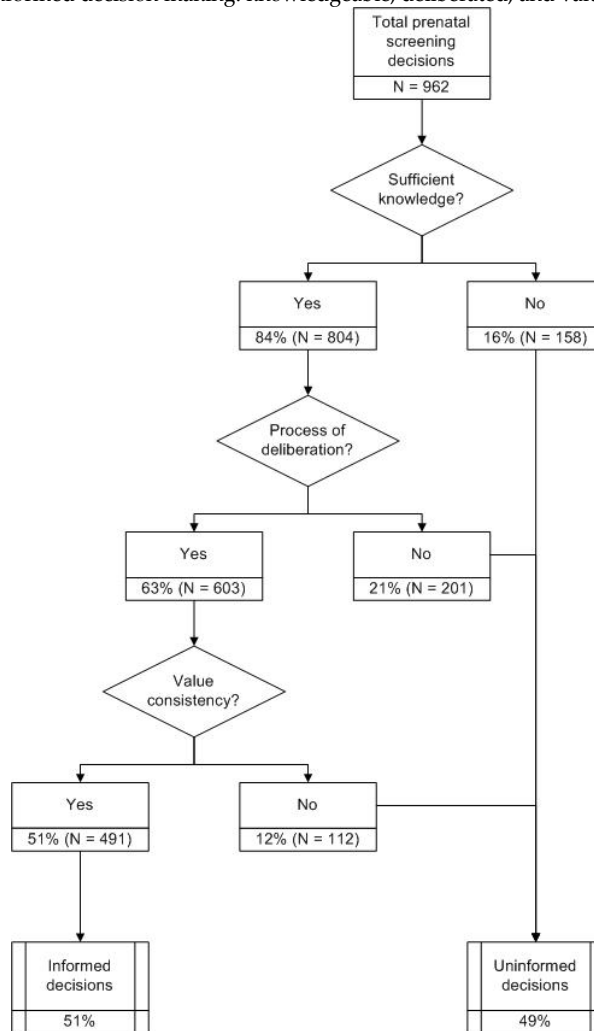
^e A decision was labelled as an informed decision only when all of the three criteria were fulfilled: knowledgeable, deliberate, and value-consistent. In all other cases, the decision was labelled uninformed.

Results

Eighty-four percent of the participants were assigned to the group with sufficient knowledge about the relevant aspects of prenatal screening for DS and NTD, 75% of the decisions were the result of a process of deliberation, and value-consistency was determined for 82% of the women (Table 2).

As can be seen from Figure 1, 84% of the women made a knowledge-based decision, 63% made a decision that was knowledge-based and deliberated, and 51% made a decision that was knowledge-based and deliberated and value-

Figure 1. Informed decision making: knowledgeable, deliberated, and value-consistent.



consistent. In other words, according to our definition, 51% of the women met all three requisites, and thus made informed decisions. Consequently, 49% of them could be classified as not having made an informed decision: 16% due to insufficient knowledge, 21% due to lack of deliberation, and 12% due to value-inconsistency (Table 3).

Comparison of test acceptors with test decliners revealed that women who accepted the prenatal screening test offer made significantly less informed decisions

as compared to test decliners (42% vs. 58%, $\chi^2 = 26.9$, $p < 0.001$) (Table 3). Partitioning (28) identified which of the informed decision variables (knowledge, deliberation, or value-consistency) was, or were, as the case may be, responsible for this difference. The overall χ^2 , having 3 degrees of freedom ($\chi^2 = 43.47$, 3 df, $p < 0.001$), could be partitioned into three χ^2 s with 1 degree of freedom: knowledge vs. value consistency ($\chi^2 = 4.03$, 1 df, $p = 0.05$), deliberation vs. informed decision making ($\chi^2 = 39.07$, 1 df, $p < 0.001$), and knowledge and value consistency vs. deliberation and informed decision making ($\chi^2 = 0.37$, 1 df, $p > 0.5$). Thus, the association between informed decision making and test uptake reflected primarily an association between the level of deliberation and test uptake.

Table 3: Informed and uninformed decision making divided by test acceptors and test decliners

		Test acceptors	Test decliners	Total
		N (%)	N (%)	N (%)
Informed decisions		185 (42)	306 (58)	491 (51)
Uninformed decisions	Due to:			
	Insufficient knowledge	83 (19)	75 (14)	158 (16)
	Lack of deliberation	128 (29)	73 (14)	201 (21)
	Value inconsistency	45 (10)	67 (13)	112 (12)
Total		441 (100)	521 (100)	962 (100)

Discussion and conclusion

If enabling informed decision making is the main objective of counselling on many health-care decisions, instruments need to be developed that assess whether or not informed decision making has occurred (29). As there is a shortage of instruments that measure the multidimensional concept of informed decision making, we tried to compose an integrated instrument that assesses decisions as being informed when they are knowledgeable, value-consistent, and deliberated. According to this operationalization, only half of the participants made a knowledgeable, value-consistent, deliberated decision about prenatal screening, and thus, could be classified as having made an informed decision. There were no major differences in the cause of uninformed decision making: 16% was due to insufficient knowledge, 21% was due to lack of deliberation, and 12% was due to value inconsistency. Of the test acceptors, 42% made informed decisions, and of the test decliners 59%. The lower rate of informed decisions among test acceptors was mainly caused by the lower rate of deliberation in this group. Several possible explanations could be found for this. Firstly, for many participants accepting prenatal screening may not

be a decision at all, but more perceived as something self-evident (25,34). This may be a reasonable explanation in countries where screening is part of standard antenatal care, and where accepting screening has become routine (24). However, for this study this is not a likely explanation, because up till now prenatal screening has not been part of standard antenatal care in the Netherlands. Secondly, deliberating about the different options and their pros and cons may cause participants to decline prenatal screening. It could be that they become conscious of some disadvantages of these prenatal screening tests, while deliberating about it (34). This possible explanation is supported by our earlier finding that the unfavourable screening test characteristics and the adverse characteristics of invasive testing were indeed important reasons for declining prenatal screening(35). This reasoning might explain why routinisation of prenatal screening, which implies less deliberation about the decision, has led to high uptake rates in many countries(24).

The strength of our operationalization of measuring informed decision making as opposed to the MMIC, is that it does not only classify someone as knowledgeable or not, but —by assessing the decision making process— it also takes into consideration whether or not this knowledge is used in the decision. A limitation of this study is that the three elements of our instrument were dichotomized and given equal weight. This resulted in a rough classification and, ultimately, a very crude distinction between informed and uninformed decision making. Another limitation concerns the deliberation scale: a more objective and extensive measurement of the decision making process (e.g. interviews or think-aloud methods) might be preferred above our self-reported measure.

Considering informed decision making, many questions still remain to be answered. Which knowledge is necessary to make an informed decision? Does everybody need the same amount of information to be able to make an informed decision? For instance, is it necessary to know the exact test characteristics for someone who is against prenatal testing because she is ethically opposed to abortion? Women with a neutral or ambivalent attitude were excluded from the analysis, but in what way should value consistency and informed decision making be measured among these women? Should counselees be pushed to deliberate and consider every available option? Or should this come into the picture only when someone is at risk of making a value-inconsistent decision? Is informed decision making necessary for every decision concerning health care or screening? Or does it apply to preference-sensitive or value-sensitive decisions only? Further research is needed to answer these questions, and to examine the validity and feasibility of this method of measuring informed decision making in decisions concerning treatment and screening.

According to our operationalization of informed decision making, only half of the pregnant women made an informed decision to accept or decline prenatal screening. This implicates that for many women the aim of offering screening, which is to enable people to make informed decision, is not achieved. Therefore, interventions that facilitate informed decision making should be implemented.

Practice implications

It appears from this study that decisions to accept or decline prenatal screening are often not completely informed decisions. This seems to be the international status quo about prenatal screening, since this finding is in accordance with many other studies (8). Nevertheless, it is inconsistent with the main objective of offering prenatal screening, which is to enable people to make informed decisions. Therefore, to resolve this contradiction, interventions to enhance informed decision making might need to be developed and implemented. Decision aids (DAs) are interventions that help people to make deliberative decisions between the available options (36,37), by providing a framework in which treatment alternatives and their potential outcomes can be systematically analysed (38). Although not unambiguous(29,39), research has shown that DAs are able to facilitate and improve informed decision making (13,36,37). As implementation of a DA requires the availability of a well-validated instrument, and makes many demands on those counselling, a less-demanding approach is suggested: instead of using a full-blown DA, decision makers should be guided and encouraged during the counselling to deliberate about the various alternatives, their pros and cons, and their consequences. In the case of prenatal screening, pregnant women might be encouraged by their midwife or gynaecologist to consider the possibility of an unfavourable test result and the potential consequences (40). In this way, informed decision making about prenatal screening will probably enhance. Further research should be performed to investigate this supposition.

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Appendix

Items of the deliberation scale

Items

1. I have tried to mentally visualize the situation.
 2. I have imagined how I would feel if I did not participate in the NTM/MST.
 3. I have imagined how I would feel if I participated in the NTM/MST.
 4. I have tried to think through the consequences of not accepting the NTM/MST.
 5. I have tried to think through the consequences of accepting the NTM/MST.
 6. I have made a (mental) list of the pros and cons of the NTM/MST
-

Chapter 8

General discussion

Introduction

The central objective of this study was to gain more insight into the decision making of pregnant women concerning prenatal screening for congenital defects. The three central themes were: uptake of prenatal screening and women's motivations for decision making, factors influencing prenatal screening decisions, and informed decision making about prenatal screening. The main findings are summarised and discussed in the present chapter. Furthermore, the strengths and limitations of this study are discussed, and recommendations for policy, practice, and further research are made. The chapter ends with a final conclusion.

Main findings

Uptake and motivations

Offering prenatal screening to an unselected population of pregnant women in the Netherlands resulted in an uptake rate of less than 50%. Utilisation of prenatal screening appeared to be inversely related to level of education and degree of religiosity. Moreover, test uptake depended on which test was offered: fewer women accepted the maternal serum test, as compared to the nuchal translucency measurement (38% vs. 53%). The participants accepted prenatal screening primarily because they wanted to gain more knowledge about the health of the foetus, and because of the favourable test characteristics (the test does not involve a risk for the baby). The main reasons for declining a prenatal test were: the unfavourable test characteristics (the test gives no certainty), the adverse characteristics of invasive diagnostic testing, anxiety/uncertainty, or that screening was not considered necessary or applicable.

Factors influencing decision making

From a multivariate path analysis, it appeared that a pregnant woman's attitude toward prenatal screening and subjective norm to accept or decline the prenatal test determined her intention for undergoing prenatal screening or not. Attitude toward prenatal screening was explained by attitude toward termination of pregnancy, and by perceived test efficacy. Pregnant women's perceptions of risk and severity were associated with their level of child-related anxiety, which, subsequently, only weakly influenced intention to test. Pregnant women's risk perception of having a child with Down syndrome, for example, consists of both a cognitive component and an emotional component. However, the role of these risk perception components in the decision making on prenatal screening was limited. Finally, the decision making process concerning prenatal screening was not influ-

enced by the prenatal counsellor's (midwife or gynaecologist) attitude toward prenatal screening, but rather by the pregnant women's perception of the counsellors' advice.

Informed decision making

When informed choice is conceptualised as a knowledge-based and value-consistent choice, it was found that over two-thirds of the participants could be classified as having made such an informed choice. It appeared that informed choice was associated with better psychological outcomes. When this definition was extended by a third requisite that addresses the process of decision making (i.e. a process of deliberation about the alternatives), only half of the participants made an informed decision. It was found that test acceptors made fewer informed decisions as compared to test decliners. This difference mainly arose from the fact that test acceptors made fewer deliberated decisions.

Discussion of main findings

Test uptake

One of the main findings from this study is that only 46% of the pregnant women accepted the prenatal screening test they were offered. This percentage is much lower than that found in many international studies on the utilisation of prenatal screening (1-10), and lower than the uptake rates of a few national studies as well (11-13). In chapter 2, several possible explanations for these differences are presented. The difference with most international studies is possibly related to the fact that prenatal screening has been part of standard antenatal care for years in the countries concerned. In such contexts, uptake rates are expected to be higher, as compared to a situation in which prenatal screening is relatively unknown and exceptional (this was the case in the Netherlands during our study). This might explain why studies in those countries where prenatal screening has become standard antenatal care (e.g. United States, United Kingdom, Israel, Denmark, France) generally report uptake rates between 75% and 95%. A recent study in the UK, however, reported test uptake in less than 50% of the participants (14). This indicates that, although many international studies suggest this relation between a standard offer of prenatal screening and high percentages of acceptance of the test, offering prenatal screening as a standard part of antenatal care does not inevitably have to lead to high uptake rates. Apparently, other factors also play a role here. How such differences within countries occur should be subject to further research.

In contrast to the present study, two other Dutch studies reported high uptake of prenatal screening (about 80%) (11-13;15). The papers of Kamerbeek et al. and Roelofsen et al. draw on the same study that was performed in the Northern Netherlands, where prenatal screening has been offered to pregnant women for many years (12;16). Although this was not allowed formally, it probably has resulted in a situation where prenatal screening was more routinised than in other parts of the Netherlands where prenatal screening is not being offered to all pregnant women. This might explain the high uptake of prenatal screening in the Northern Netherlands. The papers of Muller et al. are based on a study on the performance and acceptance of the nuchal translucency measurement in the Netherlands (17). In this study, pregnant women were offered a prenatal screening test in the context of a routine visit (together with a dating scan). This might have resulted in higher test uptake as compared to our study in which a separate visit was required (18). In their paper, Muller et al. suggested some additional reasons that might have caused the differences in uptake between the two studies. It is suggested that in our study participants had to consent to randomisation between screening and non-screening, and that women consenting to randomisation –and thereby accepting the possibility of non-screening– may feel less need for screening in the first place. This potential bias was however excluded since on inclusion, women did not know that the study was about prenatal screening (19).

Factors influencing decision making on prenatal screening

Education

We found a relation of education with knowledge about prenatal screening and, consequently, with informed choice (chapter 6), and an inverse relation of education with test uptake (chapter 2). Lower educated women were less knowledgeable, made fewer informed choices, and had higher uptake rates of prenatal screening, as compared to higher educated women. The finding that knowledge and informed decision making are dependent on level of education is in line with other studies (20-23). The negative association between level of education and test uptake is a more uncommon finding. For instance, Khoshnood et al. found that lower educated women showed reduced use of prenatal screening, and the main reason for foregoing prenatal screening was that the test was not offered. In our study, every pregnant woman was offered the test, and declining was largely related to test characteristics. It is plausible that better comprehension of the test characteristics is associated with higher levels of education, whereas limits in access or information are associated with lower socioeconomic status (20).

Interestingly, contrary to pregnant women's knowledge levels, their levels of value consistency did not differ significantly with education. Thus, although lower

educated women have less knowledge about prenatal screening, their ability to make decisions in line with their own values is as good as higher educated women. Higher educated decision makers have, on average, more knowledge and less positive attitudes toward prenatal screening, as compared to lower educated women. This might indicate that those women who are better able to comprehend (i.e. both knowledge and deliberation) the possibly disadvantageous characteristics of prenatal screening, consequently have more negative attitudes toward prenatal screening, which in turn leads to less acceptance of prenatal screening. This suggests that if pregnant women were to be offered adequate counselling and decision support, this might lead to them making more knowledgeable and deliberated decisions about prenatal screening, to more negative attitudes toward having a prenatal test done, and to a lower test uptake rate. More concretely, it is anticipated that the implementation of a decision aid for prenatal screening would lower the number of women who will have prenatal screening done.

Test characteristics

About one in three pregnant women who reported that they would not undergo prenatal screening, mentioned as the decisive reason for their decision to reject the test offer: the bad characteristics of the screening test or the diagnostic tests. These unfavourable characteristics of prenatal screening refer to the fact that the test provides only a risk estimation and can not give certainty about the presence or absence of the screened disorder. The adverse characteristics of invasive diagnostic testing refer to the risk of an iatrogenic abortion as a result of the testing procedure. The relevance of test characteristics in pregnant women's decision making on prenatal screening is also recognized by the results described in chapter 3 in which perceived test efficacy appeared to be an important predictor of a woman's attitude toward prenatal screening, which in turn was a main determinant of her intention to have the test done. This implies that women who have a negative picture of the test (i.e. not able to give certainty, risky invasive diagnostic testing necessary for certainty) acquire a negative attitude toward prenatal screening, and women who evaluate the test as positive (i.e. more information, no risk for the foetus) become positive about prenatal screening. These findings provide firm evidence for the prominent role of pregnant women's considerations about the quality of both screening tests and diagnostic tests in their decision making on prenatal screening. Green et al.'s systematic review on the psychosocial aspects of genetic screening also found that significant numbers of women who declined prenatal screening did so because the screening test did not provide a definite answer, and was unreliable (24).

The above-mentioned unfavourable test characteristics address uncertainties and risks of the prenatal tests and were frequently mentioned as decisive reasons for rejecting prenatal screening. It is therefore anticipated that future tests with better characteristics –for instance screening tests that will give more certainty (i.e. better sensitivity and specificity), and less risky diagnostic tests– will be more acceptable for pregnant women, and will consequently result in higher uptake rates. However as long as a prenatal test does not have 100% sensitivity and 100% specificity, there will be false-negative and false-positive test results. Although these risks of being wrongfully reassured or wrongfully worried may become very small, it is expected that the very possibility of a false result will keep people refraining from having the test done for this specific reason. This relates to the certainty effect, described by Tversky and Kahneman (25). The certainty effect implies that people overweigh outcomes that are considered certain, in relation to outcomes that are merely probable. For example, people generally prefer a 100% chance of winning a one-week tour of England above a 50% chance of winning a three-week tour; while people prefer a 5% chance of winning a three-week tour above a 10% chance of winning a one-week tour (25). So, changing the specificity of a prenatal screening test from 95% to 99% would result in a less positive attitude toward the test, than changing it from 99% to 100%. In other words, as long as the test is unable to give certainty, women will keep preferring non-testing above testing.

Attitude toward termination of pregnancy

When prenatal testing has detected the presence of Down syndrome or neural tube defects, the two alternative options are to either continue the pregnancy, or to terminate the pregnancy, since no treatment for these disorders is currently available. However, the alternative of continuing the pregnancy and preparing for the birth of a child with Down syndrome, for example, is a scarcely chosen option, since many studies found that the vast majority of prenatally diagnosed cases of Down syndrome are aborted (26-28). This might explain our finding that pregnant women's attitudes toward prenatal screening were strongly related to their attitude toward termination of pregnancy (chapter 3). In other words, women who did not consider termination of pregnancy in the case of Down syndrome or neural tube defects as an option for themselves, demonstrated negative attitudes toward prenatal screening, and were less keen on having a prenatal test done. Other studies also found this association between prenatal screening behaviour and one's moral value concerning termination of pregnancy (24). Moreover, the relevance of ethical considerations concerning termination of pregnancy in the decision making on prenatal screening is also indicated by the finding in chapter 2 that 12% de-

clined prenatal screening primarily because they were opposed to termination of pregnancy. This implies that prenatal counselling should also address these moral issues and should not be limited to information transfer about risks and test characteristics.

Subjective norm

A subjective norm to a certain behaviour is one's belief that specific individuals think she or he should perform that behaviour and her/his motivation to comply with this these individuals (29). This thesis addressed the role of subjective norm in pregnant women's decision making on prenatal screening at two different levels. In the first place, subjective norm was studied in the context of a hypothesised behavioural model that was based on decision theory and health behaviour theory. The core of this model consisted of the relevant constructs of theory of planned behaviour, including subjective norm and attitude toward the behaviour (29). In chapter 3 it appeared that both attitude and subjective norm were main determinants of prenatal screening behaviour. Using path modelling, it was found that subjective norm had a significant influence on pregnant women's attitudes toward prenatal screening as well. The subjective norm scale included the opinion of the pregnant woman's partner, her prenatal counsellor, and that of another important person (e.g. her mother or a friend). Initially, these findings were interpreted as worrisome, as they suggest that pregnant women seem to accept or decline prenatal screening because other people want them to do so. This would conflict with the principle of respect for autonomy and with the aim of prenatal screening, which is to enable people to make their own, informed decisions.

Subsequent analyses to explore which mechanisms lay behind this subjective norm focused on the opinions and advice of the prenatal counsellors. So, the attitude of the prenatal counsellors was regressed on different aspects of pregnant women's decision making processes (i.e. test uptake, attitude toward prenatal screening, and pregnant women's change in this attitude) (chapter 5). No influence of the counsellor on pregnant women's decisions could be found. Taking the findings of chapters 3 and 5 together, this suggests that what we initially called subjective norm is probably an indication of social support. It seems that pregnant women seek support from others (e.g. from their counsellor) for their intended choices, rather than behaving in accordance with their counsellor. This suggests that the association between subjective norm and attitude (subjective norm predicting attitude) should actually have been modelled the other way around. In other words, the subjective norm that a pregnant woman perceives from important others is likely to depend on her attitude (and intended behaviour).

Factors not influencing decision making on prenatal screening

Risk perception

Chapter 2 described the reasons pregnant women gave concerning whether or not to have the prenatal test done. It appeared that the concept of risk took up a prominent position among their justifications. Reasons that implicate perceptions of the risk of having a child with the screened disorder were mentioned by both test acceptors and test decliners. About one in eight pregnant women who reported that they would have the test done mentioned their increased risk (e.g. due to age or family history) as the decisive reason for accepting the test. Also about one in eight of those who said they would decline the test, did so because prenatal screening was not applicable or not necessary. This category included reasons like 'I am not in a risk group' (primarily because of young age), which clearly points to a low perceived risk of having a child with, for example, Down syndrome. However, although chapter 2 suggests that risk perception plays a significant role in the decision making process about prenatal screening, chapters 3 and 4 give a different picture. It appeared from both the multivariable path analysis in chapter 3, and the bivariate analysis in chapter 4 that risk perception plays a very limited role in the decision concerning whether to accept or decline the offered prenatal test. It remains a subject for further research to gain more insight into how and why these differences occur. It might possibly be related to the psychological principle of failure of introspection (30). This principle assumes that people have little introspective access to their own cognitive processes, and may be unaware of the role that various factors played in their decision making. Because of this, pregnant women's post-choice justifications might not accurately represent the cognitions that underpinned their testing behaviour. This is in accordance with Green et al.'s review of psychosocial aspects of prenatal screening. They found that the so-called 'reason-studies' generally gave a different picture of the decision making process than the 'predictor-studies' (24).

Risk perception takes up a prominent place in most health behaviour and decision making theories (31). It is a concept that has received much attention in research and counselling, and studies that investigated the role of risk perception in decision making about (prenatal) screening, generally found that risk perception does play a role in decision making (although several of those underline that it played only a moderate role) (32-37). This, however, was not reflected in the results of the multiple regression analysis in chapter 3. Why does the study presented in this thesis give such a different picture? This study concerns decision making about prenatal screening for Down syndrome and neural tube defects, which involves low risks, while many of the cited studies address decision making in high-risk contexts, e.g. breast cancer (Down syndrome: around 1:500 and neural tube de-

fects: around 1:800 vs. breast cancer: around 1:10). Moreover, the present study included an unselected population of pregnant women, whereas some other studies used selected populations (e.g. only women of advanced maternal age) (34). As the risk of Down syndrome increases with age, a sample of older respondents would also exhibit higher risks as compared to our study. These aspects suggest that in studies concerning decisions in the context of low risks, the perceptions of those risks play a less prominent role in the decision making process, as compared to studies that involve high risk decisions. This implies that the perception of the Down syndrome risk as either 1 in 10000 or 1 in 100 might not really make a difference in the attitude or intention to undergo a test, whereas the perception of a breast cancer risk as 1 in 2 or 1 in 20 might have an impact on the decision making. This hypothesis is supported by the principle of probability insensitivity, which poses that complex, emotional, and time-limited decisions are insensitive to changes in probability (38;39). It is suggested that probabilities and changes in probabilities are difficult to incorporate into a mental image (39-41). For instance, one's image of a car accident remains equally vivid and scary whether its probability of occurring is 10% or 1% (38). Studies have found that this principle of probability insensitivity particularly applies to small probabilities (39;40).

Risk communication is a standard part of counselling on (prenatal) genetic testing. However, it is a critical component of the counselling process, as counsellors find it difficult to adequately understand risks, even after being given the counselling (40;42-46). Given the limited role of risk perception in decision making on prenatal screening, the question arises as to why all these risks need to be communicated. What is the necessity for prenatal counsellors to take pains to make counsees understand these risks? It may be that these efforts are indeed unnecessary, and that just telling pregnant couples that there is a risk that the baby has a congenital defect, and that prenatal screening tests can give an indication of their personal risk of having a child with Down syndrome or neural tube defects, but that these test are not perfect as they might unnecessarily reassure or worry people, may be sufficient information for them to make an adequate decision. On the other hand, the knowledge of the risks and benefits is considered as a prerequisite for informed decision making (47;48). Risk information is assumed to be necessary for an adequate weighing of the risks and benefits. It might be that the limited role of perceptions of risk in pregnant women's decisions rather indicates a lack of comprehension of those risks. Accordingly, better risk communication would lead to decision making that is more in accordance with pregnant women's risk perceptions. However, there is no consensus on what constitutes effective, understandable risk communication, and how adequate, reasoned decision making is best facilitated (49). At the very least, it might be useful if counsellors were to stimulate

active processing of the information, as it has been shown that the active processing of risk information leads to better understanding and to more accurate risk perceptions (46). It is suggested that “risk information should be presented in the way that encourages people to think about it in an active and deliberate way” (46). These issues of the role of risk communication in prenatal counselling, and risk perception in people’s decisions needs further research.

Perceived threat

This thesis showed that neither perceived risk nor perceived severity of the screened disorder played an important role in pregnant women’s decision making process on prenatal screening (chapter 3). This is in conflict with health behaviour theory, since most social cognitive models assume that perceived threat directly or indirectly determines one’s intention to execute a certain behaviour (the theory of planned behaviour involves considerations of seriousness and probability as determinants of attitude) (31). Perceived threat was not a main predictor of prenatal screening behaviour, which indicates that pregnant women accept or decline prenatal screening irrespective of whether they see Down syndrome or neural tube defects as a threat. For instance, a pregnant woman accepts prenatal screening according to her positive attitude, although she does not perceive Down syndrome as a probable, serious disorder. From a theoretical point of view, however, perceived threat can be considered a requisite of one’s interest in prenatal screening. In the case of Down syndrome screening, it is assumed that if a pregnant woman perceives having a child with Down syndrome as a probable, serious disorder, then she will be interested in prenatal screening, and if not, she will perceive no rationale for prenatal screening. However, the results described in this thesis indicate that many pregnant women do not make decisions in this rational, deliberated manner. A low perceived threat does not lead to a low intention to undergo prenatal screening, and conversely, a high perceived threat does not make pregnant women more inclined to have prenatal screening performed. Undergoing prenatal screening to detect whether the unborn child has Down syndrome or a neural tube defect seems to be primarily based on grounds other than perceived risk and severity. These factors include pregnant women’s evaluation of the quality of the screening test, their attitudes toward termination of pregnancy and toward prenatal screening, and whether they think that important others want them to have the test done or not (chapter 3).

Emotions

The role of emotions in decision making is increasingly being recognised in recent years (41;42). The present study tried to investigate the role of emotions in prenatal

screening behaviour. Several chapters addressed different aspects of this issue. In the first place, it was shown in chapter 2 that few pregnant women gave emotional reasons for declining or accepting prenatal screening, hardly any of the test acceptors had emotional motivations, like: 'I want to have prenatal screening done because I am anxious about the health of the foetus.' Among the test decliners, however, there were more women who had emotional motivations. Over one third of them indicated anxiety/uncertainty as one of their reasons for declining. However, a closer look at this category of reasons, revealed that the reasons were of the type: 'I do not want to become unnecessarily anxious', 'the test result will make me uncertain and anxious', 'I am afraid of a positive test result'. In fact, these reasons are anticipated emotions, rather than anticipatory emotions (41). They do not reflect affective reactions to prenatal screening, which make pregnant women decline the test. It rather reveals a rational anticipation of a situation that might arouse undesirable emotions. These emotions include uncertainty and anxiety, since the screening test is unable to provide a definite diagnosis. Irrespective of receiving either a positive or a negative test result, one might feel uncertainty about the health of the foetus. A positive test result particularly can cause anxiety (19).

The anticipation of negative emotions (anxiety, uncertainty, worry) may be closely related to one's evaluation of the quality of the test. It is plausible that a pregnant woman who perceives that the test result gives too little information (as it is only a risk estimation), will be more inclined to decline the test by anticipating the uncertainty and anxiety that would remain after each test result. In contrast, a pregnant woman who perceives that the test gives enough information (as it will show her specific risk, and thus give her more certainty than before the test), will probably see no uncertainty or anxiety to anticipate upon. It is expected that future prenatal screening tests with better test characteristics (higher sensitivity and specificity, and consequently less uncertainty) will eliminate these anticipated emotions as a reason for declining the test. This reasoning supports the earlier mentioned expected increase of test uptake if better tests become available.

As risk perception has been described as a multidimensional construct containing both cognitive and affective components, chapter 4 explored whether such components of risk perception could be discerned in the context of the risk of having a child with a congenital disorder. Although the analysis confirmed the existence of these components, the hypothesis that the emotional risk perception was the better predictor of prenatal screening behaviour as compared to the cognitive risk perception could not be confirmed, since both components were bad predictors of test uptake. This hypothesis was based on the affect heuristic, which poses that people tend to use their emotional evaluation of a risk for decision making, rather than a deliberate, cognitive evaluation (50;51). This affect heuristic is as-

sumed to be more salient in complex, dreaded, time-limited situations. As a prenatal test can only be done in a specific gestational age frame of a few weeks, and pregnant women are confronted with much information about different risks, disorders, tests, and outcomes, the decision concerning whether to accept or decline prenatal screening can be considered as time-limited and complex. Thus, it was expected that the emotional risk perception would predict pregnant women's choices better than the cognitive risk perception. As none of the risk perception components were found to play an important role in the decision making, we could not confirm that pregnant women's prenatal screening decisions relied on the affect heuristic.

Finally, in chapter 3 emotions were included in the path model that was to be tested. We hypothesised that emotions concerning the health of the unborn baby determined both pregnant women's attitudes toward prenatal screening and their intentions to have a prenatal test done. The relation between emotion and attitude was not confirmed, while a statistically significant, but very small association between emotion and intention was found. Interestingly, pregnant women's worries were determined by their perceived risk and their perceived severity of Down syndrome. So, pregnant women perceiving having a child with Down syndrome as a real threat, will correspondingly be worried about the health of this child. However, these worries subsequently were not substantially related to their intentions of undergoing a prenatal test. This was an unexpected finding, as these tests are offered precisely to provide more information about the health of the child.

Summarizing then, anticipated emotions seem to play a role in the decision to decline prenatal screening, but anticipatory emotions do not appear to play a major role in pregnant women's decision making process on prenatal screening. This is in line with the analyses of Kleinveld et al. who found –in the same sample– that offering prenatal screening did not influence general feelings of anxiety or specific child-related anxiety, and that those who decided to have prenatal screening done were only slightly more anxious about the health of their child in advance (19). It appeared that emotions play a less prominent role in pregnant women's decision making processes regarding prenatal screening than was expected.

Informed decision making

In this section the findings with regard to informed decision making will be discussed. Several aspects of the assessment of informed decision making will be addressed. Finally, some ways to improve the measurement of informed decision making are presented.

Are pregnant women making informed decisions on prenatal screening?

As prenatal screening is offered to enable pregnant couples to make informed reproductive decisions, it is relevant to assess to what extent pregnant women's prenatal screening behaviour actually is grounded in informed decision making. There is ample evidence suggesting that substantial proportions of pregnant women are currently not making fully informed decisions on prenatal screening (24;47;52-54). It is even stated that informed choice in health care has been evident more as rhetoric than reality (55). Given this international status quo it was considered particularly interesting to assess informed decision making on prenatal screening in the Netherlands, where it is currently not part of standard antenatal care. It was expected that in such a context informed decision making would be more prevalent than in a context where prenatal screening is normalised. As described in the general introduction of this thesis, informed choice and informed decision making were operationalised in two different ways.

In chapter 6, informed choice was measured according to the operationalisation of the multidimensional measure of informed choice (MMIC) (56;57). This measure classifies decisions that are knowledge-based and value-consistent as informed choices. It appeared that almost one-third of the participants did not make an informed choice to accept or decline prenatal screening. In chapter 7 a measure of the decision making process was incorporated in the classification system. According to this new operationalisation, one in two pregnant women made an informed decision on prenatal screening. In sum, the chapters 6 and 7 of this thesis showed that pregnant women frequently do not make informed decisions on prenatal screening, as one in two or three pregnant women does not base her acceptance or rejection of prenatal screening on a process of informed decision making. These findings are in accordance with existing literature on informed decision making. Several studies reported percentages of informed choice of about 40-70% (14;58;59). However, it should be noted that in our study different scales for knowledge and attitude were used, so comparison of these percentages of informed choice should be treated with caution. Nonetheless, these findings indicate that there is an urgent need for interventions that facilitate well-informed, deliberated decision making.

The role of deliberation in informed decision making

As presented in the general introduction, many definitions for informed decision making exist. Some of them define an informed choice as knowledge-based, value-consistent, and behaviourally implemented, while others include a thorough decision making process as one of the aspects of an informed decision. In this thesis, prenatal screening decisions were analysed in two chapters, each with one defini-

tion as point of departure. The MMIC (based on the former definition) appeared to be a valuable and useful instrument in assessing whether pregnant women's prenatal screening behaviour is knowledgeable and consistent with their attitudes toward prenatal screening (chapter 6). Nevertheless, in our view, it disregards the decision making process, as it is restricted to the assessment of knowledge –which is a condition before a decision– and value-behaviour consistency –which is in fact an end result of a decision. Whether and how the knowledge is used in a process of deliberation remains unanswered. The relevance of deliberation for informed decision making is recognized by many researchers (3;24;46-48;60). Lerman et al. state: "While risk communication is an important component of genetic counselling, deliberation about the potential positive and negative consequences of alternate choices is often considered the more essential feature of informed decision making." This is why we tried to develop a measure that extended the MMIC by incorporating a measure of the decision making process (i.e. a measure of deliberation about the alternatives, and weighing up their pros and cons) (chapter 7).

From a theoretical point of view, decision making is inherent to the trade-off of pros and cons and weighing of the various alternatives. Both rational decision theory and health behaviour theories consider decisions to be based on deliberative processing of the available information (61-64). For instance, in subjective expected utility (SEU) theory, decision making is based on the weighing of the perceived probabilities and utilities of the possible outcomes of every alternative. The theory of planned behaviour also assumes reasoned and deliberated decision making. Furthermore, in line with many other authors, Kahneman and Tversky distinguish between two different modes of information processing: systematic, analytic processing –in which different alternatives and their pros and cons are weighed against each other– versus heuristic, intuitive processing –in which simple decision rules (heuristics) are used (65;66). The latter, called 'system 1' by Kahneman, generally operates fast, effortlessly, and implicitly, while the former mode of information processing, 'system 2', operates slowly, effortful, and deliberately controlled (65). As heuristic processing is less effortful, people tend to use this mode of processing in decision making. However, although heuristic decision making is very adequate in certain circumstances, heuristics "can systematically lead us to make poorer judgements and choices than we would by thinking in a more controlled manner about our decisions." (64). In the context of prenatal screening, system 1 involves reliance on heuristics, like: 'This is what is expected of me', 'I will do what the experts recommend', 'I'll take all the tests I'm offered' (3;24;47). Informed decision making requires deliberation in order to arm against these cognitive errors due to heuristic decision making. Precautions need to be taken, like Ulysses took precautions against himself by letting his crew chain him to the ship's mast before coming

within earshot of the Sirens (64). As it was shown that many pregnant women did not make informed decisions, partly due to lack of active, systematic information processing, interventions that enable people to make informed, deliberated decisions need to be implemented.

Limitations of present approach to informed decision making

All three components of informed decision making entail limitations. In the first place, knowledge was measured using a fixed set of questions, and a fixed cut-off point. This implies that for all pregnant women, equal amounts of knowledge determine whether it is sufficient or insufficient knowledge for informed decision making. However, people differ in the amount and type of information they want for decision making (59). For instance, is it necessary to know every detail about prenatal screening for someone who declines prenatal screening because she is ethically opposed to termination of pregnancy? The answer is: probably not. Nevertheless, although this example shows a value-consistent rejection of prenatal screening, one might argue that it is not a fully informed decision (e.g. she might lack knowledge about test characteristics). This relates to a recent proposal concerning measuring informed choice for screening (67). This proposal implies that, although everyone receives an agreed minimum of information, it is left to the counsellee to decide whether she or he wants to make an informed decision about screening. Pregnant women who do not want to make an informed decision base their decision on their own values and preferences, or those of others (67). This comes close to the proposal for the evaluation of a national prenatal screening programme in the Netherlands, which assumes that the counselling begins with the question whether the pregnant woman wants to be informed about prenatal screening (68). If not (e.g. because she is opposed to termination of pregnancy), the question whether these women make fully informed decisions will remain unanswered.

Secondly, value-consistency was assessed by checking the concordance between a pregnant woman's attitude toward prenatal screening, and her actual prenatal screening behaviour. It is assumed that these attitudes reflect underlying values. Nevertheless, as value-consistency was measured with only one attitude (toward prenatal screening), it might be that decisions that are inconsistent with this attitude are in fact consistent with other attitudes, which were not measured. For instance, a pregnant woman who is positive toward undergoing prenatal screening might decline the test because she has a negative attitude toward undergoing invasive diagnostic testing. Such a test decliner is in fact demonstrating perfectly value-consistent behaviour.

Thirdly, the six-point scale that was used to assess the level of deliberation is not the optimal measure of the actual decision making process. Although it is a short and simple instrument for gathering information about the decision making process, a more objective and extensive measure might give a more reliable picture of the process (47;69).

Another point of discussion in the context of the measurement and classification of attitude-behaviour consistency is the fact that these attitudes are usually dichotomised in either positive or negative attitudes. In general, attitudes are measured using a number of semantic differentials concerning undergoing prenatal screening. Thus, total attitude scores take up the whole range between totally positive and totally negative. Dichotomising this range of attitudes implies labelling attitudes as either positive or negative, including those scores around the midpoint of the scale.

However, as attitudes around the midpoint of the scale reflect neutral or ambivalent attitudes, it may be incorrect to consider these attitudes as positive or negative. Pregnant women with neutral attitudes toward prenatal screening decide on grounds other than attitude, as their attitude does not indicate a preference or direction for decision making. These other factors concern, for instance, the opinion of the partner or midwife/gynaecologist on prenatal screening. Additionally, aggregate attitude scores around the midpoint of the scale might also reflect ambivalence. Ambivalence toward prenatal screening indicates that a pregnant woman has conflicting attitudes toward prenatal screening. So, being negative on some items while being positive on others, thereby resulting in an aggregate score around the midpoint of the scale, might indicate ambivalence. As ambivalence involves both positive and negative feelings toward prenatal screening, attitude-behaviour consistency might be problematic in ambivalent cases as well. Because of these reasons, attitude scores around the midpoint of the scale were excluded from the analyses in chapter 6 and 7. Further research is needed to explore these issues of neutrality and ambivalence.

Measuring degree of informed decision making

The dichotomisation of criteria for informed decision making implies a very crude classification of either informed or uninformed decisions. It would be preferable to develop a more sensitive measure that assesses people's degree of informed decision making. The dichotomisations are necessary primarily because of the criterion of value-consistency. Although all scales are continuous variables, test uptake is the only variable that is dichotomous. Whether a choice is consistent with the decision maker's attitude can therefore only be assessed by dichotomising this attitude,

and classifying test accepters with a positive attitude, and test decliners with a negative attitude as value-consistent, and the opposites as value-inconsistent.

A possible solution for this problem might be that a measure of informed decision making would not include actual test uptake (being a dichotomous variable by definition), but the decision maker's intention to undergo a test. It is assumed that, for value-consistent decisions, stronger attitudes will result in stronger intentions, and more neutral attitudes will result in weaker intentions. Value-inconsistent decisions will show a discrepancy between attitude and intention. The concordance between attitude and intention (both continuous variables) can be assessed by, for example, subtracting the two scores. This would result in another continuous variable with zero indicating perfect attitude-intention consistency, and higher scores indicating larger discrepancies between attitude and intention. Finally, this variable can be combined together with the continuous variables knowledge and deliberation into a continuous measure that assesses the degree of informed decision making. This measure would range from very uninformed (less knowledge, less deliberation, less attitude-intention consistency), to very well-informed decisions. This results in a more sensitive measure. Moreover, as some components might be considered more important than others, different weights can be assigned.

A drawback of this approach is that it uses intention rather than actual behaviour. However, in the present study, it was shown that intention to undergo prenatal screening was strongly correlated with actual test uptake, so intended choice can be considered as a reliable measure of actual choice. Furthermore, it can be questioned whether the level of informedness of a decision is dependent on the actual implementation of a decision. For instance, practical barriers might cause a woman to forego prenatal screening, while she actually made an informed decision to have it done. Incongruency between attitude and the actual implementation of the intended choice does not necessarily indicate ill-informed decisions (52).

Routinisation

In many countries, prenatal screening has been part of standard antenatal care for years. Many authors have observed a process of routinisation of prenatal screening in those countries. In this section we address this issue in relation to the situation in the Netherlands, and the extent to which insights from the present study can contribute to this area. In the context of prenatal screening, routinisation or normalisation implies that prenatal screening has become accepted as a normal thing to do during pregnancy, not requiring an explicit, informed decision. Accepting

prenatal screening is not a result of a considered, conscious decision any more, but rather perceived as something self-evident.

In parts of the US, for instance, it is convincingly demonstrated that undergoing prenatal screening has become normalized (2;70-72). The same picture arises from several studies in other countries, including Israel, France, the United Kingdom, Australia, and Taiwan (53;73-78). Observing the practice of prenatal screening in California, it is concluded that prenatal screening ceases to be something about which a deliberate decision needs to be made (71;79). It is even stated that "genetic screening for all pregnant women has become almost as routine as forsaking alcohol and caffeine during pregnancy." (72). Prenatal screening is more and more considered as the normative conduct of pregnant women: "Compliance with the use of all available technologies to ensure a positive pregnancy outcome is emerging as a norm of good motherhood." (73). However, as routinisation of the utilisation of prenatal screening conflicts with informed decision making, researchers have agitated against it: "Women need to be given a choice rather than being pressured and made to conform to the routinisation of prenatal testing." (80). The consequence of the routinisation of prenatal screening is that pregnant women are often uninformed about its possible implications, which can result in anxiety or in a decision that is inconsistent with their values or preferences (72).

The present study does not provide evidence for such a practice in the Netherlands at the moment. For instance, it was found that only half of the pregnant women decided to undergo prenatal screening, that most women had clear motives for accepting or declining the test, and that prenatal counsellors did not impose their views on pregnant women's decisions. This is not surprising, as prenatal screening was not part of standard care, and only offered in a research setting. Moreover, the information booklet that pregnant women received paid special attention to an informed decision making process, also stressing that they should make their own decision. Nevertheless, the results do indicate that acceptance of prenatal screening is less based on explicit reasons (chapter 2) and informed decisions (chapter 7) as compared to test decliners. This suggests that pregnant women who undergo prenatal screening make less reasoned and deliberated decisions. While prenatal screening is currently not routinised in the Netherlands, it is anticipated that the introduction of a nationwide prenatal screening programme would accelerate its routinisation.

The issue of the routinisation of prenatal screening relates to the situation of neonatal screening in the Netherlands. Neonatal screening has become standard (utilisation of almost 100%), and having neonatal screening done does not require an informed decision based on a process of deliberation about its pros and cons. The healthcare system simply presumes that every newborn undergoes the genetic

test. As for neonatal screening, the advantages so evidently outweigh any possible disadvantages, so its normalisation might be considered less problematic than prenatal screening. As argued for prenatal screening, values and attitudes toward pregnancy, handicap, termination of pregnancy, etc. play an important role. The acceptance of a prenatal test depends on these values, and should not be assumed a priori. Many decisions about treatment of screening involve such values, and therefore are preferably based on people's active, informed decision making.

Strengths and limitations

Strengths

The first strength of this study was that it was performed at a time when neither the offer nor the acceptance of prenatal screening was normalised in the Netherlands. This provided an ideal climate for studying pregnant women's considerations and decision making concerning prenatal screening in an unbiased and unprejudiced way. A second strength of the study is that the participants constitute a large, unselected sample of the Dutch pregnant population, since participants of all ages, and from various areas of the Netherlands were included. A third strength of the study is its longitudinal design. The participants filled in several questionnaires during their pregnancy. So, variables were measured before the information about prenatal screening was provided and the prenatal test was offered, after the test offer but before the possible test result, and after the test result or a comparable point in time for those who declined the test. This enabled us to study the development and changes of attitudes, risk perceptions, emotional variables, etc. over time. The accordance or discordance between intentions to have prenatal screening done and actual prenatal screening behaviour were also able to be assessed. Furthermore, the participants were asked for their considerations and decisive reasons for accepting or declining prenatal screening at the time the decision was actually made and before the possible test result. In this way, hindsight bias was prevented, and a reliable picture of relevant motives that play a role in the decision making process was created.

Limitations

A first limitation of this study was that the decision making process was examined through self-reporting. Participants were asked to fill in and return postal questionnaires. This method of data collection is susceptible to socially desirable responses. Moreover, it appeared, for instance, in the genetic counselling context, that people's reports about their motives for, and influences on, their decisions are

not always reliable (81). So, to further analyse the decision making process about prenatal screening, process tracing techniques might be useful. For instance, pregnant couples could be stimulated to think and deliberate aloud about the prenatal screening offer and their intentions. By recording, transcribing and analysing these decision making processes, a more complete and valid picture of what motivates their intentions and what factors influence their decisions can be created. This also applies to the pregnant women's consultations with their midwife or gynaecologist.

Secondly, the questionnaires only contained questions for the pregnant women herself, and not for her partner. However, a woman's partner, who generally is one of the parents of the unborn child, must be considered as part of the decision making unit. Together they decide whether or not they want to have a prenatal test to get more information about the health of their child. As the present study did not assess the knowledge, attitudes, emotions, values, etc., of the women's partners, our insight into the decision making process is limited to that of the pregnant women. This may particularly be important because the partner may have different views about prenatal screening than the pregnant mate. That these views could diverge from each other appeared from the analysis in chapter 5. The assessment of subjective norm included a question about whether or not the woman's partner wanted her to have the test done. It was found that, although the women's attitude toward prenatal screening and their partners' opinions about having a prenatal test done significantly correlated with each other, they sometimes had different opinions about prenatal screening. Further research should include the assessment of the partners' attitudes and preferences.

Selection bias

Different types of selection bias were assessed. The recruitment of participants for the study was done by midwives and gynaecologists. They were instructed to ask every new pregnant woman who called to their practice whether she wanted to receive information about the study. A survey among the practitioners revealed that this question was asked to almost every pregnant woman who entered their practice (unpublished data). This survey also showed that practically every woman consented to receive the information letter. Thus, referral bias could not be detected. After they had read the information letter, 74% of the pregnant women gave their informed consent to participate in the study. The possibility of selective non-response was investigated through a survey among the women who did not return their informed consent form (unpublished data). This survey revealed that the main reasons for not participating in the study were lack of time, or lack of interest, suggesting that non-response bias probably did not influence the results.

However, comparison of the background characteristics of those who gave informed consent with the general pregnant population revealed that more higher educated women participated in the study. Finally, to address any withdrawal bias, the background characteristics of the sample of participants at the first measurement was compared to the follow-up measurements (unpublished data). It appeared that lower educated women dropped out more frequently. These findings are in line with the assessment of the representativeness of the sample that was used in chapter 2. Comparison of several demographic characteristics with those of the general pregnant population in the Netherlands revealed no relevant differences, except for educational level. So, both selective non-response and selective withdrawal cause bias with regard to educational level. Higher educated women have higher levels of knowledge, more negative attitudes toward prenatal screening, and make more informed decisions on prenatal screening. Moreover, higher educated women accept prenatal screening less frequently. This implies that this study might overestimate the levels of knowledge, negative attitude, informed choice, and underestimate the uptake of prenatal screening, in relation to the general pregnant population in the Netherlands. As mentioned in the general introduction, the inclusion criteria of the study were: command of the Dutch language, and less than 16 weeks of gestation. Especially the language criterion might create selection bias. Those who lack command of the Dutch language include ethnic minorities. Research in the context of prenatal screening has shown that the utilisation of prenatal screening is lower among pregnant women from minority ethnic groups (82). This might have caused an overestimation of the uptake of prenatal screening, as compared to the general Dutch pregnant population. However, this effect might be counterbalanced by the effect of educational level on test uptake.

Recommendations

Policy

During the period this study was performed, prenatal screening was not being offered to pregnant women as a standard part of antenatal care. As described in the general introduction, a nationwide prenatal screening programme is against the regulations as laid down by the Prenatal Screening Act (PSA). Recently however, the government decided that every pregnant woman should be informed about prenatal screening, and that prenatal screening should be actively offered only to pregnant women of advanced maternal age (83). Although informing about prenatal screening comes close to offering it, and the current policy in the Netherlands de facto comes down to a nationwide screening programme, the government

makes a distinction between informing and offering. In fact, this policy implies that prenatal screening no longer comes under the PSA. This is considered problematic, since it implies that the quality of prenatal screening tests, and whether they might cause a threat to the physical or physiological health of the population is not being checked (84). Accordingly, the population will not be protected by the regulations of the PSA against being informed about any prenatal test. As more genetic tests are becoming available for more disorders (of varying seriousness), genetic screening and prenatal genetic screening will continue to increase. It has to be guaranteed that informing pregnant women about these future screening tests falls under the regulations of the PSA.

Critics of prenatal screening state that offering prenatal screening entails an implicit recommendation to have the test done, and that women perceive it as an offer they can not refuse (85). In the Netherlands this is a frequently heard argument against a nationwide prenatal screening programme. However, the present study shows that less than 50 percent accept an unsolicited offer of prenatal screening, and most pregnant women make a choice that is in line with their attitude toward prenatal screening. These findings refute the suggestion that offering prenatal screening makes women feel that they should have the test done, and that it automatically leads to a high acceptance.

On the other hand, it appears that offering prenatal screening through an information booklet implies that substantial proportions of pregnant women will not make well-informed decisions. This conflicts with the objective of prenatal screening, which is to facilitate informed decision making. Since informed decision making is so frequently lacking, and the practice of prenatal screening does not meet its central objective, the question arises as to whether there is a basis for the implementation of a national prenatal screening programme. At the very least, the results of this study indicate that offering prenatal screening (i.e. informing and counselling) should go hand in hand with the implementation of some kind of decision support. Research has shown that decision aids are able to facilitate and improve informed decision making (86). Although, specific prenatal screening decision aids are being developed in the Netherlands at the moment, it is unknown to what extent such a decision aid will guarantee informed decision making. Nevertheless, without the use of decision aids, it is feared that a nationwide prenatal screening programme will lead to the routinisation of prenatal screening (i.e. almost universal utilisation without informed decision making), as has been shown in other countries and other contexts.

The practice of prenatal screening should be monitored regularly to detect undesirable developments at an early stage. This evaluation should include the assessment of the extent to which pregnant couple's prenatal screening behaviours

are grounded in their own, informed decisions (68). It should also include the assessment of the quality of the screening tests, which, in the case of the nuchal translucency measurement, can not be taken for granted (87).

Practice

The findings of this study produce several recommendations for the prenatal counselling practice.

Informing and counselling pregnant women about prenatal screening is aimed at informed decision making. As this study revealed a discrepancy between this objective and everyday practice, prenatal counsellors (i.e. midwives and gynaecologists) should make efforts to bring reality closer to that ideal. As informed decision making was defined by three criteria, these efforts should focus on those criteria. On average, about 2 out of every 10 pregnant women had insufficient knowledge about prenatal screening to make a fully informed decision. This proportion ranges from 1 out of 10 for higher educated women, to 4 out of 10 for lower educated women. So, endeavours of prenatal counsellors to comprehensibly communicate the disorders, risks, tests, outcomes, and decision alternatives may be worthwhile especially for lower educated women.

Secondly, substantial proportions of pregnant women made choices that were inconsistent with their attitude toward prenatal screening. As this may be the result of barriers (positive decliners), or subjective norms (negative acceptors), counselling on prenatal screening should be attuned to signals that indicate such barriers or pressures. Moreover, addressing attitude-behaviour consistency includes communication about underlying factors, e.g. attitude toward termination of pregnancy. As a pregnant woman's views on termination of pregnancy are closely related to her moral values, the counselling on prenatal screening should include these moral issues (i.e. value toward pregnancy, handicap, termination of pregnancy, etc.). Several researchers suggest that prenatal counselling is focussed too much on risk communication, while these risks play a very limited role in pregnant women's decision making: "Although clinicians spent a lot of time explaining the percent risk indicated by the screening test, this was not important to the way patients recalled the consultation. They did not focus on risk figures but more on a generalised notion that their baby may be in jeopardy." (88). Other studies found that counselling about prenatal screening focussed too much on the technical aspects, and less on the screened disorders, and the relation with termination of pregnancy (71;89). In short, it is recommended that counselling about prenatal screening should include discussion about both moral and social norms, and is not limited to the communication of the epidemiological risks, and the technical aspects of screening tests.

Finally, to enhance the decision making process, the counsellor should facilitate deliberation about the pros and cons of the alternatives, and about the possible test outcomes and their consequences. For instance, several authors suggest that counsellors should encourage pregnant women to deliberate about and anticipate on the possible test outcomes (72;75;90). "Although one cannot fully know how one will react to the terrible news that their foetus has an untreatable abnormality, a fully informed choice should contemplate that scenario" (72). Chiang adds: "pregnant women should be supported to think deliberately rather than just be informed" (75). This can be done by simply asking pregnant couples 'what if' questions (e.g. "What would you decide if the test results in an increased risk?")

Further research

Although many questions regarding decision making on prenatal screening were addressed in this thesis, even more questions for further research came to the fore.

Test uptake

The present study found a relatively low uptake of prenatal screening, while other national and international studies report much higher uptake percentages. Large differences in uptake rates are shown even within countries and regions. However, pregnant women's attitudes probably do not differ that much between samples within one country or region. Therefore, differences in pregnant women's attitudes toward prenatal screening only partly explain the differences in test uptake. Further research should investigate what other factors play a role here.

The relation between test uptake and informed decision making should be measured and monitored over time. Will the introduction of a nationwide prenatal screening programme decrease or increase test uptake, and will it decrease or increase informed decision making over time? This relates to the issue of routinisation of prenatal screening, which should also be a matter of further research. Should prenatal screening be prevented from becoming as normal as, for example, a dating scan? And if so, how?

Furthermore, the role of ambivalence in prenatal screening decision making should be investigated. An indication that substantial proportions of pregnant women had ambivalent feelings toward prenatal screening can be seen in the fact that many women mentioned reasons for both accepting and declining prenatal screening (chapter 2).

Factors influencing prenatal screening decisions

The role of emotions in prenatal screening making should be studied. Why do perceptions of risk and severity cause feelings of anxiety about the health of the child,

while this anxiety is largely unrelated to decision making on prenatal screening? Although many reasons for accepting or declining the test were related to risks, it appeared that risk perception did not influence decision making on prenatal screening. Further research should address this apparent discrepancy.

Subjective norm appeared to play an important role in pregnant women's decision making. Although in terms of the theory of planned behaviour subjective norm should be interpreted as social pressure to perform a certain behaviour, it was found that it might rather be interpreted as social support. Further research is needed to clarify these different interpretations. Pregnant women's decisions were highly influenced by the advice they perceived to have received from their counsellor. Did the counsellors indeed give this advice or are they retrospectively constructed by pregnant women? If so, do these directive statements reflect prenatal counsellor's own views or are they based on the counselees' preferences?

Informed decision making

With regard to informed decision making, its measurement should be further developed and validated. As described above, the measures of the three criteria of informed decision making can be improved. The validation should include its application in other contexts (e.g. prostate cancer screening). The proposed measure of the degree of informed decision making, rather than the crude dichotomous categorization should be further developed, operationalized, and validated.

Furthermore, since this study measured people's attitudes and not actual values, it should be investigated to what extent these attitudes reflect values, and if so, what values play a role in prenatal screening behaviour (values toward pregnancy, life, termination of pregnancy, handicap, etc.).

More research is needed on how to comprehensibly communicate the relevant information that is needed to make informed decisions. Moreover, what information is actually needed for informed decision making?

Finally, further research is needed to investigate methods that might resolve the discrepancy between objective and practice. Decision aids should not be limited to the improved and better structured provision of information, but should focus on all three elements of informed decision making. The effectiveness of such a decision aid should be examined before implementation. The primary outcome measure of this evaluation is the degree of informed decision making; secondary outcome measures should include satisfaction with the decision, and decisional conflict.

Conclusion

The study presented in this thesis gives more insight into the decision making process concerning whether to accept or decline prenatal screening for congenital defects. Offering prenatal screening to an unselected population of pregnant women results in a test uptake of less than 50%. Prenatal screening utilisation depends on education, religion, and the type of test that was offered. Moreover, test characteristics, attitudes, and subjective norms appeared to play important roles in pregnant women's prenatal screening decisions. Finally, the present study has demonstrated that the generally accepted aim of a prenatal screening programme, which is to enable expectant couples to make their own well-informed decisions, is frequently not achieved in the Netherlands.

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Summary

Introduction

About 2-4% of all babies are born with a major congenital abnormality. In fifteen percent of the cases there is a genetic cause, 20-30% have multifactorial causes, and the most common examples are Down syndrome (total birth prevalence: 1 in 500) and neural tube defects (total birth prevalence: 1 in 1000), respectively. Prenatal screening for these disorders has been available since the 1980s (maternal serum screening, triple test), and 1990s (nuchal translucency measurement). Prenatal screening identifies a high-risk group within a population of pregnant women. Invasive diagnostic testing is required to provide certainty about the presence or absence of the screened disorder.

Prenatal screening has become standard practice in many western countries, and an increasing proportion of foetuses with Down syndrome or neural tube defects are being diagnosed prenatally. In the Netherlands, however, prenatal screening was not offered as a routine part of antenatal care at the time of the present study. The introduction of a nationwide prenatal screening programme has been a matter of debate for over two decades. Proponents of prenatal screening argue that it gives pregnant women of all ages the opportunity to gain more knowledge about the health of the foetus in a risk-free, painless way, that it gives women reassurance, and that the prevention of grief and suffering justifies termination of pregnancy. By contrast, opponents of prenatal screening argue that prenatal screening will lead to medicalisation of uncomplicated pregnancies, uninformed choices, unnecessary anxiety and worry in pregnant women, and to social pressure.

Notwithstanding these many suppositions in relation to prenatal screening, little is known about the actual advantages and disadvantages of prenatal screening, about pregnant women's reasons for accepting or declining prenatal screening, about their decision-making processes, and about the extent to which pregnant women actually make informed decisions on prenatal screening. The study presented in this thesis aimed to give more insight into the decision making of pregnant women who are offered prenatal screening for congenital defects. This thesis addressed three central themes:

1. Uptake of prenatal screening and women's motivations for decision making
2. Factors influencing prenatal screening decisions
3. Informed decision making about prenatal screening

Methods

The study presented in this thesis forms part of a larger research project, which was a randomised controlled trial aimed to investigate risk perception, decision making and psychological wellbeing of pregnant women who are offered prenatal screening. The recruitment of study participants was done by midwives and gynaecologists, who were instructed to ask permission from every pregnant woman who entered the practice during the inclusion period to be sent a research information letter. Those who gave informed consent were randomised into the intervention group (the offer of a prenatal screening test, either the maternal serum test, or the nuchal translucency measurement), or into the control group (standard antenatal care). The test offer consisted of an information booklet, and an oral explanation. The data collection took place through postal questionnaires at different points in time during and after pregnancy. The inclusion period lasted from May 2001 to May 2003, and the very last follow-up measurement was performed in April 2004. During the inclusion period, 4077 pregnant women were asked to participate in the study, of whom 2986 (73%) gave informed consent. As this thesis involves decision making on prenatal screening, and as the participants in the control group were not offered prenatal screening, only data of pregnant women in the intervention group were used.

Uptake and motivations

Chapter 2 described the uptake of prenatal screening offered to an unselected population of pregnant women. It examined how uptake varies across the background variables. Furthermore, this chapter aimed to investigate pregnant women's reasons for either accepting or declining prenatal screening. The overall test uptake was 46%. Test uptake was significantly higher among women who were offered the nuchal translucency measurement, than among those who were offered the maternal serum test (53% vs. 38%, respectively). Test uptake appeared to be significantly lower for women who are actively religious, and for those who are higher educated. The main reasons for accepting prenatal screening were 'gaining knowledge about the health of the foetus' (50%), 'favourable characteristics of the screening test' (18%), 'increased risk of having a child with DS' (15%). The main reasons for declining were 'unfavourable characteristics of the screening test' (42%), 'not applicable/not necessary' (35%), 'anxiety/uncertainty' (36%), 'adverse characteristics of the invasive tests' (32%), and 'being against abortion' (15%). In sum, the uptake of prenatal screening was relatively low, and different distributions of reasons were reported, compared to other studies. These differences may

be due to the specific Dutch situation, in which prenatal screening is not part of standard antenatal care.

Factors influencing prenatal screening decisions

Chapters 3-5 examined what factors influenced the decision making about whether or not to undergo prenatal screening. Chapter 3 described the development and testing of a hypothetical prenatal screening model. This model was based on rational decision theory, the theory of planned behaviour, emotional decision theory, and on previous findings. It was hypothesised that a pregnant woman's perceived risk and perceived severity of having a child with Down syndrome, her attitude toward termination of pregnancy, her evaluation of the test (response efficacy), and whether other people want her to have the test done (subjective norm), all influence one's attitude toward prenatal screening. It was also hypothesised that attitude and subjective norm would directly affect prenatal screening intention, as it was expected that both a positive attitude and a positive subjective norm will increase a pregnant woman's intention to undergo prenatal screening. The fear of bearing a physically or mentally handicapped child (child-related anxiety) was assumed to be related to both attitude and intention. Path analysis showed that, after a few modifications, the model fitted reasonably to the data, implying that the hypothesised model was largely confirmed. It appeared that attitude toward termination of pregnancy, perceived test efficacy, and subjective norm determined a woman's attitude toward prenatal screening. Anxiety was influenced by perceptions of risk and severity, and by subjective norm, but appeared to be a weak predictor of intention to test. Finally, attitude and subjective norm were strongly related to pregnant women's intentions to undergo prenatal screening. In sum, undergoing prenatal screening to detect whether the unborn child has Down syndrome or a neural tube defect seems to be primarily based on grounds other than perceived risk and severity. These factors include pregnant women's evaluation of the quality of the screening test, their attitudes toward termination of pregnancy and toward prenatal screening, and whether they think that important others want them to have the test done or not.

Chapter 4 focused on risk perception and aimed to investigate whether a cognitive component and an affective component of risk perception could be identified, using two different risk perception scales (numerical and verbal). Furthermore, the role of the different risk perception components in the decision to accept or decline prenatal screening was assessed. Factor analysis revealed two factors. The first factor consisted of numerical risk perception and age-related risk, and

was discerned as the cognitive component; the second factor consisted of verbal risk perception and child-related anxiety, and was identified as the affective component. Both risk perceptions correlated very weakly with intended choice. In sum, the hypothesis that the perception of the risk of having a child with a congenital disorder can be separated into a cognitive and an affective component was confirmed, while both components seem to play only a limited role in the decision-making process.

Chapter 5 focused on the possible influence of the prenatal counsellors (i.e. midwives, gynaecologists) on pregnant women's decisions concerning prenatal screening. The three research questions were: Do prenatal counsellors' attitudes toward prenatal screening predict pregnant women's intended test uptake? Do prenatal counsellors' attitudes predict pregnant women's post-counselling attitudes toward prenatal screening? Do prenatal counsellors' attitudes predict pregnant women's attitude change from pre- to post-counselling? It was hypothesized that the influence of the counsellor can be detected at these three levels of the decision-making process. Furthermore, it was hypothesized that pregnant women's perception of the counsellor's advice (to accept or decline the prenatal screening test) would influence their decision-making process. The majority of the pregnant women had made their decision before the consultation with their prenatal counsellor, and most women reported that their counsellor did not reveal his/her opinion about prenatal screening. However, substantial minorities of participants perceived that their counsellor deviated from non-directiveness by either hinting at or advising positively or negatively about undergoing prenatal screening (9% and 19%, respectively). Multilevel regression analyses revealed that neither uptake rates, nor attitude toward prenatal screening were significantly predicted by the counsellors' attitudes toward prenatal screening. In conclusion, although in our study substantial numbers of counselees reported having been given advice, either implicitly or explicitly, the regression analyses did not confirm the frequently mentioned influence of genetic counsellors' attitudes on counselees' decision making.

Informed decision making

It is generally accepted that the central objective of offering pregnant women information about prenatal screening is to enable them to make informed choices, rather than to achieve as high uptake rates as possible. Chapters 6 and 7 aimed to investigate informed decision making among women who are offered prenatal screening. According to the different definitions of informed decision making, and

the different approaches in the measurement of informed decision making, this thesis used two different operationalisations of informed decision making.

In chapter 6, informed choice was measured according to the operationalisation of the multi-dimensional measure of informed choice (MMIC). This measure classifies decisions that are based on sufficient knowledge about prenatal screening, and that are consistent with the decision maker's values as informed choices. Chapter 6 aimed to assess to what extent pregnant women who are offered prenatal screening for congenital defects make informed choices. In addition, it was aimed to assess the psychological effects of informed decision making. Of the participants, 83% had sufficient knowledge about prenatal screening. Knowledge was associated with age, and with educational level. Of the pregnant women with a positive attitude toward prenatal screening, 76% decided to have the test done; of those with a negative attitude, 88% declined prenatal screening. Overall, 82% of the pregnant women made value-consistent choices. Sixty-eight percent of the pregnant women were knowledgeable and value-consistent, i.e. made an informed choice. Informed choice was associated with less decisional conflict (this applied only to test acceptors), and more decisional satisfaction. Anxiety scores did not differ between women who made an informed choice and those who did not make an informed choice. It was concluded that the majority of the pregnant women possessed sufficient knowledge to make an informed choice about prenatal screening, and most of them had values that were consistent with the choices they made. However, almost one-third of the women could not be classified as having made an informed choice about prenatal screening. Informed choice appeared to be psychologically beneficial.

Chapter 7 aimed to construct a measure of informed decision making that incorporated the assessment of the decision-making process in the classification system described above. To measure the decision-making process, a scale that assessed the extent of deliberation about the alternatives and their pros and cons was used. So, an informed decision was defined by knowledge, deliberation, and value-consistency. Furthermore, this chapter aimed to investigate differences in the level of informed decision making between test acceptors and test decliners. It appeared that only 51% of the pregnant women fulfilled all three criteria and were classified as having made an informed decision about prenatal screening. Women who accepted the prenatal screening offer made significantly less informed decisions as compared to those who declined the test. This difference was mainly caused by the lower level of deliberation among test acceptors.

Discussion

In the general discussion (chapter 8), it is mentioned that the percentage of pregnant women who accepted the prenatal screening test they were offered (less than 50%) is much lower when compared to studies in other national and international studies. This difference may partly be explained by the degree of routinisation of prenatal screening. However, other factors probably also play a role here (e.g. study design, regional differences). Characteristics of both prenatal screening and prenatal diagnostic tests were frequently mentioned reasons for accepting or declining prenatal screening. Moreover, perceived test efficacy appeared to be a determinant of a woman's attitude toward prenatal screening. It is therefore expected that future tests with better characteristics (i.e. screening tests with higher sensitivity and specificity, and less risky diagnostic tests) will result in a higher uptake of prenatal screening. Attitude toward prenatal screening (primarily determined by a pregnant woman's attitude toward termination of pregnancy) and subjective norm were the most important factors in explaining prenatal screening behaviour. This indicates that moral and social considerations play important roles in the decision making on prenatal screening. It was concluded that pregnant women frequently do not make fully informed decisions on prenatal screening. So, for many pregnant women the aim of offering prenatal screening, which is to enable people to make informed decisions, is not achieved. This finding is in line with many international studies. This indicates that there is urgent need for interventions that facilitate informed, deliberated decision making. Such decision aids show promising results, although their ability to enhance informed decision making in the context of prenatal screening still has to be evaluated.

In many countries where prenatal screening has been part of standard antenatal care for some years, a process of routinisation of prenatal screening has taken place. This implies that it has become normal and self-evident to undergo prenatal screening during pregnancy. This conflicts with the concept of informed decision making, which implies considered, conscious decision making. Although the present study does not provide evidence for such a practice in the Netherlands, the high percentage of pregnant women who did not make fully informed decisions, especially among test acceptors, is an alarming finding.

Recommendations

Since informed decision making is so frequently lacking, implying that prenatal screening does not meet its central objective, this study poses the question of whether there is a basis for the implementation of a national prenatal screening

programme. At the very least, it indicates that offering prenatal screening should go hand in hand with the implementation of some kind of decision support. Besides the use of decision aids (e.g. website, booklet), prenatal counsellors should encourage pregnant women to deliberate about, and anticipate, the possible test outcomes. Moreover, the findings of the study suggest that prenatal counselling should also discuss moral and social issues and should not be limited to information transfer about risks and test characteristics.

Further research should evaluate (prenatal) screening programmes by investigating the degree of informed decision making. The measurement of informed decision making should be further developed and validated. More research is needed on how to comprehensibly communicate the relevant information that is needed to make informed decisions. Finally, further research is needed to investigate methods that might resolve the discrepancy between the objective and the practice of prenatal screening.

Samenvatting

Inleiding

Twee tot vier procent van alle baby's wordt geboren met een aangeboren afwijking. In 15% van de gevallen is er alleen een genetische oorzaak, terwijl het in 20 tot 30% van de gevallen om multifactoriële aandoeningen gaat. Downsyndroom en neuraalbuisdefecten behoren tot de meest voorkomende aangeboren aandoeningen (totale geboorteprevalentie: 1 op 500 en 1 op 1000, respectievelijk). Prenatale screening naar deze aandoeningen is beschikbaar sinds de jaren tachtig (maternale serum test, triplettest) en negentig (nekplooiemeting) van de vorige eeuw. Prenatale screening identificeert een hoogrisicogroep binnen een populatie zwangere vrouwen. Invasieve diagnostiek is nodig om zekerheid over de aan- of afwezigheid van de aandoening waarop gescreend werd, te krijgen.

Prenatale screening is in veel Westerse landen onderdeel van de standaard prenatale zorg geworden en een toenemend percentage van de foetussen met Downsyndroom of neuraalbuisdefecten wordt prenataal gediagnosticeerd. In Nederland echter, werd, tijdens de uitvoering van deze studie, prenatale screening niet standaard aangeboden aan alle zwangere vrouwen. Over de invoering van een dergelijk nationaal screeningsprogramma wordt al meer dan twintig jaar gedebatteerd in ons land. Voorstanders van prenatale screening vinden dat het zwangere vrouwen de mogelijkheid geeft om op een risicoloze en pijnloze manier, meer informatie over de gezondheid van de foetus te verkrijgen, dat het voor geruststelling kan zorgen, en dat het voorkómen van verdriet en leed een eventuele selectieve abortus rechtvaardigt. Aan de andere kant, vinden tegenstanders van het standaard aanbieden van prenatale screening dat het zal lijden tot medicalisering van ongecompliceerde zwangerschappen, tot slecht geïnformeerde keuzes, tot onnodige angsten en zorgen, en tot sociale druk.

Ondanks de vele meningen en veronderstellingen van zowel voor- als tegenstanders van prenatale screening, is er maar weinig bekend over de daadwerkelijke voor- en nadelen van een prenataal screeningsprogramma, over de redenen van zwangere vrouwen om wel of geen prenatale screening te ondergaan, over hun besluitvormingsproces, en over de mate waarin zwangere vrouwen geïnformeerde beslissingen nemen over prenatale screening. De studie waarover in dit proefschrift verslag wordt gedaan, had tot doel om meer inzicht te verkrijgen in de besluitvorming van zwangere vrouwen die prenatale screening naar aangeboren afwijkingen van de foetus aangeboden krijgen. Dit proefschrift behandelt drie centrale thema's:

1. Het percentage zwangeren dat prenatale screening laat doen en de motivering van de besluitvorming

2. Factoren die beslissingen over prenatale screening beïnvloeden
3. Geïnformeerde besluitvorming over prenatale screening

Methode

Deze studie was onderdeel van een groot onderzoeksproject. Dit project bestond uit een gerandomiseerde gecontroleerde trial, en was erop gericht om de risicoperceptie, de besluitvorming, en het psychisch welbevinden van zwangere vrouwen die prenatale screening aangeboden krijgen, te onderzoeken. De rekrutering van deelnemers aan de studie werd uitgevoerd door 44 verloskundige en gynaecologische praktijken. De verloskundigen en gynaecologen waren geïnstrueerd om alle zwangere vrouwen die zich gedurende de inclusieperiode aanmeldden in de praktijk te vragen of zij een onderzoeksinformatiebrief opgestuurd wilden krijgen. De deelnemers werden vervolgens via randomisatie ingedeeld in de interventiegroep of in de controlegroep (gebruikelijke zorg). De deelnemers in de interventiegroep kregen een prenatale screeningstest aangeboden: ofwel the maternale serumtest, ofwel de nekpluoi meting. Het testaanbod bestond uit een informatieboekje over de screeningstest, en een mondelinge toelichting door de verloskundige of gynaecoloog. De dataverzameling vond plaats middels vragenlijsten die de deelnemers op 5 tijdstippen tijdens en na de zwangerschap thuisgestuurd kregen.

De inclusieperiode duurde van mei 2001 tot mei 2003, en de laatste vragenlijst werd ingevuld in april 2004. Gedurende de inclusieperiode werden 4077 zwangere vrouwen gevraagd om deel te nemen aan de studie, waarvan er 2986 (73%) *informed consent* gaven. Omdat dit proefschrift handelt over besluitvorming over prenatale screening, en de deelnemers in de controlegroep geen prenatale screening aangeboden kregen, werden alleen gegevens van vrouwen uit de interventiegroep gebruikt.

Het percentage zwangeren dat prenatale screening laat doen en de motivering van de besluitvorming

In hoofdstuk 2 werd onderzocht welk percentage van de zwangere vrouwen die prenatale screening aangeboden kregen, de test daadwerkelijk liet doen, en of dat percentage verschilde tussen bepaalde subgroepen. Ook werd onderzocht wat de redenen van zwangere vrouwen zijn om wel of geen prenatale screening te laten doen. Het percentage zwangeren dat inging op het screeningsaanbod was 46%. Dit percentage was significant hoger onder zwangeren die de nekpluoi meting aange-

boden kregen, dan onder hen die de maternale serumtest aangeboden kregen (resp. 53% en 38%). Vrouwen die actief religieus waren, en vrouwen die hoger waren opgeleid, lieten significant minder vaak prenatale screening doen. De belangrijkste redenen om de test te laten doen waren: 'kennis/nieuwsgierigheid' (50%), 'gunstige screeningsteigenschappen' (18%), en 'verhoogd risico (leeftijd of familiegeschiedenis)' (15%). De voornaamste redenen om geen prenatale screening te laten doen waren: 'ongunstige screeningsteigenschappen' (42%), 'angst/onzekerheid' (36%), 'niet nodig/niet van toepassing' (35%), en 'negatieve eigenschappen van vervolgdagnostiek' (32%). Vergelijken met andere studies is het percentage zwangeren dat koos voor prenatale screening relatief laag, en gaven zwangeren andere redenen voor hun beslissing. Deze verschillen kunnen te maken hebben met de specifieke Nederlandse situatie waarbij prenatale screening geen onderdeel van de standaard prenatale zorg is.

Factoren die beslissingen over prenatale screening beïnvloeden

In hoofdstuk 3 t/m 5 werd onderzocht welke factoren de besluitvorming over prenatale screening beïnvloedden. Hoofdstuk 3 beschrijft hoe een hypothetisch beslissingsmodel voor prenatale screening werd ontwikkeld en getest. Dit model was gebaseerd op verschillende beslissingstheorieën en op eerdere bevindingen. De volgende hypothesen lagen ten grondslag aan het model: (1) De attitude van een zwangere vrouw ten opzichte van prenatale screening wordt bepaald door haar perceptie van het risico op en de ernst van het krijgen van een kind met Downsyndroom, haar attitude tegenover selectieve abortus, haar mening over de kwaliteit van de test, en of anderen vinden dat zij de test al dan niet moet doen (subjectieve norm). (2) Attitude t.o.v. prenatale screening en subjectieve norm zouden direct van invloed zijn op de intentie om een prenatale test te laten doen. (3) De laatste hypothese was dat de angst om een fysiek of mentaal gehandicapt kind te krijgen van invloed zou zijn op de attitude van de zwangere vrouwen tegenover prenatale screening, en op haar intentie om de test te gaan doen. Padanalyse liet zien dat het model na enkele modificaties redelijk bij de data paste. Met andere woorden, de hypothesen over de verschillende relaties tussen factoren werden grotendeels bevestigd. Het bleek dat attitude tegenover abortus, de mening over de testkwaliteit, en subjectieve norm gerelateerd waren aan de attitude van een zwangere tegenover prenatale screening. Gevoelens van angst werden beïnvloed door de risicoperceptie en de perceptie van de ernst van de aandoening, en door subjectieve norm, maar angst bleek een slechte voorspeller van de intentie om de test te laten

doen. Tenslotte, subjectieve norm en attitude hadden een sterke invloed op de intentie van een zwangere om prenatale screening te laten doen. Kortom, de intentie om prenatale screening naar het risico dat het ongeboren kind Downsyndroom of neuraalbuisdefecten heeft, te laten doen, lijkt primair te zijn gebaseerd op andere gronden dan de perceptie van het risico en de ernst. De besluitvorming is vooral gebaseerd op de evaluatie van de kwaliteit van de test, de attitude tegenover abortus, en op de mening van andere belangrijke mensen (zoals verloskundige/gynaecoloog, partner, vriendin, moeder, etc).

Hoofdstuk 4 focust op risicoperceptie. Onderzocht werd of risicoperceptie, door middel van twee verschillende risicoperceptieschalen (numeriek en verbaal), onderscheiden kon worden in verschillende componenten (cognitief en emotioneel). Ook werd geanalyseerd wat de rol van deze beide risicopercepties was in de besluitvorming over prenatale screening. Uit factoranalyse bleek dat er inderdaad een cognitieve component en een emotionele component te onderscheiden waren. Uit regressieanalyse bleek dat de verbale risicoperceptie (de emotionele component) bleek niet voorspellend was voor de intentie om prenatale screening te laten doen, en de numerieke risicoperceptie (de cognitieve component) wel, maar dit effect was erg klein. Beide risicopercepties lijken dus een beperkte rol te spelen in de beslissing om wel of geen prenatale screening te laten doen.

Hoofdstuk 5 behandelt de mogelijke invloed van de prenatale counselor (verloskundige, gynaecoloog) op de beslissing over prenatale screening van een zwangere. De drie onderzoeksvragen waren: is de attitude tegenover prenatale screening van een counselor voorspellend voor de keuze van een zwangere vrouw om wel of geen prenatale screening te laten doen? Is deze attitude van de counselor van invloed op de attitude van een zwangere vrouw tegenover prenatale screening? Is de attitude van de counselor voorspellend voor de eventuele attitudeverandering van een zwangere? De meerderheid van de deelnemers had vóór het consult met de verloskundige of gynaecoloog de beslissing om wel of geen prenatale screening te laten doen al genomen. Hoewel de meeste zwangeren rapporteerden dat de counselor zijn of haar mening over prenatale screening niet liet merken, zei een substantiële minderheid dat de counselor de eigen mening liet doorschermen of zelfs een advies gaf om de test wel of niet te doen. Multipelle regressie toonde aan dat noch de uiteindelijke keuze, noch de attitude(-verandering) significant voorspeld werd door de attitude van de counselor. De vaakgehoorde suggestie dat counselors de besluitvorming van hun cliënten beïnvloeden, werd dus niet bevestigd door deze studie.

Geïnformeerde besluitvorming over prenatale screening

Het aanbieden van prenatale screening heeft niet tot doel om zoveel mogelijk zwangeren een prenatale test te laten ondergaan, of om zoveel mogelijk geboortes van kinderen met Downsyndroom of neuraalbuisdefecten te voorkomen, maar om zwangeren een staat te stellen een geïnformeerde beslissing hierover te nemen. Het thema geïnformeerde besluitvorming wordt in de hoofdstukken 6 en 7 behandeld. Er zijn verschillende definities van geïnformeerde besluitvorming, en dus ook verschillende benaderingen om geïnformeerde besluitvorming te meten. Daarom worden in hoofdstuk 6 en 7 verschillende operationalisaties van geïnformeerde besluitvorming gebruikt. In hoofdstuk 6 werd gebruik gemaakt van de operationalisatie van de *multidimensional measure of informed choice* (MMIC). Dit instrument classificeert beslissingen aan de hand van de hoeveelheid kennis over prenatale screening en van de mate van overeenstemming tussen de attitude tegenover prenatale screening en de uiteindelijke keuze. Een beslissing die op voldoende kennis is gebaseerd, en die overeenkomt met de attitude van de zwangere wordt gedefinieerd als een geïnformeerde keuze.

Hoofdstuk 6 had tot doel om vast te stellen hoeveel zwangeren zo'n geïnformeerde keuze over prenatale screening maakten, en wat de psychische effecten zijn van een geïnformeerde keuze ten opzichte van een ongeïnformeerde keuze. Het bleek dat 83% van de deelnemers voldoende kennis had over prenatale screening, en het kennisniveau gerelateerd was aan leeftijd en opleiding. Ongeveer eenderde van de deelnemers had een positieve attitude tegenover prenatale screening, en driekwart van hen liet ook daadwerkelijk prenatale screening doen. Ook eenderde van de deelnemers was negatief over prenatale screening, en 88% van deze groep liet de test niet doen. In totaal nam 82% van de zwangere vrouwen in het onderzoek een beslissing over prenatale screening die overeenkwam met de eigen attitude. 68% van de deelnemers had én voldoende kennis, én nam een beslissing in lijn met de eigen attitude. Met andere woorden, tweederde maakte een geïnformeerde keuze. De zwangeren die een geïnformeerde keuze hadden gemaakt ervoeren minder beslissingsconflict en waren meer tevreden met de beslissing, dan degenen die geen geïnformeerde keuze hadden gemaakt. Er waren geen significante verschillen in de hoeveelheid angst die men ervoer. Kortom, hoewel de meerderheid van de deelnemers voldoende op de hoogte was van de belangrijke aspecten van prenatale screening en beslissing nam die in overeenstemming was met de eigen mening/visie op prenatale screening, maakte eenderde van de deelnemers geen goedgeïnformeerde keuze over prenatale screening.

In hoofdstuk 7 werd aan het hierboven beschreven classificatiesysteem een derde element toegevoegd, te weten: het beslissingsproces. Met de MMIC wordt informatie verkregen over het kennisniveau en of de keuze overeenstemt met de attitude van de zwangere in kwestie, maar er is niets bekend over het beslissingsproces. Hoe is de keuze tot stand gekomen? Is er een afweging van voor- en nadelen geweest? Heeft men de consequenties van de verschillende keuzemogelijkheden doordacht? Om dit beslissingsproces te meten is gebruik gemaakt van een vragenlijst die naar bovenstaande aspecten vraagt. In hoofdstuk 7 werd een goed geïnformeerde beslissing dus gedefinieerd door voldoende kennis, consistentie tussen attitude en gedrag, én deliberatie. Het bleek dat slechts de helft van de deelnemers voldeden aan al deze drie criteria, en dus slechts de helft aangemerkt kon worden als geïnformeerde beslissers. De keuze om de prenatale test te laten doen was minder vaak een geïnformeerde beslissing dan de keuze om de test niet te laten doen. Dit verschil werd voornamelijk veroorzaakt doordat zwangeren die de test lieten doen lager scoorden op de vragenlijst over het beslissingsproces en dus een minder weloverwogen beslissing namen, dan de vrouwen die besloten om de test niet te laten doen.

Discussie

Het percentage zwangeren dat de aangeboden prenatale screeningstest liet doen (minder dan 50%), is lager dan in veel andere studies. Dit verschil zou verklaard kunnen worden doordat in veel landen prenatale screening routinematig wordt aangeboden aan zwangeren, en daardoor een standaard onderdeel is geworden van de prenatale zorg. Aan de andere kant kunnen ook andere factoren een rol spelen (zoals bijvoorbeeld de opzet van de studie of regionale verschillen). De eigenschappen van de prenatale test werden vaak genoemd als reden om de test niet te doen (bijv. 'de test geeft geen zekerheid'). Daarom is de verwachting dat toekomstige testen met betere testeigenschappen (bijv. screeningstesten die meer zekerheid kunnen geven over de aan- of afwezigheid van de aandoening bij het ongeboren kind) ertoe zullen leiden dat meer zwangeren de prenatale screening zullen laten doen. De attitude ten opzichte van prenatale screening (voornamelijk bepaald door de opvatting over abortus) en subjectieve norm (of mensen in de omgeving vinden dat ze de test al dan niet moet laten doen) waren de belangrijkste factoren die de keuze voor of tegen prenatale screening verklaren. Dit wijst erop dat morele en sociale overwegingen een belangrijke rol spelen in de besluitvorming over prenatale screening. Veel deelnemers bleken geen volledig geïnformeerde keuze te maken over prenatale screening. Dit betekent dat voor veel zwangeren het doel

van het aanbieden van prenatale screening –mensen in staat stellen een geïnformeerde beslissing te nemen– niet bereikt wordt. Dit betekent dat er een urgente noodzaak is om interventies te ontwikkelen die mensen helpen bij het nemen van een goedgeïnformeerde, weloverwogen keuze. Zulke zogenoemde *decision aids* geven veelbelovende resultaten, maar of ze het aantal geïnformeerde prenatale screeningsbeslissingen kunnen doen toenemen, moet verder onderzocht worden.

In veel landen waar prenatale screening jarenlang onderdeel is van de standaard prenatale zorg, is een proces van routinisatie opgetreden. Dit betekent dat het normaal en vanzelfsprekend is geworden om prenatale screening te laten doen tijdens je zwangerschap, iets waar geen expliciete beslissing meer over genomen wordt. Deze routinisatie is echter in strijd met het doel van geïnformeerde besluitvorming (het maken van een bewuste, weloverwogen keuze). Hoewel deze studie geen bewijs levert voor het optreden van routinisatie van prenatale screening in Nederland, is het hoge percentage zwangeren dat geen geïnformeerde beslissing nam (met name onder zwangeren die de test lieten doen) een verontrustende bevinding.

Aanbevelingen

Aangezien goedgeïnformeerde, weloverwogen besluitvorming over prenatale screening vaak achterwege blijft, en het doel van prenatale screening dus niet gehaald wordt, komt de vraag naar voren of dit een goede basis is voor de introductie van een nationaal programma voor prenatale screening naar aangeboren afwijkingen in Nederland. Het aanbieden van prenatale screening zou tenminste gepaard moeten gaan met de implementatie van een vorm van beslissingsondersteuning. Verloskundigen en gynaecologen zouden zwangeren moeten stimuleren om de voor- en nadelen van prenatale screening af te wegen en te anticiperen op de mogelijke testuitkomsten. Verder wijzen de resultaten van deze studie erop dat de prenatale counseling zich niet alleen zou moeten richten op informatieoverdracht van risico's en testeigenschappen, maar ook op morele en sociale aspecten van prenatale screening.

Evaluatieonderzoek van (prenatale) screeningsprogramma's zou zich moeten richten op de mate van geïnformeerde besluitvorming. Verder onderzoek is nodig naar de ontwikkeling en validering van meetinstrumenten van geïnformeerde besluitvorming. Ook is nog veel onbekend over hoe de complexe informatie over screening het best op een begrijpelijke wijze gecommuniceerd kan worden. Ten slotte is verder onderzoek nodig naar methoden die de discrepantie tussen doel en praktijk van prenatale screening zouden kunnen dichten.

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*De naam van de HEER zij geprezen,
Van nu tot in eeuwigheid.
Van waar de zon opgaat tot waar zij ondergaat,
Zij geloofd de naam van de HEER.
Psalm 113 vers 2 en 3*

About the author

Matthijs van den Berg was born on July 14th 1978, in Dordrecht, the Netherlands. In 1996, he completed his secondary school education at Scholengemeenschap Het Wartburg College in Rotterdam, and started studying biomedical sciences at the University of Leiden. He graduated in 2001 after a final internship period at the department of Medical Decision Making of the Leiden University Medical Center, where he studied patient autonomy in the physician-patient relation. In that same year he started working for the ChristenUnie/SGP party in the European Parliament for the temporary committee on human genetics and other new technologies in modern medicine. In January 2002 he began as a PhD student at the EMGO Institute of the VU University Medical Center in Amsterdam, where he conducted the study described in this thesis. In the five years till 2006 he followed the post-graduate epidemiology programme, resulting in a Masters degree in Epidemiology. He is currently working as a researcher at the centre for public health forecasting of the National Institute for Public Health and the Environment (RIVM). Matthijs van den Berg is married to Sigrid Visser, and they have one daughter (Meike).