Determinants affecting pregnant women’s utilization of prenatal screening for Down syndrome: a review of the literature

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ABSTRACT

Objective: Uptake rates for Down syndrome screening (DSS) in the Netherlands are low compared with those in Northern European countries (27% versus 61% in the United Kingdom and 90% in Denmark). These differences are unexpected, especially since the countries have similar cultural and social values.

Method: To analyse factors that underlie differences in uptake we reviewed current literature on individual characteristics and healthcare system factors, which determine potential influential factors on utilization of DSS.

Results: Arguments “Against abortion” and “Down syndrome (DS) not severe enough condition to terminate pregnancy” correlated with declining DSS, whereas “Perceived guidance of healthcare professional,” “Perceived negative attitude of society towards DS” and “Preparation/ Early termination” correlated with accepting DSS. However, the majority of determinants were used both in favour of accepting or declining DSS.

Conclusions: Decision making regarding DSS seems a process with influences on different levels and subtle interactions rather than a dichotomous process. Utilization may also be influenced by the implicit way it is being offered, as an extra option or as part of routine care and how this offer is perceived by the social environment. Finally, national healthcare system characteristics, although underrepresented in the examined studies, may also influence participation in DSS.
INTRODUCTION

Down syndrome screening (DSS) is performed in the first trimester of pregnancy by maternal serum screening and by ultrasound measurement of the fetal nuchal translucency. Its aim is to inform prospective parents on the risk of Down syndrome (DS) in the ongoing pregnancy, to provide them with timely options, including invasive diagnostic procedures in case of an increased risk for DS, and, if diagnosed for DS, preparation for a disabled child or termination of pregnancy (TOP) [1]. In the Netherlands, DSS is offered to all pregnant women. Accepting or declining should be an informed and autonomous choice as consequences of a screen positive result and knowledge about the condition might lead to complex ethical, emotional and social dilemmas. Therefore, the choice should be based on accurate and balanced information and in concordance with the client’s attitude and values [2].

Uptake rates of DSS in the Netherlands are low compared to those of other Northern European countries (27% versus 61% in the UK and 90% in Denmark [3–7]). As these countries show close resemblance regarding social and cultural factors, have an extensive screening program and are comparable both in content, execution and basic values of informed and autonomous choice, these findings are unexpected [8–11].

Preceding a larger study on those factors that underlie the differences in uptake, this article aims to study the current literature on factors that affect participation in DSS, including individual characteristics and healthcare characteristics, and their mutual interaction.

A general framework to study differences in uptake is the Andersen Model of Health Care Utilization (Figure 1). This model suggests that people’s use of health services is a function of their predisposition to use services, factors which enable or impede use, and their need for care. The predisposing characteristics are subcategorized in demographic, social and health beliefs factors. Demographic factors represent the biological imperatives people need to use health services. Social structure represents factors that determine the status of a person in the community, the ability to cope with problems and to command resources to deal with these problems, such as existing social network, social interaction and culture. Health beliefs stand for attitudes, beliefs and knowledge that people have on health and health service and that might influence their subsequent perceptions of need and use of healthcare services. The enabling or impeding factors consist of health system characteristics (availability of facilities and healthcare personnel), organizational characteristics (accessibility of health services, waiting time), financial factors (health insurance and reimbursement) and personal factors (means and knowledge to access and use services, travel time). Need factors can be classified in perceived and evaluated need. Perceived need represents the imperative that accounts for personal care-seeking, consumption of services and adherence to a medical regimen while evaluated need represents professional judgement of health status [12].

To our best knowledge, a narrative review of the literature integrating the effect of both the individual and healthcare characteristics on uptake has not been published before. Therefore, we searched the literature on studies that reported determinants of influence on screening behaviour and classified these determinants according to the Andersen model. The purpose of this study was to answer the following question:
• Which are the individual and environmental factors (predisposing, enabling and need), known from the current literature that correlate with participation in DSS?

METHOD

Literature research
We have identified full text articles on studies in the peer-reviewed literature in English regarding determinants of influence on utilization of prenatal screening for DS. Studies were obtained from a search in Medline, Embase and PsycINFO from January 1990 up to March 2012. The starting date corresponded with the introduction of the first DSS program in the early nineties. The syntax is listed in Table 1.

Inclusion criteria:
• Participants included pregnant women, or women with a history of past pregnancy
• Participants regarded participation in prenatal screening
• Studies which described factors of influence on decline or acceptance of prenatal screening
• Both qualitative and quantitative studies
• Studies that considered pregnant women’s views
• Studies from Western countries
• Studies describing prenatal diagnosis, TOP, expert opinions or the supply side were excluded.

Mixed method
We explicitly chose a mixed method approach, integrating both quantitative and qualitative studies. The analysis of quantitative data provides a general understanding of the research problem and a representative picture of the situation, while the analyses of qualitative data complement and refine those statistical results by exploring participants’ views in more depth. Synthesis of either qualitative or quantitative data alone may obscure factors that are potentially important in altering the uptake [13,14]. The conclusions of a review can be substantially altered by the inclusion of qualitative data, which are more likely to reflect the experiences of the target group [15].

[TABLE 2]
For quantitative studies significant results were collected, for qualitative studies we collected author’s findings and created relevant themes. Subsequently, the results of the studies were grouped according to the Andersen classification as perceived by pregnant women. Finally, the results were categorized in major topics and subdivided into qualitative or quantitative results.
RESULTS

Studies included
The literature search (Table 1) identified 422 publications. Study of the title showed that 124 of these met the inclusion criteria. Screening the abstracts of these publications revealed that 52 abstracts met all the inclusion criteria and were assessed for the full publication. Thirteen studies of the 422 met all inclusion criteria. These are listed in Supplementary table. Five studies had a quantitative design, and eight had a qualitative design. The results were categorized according to their effect on acceptance or decline of DSS. The subdivision is summarized in Table 2. A description per subcategory is given below.

Decline

Predisposing
Health beliefs and social factors were the categories mentioned most often as influential in declining DSS. Health beliefs could be subdivided into attitudes towards DS, disability, abortion, nature and experience. Women who declined DSS weighed the possibilities of an adverse screening result in relation to their moral beliefs and values [16]. Women with most favourable attitudes towards people with DS were the least likely to intend to test and terminate [17]. In both qualitative and quantitative studies, women mentioned that DS was a condition, not severe enough to terminate pregnancy [18–23].

The qualitative data showed a more in depth explanation of women’s values and beliefs. The attributed value to life with a disability, and their expected ability to cope emotionally with a disabled child, were important reasons for declining [20]. The moral authority of nature, in which nature has to take its course and whereby having a healthy or handicapped child is destiny, has been mentioned in qualitative studies as a reason to decline [18,20]. Frequently mentioned reasons to decline were “being against abortion” or “not wanting to have an abortion,” and were found in qualitative as well as in quantitative studies [22,23]. Qualitative data illustrated that views on abortion were directly related to screening choices, showing that women who were reluctant to consider termination, did not consider participating in DSS [22]. Personal experience with DS and DSS was associated with either being in favour or against testing [18,23,24]. In both qualitative and quantitative studies, negative experiences of friends with prenatal testing, such as stress and grief [19,23,24], particularly with false positive results, influenced women towards declining DSS. Test decliners more often lacked embodied knowledge (e.g. they had not experienced serious illnesses themselves, nor were they aware of illness in their families), and therefore, presumed that DSS was unnecessary [23].

Decliners acknowledged that all parents want a healthy child but at the same time felt that variability and complexity in ability and health should be maintained in society. Besides, they felt the need to defend their decision towards society [18].

Enabling
Determinants categorized as enabling factors concerned mainly organizational factors. Unfavourable characteristics, for example, unreliability of the test, and adverse characteristics of a possible subsequent invasive test, were mentioned as reasons to

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Decline DSS, both in qualitative and quantitative studies [18,22]. In quantitative studies, practical reasons, such as unsuitable visiting hours for test performance, were also reported to influence the utilization of DSS [19].

Need
Unjust anxiety or uncertainty, due to possible false positive results, were frequently mentioned as reasons to decline DSS, both in qualitative and quantitative studies [19,20,22]. Additionally, in qualitative studies, decliners considered avoiding unnecessary risks for the fetus as the most rational action in the best interest of the child [20]. A number of women declined because testing was perceived as not applicable (e.g. “I am too young”) [22].

Accept
Predisposing. The determinants of accepting DSS can be roughly divided into two categories; health beliefs and social factors. The attitude towards DS or parenting a child with DS, was directly related to accepting DSS. Almost all women holding a negative attitude towards parenting a child with DS tended to accept screening [17]. In both qualitative and quantitative data, screening was accepted without direct association with abortion, but as a step towards further diagnosis [17,20]. The qualitative data gave a more detailed explanation of attitudes. Women mentioned their “philosophy of life” as the determining factor in the decision to have DSS [24]. Although a disabled child might help family members to cope with adversity and to appreciate the value of little and irrelevant things, the expected unbearable emotional and physical burden of a child with DS had been decisive in accepting testing [20]. However, for some acceptors the attitudes towards disability were not always based on knowledge, but were a reflection of their wish for a healthy baby [25]. Acceptors considered testing as a possibility to control nature and to interfere with the pregnancy outcome in order to guarantee a good life for themselves and their children. This attitude was only found in qualitative data [20,24]. Professional experience with children with DS could become a factor either in favour or against testing [23]. Women with personal experience, from close associations with others, on raising a child with DS, had vividly anticipated a life with a child with a disability. These women chose testing to be prepared or have the option to terminate [23]. With regards to the external environment of pregnant women, qualitative studies have reported that women who accepted screening more often mentioned guidance from their healthcare professional. Perceived negative attitude of society towards DS or social discrimination have also been mentioned as influencing factors on accepting DSS [21,25].

Enabling. The enabling factors could be subdivided into financial and organisational factors. In qualitative studies the financial burden of a child with DS has been mentioned as a reason to accept testing, especially by prospective fathers [16,25]. Quantitative and qualitative data showed that favourable characteristics and technical advances of the screening test were also factors in accepting testing [16,19,22,25].

Need. Need factors could be divided into objective need and subjective need. A priori increased risk for DS (e.g. advanced maternal age) is frequently considered as
a medical reason to perform a screening test (‘objective need’), and therefore, mentioned as a reason why women decide to accept testing [22]. For some women accepting testing was a self-evident act, and considered as part of routine care. This was confirmed in both qualitative and quantitative, studies [19,25]. Among the subjective or personal need factors, ‘seeking reassurance,’ ‘hope for a healthy baby’ and ‘reduction of anxiety’ were often mentioned as reasons to accept DSS, both in qualitative and quantitative studies [17,19,20,23,25]. The possibility to prepare in case of a determined disability and to terminate early were also reported reasons to accept [16,19,23]. In addition, a number of women mentioned becoming knowledgeable or curious [22]. Best interest of the child, and ensuring a good life for their child (prevention of life with a major disability) were other reasons to accept testing [20].

**DISCUSSION**

In this review, determinants of influence on uptake of DSS, as perceived by pregnant women were studied according to their effect on participation (Table 2). We categorized the determinants according to the Andersen model of Health Care Utilization, thus covering factors at the individual and at the health system level. Our results indicate that the factors ‘Against abortion’ and ‘DS not severe enough condition to terminate pregnancy’ correlated with declining whereas ‘perceived guidance of healthcare professional,’ ‘perceived negative attitude of society towards DS’ and ‘preparation/early termination’ correlated with accepting DSS [16,19–23,25]. Most factors mentioned by decliners could be categorized as predisposing (dominated by health beliefs). Decliners often had strong personal opinions, which hardly seem to be influenced by the guidance of health professionals or existing enabling factors in the healthcare system. Acceptors also relied on predisposing factors, but their attitudes were socially more diverse.

In contrast to the identified differences, we were also able to identify nine major similarities in arguments concerning participation in screening. Our results indicate that ‘nature,’ ‘attitude towards DS,’ ‘experience,’ ‘supportive influence of others,’ ‘test characteristics,’ ‘reassurance,’ ‘anxiety,’ ‘best interest of the child’ and ‘participation’ were mentioned both in favour of and against screening. They comprised predisposing, need and enabling factors. By including both qualitative and quantitative studies we were able to complement, confirm and refine the statistical results. ‘Perceived negative attitude of society,’ ‘perceived guidance from healthcare professionals,’ ‘nature’ and ‘best interest of the child’ were factors extracted from qualitative data alone and may be a more specific explanation of the wish to reduce anxiety and seek reassurance [19,22,25].

First, we can conclude that both women accepting and those declining refer to the same factors to support their choice (Table 3), which has also been mentioned by others [20,22,26]. A possible explanation is that the process of decision making, particularly with regard to ethical, emotional and social dilemma’s, is not a rational and dichotomous process. Perhaps, decisions on DSS are made on different levels, reflected in predisposing and need factors, and are justified on rational grounds (need and enabling). Women decide on prenatal testing by balancing the information provided by the test against the risk of further investigation, the emotional burden of a disabled child on their well-being and their life perspective, as well as that of...

family members. Normative moral principles are introduced when the choice is made, namely, as factors justifying and supporting the decision [20]. As consequences of a screen positive result and knowledge about the condition emerge complex ethical, emotional and social dilemmas, informed decision making remains import- ant, but one should realize that women might use the information to justify their intuitive choice, rather than making a choice after non-directive counseling. The finding that the majority of women (60–70%) decide before consult-

ation could also be a reflection of this [27,28].

The emphasize of “healthcare beliefs” factors affecting utilization is in line with previous studies on women’s reasons for participation in DSS which have shown relationships between attitudes (health beliefs) and actual testing behaviour [2,17,27– 30]. A recent Dutch study demonstrated that a relatively positive attitude toward DS and a negative attitude toward TOP were related to the low uptake in this country. Most women would not consider TOP in case of a diagnosed DS [30]. Healthcare beliefs (attitudes and values) remain useful in explaining the individual use of healthcare, but cannot explain the large differences in uptake between Northern European countries. The existing uptake differences suggest correlations not only with individual characteristics, such as health beliefs and need, but also with healthcare system characteristics and their mutual interaction. Therefore, second, we conclude that system characteristics, such as (perceived) enabling or impeding factors (e.g. universal offer, reimbursement, informed choice), which are underexposed in literature, could be of influence on national participation in DSS. In the included studies, DSS is formally offered in an opt-in fashion. In practice, utilization may be influenced by the implicit way it is offered, as an extra option or as part of routine care. Moreover, it may not be directly influenced by the way it is offered, but rather by how utilization is perceived by the social environment. The moral significance of prenatal testing is inseparably bounded by the social context in which it is practiced. This effect of national healthcare policies has not been addressed before.

The effect of both individual and healthcare characteristics on utilization of DSS has not been extensively studied and this article summarizes the available data for the first time. Due to low number of studies, originating from only five countries, the general applicability of the study results may be limited. Restriction to English language studies could have excluded locally published studies on influential factors in European countries. The mixed method approach strengthens the findings of this study, as the more general results were confirmed and broadened by the more specific results of the qualitative studies.

In conclusion, with this review we suggest that decision making regarding DSS is rather a subtle process of influences on different levels than a dichotomous process. Decision making might be justified later, on rational grounds. Another factor of influence might be whether it is offered as an option or as part of routine care. Future research should focus on the subtle process of decision making and on the influence and role of different levels. Moreover, more research is needed on the influence of healthcare system characteristics, such as enabling and impeding factors, and on the mutual interaction of these factors.

|TABLE 3|
Declaration of interest

The author is supported by foundation of Friends of the UMC Utrecht (UMC Utrecht grant organisation).

REFERENCES


**FIGURE AND TABLES**

![Figure 1](image-url)  
*Figure 1. The Andersen model of health behaviour [12]. Reprinted with permission from the American Sociological Association.*
### Table 1. Search syntax.

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<thead>
<tr>
<th>Topic</th>
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<th>Quantitative</th>
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<td></td>
</tr>
<tr>
<td>DS not being severe enough to terminate pregnancy</td>
<td>Gottfredsdottir 2009, Garcia 2008</td>
<td>Santalahti 1998</td>
</tr>
<tr>
<td>Perceived guidance from healthcare professionals</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Perceived negative attitudes of society towards DS</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Enabling</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Need</td>
<td></td>
<td></td>
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<tr>
<td>Part of routine care</td>
<td></td>
<td></td>
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<tr>
<td>Abortion/prepare</td>
<td></td>
<td></td>
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</tbody>
</table>
Table 3. Differences and similarities between arguments against or in favour regarding the utilization of prenatal screening for Down’s syndrome, grouped according to the Andersen model and subdivided in qualitative and quantitative studies.

<table>
<thead>
<tr>
<th>Topic</th>
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<th>Decline Quantitative</th>
<th>Accept Qualitative</th>
<th>Accept Quantitative</th>
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<td>-</td>
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<tr>
<td>Perceived guidance from healthcare professionals</td>
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<td>Perceived negative attitudes of society towards DS</td>
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<tr>
<td>Abortion/prepare</td>
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<td>-</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Similarities</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Attitudes, beliefs and values</td>
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<tr>
<td>Participation</td>
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<td>+</td>
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<tr>
<td>Nature</td>
<td>+</td>
<td>-</td>
<td>+</td>
<td>-</td>
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<tr>
<td>Experience</td>
<td>+</td>
<td>+</td>
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<td>-</td>
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<td>Supportive influence of significant others</td>
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