



A meta-synthesis of pregnant women's decision-making processes with regard to antenatal screening for Down syndrome

Bernie Reid ^{a,*}, Marlene Sinclair ^a, Owen Barr ^a, Frank Dobbs ^b, Grainne Crealey ^c

^a Institute of Nursing Research, University of Ulster, Magee Campus, United Kingdom

^b Institute of Postgraduate Medicine and Primary Care, University of Ulster, Coleraine, United Kingdom

^c Clinical Research Support Centre, Belfast, United Kingdom

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ABSTRACT

The diffusion of antenatal screening programmes for Down syndrome has triggered much discussion about their powerful potential to enhance pregnant women's autonomy and reproductive choices. Simultaneously, considerable debate has been engendered by concerns that such programmes may directly contribute to the emergence of new and complex ethical, legal and social dilemmas for women. Given such discussion and debate, an examination of women's decision-making within the context of antenatal screening for Down syndrome is timely. This paper aims to undertake a meta-synthesis of qualitative studies examining the factors influencing pregnant women's decisions to accept or decline antenatal screening for Down syndrome. The meta-synthesis aims to create more comprehensive understandings and to develop theory which might enable midwives and other healthcare professionals to better meet the needs of pregnant women as they make their screening decisions. Ten electronic health and social science databases were searched together with a hand-search of eleven journals for papers published in English between 1999 and 2008, using predefined search terms, inclusion and exclusion criteria, and a quality appraisal framework. Nine papers met the criteria for this meta-synthesis, providing an international perspective on pregnant women's decision-making. Twelve themes were identified by consensus and combined into five core concepts. These core concepts were: destination unknown; to choose or not to choose; risk is rarely pure and never simple; treading on dreams, and betwixt and between. A conceptual framework is proposed which incorporates these themes and core concepts, and provides a new insight into pregnant women's complex decision-making processes with regard to antenatal screening for Down syndrome. However, further research is necessary to determine whether or not the development of a model of decision-making may empower pregnant women in making choices about screening.

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Introduction

The development and implementation of new reproductive technologies, including antenatal screening for Down syndrome, has triggered much discussion about their powerful potential to enhance pregnant women's autonomy and reproductive choices (Brown & Webster, 2004; Buchanan, Brock, Daniels, & Wilker, 2000; Scully, Banks, & Shakespeare, 2006). Concurrently, considerable debate is engendered by concerns that such technologies may directly contribute to the emergence of new and complex ethical, legal and social dilemmas for women (Brown & Webster, 2004; Getz & Kirkengen, 2003). In analyzing such contradictory

perspectives, Rapp (2000, page 3) points to the complexity of the decision-making processes confronting women, arguing that:

'.... the construction and routinization of this technology is turning women to whom it is offered into moral pioneers: situated on a research frontier of the expanding capacity for prenatal genetic diagnosis, they are forced to judge the quality of their own fetuses, making concrete and embodied decisions about the standards for entry into the human community.'

Consequently, there has been a proliferation of studies focusing on the decision-making processes of pregnant women with regard to antenatal screening for Down syndrome, many of them qualitative. With more qualitative studies focusing on women's experiences of screening, the ways in which they decide to accept or decline screening, their responses to specific tests and the influence of screening on women's attachment to their fetuses (Garcia et al.,

* Corresponding author. Tel.: +44 02871375191.

E-mail address: reid-b@email.ulster.ac.uk (B. Reid).

2002), one must ask: What generalizations can be made from the findings of these qualitative studies that will enable midwives and other healthcare professionals to better meet the needs of women as they make their decisions about screening, and how can these studies contribute to the development of theory in the area of decision-making? Separately, the studies may provide glimpses into the experiences of women as they ascribe meaning and reason to their decisions yet do not always contribute significantly to a full understanding of women's decision-making processes (Jenson & Allen, 1996; Paterson, Thorne, Canam, & Jillings, 2001; Zimmer, 2006). To create a more comprehensive understanding of study findings, single studies may be synthesized (Bondas & Hall, 2007; Paterson et al., 2001; Sandelowski & Barroso, 2003a; Zimmer, 2006). In other words, the findings from existing studies may be clarified and filled out into thickly descriptive and comprehensive understandings of women's decision-making processes which, in conceptual terms, are greater than the sum of the parts (Schrieber, Crooks, & Stern, 1997; Zimmer, 2006). Thus, attaining a level of conceptual or theoretical development beyond that achieved by any individual study may increase the usefulness of study findings.

Moreover, such theoretical developments may subsequently be tested empirically or, alternatively, used to explicate a number of existing theoretical frameworks tentatively called upon to explain women's decision-making processes. Such theoretical frameworks include, for example; rational choice (The Theory of Planned Behaviour, Ajzen & Fishbein, 1980) and role adaptation theories (Maternal Role Attainment, Mercer, 1995). The Theory of Planned Behaviour (Ajzen & Fishbein, 1980), for example, posits that in making a decision about screening, women evaluate the potential outcomes of their decision, their own beliefs and attitudes, their perceptions of what they think significant others think they should do, and how difficult it will be to accomplish the decision and then form the intention to carry out the decision if these components converge to support it. In contrast, Mercer's (1995) theory of 'maternal role attainment' tentatively suggests that women's decision-making processes may journey through four stages: anticipatory, formal, informal and personal identity. The anticipatory stage is a time of expectation about pregnancy and future motherhood. During the formal stage women may be guided by formal, consensual advice from experts and significant others in the social system. During the informal stage, women may progress from following the guidance of others and begin to structure their decisions to fit themselves and their families according to their past experiences and future goals. The stage of personal identity is characterized by a sense of confidence and satisfaction as women may accept and integrate their decisions. Yet, there is a demonstrable lack of existing literature examining the potential application of such theoretical frameworks to decision-making processes about screening (Wohlgemuth, 2006).

Summarizing, Estabrooks, Field, and Morse (1994) and Paterson et al. (2001) point to the significance of synthesis in presenting more comprehensive understandings and developing theory as a means towards strengthening and facilitating the greater use of qualitative findings in midwifery and healthcare practice. This paper therefore reports on a synthesis of qualitative research studies relating to pregnant women's decision-making processes with regard to antenatal screening for Down syndrome.

Methodology

Meta-synthesis is perhaps the most well developed methodology for synthesizing qualitative studies (Britten, Campbell, Pope, Donovan, & Morgan, 2002). Meta-synthesis is 'research of research' (Paterson et al., 2001, page 5) which incorporates analysis of the theory, methods and findings of existing qualitative research

studies and subsequent synthesis of these insights to provide new understandings of the phenomenon (Jenson & Allen, 1996; Nelson, 2002; Paterson et al., 2001; Sandelowski, Docherty, & Emden, 1997). In contrast to an aggregated review of qualitative literature on a given phenomenon (Zimmer, 2006) or to secondary analysis of primary data from previous studies (Sandelowski & Barroso, 2003a), meta-synthesis represents a discrete and distinct approach to new inquiry based on a critical re-interpretation of existing studies (Paterson, Thorne, & Dewis, 1998; Paterson et al., 2001). It creates a process by which the nature of re-interpretation is explicit and meanings that extend well beyond those presented in existing studies are distilled (Fingeld, 2003; Jenson & Allen, 1996; Paterson et al., 2001). Yet the process is not one in which summation of meanings are valued over the rich and thick descriptions of individual qualitative studies (Walsh & Downe, 2005), but rather seeks to 'carefully peel away the surface layers of studies to find their hearts and souls in a way that does the least damage to them' (Sandelowski et al., 1997, page 370). Moreover, in undertaking such a process, studies may relate to each other in one of three ways: they may be directly comparable as reciprocal translations; they may stand in opposition to each other as refutational translations; or taken together they may represent a line of argument (Britten et al., 2002; Noblit & Hare, 1988).

As an interpretative qualitative research approach, the philosophical underpinnings of meta-synthesis are firmly rooted in the tenets of a constructivist orientation to epistemology and geared towards an understanding of how individuals construct and reconstruct knowledge about a phenomenon (Denzin, 1989; Guba & Lincoln, 1994; Paterson et al., 2001; Zimmer, 2006). It is therefore important to understand that in any meta-synthesis approach, the construction of research findings occurs at three levels (Schutz, 1962). Firstly, qualitative study participants construct their own understandings and meanings related to the phenomenon of interest. Schutz (1962) refers to this as first-order constructs. Secondly, the authors of existing studies have constructed their research findings in accordance with their own understanding and interpretation of the data. Such secondary-order constructs are influenced not only by the authors' own lived experiences but also framed within the epistemological assumptions of the chosen methodology (Paterson et al., 2001; Schutz, 1962). Thirdly, those undertaking meta-synthesis bring together a number of such second-order constructs with all their methodological and contextual complexity, to construct third-level constructs or interpretations, once again, with their own epistemological slant (Paterson et al., 2001; Schutz, 1962; Zimmer, 2006). Consequently, they deal with re-constructions of re-constructions of constructions. In this way, meta-synthesis connects the 'common sense' understandings and meanings of study participants with the 'scientific world of concepts and theories' in order that an underlying reality might be found, however fragile such a reality might be (Aspers, 2004, page 4).

Method

Concise outlines of the processes followed by the meta-synthetic method are provided by several authors and offer many common approaches (Jenson & Allen, 1994; Noblit & Hare, 1988; Sandelowski & Barroso, 2003b). The meta-synthesis presented in this paper was based on the method of Noblit and Hare (1988). This classic method, developed to reconcile different ethnographers' interpretations of the same phenomena in studies of educational institutions, facilitates a rigorous procedure for deriving substantive interpretations about any set of ethnographic or interpretative studies (Bondas & Hall, 2007). Moreover, it is the qualitative method most commonly cited in meta-synthesis publications

(Bondas & Hall, 2007). It is nonetheless important to acknowledge that this paper represents our reading of the studies selected; other authors with dissimilar interests may read the studies differently. However, recording an audit trail, which enables the reader to follow the actions taken, will ensure that whilst an alternative reading is possible the basis for this meta-synthesis is transparent, the nature of re-interpretation explicit and the steps repeatable.

Defining the meta-synthesis question

This meta-synthesis formed an initial exploratory phase of a more extensive research study which aims to examine holistically the decision-making processes of pregnant women with regard to antenatal screening for Down syndrome. The meta-synthesis question was therefore defined as:

'What factors influence pregnant women's decisions to accept or decline maternal serum and/or nuchal translucency screening for Down syndrome?'

A preliminary scoping of the literature by one member of the research team (BR) led us to consider that this question was broad enough to capture the attributes of women's decision-making within the context of antenatal screening for Down syndrome but narrow enough to create a feasible limit to the number of existing qualitative studies, to be included in the meta-synthesis (Paterson et al., 2001). In addition, the question allowed the meta-synthesis to be inclusive of both women who had accepted and women who had declined screening.

Searching for studies

Any qualitative research study involving pregnant women of any nationality or ethnicity who had made decisions about whether to accept or decline maternal serum and/or nuchal translucency screening for Down syndrome at any stage during pregnancy was eligible for inclusion. Qualitative research was liberally defined as encompassing a highly diverse array of orientations to, and strategies for, inquiry that generally concern themselves with how the social world is interpreted, understood, experienced and produced (Barroso et al., 2003). Noblit and Hare (1988) question the blending of such different qualitative methodologies because of the potential to complicate the meta-synthesis with substantively different types of knowledge that cannot be easily translated into one another. However, in keeping with the viewpoint of Sandelowski et al. (1997) and Paterson et al. (2001), this meta-synthesis sought to include the fullest possible range of qualitative methodologies. The search for studies was limited to those which focused on decision-making about antenatal screening for Down syndrome from the perspective of individual pregnant women themselves, and not as predefined by professionals, researchers or policymakers. It was considered that this would facilitate an interpretation of decision-making processes that was as unimpeded as possible by taken-for-granted assumptions, as well as offering the potential to elucidate any possible differences in decision-making within and between different groups of pregnant women. Nonetheless, such selective searching proved both laborious and time-consuming.

All relevant English language qualitative research studies published in peer-reviewed journals between January 1999 and January 2008 were retrieved. The decision to commence the search in January 1999 coincided with a subsequently reported increased momentum towards offering all pregnant women, in Western-industrialised countries, antenatal screening for Down syndrome (Rapp, 2000; Sher, Romano-Zelekha, Green, & Shohat, 2003; Vassy, 2006; Ward & Muir Gray, 2002). Adopting the approach of Sandelowski and Barroso (2005), search terms were truncated where

appropriate and included combinations of topic, theme and method terms (Table 1). Ten electronic databases were searched together with a hand-search of eleven journals (Table 2). Hand-searching involved one of the authors (BR) manually searching the content pages of the print version of all eleven journals. Despite our efforts to undertake a comprehensive search, we acknowledge that our strategy focused primarily on health related databases and journals and therefore may have omitted some relevant studies published in English. It is also possible that our use of search terms, for example; 'narrative/narrative analysis' may have predisposed to the omission of studies termed 'conversational' or 'discourse analysis'.

Selecting studies

The process of selecting the studies was highly iterative and closely aligned to constructionist epistemologies. One member of the research team (BR) independently undertook the search for studies. The total number of published works retrieved amounted to over twelve thousand. The overwhelming majority of these works were excluded on the basis of the title alone which indicated that they were either not relevant to the meta-synthesis or were not qualitative research studies. Where this was not clear, the abstract was reviewed. A total of one-hundred-and-eighty-nine abstracts were reviewed and one-hundred-and-eighty were subsequently excluded on the basis that they did not include pregnant women as participants (as in explorations of healthcare professionals perceptions of women's decision-making processes); no human beings were participants (as in discourse or content analyses of media representations of why women decide to accept or decline screening); studies focusing on women's health screening where distinctions could not be made between decision-making about antenatal screening and cancer screening; and mixed-method studies in which qualitative findings could not be separated from quantitative findings.

Characteristics of the selected studies

The nine studies selected for inclusion in the meta-synthesis reflected an international perspective on pregnant women's decision-making with regard to antenatal screening for Down syndrome. Two of the three UK-based studies shared some common authors and used the same data set to focus on women's perceptions of first trimester ultrasound screening (Williams et al., 2005) and of being at higher risk (Heyman et al., 2006). A co-author of the two UK-based studies had previously authored one of the two Israeli studies. These studies explored the perspectives of two very different groups of women living in Israel, namely Bedouin women (Lewando-Hundt et al., 2001) and Jewish women of European origin (Reminick, 2006). Further studies originated from the US (Markens, Browner, & Press, 1999), Australia (Liamputtong, Halliday, Warren, Watson, & Bell, 2003),

Table 1
Search terms.

Topic	Prenatal screening, prenatal testing, antenatal screening, prenatal screening, fetal screening, fetal abnormality, Down syndrome, trisomy 21, maternal serum screening, nuchal translucency, ultrasound screening, genetic screening, genetic testing.
Theme	Decision-making, women's decisions, experiences, women's experiences, factors influencing, psychological aspects, ethical aspects, socio-cultural aspects, legal aspects, spiritual aspects.
Method	Case study, constant comparative analysis, descriptive study, ethnography, exploratory, field study, focus group, grounded theory, hermeneutic, interview/interview study, narrative/narrative analysis, naturalistic inquiry, phenomenology, purposive/purposeful sample, qualitative study/qualitative research, thematic analysis.

Table 2
Sources for search.

Electronic databases	Journals
Allied and Complementary Medicine	Birth
British Nursing index	British Journal of Midwifery
CINAHL	Ethnography
EMBASE	Health Care for Women International
Genetics Abstracts	Journal of Advanced Nursing
MEDLINE	Journal of Contemporary Ethnography
National Research Register	Journal of Midwifery and Women's Health
Ovid MyJournals	Midwifery
PsycInfo	Social Science & Medicine
Sociological Abstracts	Women's Health Issues

Taiwan (Chiang, Chao, & Yuh, 2006) and the Netherlands (Garcia, Timmermans, & van Leeuwen, 2008). Yet despite reference to the US study in three of the subsequent studies, no study had explicitly built upon the findings of those published before it and hence findings had not been brought together. Moreover, it is acknowledged that the studies selected present qualitative findings at different times during pregnancy in relation to women's decision-making processes: pre-screening (four studies); post-screening (two studies); after twenty-four weeks gestation (two studies) and at an unknown time (one study). Some studies considered screening only whilst other studies included diagnostic testing; some considered maternal serum screening whilst others considered nuchal translucency screening; and some were reflective of screening as routine practice whilst others were not. The reader therefore needs to place the findings of this meta-synthesis within such a diverse context.

Appraising the quality of the selected studies

Existing literature relating to the appraisal for quality of individual studies is equivocal (Walsh & Downe, 2006). Britten et al. (2002) adopted a pragmatic approach, prioritizing the need for a worked example of the stages of meta-synthesis over a rigorous quality assessment of studies. Despite the value of such an approach, it nonetheless raises the possibility that meta-synthesis of methodologically flawed studies may result in flawed meta-synthesis (Walsh & Downe, 2006). In keeping with such a viewpoint, the studies in this meta-synthesis were subjected to a detailed quality assessment using a framework developed specifically to appraise qualitative research (Walsh & Downe, 2006). This framework assessed the appropriateness and coherence of the study scope and purpose; methodology; methods including sampling strategy, data collection methods, ethical dimensions, analysis and interpretation; relevance and transferability; and researcher reflexivity. A summary quality grade was then allocated to each study (see Table 3) with grade **A** denoting no or few flaws, the study credibility, transferability and confirmability are high; grade **B** having some flaws, unlikely to affect the credibility, transferability, dependability, and/or confirmability of the study; grade **C** having some flaws which may affect the credibility, transferability, dependability and confirmability of the study; and grade **D** having significant flaws which are likely to affect the credibility, transferability, dependability and/or confirmability of the study. The quality of the studies was generally fairly good, with some weaknesses in the use of techniques to ensure the transparency of methodology; sampling strategy and analysis; and in accounting for ethical dimensions and reflexivity. A note of caution is nonetheless warranted as journals may not include all such considerations on publication despite them being part of the relevant study. Indeed one of the welcome by-products of

undertaking meta-synthesis may be an improvement in the quality of published reporting of qualitative research (Campbell et al., 2003).

Analyzing and synthesizing the selected studies

Appraising quality was essentially the first phase of repeated reading of the studies leading to the identification of recurring key themes across studies. This stage helped us to identify how the studies were related. The first iteration of key themes was derived from both the verbatim text of participants in the studies and from the themes identified by authors of the studies (Table 4, Column 1). In Schutz's (1962) terms, such key themes are representative of first- and second-order constructs respectively. In order to remain faithful to the themes arising from the studies, the original terminology used was preserved in identifying key themes.

Translating the studies into one another entailed examining the key themes in relation to others in the selected study, and across studies, and was analogous to the method of constant comparison used in qualitative data analysis. The translation of key themes from one study to another was idiomatic rather than word for word translation. This led to the final iteration of key themes which reflects our terminology (Table 4, Column 2). Again, these may be considered as second-level constructs (Schutz, 1962).

By translating the studies into one another, it was possible to establish relationships between the nine studies. The studies were not refutations of one another. Indeed the relationship between them seemed to be reciprocal, from which a line of argument could be developed. By considering each key theme in turn and using them as building blocks, we derived a first iteration of five core concepts (Table 4, Column 3). A final iteration of core concepts was developed using key phrases borrowed from English literature (Table 4, Column 4). Taken together, these five core concepts represent the line of argument, constituting the synthesis achieved. Such core concepts may be considered third-order constructs (Schutz, 1962) and it is these that represent a level of conceptual development beyond that attained by individual existing studies. Table 5 outlines the relationship between terms used in the meta-synthesis process.

The process of appraising quality, analysis and synthesis began with the oldest study (Markens et al., 1999). Such an approach was consistent with Noblit and Hare's (1988, page 62) conception of synthesis as being '... historical in that it uses time to give order and history-in-use to give context'. Nevertheless, it is possible that the order in which the studies were appraised, analysed and synthesized may have affected the outcome of the synthesis.

Furthermore, the process of appraising quality, analysis and synthesis was led by one member of the research team (BR). Findings were fed back to other team members, each member of the research team having read each of the nine studies. Differences between team members emerged particularly with regard to the utility of appraising the quality of studies; the degree of importance attributed to particular key themes and the identification of core concepts. Such divergent views may be considered reflective of the diverse professional and research backgrounds of individual team members. Nevertheless, building upon such diversity and by means of a series of discursive meetings, the final composite analysis and synthesis were agreed by consensus.

Findings

The findings of this meta-synthesis are presented in terms of the five emergent core concepts: destination unknown; to choose or not to choose; risk is rarely pure and never simple; treading on dreams and betwixt and between. The key themes constituting

Table 3
Profile of studies.

Study	Markens et al., 1999, US	Lewando-Hundt et al., 2001, Israel	Liamputtong et al., 2003, Australia	Pilnick et al., 2004, UK	Williams et al., 2005, UK	Heyman et al., 2006, UK	Chiang et al., 2006, Taiwan.	Reminnick, 2006, Israel.	Garcia et al., 2008, Netherlands
Discipline	Sociology/psychiatry/anthropology	Sociology	Public health	Sociology/midwifery/fetomaternal medicine	Sociology/midwifery/anthropology	Sociology/anthropology/midwifery	Nursing	Sociology	Medicine/ethics/philosophy
Scope and purpose	To compare the explanations of women who declined maternal serum screening with those of women who accepted	To gain an understanding of the selective uptake of maternal serum screening by Palestinian Bedouin women	To examine women's reasons for declining both ultrasound and maternal serum screening, and invasive diagnostic tests	To investigate the relationship between information given by midwives and decision-making by pregnant women offered nuchal translucency screening.	To explore the experiences of women offered first trimester maternal serum and nuchal translucency (NT) screening	To explore the meaning of higher risk status to women undergoing first trimester maternal serum and nuchal translucency (NT), and second trimester maternal serum screening	To describe women's decisions with regard to second trimester maternal serum screening	To explore the key social influences involved in the growing range and prevalence of antenatal genetic tests from the perspective of Israeli Jewish women of Ashkenazi (European) origin	To gain insight into the impact of ethical views on women's decision-making processes regarding maternal serum and nuchal translucency screening
Methodology	Not stated	Not stated	Not stated	Not stated	Not stated	Not stated	Grounded theory	Social construction of 'good' motherhood; medicalisation of reproduction; consumerist paradigm of childbearing.	Qualitative
Methods									
Sampling strategy	Not stated	Not stated	Not stated	Not stated	Not stated	Purposive sampling	Convenience sampling	Not stated	Random sampling from RCT intervention group
Sample	25 low-risk women aged 18–35 years who declined screening recruited after 24th week of pregnancy. Comparison sample of women who accepted screening not described	16 women offered screening but unclear if they accepted or declined. Unclear if women recruited during pregnancy and/or during postnatal period	28 women aged 37 years and older who declined both screening and diagnostic tests recruited at or after 24th week of pregnancy	14 women offered and accepting of nuchal translucency screening recruited pre-screening	14 women recruited post-screening. 11 women accepted serum and NT screening (9 screened 'low risk' and 2 'high risk'); 2 women declined serum but accepted NT screening and 1 woman declined all screening	27 women (22 < 34 years, 5 > 34 years) recruited pre-screening, post-screening/ diagnostic testing and postnatally (5 declined screening, 14 screened low risk, 7 screened high risk, and 1 miscarried before screening	26 women aged 22–35 years who accepted screening, received a high risk result and were awaiting diagnostic testing	50 women recruited pre-screening (27 women accepted screening and 23 women declined screening)	59 women in the process of deciding to accept or decline screening
Data collection methods	Tape-recorded interviews	In-depth interviews	Questionnaire containing open questions	Tape-recorded consultations and interviews	Tape-recorded interviews conducted as guided conversations	Tape-recorded interviews	Participant observation and tape-recorded interviews	Tape-recorded interviews	Semi-structured tape-recorded interviews
Ethical considerations	Not stated	Not stated	Ethical approval obtained	Ethical approval obtained	Not stated	Not stated	Consent to record interviews obtained	Not stated	Reference to protecting participant anonymity. Ethical approval obtained

(continued on next page)

Table 3 (continued)

Study	Markens et al., 1999, US	Lewando-Hundt et al., 2001, Israel	Liamputtong et al., 2003, Australia	Pilnick et al., 2004, UK	Williams et al., 2005, UK	Heyman et al., 2006, UK	Chiang et al., 2006, Taiwan.	Reminnick, 2006, Israel.	Garcia et al., 2008, Netherlands
Relevance/transference	Yes	Inadequate discussion to establish plausibility	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Provides new insights/understandings	Yes	Yes	Yes	Yes	Yes	Yes	Some	Some	Yes
Findings interwoven with relevant literature/theory	Limited	Limited	Yes	Some discussion of how findings refute those of other studies	Findings interwoven with relevant explanatory literature and existing theories	Findings interwoven with relevant explanatory literature and existing theories.	Findings interwoven with relevant explanatory literature and existing theories.	Findings interwoven with relevant explanatory theories and socio-cultural considerations.	Findings interwoven with some relevant explanatory literature and moral theory
Limitations of study	Not outlined	Not outlined	Not outlined	Limitations outlined.	Limitations clearly outlined	Limitations of study clearly outlined.	Not outlined	Not outlined	Not outlined
Implications for practice/research/education	Not outlined	Implications for further research outlined	Implications for practice outlined	Significance for practice outlined	Significance for current policy outlined.	Practice and policy implications outlined.	Practice implications outlined	Implications for practice, policy and public debate outlined.	Significance for practice outlined.
Researcher reflexivity	Not demonstrated	Not demonstrated	Not demonstrated	Not demonstrated	Not demonstrated	Not demonstrated	Not demonstrated	Not demonstrated	Not demonstrated
Quality rating	C	C	C	C	B	B	D	B	B

each core concept are outlined in detail. Each core concept also has a section entitled 'synthesis' which is a summary statement clarifying the rationale for, and the essence of, each concept. Finally, the five core concepts are taken together to tentatively propose a conceptual framework which may provide a new insight into pregnant women's decision-making processes with regard to antenatal screening for Down syndrome.

Destination unknown

Anxiety

Pilnick, Fraser, and James (2004, page 90) reported that women engaged with screening in anticipation of abating their anxiety and receiving reassurance about the health and well-being of the fetus. Yet a dichotomy existed with other women pointing to the potential anxiety provoked by screening; anxiety focusing on the stress and worry experienced whilst awaiting screening results, distressing decisions about diagnostic testing, and perceptions that anxiety will continue through pregnancy even in the event of low risk result. As one participant in Reminnick's (2006, page 33) study stated:

'Truth is, I will worry till I get the results, only then I'll be able to sleep quietly ... for some time at least, since I have many other tests waiting ahead ...'.

Indeed (Markens et al., 1999, page 364) noted that women decided to decline screening because of the potential anxiety it may pose for them:

'I have my reasons for not having certain testing ... An unnecessary anxiety in a lot of cases. I think that it can cause anxiety rather than reassure'.

Rapp (2000) has suggested that such anxiety is composed of several layers. Most obviously, anxiety fed upon women's personal and social aspirations embodied in producing a 'normal' healthy baby whilst fearing that 'the baby is ...deformed ...' (Williams et al., 2005, page 1897). The desire to benefit from screening technologies as well as rejecting such technologies in favour of trusting the natural processes of pregnancy, and the struggle to make personal meaning out of screening results further reflected the multi-layered nature of women's anxiety (Markens et al., 1999, page 366):

'... I think that a false positive can cause a lot of stress ... I think you were worrying about the baby and were all uptight and you couldn't enjoy your pregnancy, I think that if you enjoy your pregnancy, the fetus is doing better too'.

Views on abortion

Women tenuously framed their decisions about screening within the context of potential future decisions about abortion. Indeed, it is this tenuous relationship between screening and abortion that led Rapp (2000, page 131) to view women as 'normalising gatekeepers' adjudicating on 'entry into the human community'.

Williams et al. (2005, page 1988) highlighted most women's uncertainty about what they would do:

'... but that's why the test would be so hard for me, because I wouldn't be straightaway, oh, if there's something wrong, I'm not having it'.

Yet some women were very clear that they would not choose to have an abortion, thereby perceiving screening as pointless (Heyman et al., 2006; Markens et al., 1999; Reminnick, 2006). As one participant stated (Markens et al., 1999, page 365):

'... the test might as well not even exist because I'm not going to have an abortion anyway'.

Table 4
Emerging themes and concepts.

Key themes, first iteration	Key themes, final iteration	Core concept, first iteration	Core concept, final iteration	Relevant papers
Stress/distress/worry/anxiety Attitudes towards/decisions about abortion/views on abortion Coping with Down syndrome/having a child with Down syndrome	Anxiety Views on abortion Perceptions of having a child with Down syndrome	Anticipation	Destination unknown	Markens et al., 1999 Lewando-Hundt et al., 2001 Liamputtong et al., 2003 Pilnick et al., 2004 Williams et al., 2005 Chiang et al., 2006 Heyman et al., 2006 Reminnick, 2006 Garcia et al., 2008
Routinization/routine nature of screening Professional knowledge/trust in professionals/expert authority	Routine nature of screening Expert authority	Impoverished choice	To choose or not to choose	Markens et al., 1999 Liamputtong et al., 2003 Pilnick et al., 2004 Williams et al., 2005 Chiang et al., 2006 Heyman et al., 2006 Reminnick, 2006 Garcia et al., 2008
Perceiving risk/risk information Other factors influencing risk/embedded risk/contextual risk Risk and diagnostic testing	Information about risk Contextualising risk Risk and diagnostic testing	Instability of risk	Risk is rarely pure and never simple	Markens et al., 1999 Lewando-Hundt et al., 2001 Liamputtong et al., 2003 Pilnick et al., 2004 Heyman et al., 2006 Reminnick, 2006 Garcia et al., 2008
Perceptions of the fetus/images of the fetus 'Tentative' pregnancy/just in case Women's sense of responsibility/responsible motherhood	Imaging the fetus Just in case Responsible motherhood	Maternal-fetal relatedness	Treading on dreams	Markens et al., 1999 Liamputtong et al., 2003 Williams et al., 2005 Heyman et al., 2006 Reminnick, 2006 Garcia et al., 2008
'Right to know' versus 'right not to know'	'Right to know' versus 'right not to know'	Betwixt and between	Betwixt and between	Markens et al., 1999 Liamputtong et al., 2003 Reminnick, 2006

Interestingly, a few women who had ruled out abortion accepted screening on a 'gamble' that it will generate a low risk result (Heyman et al., 2006).

Garcia et al. (2008, page 759) suggested that screening was only useful for women who would choose to have an abortion:

'If you take a test you make beforehand the decision to terminate pregnancy if you hear that you'll get a child with Down syndrome. I think that you take a test with the purpose to terminate your pregnancy'.

Yet this suggestion was latterly refuted by both Garcia et al. (2008) and Markens et al. (1999) who argued that, although women who accepted screening did not exclude abortion in the event of a confirmed diagnosis, they did not always directly associate screening with the possibility of abortion (Garcia et al., 2008; Markens et al., 1999). For women considering abortion, there was recognition that this was a highly charged moral area and they expressed a preference for early pregnancy screening in order that decisions to choose abortion might remain private from wider society (Williams et al., 2005).

While some women's views on abortion were based on religious beliefs (Lewando-Hundt et al., 2001; Liamputtong et al., 2003), other women's views were based on personal beliefs and values (Williams et al., 2005). Yet, regardless of their origin, such views were inextricably intertwined with women's commitment to being pregnant; perceptions of the fetus and of having a child with Down syndrome (Reminnick, 2006; Williams et al., 2005).

Perceptions of having a child with Down syndrome

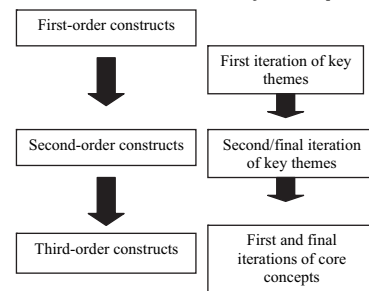
Women's perceptions of having a child with Down syndrome shadowed their decision-making about screening. Such

perceptions – articulated in terms of disability; ability to mother a child with Down syndrome; potential impact the child may have on the family and concerns about social support – were deeply intertwined and not easily reducible.

Ambiguity appeared to surround women's perceptions of a child with Down syndrome as being a 'burden' for themselves, their families and society (Chiang et al., 2006; Garcia et al., 2008; Reminnick, 2006). Indeed, whilst some women pointed to their low acceptance of disability and lack of will to mother a child with Down syndrome (Reminnick, 2006), others believed that (Liamputtong et al., 2003, page 95):

'there is a place in society for 'special' children as well as 'normal' children. If my baby is 'special' then I'll do all I can to look after him/her, and offer him/her the best possible life'.

Table 5
Relationships between terms used in the meta-synthesis process.



Women agreed that a child with Down syndrome might 'help family members to cope with adversity and to appreciate the worth of little and irrelevant things' (Garcia et al., 2008, page 759) yet shared concerns that the special attention required by such a child could predispose to more family worries and costs, together with lack of attention for other children (Garcia et al., 2008). Interestingly, more religious and less educated women perceived immediate or extended family as source of support in case of child with Down syndrome whilst middle-class and more educated women perceived that they themselves might be the main carer with social support considered less extensive and accessible (Reminnick, 2006). Yet women's fears of '... getting old, who will take care of the child? Will he or she become a burden on others?' (Chiang et al., 2006, page 275) appeared universal.

It was not possible to discern from most of the studies if women's perceptions reflected actual prior experiences of children with Down syndrome. But for one participant in Williams et al.'s (2005, page 1988) study:

'... having looked after a lot of children with special needs and having a reasonable understanding of the impact without obviously being in that situation myself, of the implications for the whole family, I personally would not choose to have a child if they were to have Down's syndrome ...'.

Interestingly, only one study referred to the role of screening in providing women with information which might enable them to prepare for having a child with Down syndrome (Garcia et al., 2008). The use of the word 'child' may also have been significant, being defined by the Oxford English Dictionary as, 'very young person, member of family' and thereby appearing to construct the nature of Down syndrome as both lifelong and relational.

Synthesis

The three themes in this section seemed to coalesce into something that was beyond anxiety, views on abortion and perceptions of having a child with Down syndrome. This led to the concept of 'destinations unknown'. Women's decisions about screening were incumbent upon a range of expectations based on current images of future psychological states and ethical dilemmas (Giddens, 1991). Women engaged in the processes of introspection and reflection (Sallach, 2003), processes which involved subtle interaction between individual and perceived external expectations. Therefore, despite the expectations generated being reflective of what women actually thought and felt, they were also coloured by relevant external sources. Thus, for example, women's expectations of having a child with Down syndrome were indicative of wider society's misconceptions about the personality, behaviour and developmental potential of such children (Gilmore, 2006). Given that expectations were influenced in this way, they may have proved incongruent with what women actually experienced. Hence, Thornton and Nardi (1975) consider anticipatory expectations to be incomplete and ambiguous, helping women's decision-making about screening only to the extent that they were accurate in drawing inferences about future states relating to anxiety, abortion and having a child with Down syndrome.

To choose or not to choose

The routine nature of screening

For most women, screening was viewed as routine in nature. One explanation for such a viewpoint was quite simply that 'The technology is available so why not use it' (Markens et al., 1999, page 364). However, this did not fully explain women's views relating to the routinization of screening. Accordingly, (Chiang et al., 2006,

page 274) pointed to women's acceptance of screening as an integral part of routine antenatal care:

'The examining process made you feel that the test is a necessary part of maternity care.'

Markens et al. (1999) suggested that women were more likely to accept screening when it was presented within the context of non-controversial routine antenatal care. A few women accepted screening in order to engage with maternity care services (Heyman et al., 2006). Moreover, women followed prescribed screening pathways order to ensure positive birth outcomes (Markens et al., 1999).

Indeed, women may not have been aware that they were in fact, consenting to an optional test, but rather framing their decisions to accept screening in terms of it being 'a formality' (Pilnick et al., 2004, page 90); and articulating their acceptance of screening as 'there's no reason to say no' (Williams et al., 2005, page 1987) or questioning 'Why would I not want to have this um really un-intrusive test?' (Pilnick et al., 2004, page 90).

Expert authority

A further consideration influencing the routinization of screening appeared to be women's acceptance of its strong endorsement by expert authority (Reminnick, 2006). Such expert authority was built upon trust and knowledge. Brown and Webster (2004) have pointed to the importance of trust in the expert-woman relationship and comment that routinization of screening is not possible when trust fails. The unlimited trust which some women placed in the expert is captured by the comments of one participant in Heyman's et al. (2006, page 2364) study:

'But you just ... basically trust (name of consultant), and whatever is to be done, then just do it.'

In turn, trust was predicated by women's belief in the beneficence of the expert (Heyman et al., 2006). However, such beneficence was challenged by a minority of women who argued that screening was against the true interests of women and their babies, and that the main beneficiaries of the expansion of the 'genetic testing race' were doctors and the biomedical industry (Reminnick, 2006).

The authority of the expert also rested upon his/her ability to interpret the exclusive and complex biomedical knowledge surrounding screening. The consequence of this orientation was an asymmetrical relationship between women's information about screening and expert knowledge (Reminnick, 2006), alluding to what Arrow (1963) has described as the concept of moral hazard. Such asymmetric information may have adversely affected women's decision-making by rendering them (Chiang et al., 2006, page 275):

'...powerless when we encounter medical science'.

Moreover, reliance on expert knowledge tended to negate what Katz Rothman (1994) has described as women's intuitive knowledge processes. As one participant in Williams et al.'s (2005, page 1988) study commented:

'see, I think we're not experts in this baby thing anyway, so it's like whatever they offer, we would just take ...'.

Yet a few women voiced their resistance to values perceived as inherent within expert knowledge: values about the normality/ abnormality of babies and the assessment of risk (Liamputtong et al., 2003).

Synthesis

The most striking effect of women's views about the routine nature of screening and expert authority was the reduction, rather than the

expansion, of choice. The combination of the context in which screening was presented, and the value placed upon expert authority not only routinized screening, but also made choice largely illusory. Women's reaction to what they encountered tended to be one of conformity. The result was deeply problematic because, without a sense of choice, women's decision-making processes became impoverished as the crucial issues that underlay such processes were obscured and bypassed (Suter, 2002). Garcia et al. (2008) highlighted concerns regarding 'standard testing' and the potential to make very-poorly considered decisions which fail to carefully examine women's motivations for screening and the meaning of screening information to them. Thus, women who believed they had no choice may have found themselves unexpectedly on a screening path for which they were completely unprepared.

Risk is rarely pure and never simple

Information about risk

Women perceived risk information as being presented in terms of complex statistics, risk analysis, and arcane, rapidly expanding screening technologies. Women either declined screening because of complex and inconclusive risk information (Liamputtong et al., 2003) or accepted screening because they felt intimidated (Reminnick, 2006, page 37):

'Who do they think I am – an MD or a scientist? I don't know every other word in this flyer; what's the point in reading it? It only gets scarier ...'.

Risk information took for granted that a rational woman's individual attitude to the uncertain possibility of having a baby with Down syndrome was representable in terms of probability. Whilst some women grasped the significance of probabilistic risk figures (Heyman et al., 2006, page 2367):

'So 1 in 174, or whatever it was, I just imagined 174 women, and I could be that one person', others did not (Pilnick et al., 2004). In some instances, more descriptive categories also proved problematic (Pilnick et al., 2004, page 90):

'You know, there's no sort of middle ground. There's either ... high risk or low risk, and um you know I would definitely say I wasn't low risk, but then I didn't fall into their high risk'.

Thus, some women employed trust in the expert as a substitute for engaging with the complex process of interpreting risk (Heyman et al., 2006).

Nonetheless, some women challenged the inconclusive and inaccurate nature of screening results (Markens et al., 1999, page 364):

'... and sometimes the test is inaccurate and the chances of the test being inaccurate are higher than the chances that, something could be wrong, most likely'.

Women's expectations about the accuracy of screening were gleaned largely from the information provided by healthcare professionals (Lewando-Hundt et al., 2001). Other women declined screening on the basis that results did not give sufficient information for future decisions (Garcia et al., 2008).

Contextualising risk

Beyond the debate about probabilistic risk information, women's subjective perceptions of risk also influenced decision-making. Such perceptions were contextual and embedded in a chain of prior, unique life experiences that were 'biographically determining'. On this basis, women experienced risk differently

because they came to screening with diverse life experiences, which potentially led to different perceptions of risk and different decision-making. Thus some women declined screening because they saw themselves as 'fit and healthy' with 'no problems in earlier pregnancies' (Liamputtong et al., 2003, page 94). Other women had experienced difficulties in conceiving and did not wish to risk harming their fetus by being screened (Liamputtong et al., 2003). Lewando-Hundt et al. (2001) reported that women were well aware that Bedouin culturally preferred consanguinity may have predisposed to increased risk, but acceptance of screening was related to family experience of risk rather than consanguinity. Thus women's contextualised perceptions of risk may or may not have conflicted with biomedical interpretations of risk (Markens et al., 1999; Pilnick et al., 2004).

Risk and diagnostic testing

Some women found it difficult to differentiate between the results from screening and diagnostic testing (Pilnick et al., 2004). Indeed, high risk screening results were interpreted by some women as a definitive diagnosis (Lewando-Hundt et al., 2001). Yet for other women the unsatisfactory nature of screening results predisposed to the need for more definitive information from diagnostic testing (Pilnick et al., 2004). However, decisions about diagnostic testing further inflamed women's anxiety and ambivalence: anxiety and ambivalence related to the risk of miscarriage associated with such invasive testing (Markens et al., 1999; Pilnick et al., 2004). Despite such risks, some women perceived that there was no point in having screening if they then did not wish to proceed to diagnostic testing (Heyman et al., 2006; Liamputtong et al., 2003). Indeed, Heyman et al. (2006) noted that, even when given advice to the contrary, women felt that acceptance of screening entailed acceptance of diagnostic testing if indicated. Hence, it may be suggested that women did not view screening decisions as discrete but rather as one stage of a progressive decision-making journey; a journey that once embarked upon was difficult to disembark and may ultimately have led to decisions about abortion and/or having to cope with a child with Down syndrome.

Synthesis

Screening for Down syndrome was designed and continues to be developed from a biomedical perspective of pregnancy as a time of risk, risk avoidance and risk reduction (Pillely Edwards & Murphy Lawless, 2006). From such a perspective, risks were presented as objective, neutral probabilistic facts that were correct, precise and unambiguous. Probabilistic screening risk figures should have acted as external stimuli that were understood by all women to be of the same value. On this basis, women faced with uncertainty should have made rational decisions based on the magnitude of the risk. However, the growing credibility of medical sociology, and social constructionism in particular, has led to this approach being increasingly challenged (O'Byrne & Holmes, 2007).

Probabilistic screening risk figures were not indisputable facts with an independent reality, but were a social product that were constantly constructed, reconstructed and negotiated at multiple levels. The concept of risk was therefore unstable. At one level risk figures were constructed by expert authority who retained the power to define and interpret them. But women did not always translate such figures into similar risk categories. Indeed women were sceptical about the usefulness of risk categories and tended to simplify risks into meaningful contextual statements that were embedded in their own particular lives and experiences. To expert authority, risk retained the character of a population-based number, but to the individual woman, the population base was one, herself, and 'one in a hundred' meant that she could have been the

one (Baillie, Smith, Hewison, & Mason, 2000). Moreover, the inaccuracy of screening further blurred risk categories leaving experts and women with as much uncertainty as before screening. The idea of risk also implied activity, since it transfigured the potentiality of Down syndrome into something which could be acted upon. Thus high risk probabilistic results reshaped women's perspectives for the future of the pregnancy and fetus from a state of waiting to vexed decision-making and potential action with regard to diagnostic testing, abortion and having a baby with Down syndrome. Such complex and unstable constructions of risk were succinctly highlighted by one participant in Markens et al.'s (1999, page 366–367) study:

'The chances of it ... if it came out high or low that they would probably want an amnio and the risk of amnio was higher than the test being accurate and since I didn't have any risk factors and because I am not older, because nobody in my family ever had a problem, I just felt it was not necessary'.

Treading on dreams

Imaging the fetus

Nuchal translucency ultrasound screening focused women on images of the fetus. For some women, nuchal translucency was about making the pregnancy 'real' at a much earlier stage than the moment of quickening (Heyman et al., 2006, page 2365):

'I can't wait. I will feel better when I go to see something on that screen because you still don't feel like you are pregnant', whilst for other women, nuchal translucency (Williams et al., 2005, page 1987):

'... wasn't about screening, it was just something I went along to, to have the scan and see the baby. Screening wasn't important for me until I got the results'.

Such perceptions appeared to point towards women's bypassing of decision-making processes in relation to screening.

Yet, even as nuchal translucency screening personified the fetus, it also put its situation into question. Its power to redefine women's perceptions of the fetus towards 'perfect personhood' by '...actually seeing the outline of the nose and everything ...' (Williams et al., 2005, page 1986) also had the potential to present women with further decision-making dilemmas in the event of adverse screening findings. Such dilemmas related primarily to abortion (Williams et al., 2005, page 1987):

'... but when you see that baby on the screen, you don't care what it's got wrong with it, you just see that it's there and you know it's inside you ... it must be a horrible decision once you've actually seen that this is a baby inside you, to suddenly say 'no', I don't want to carry on with it'.

Indeed, Garcia et al. (2008) reported that, despite differing views about when the fetus may be considered a person, women defined abortion in the first few months of pregnancy as the loss of a real child.

Just in case

Moreover, some women's growing awareness of the fetus was reshaped by their need to maintain a distance from it 'just in case' Down syndrome should be discovered, and they should be confronted with the necessity to choose to end or continue the pregnancy (Williams et al., 2005, page 1987):

'And she just said, keep looking at the screen, just concentrate on the screen, but I just looked away ... because I didn't want to

get attached to this thing (fetus) that was moving, you know (tearful)'.

Thus simultaneously substantiated and distanced, the fetus was suspended in time and status, until fully screened and proved 'normal' (Reminnick, 2006). Yet for other women, the shadow cast by screening over their ongoing adaptation to pregnancy and emotional involvement with the fetus continued for longer periods of time (Heyman et al., 2006, page 2369):

'And it probably took me about another month after having the results to actually finally think to myself, I am pregnant. I want to have this child'.

Responsible motherhood

Women's perceptions of motherhood appeared to be based on responsibility for ensuring the health and well-being of fetus and child, with such perceptions impacting upon their decision-making about screening (Garcia et al., 2008; Reminnick, 2006). Some women accepted screening as they wished to live up to perceived expectations of a caring and enlightened mother as early in the pregnancy as possible (Reminnick, 2006, page 31):

'This is my first baby and I want to do all that I can to ensure its good health'.

Other women argued that they were doing all that is necessary to ensure the birth of a healthy baby by avoiding the risks posed by screening – risks posed by information, false positives and amniocentesis (Liamputtong et al., 2003; Markens et al., 1999). Indeed women who declined the 'maternally responsible course of action' (Markens et al., 1999, page 362) felt labelled by colleagues and peers as irresponsible or not caring enough about their future babies (Reminnick, 2006). Nonetheless, such divergent viewpoints appeared to link women's responsibility for ensuring the health and well-being of the fetus with healthy pregnancy behaviours, with limited recognition that chromosomal abnormalities such as Down syndrome are unaffected by such behaviours (Reminnick, 2006).

Moreover, responsible motherhood meant preventing the perceived suffering and misery that a child's disability might entail (Reminnick, 2006, page 34):

'... I will never stop blaming myself for not doing the tests, all that are available at the moment. It is just so hard to raise a child with disability; I am not prepared for that'.

There was also recognition that responsibility for raising a child with a disability would fall to the mother, contrasting sharply with notions of responsible fatherhood (Reminnick, 2006). Interestingly, both women who accepted and declined screening agreed that a child with Down syndrome would be an impediment for completely realising their values of motherhood (Garcia et al., 2008).

Synthesis

The concept of 'treading on dreams' related to how continuing technological advances in screening in conjunction with expectations of responsible motherhood impacted upon the quest for a 'perfect' baby (Reminnick, 2006). Such a quest was integral to women's decision-making processes about screening. Technological advances in screening served to hasten women's sense of relatedness to the fetus as a separate entity or 'perfect person' from its earliest stage, thereby bypassing women's embodied experiences (Oakley, 1984). Yet, whilst screening invoked the 'perfect personness' of the fetus it also contributed to the ambiguity and uncertainty which women experienced when trying to make decisions following adverse screening findings. Consequently, for

some women the status of the fetus became provisional; precluding maternal-fetal relatedness and turning the screening experience into what Katz Rothman (1994) cogently referred to as a ‘tentative pregnancy’. Women’s expectations of themselves, as ultimately accepting responsibility for the production of a ‘perfect baby’, not only encouraged compliance with screening but also perceptions of the fetus as an expendable consumer object, subject to quality control on the basis of its future potential (Lippman, 1994; Rapp, 2000). Interestingly, women did not describe their quest for the ‘perfect baby’ as a series of developmental stages, as is common in some theoretical and empirical literature (Cranley, 1981; Rubin, 1984) but rather as something that fluctuated throughout the screening experience from the baby on the screen to the child they anticipated being responsible for.

Betwixt and between

The sense of liminality associated with screening was etched into women’s perceptions about their ‘right to know’ versus their ‘right not to know’. In other words, women existed betwixt and between (Turner, 1969) wishing to know and not wishing to know. Women’s perceptions rested upon the ‘risk’ of not having information as opposed to the ‘risk’ posed by information (Markens et al., 1999). The right to know was associated with women’s aspirations to have sufficient information in order to attain control over their future motherhood by ruling out Down syndrome (Reminnick, 2006). On the other hand, the right not to know was associated with the risk that information obtained from screening results could cause emotional distress and lead to further difficult decisions (Liamputtong et al., 2003; Markens et al., 1999). The interplay of women’s ‘right to know’ versus ‘right not to know’ was succinctly highlighted by one participant in Liamputtong et al.’s (2003, page 99) study:

‘I would like to have had the reassurance that chances of Down Syndrome were low – but decided against the risk of finding out that things were not ok’.

Taking the five core concepts together, the line of argument emergent from this synthesis is as follows: women’s decision-

making processes regarding antenatal screening for Down syndrome were complex in that they take into consideration a range of expectations based on current images of future states (destination unknown); perceptions of maternal-fetal relatedness (treading on dreams) and interpretation of risk (risk is rarely pure and never simple). Such decisions were also impacted upon by the way in which screening was offered (to choose or not to choose). In making such decisions, women often existed in a state of liminality (betwixt and between).

Moreover, it is important to emphasise that there was no evidence from the studies synthesized that women’s decision-making processes were either discrete or sequential, but rather interwoven and over-lapping. So, for example, women trying to grapple with the risks associated with diagnostic testing were also engaged in anticipating how they might cope with a child with Down syndrome. Some women adamantly declined screening and felt no difficulty in asserting choice if they rejected abortion. Similarly, women used nuchal translucency scanning to ‘see’ their baby and obtain reassurance without perhaps considering the possibility of a high risk result. So rather than locating women on a particular decision-making trajectory, a framework is proposed which recognises the intersecting and juxtaposing nature of women’s decision-making with regard to antenatal screening for Down syndrome. As already indicated, the purpose of a meta-synthesis is to achieve a level of conceptual development beyond that attained in the individual existing studies: in effect to move from second-order to third-order constructs. It is therefore considered that the framework proposed in Fig. 1, which embraces all the studies included in the meta-synthesis, represents this further level of conceptual development.

Discussion

There is a growing interest in the synthesis of qualitative studies. Ours is the first such meta-synthesis of the decision-making processes of pregnant women with regard to antenatal screening for Down syndrome, either of UK studies or studies conducted elsewhere. Building on critical re-interpretations of selected studies, we developed third-order constructs, which

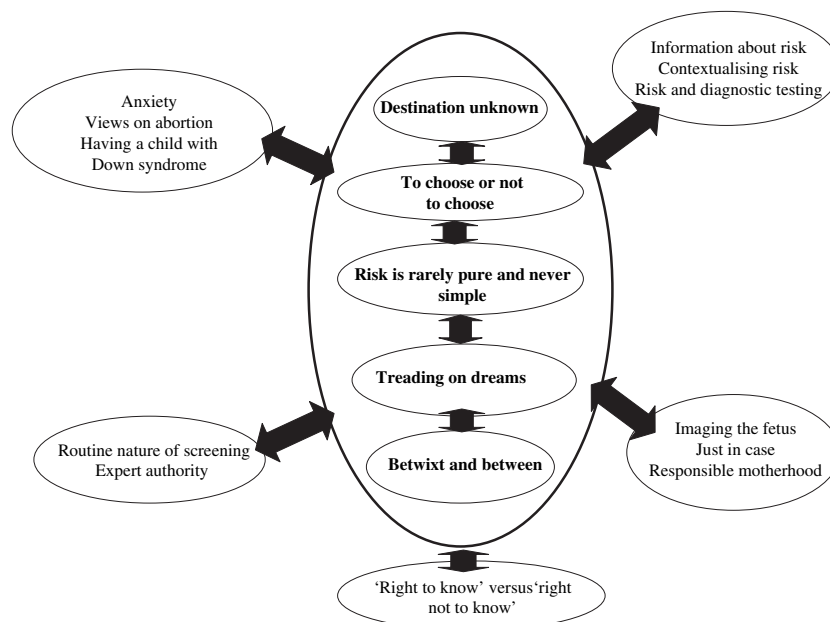


Fig. 1. Pregnant women’s decision-making processes with regard to antenatal screening for Down syndrome.

whilst consistent with the findings of individual studies, also extended beyond them. Consequently, more comprehensive understandings have been generated; understandings which point in some way towards the underlying reality of the complex processes of decision-making that were common to women from different countries and healthcare systems.

The development of third-order constructs into a conceptual framework constitutes a new and insightful level of conceptual development within the context of women's decision-making processes about screening. Such a framework has some limited congruence with existing theoretical frameworks. It points, for example; towards the inclusion of the first three stages of Mercer's (1995) theory of 'maternal role attainment' in women's decision-making but does not support viewpoint that such stages are sequential and progressive. The stage of personal identity is somewhat at odds with the uncertainty which women appear to experience: uncertainty expressed in terms of the risks and benefits of screening; vacillation between screening choices; and questioning of personal values and beliefs. The sense of ambiguity associated with risk information highlighted within our proposed framework points towards the inappropriateness of the Theory of Planned Behaviour (Ajzen & Fishbein, 1980) for examining women's decisions. Indeed Ajzen and Fishbein's (1980) theory tends to focus more on the outcomes of the decision, rather than the processes. Our proposed framework suggests that whilst women may anticipate outcomes, it is not possible to actually know all the potential outcomes.

Accordingly, we now propose to test the cogency and plausibility of the conceptual framework presented in this meta-synthesis by undertaking further ethnographic and choice modelling research. If such research verifies that the framework is indeed reflective of women's decision-making, midwives and other healthcare professionals may need to reconsider the support they provide in order to empower women in making choices about screening.

Nevertheless, this meta-synthesis has raised a number of issues which require further discussion and consideration. This meta-synthesis assumes that the findings of individual selected studies are not specific to one particular sample of pregnant women located within a particular context at a particular point in time. Britten et al. (2002) suggest that such an assumption is unlikely to be supported by all qualitative researchers. Indeed some might argue that the inevitable loss of 'some of the fine detail' and the lack of contextual detail may fail to do justice to individual studies. But, as Britten et al. (2002) suggest, the important contribution of qualitative studies will not be realized if individual studies are allowed to accumulate and are not synthesized.

As previously acknowledged, the studies selected for this meta-synthesis present qualitative findings at different times during pregnancy in relation to women's decision-making processes; report on differing screening and diagnostic tests; and were or were not reflective of screening as routine practice. This raises the question of whether the different moments in time, the characteristics of the tests analysed and the context in which screening was presented could have impacted upon the findings of the selected studies. However, it is not possible to draw such comparisons from this meta-synthesis and such issues therefore require further consideration.

Conclusion

The meta-synthesis presented in this paper has articulated a conceptual framework of pregnant women's decision-making processes with regard to antenatal screening for Down syndrome. This framework reflects a synthesis of nine selected

studies with the core concepts of destination unknown; to choose or not to choose; risk is rarely pure and never simple; treading on dreams and betwixt and between emerging as integral to women's decision-making processes. These core concepts have not previously been identified and further research has been proposed to verify such concepts. The framework points to the complexity of women's decision-making processes. It therefore seems that women do indeed act as 'moral pioneers' (Rapp, 2000, page 3) with respect to decision-making about antenatal screening for Down syndrome.

References

- Ajzen, I., & Fishbein, M. (1980). *Understanding attitudes and predicting social behaviour*. Englewood Cliffs, New Jersey: Prentice Hall.
- Arrow, K. J. (1963). Uncertainty and the welfare economics of medical care. *American Economic Review*, 53(5), 941–973.
- Aspers, P. (2004). *Interpretative phenomenology: An approach for qualitative research. Papers in social research methods qualitative series number 9*. London: London School of Economics and Political Science.
- Baillie, C., Smith, J., Hewison, J., & Mason, G. (2000). Ultrasound screening for fetal abnormality: women's reactions to false positive results. *British Journal of Health Psychology*, 5(4), 377–394.
- Barroso, J., Gollup, C. J., Sandelowski, M., Meynell, J., Pearce, P. F., & Collins, L. J. (2003). The challenges of searching for and retrieving qualitative studies. *Western Journal of Nursing Research*, 25(2), 153–178.
- Bondas, T., & Hall, E. O. C. (2007). Challenges in approaching meta-synthesis research. *Qualitative Health Research*, 17(1), 113–121.
- Britten, N., Campbell, R., Pope, C., Donovan, J., & Morgan, M. (2002). Using meta ethnography to synthesise qualitative research: a worked example. *Journal of Health Services Research and Policy*, 17(4), 209–215.
- Brown, N., & Webster, A. (2004). *New Medical Technologies and Society: Reordering life*. Cambridge: Polity Press Limited.
- Buchanan, A., Brock, D. W., Daniels, N., & Wilker, D. (2000). *From chance to choice: Genetics and justice*. Cambridge: Cambridge University Press.
- Campbell, R., Pound, P., Pope, C., Britten, N., Pill, R., Morgan, M., et al. (2003). Evaluating meta-ethnography: a synthesis of lay experiences of diabetes and diabetes care. *Social Science & Medicine*, 56(4), 671–684.
- Chiang, H.-H., Chao, Y.-M., & Yuh, Y.-S. (2006). Informed choice of pregnant women in prenatal screening tests for Down's syndrome. *Journal of Medical Ethics*, 32(5), 273–277.
- Cranley, M. S. (1981). Roots of attachment: the relationship of parents with their unborn. *Birth Defects Original Article Series*, xvii(6), 59–83.
- Denzin, N. (1989). *Interpretative interactionism*. Newbury Park, California: Sage.
- Estabrooks, C. A., Field, P. A., & Morse, J. M. (1994). Aggregating qualitative findings: an approach to theory development. *Qualitative Health Research*, 4(4), 503–511.
- Fingeld, D. (2003). Meta-synthesis: the state of the art – so far. *Qualitative Health Research*, 13(7), 393–904.
- Garcia, J., Bricker, L., Henderson, J., Martin, M.-A., Mugford, M., Nielson, J., et al. (2002). Women's views of pregnancy ultrasound: a systematic review. *Birth*, 29(4), 225–250.
- Garcia, E., Timmermans, D. R. M., & van Leeuwen, E. (2008). The impact of ethical beliefs on decisions about prenatal screening tests: searching for justification. *Social Science & Medicine*, 66(3), 753–764.
- Getz, L., & Kirkengen, A. L. (2003). Ultrasound screening in pregnancy: advancing technology, soft markers for chromosomal aberrations, and unacknowledged ethical dilemmas. *Social Science & Medicine*, 56(10), 2045–2057.
- Giddens, A. (1991). *Modernity and self-identity: Self and society in the late modern age*. Cambridge: Polity Press.
- Gilmore, L. (2006). Perceptions of Down syndrome in the Australian community. *Journal on Developmental Disabilities*, 12(1)(Suppl. 2), 59–70.
- Guba, E., & Lincoln, Y. (1994). Competing paradigms in qualitative research. In N. Denzin, & Y. Lincoln (Eds.), *Handbook of qualitative research* (1st ed.). (pp. 105–117) Thousand Oaks, California: Sage.
- Heyman, B., Hundt, G., Sandall, J., Spencer, K., Williams, C., Grellier, R., et al. (2006). On being at higher risk: a qualitative study of prenatal screening for chromosomal anomalies. *Social Science & Medicine*, 62(10), 2360–2372.
- Jenson, L. A., & Allen, M. N. (1994). A synthesis of qualitative research on wellness-illness. *Qualitative Health Research*, 4(4), 349–369.
- Jenson, L. A., & Allen, M. N. (1996). Meta-synthesis of qualitative findings. *Qualitative Health Research*, 6(4), 553–560.
- Katz Rothman, B. (1994). *The tentative pregnancy: Aminocentesis and the sexual politics of motherhood* (2nd ed.). London: Pandora.
- Lewando-Hundt, G., Shoham-Vardi, I., Beckerleg, S., Belmaker, I., Kassem, F., & Abu Jaafar, A. (2001). Knowledge, action and resistance: the selective use of prenatal screening among Bedouin women of the Negev, Israel. *Social Science & Medicine*, 52(4), 561–569.
- Liamputtong, P., Halliday, J. L., Warren, R., Watson, L. F., & Bell, R. J. (2003). Why do women decline prenatal screening and diagnosis? Australian women's perspective. *Women and Health*, 37(2), 89–108.

- Lippman, A. (1994). The genetic construction of testing. In K. Rothenberg, & E. Thomson (Eds.), *Women and prenatal testing* (pp. 9–34). Columbus: Ohio State University Press.
- Markens, S., Browner, C. H., & Press, N. (1999). 'Because of the risks': how US pregnant women account for refusing prenatal screening. *Social Science & Medicine*, 49(3), 359–369.
- Mercer, R. T. (1995). *Becoming a mother: Research on maternal identity from Rubin to the present*. New York: Springer.
- Nelson, A. M. (2002). A meta-synthesis: mothering other-than-normal children. *Qualitative Health Research*, 12(4), 515–530.
- Noblit, G., & Hare, R. (1988). *Meta-ethnography: Synthesising qualitative studies*. Newbury Park: Sage.
- Oakley, A. (1984). *The captured womb: A history of the medical care of pregnant women*. Oxford: Basil Blackwell.
- O'Byrne, P., & Holmes, D. (2007). The micro-fascism of Plato's good citizen: producing (dis)order through the construction of risk. *Nursing Philosophy*, 8, 93–102.
- Paterson, B. L., Thorne, S., & Dewis, M. (1998). Adapting to and managing diabetes. *Image: Journal of Nursing Scholarship*, 30(1), 57–62.
- Paterson, B. L., Thorne, S. E., Canam, C., & Jillings, C. (2001). *Meta-study of qualitative health research: A practical guide to meta-analysis and meta-synthesis*. Thousand Oaks, California: Sage.
- Pilley Edwards, N., & Murphy Lawless, J. (2006). The instability of risk: women's perspectives on risk and safety in birth. In A. Symon (Ed.), *Risk and choice in maternity care* (pp. 35–49). Edinburgh: Churchill Livingstone.
- Pilnick, A. M., Fraser, D. M., & James, D. K. (2004). Presenting and discussing nuchal translucency screening for fetal abnormality in the UK. *Midwifery*, 20(1), 82–93.
- Rapp, R. (2000). *Testing women, testing the fetus*. London: Routledge.
- Reminnick, L. (2006). The quest for the perfect baby: why do Israeli women seek prenatal genetic testing? *Sociology*, 28(1), 21–53.
- Rubin, R. (1984). *Maternal identity and maternal experience*. New York: Springer.
- Sallach, D. (October 2003). Interpretative agents. In *Proceedings of the agent: Challenges in social simulation conference*. University of Chicago.
- Sandelowski, M., & Barroso, J. (2003a). Focus on research methods: towards a meta-synthesis of qualitative findings on motherhood in HIV positive women. *Research in Nursing and Health*, 26(2), 153–170.
- Sandelowski, M., & Barroso, J. (2003b). Classifying the findings in qualitative studies. *Qualitative Health Research*, 13(7), 905–923.
- Sandelowski, M., & Barroso, J. (2005). The travesty of choosing after positive prenatal diagnosis. *Journal of Gynaecological and Neonatal Nursing*, 37, 307–318.
- Sandelowski, M., Docherty, S., & Emden, C. (1997). Qualitative meta-synthesis: issues and techniques. *Research in Nursing and Health*, 20, 365–371.
- Schrieber, R., Crooks, D., & Stern, P. N. (1997). Qualitative meta-analysis. In J. M. Morse (Ed.), *Completing a qualitative project: Details and dialogue* (pp. 311–326). Thousand Oaks, California: Sage.
- Schutz, A. (1962) *Collected papers, Vol. I*. The Hague: Martinus Nijhoff.
- Scully, J. L., Banks, S., & Shakespeare, T. W. (2006). Chance, choice and control: lay debate on prenatal social sex selection. *Social Science & Medicine*, 63(1), 21–31.
- Sher, C., Romano-Zelekha, O., Green, M., & Shohat, T. (2003). Factors affecting performance of prenatal genetic testing by Israeli Jewish women. *American Journal of Medical Genetics*, 120A, 418–422.
- Suter, A. M. (2002). The routinization of prenatal testing. *American Journal of Law and Medicine*, 28(2 & 3), 233–270.
- Thornton, R., & Nardi, P. M. (1975). The dynamics of role acquisition. *American Journal of Sociology*, 80(4), 870–885.
- Turner, V. (1969). *The ritual process: Structure and anti-structure*. Chicago: Aldine Publishing Company.
- Vassy, C. (2006). From a genetic innovation to mass health programmes: the diffusion of Down's syndrome prenatal screening and diagnostic techniques in France. *Social Science & Medicine*, 63(8), 2041–2051.
- Walsh, D., & Downe, S. (2005). Meta-synthesis method for qualitative research: a literature review. *Journal of Advanced Nursing*, 50(2), 204–211.
- Walsh, D., & Downe, S. (2006). Appraising the quality of qualitative research. *Midwifery*, 22(2), 108–119.
- Ward, P., & Muir Gray, J. A. (2002). *Antenatal screening service for Down syndrome in England: 2001. A report to the National Screening Committee*. London: National Screening Committee.
- Williams, C., Sandall, J., Lewando-Hundt, G., Heyman, B., Spencer, K., & Grellier, R. (2005). Women as moral pioneers? Experiences of first trimester antenatal screening. *Social Science & Medicine*, 61(9), 1983–1992.
- Wohlgemuth, N. R. (2006). *Prenatal testing decisions: Women's needs and well-being*. Saskatoon, Canada: University of Saskatchewan.
- Zimmer, L. (2006). Qualitative meta-synthesis: a question of dialoguing with texts. *Journal of Advanced Nursing*, 53(3), 311–318.