

**The influence of service organisation and delivery on  
participation in prenatal screening for Down's  
syndrome: A multiple case study.**

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The candidate confirms that the work submitted is his own and that appropriate credit has been given where reference has been made to the work of others.

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## **Abstract**

**INTRODUCTION:** The way in which NHS screening programmes are organised and delivered may influence public engagement. This is of particular interest in Down's syndrome screening where variations in uptake have not been explained by individual social characteristics. This has led to suggestion that the influence of service organisation and delivery may help to explain the variation.

**AIM:** To explore the influence of service organisation and delivery on women and partners' participation in screening in two different health districts in England with contrasting uptake rates.

**METHOD:** A multiple case study design involving document review of screening guidelines and qualitative online interviews was adopted. Purposive sampling was employed to select settings and invite in each group (n=18 – 24) community midwives, women and partners. Participants responded online to vignettes with open-ended questions and prompts, providing a range of responses. Data were analysed using content analysis.

**FINDINGS:** Participation was influenced by constraints such as the offer of screening as a routine test, influence of interpreters and ambience of the environment resulting in the routinisation of screening. Additionally, differing perceptions and the policy of nondirectiveness created tensions in the prenatal environment, within and between midwives, women and partners. The constraints, tensions and nuances in the operationalisation of the screening guidelines affected midwife - woman interaction and how screening was presented, with some midwives colluding with women to engage passively in screening. The conceptual model developed from the findings revealed a factor unaccounted for in previous research that the tensions, different relationships, decision-making models and variation in uptake rates in screening were associated with service organisational and delivery constraints.

**CONCLUSION:** The classic situation of women capable of autonomous and informed choice and midwives capable of informing nondirectively may not exist in the prenatal context. A shared decision-making process model to mitigate the constraints and tensions is proposed.

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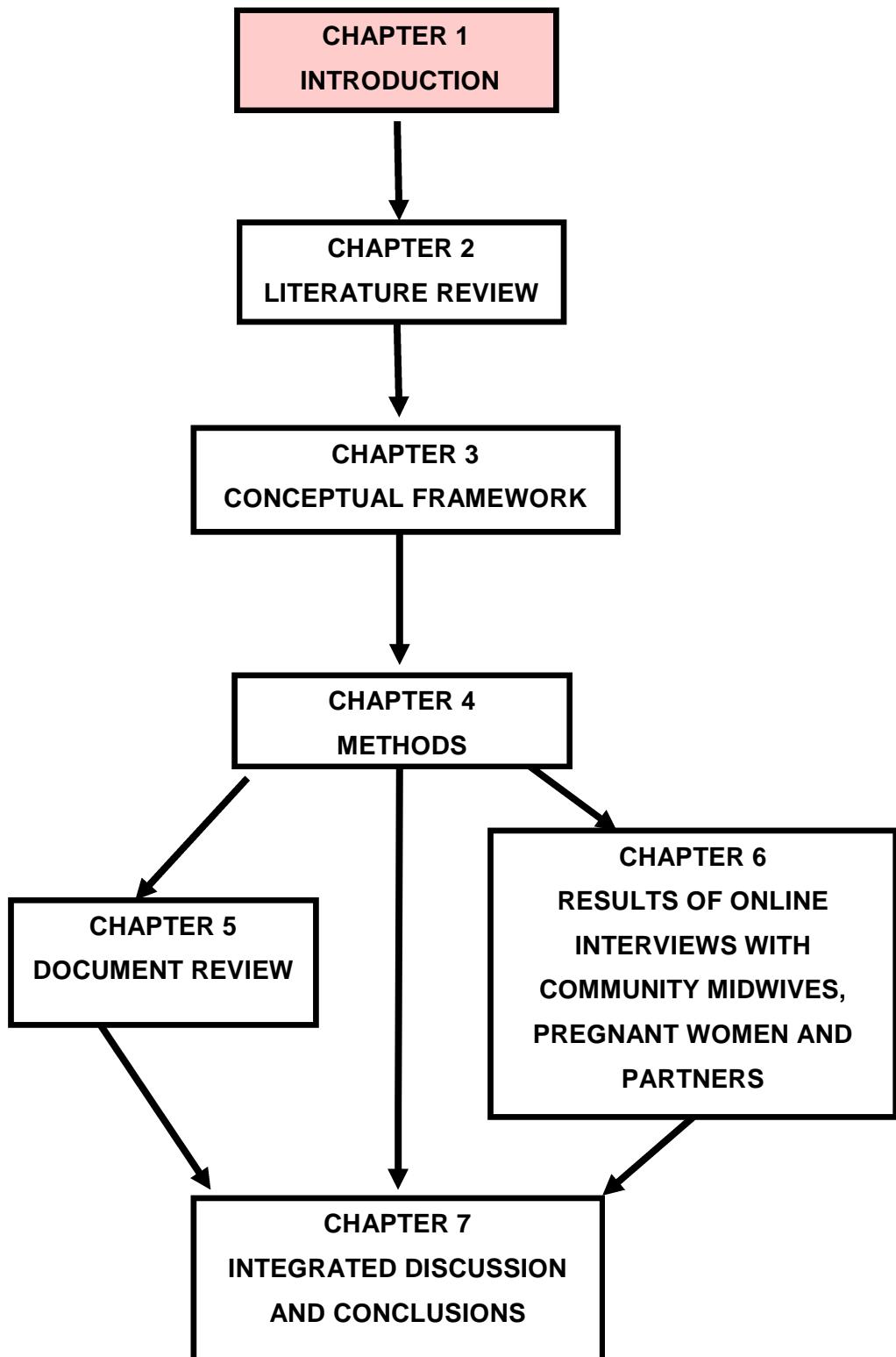
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## List of Abbreviations

AFP.....	Alpha Feto-Proteins
CRL.....	Crown Rump Length
hCG.....	Human Chorionic Gonadotrophin
NIPD.....	Non-Invasive Prenatal Diagnosis
NT.....	Nuchal Translucency
PAPP-A.....	Pregnancy Associated Placental Protein A
uE3.....	Unconjugated Estriol
TOP.....	Termination of Pregnancy
NI.....	Northern Ireland
FASP.....	Foetal Anomaly Screening Programme
NICE.....	National Institute for Health and Clinical Evidence
ISS.....	Information System Services
LCO.....	Local Screening Coordinator
PIS.....	Participant Information Sheet
SDMP.....	Shared Decision-Making Process
SDM.....	Shared Decision-Making
NSC.....	National Screening Committee
GP.....	General Practitioner
SoM.....	Supervisor of Midwives
VCT.....	Voluntary Counselling And Testing
HIV.....	Human Immunodeficiency Virus
TRA/TPB.....	Theory of Reasoned Action/ Theory of Planned Behaviour
HBM.....	Health Belief Model



## CHAPTER 1: INTRODUCTION

### 1.1 Background

*“More often one needs to ask, ‘What goes on here?’ rather than, ‘What is wrong’...This is justified by the assumption that one is interested not in the power of medical technology to achieve results, but in whether what is now known to be ‘good’ medical care has been applied”* (Donabedian, 2005, p.694,721).

These were the views expressed by Avedis Donabedian, author of evaluating the quality of medical care (Donabedian, 2005). The study was a review of reviews on contemporary methods of assessing the quality of medical care. The aim of the study was to evaluate the medical care process at the level of physician-patient interaction. These views are especially true in the area of research in prenatal Down’s syndrome screening where the quality of the programme may be difficult to assess. Outcomes such as uptake rates cannot be used as indicators of quality, because participation in screening is based on autonomous and informed choice to avoid the notion of eugenics.

Several studies have been conducted to understand women and their partners’ participation in prenatal Down’s syndrome screening and to account for the variation in uptake rates seen within and across countries with similar demography and screening services (Boyd et al., 2008). Most of the studies have examined demographic and psychosocial factors such as women’s attitudes, religion, ethnicity and socioeconomic status. The findings have largely been inconclusive. This has led to an assumption that service organisation and delivery may be responsible for the variations in uptake rates. However, no study has comprehensively examined the broader contexts in which service provider-user interaction and decision-making in first trimester combined screening programme are embedded. It is within these contexts that women and partners make or initiate decisions about screening for Down’s syndrome and consent is obtained. Further, women and their partners’ participation in screening cannot be separated from the context in which they occur.

Therefore, we know little about how pregnant women and partners’ participation in screening is affected by service organisation and delivery in the prenatal context. It is important to note that existing studies have mostly reported on second trimester screening and nuchal translucency (NT) first trimester screening. First trimester combined screening is

in the second year of implementation in some regions in England and the few published works that do exist suggests similar gaps, although have been limited in scope. In addition, this thesis will provide insight into the experiences of midwives, women and their partners in first trimester combined screening programme and address the gaps in the literature.

## **1.2 Why Down's syndrome screening programme?**

Many policies guiding healthcare programmes in the UK encourage health professionals to increase uptake rates. For example, HIV and cervical screening programmes are target driven, with healthcare professionals getting paid when they attain high levels of uptake in their practice (Jepson et al., 2005). Nevertheless, prenatal Down's syndrome screening programmes are not target driven and the aim is to enable women make an informed choice about whether or not they want to be screened. In addition, screening enables healthcare providers to offer appropriate information, support or treatment and plan for children with the condition (Gidiri et al., 2007; Skirton and Barr, 2007; Boyd et al., 2008).

The ethical principle of informed choice consisting of nondirectiveness and autonomy guides the offer of prenatal Down's syndrome screening. This makes the programme a useful lens through which to study the influence of service organisation and delivery on service users' participation in healthcare programmes. In addition, the 'booking appointment' for Down's syndrome screening was chosen to explore the influence of service organisation and delivery, as considerable information about screening is offered by midwives to women and their partners at booking. Decisions about screening are also made or initiated at booking.

## **1.3 Significance of the study**

The present goal of the screening programme is to provide nondirectional counselling to enable women and partners make informed choices (Skirton and Barr, 2007; McNeill and Alderdice, 2009; Vanstone et al., 2012). This thesis holds the potential to identify multiple avenues for facilitating informed decision-making processes across multiple service contexts. It may also highlight that, although women and partners are making more or less autonomous informed choices, their participation in screening may or may not be influenced by service organisation and delivery.

This study may reveal hidden controlling influences that need to be addressed or minimised in the prenatal context. Without a clear understanding of how service organisation and

delivery influence women and partners' participation, recommendations cannot be made for improvement in screening practices to support the informed decision-making process of women and their partners. This knowledge may have implications for research and for the development of services that are both effective and efficient in supporting decision-making processes. It may ultimately help to shape policy development and influence practice patterns. The findings may also provide a source for service providers to check their own interpretations of users' experiences.

#### **1.4 Participation in healthcare treatments or programmes**

The term 'participation' is defined as 'being involved and to take part'. However, from a health professional's perspective, it involves service users obtaining tailored information to enable them to act on it. Thus, service users experience participation as something they receive and accept, whilst the healthcare professional perceives it as something they give to users (Eldh et al., 2006; Sahlsten et al., 2008). Sahlsten et al. (2008) defined the concept of participation as consisting of an established relationship which is made up of respect, trust, mutual connected relationship with some affective elements. In addition, there is giving up of some control or power by health professionals, thus empowering and supporting service users.

Another element is the sharing of information and knowledge, through information and knowledge exchange between health professionals and patients based on their beliefs and values. Lastly, the willingness and active engagement of both health professionals and users throughout the process is required. Heggland and Hausken (2013) and Heggland et al. (2013) reported that participation in healthcare programmes involved the interaction of healthcare professionals and patients. It is pertinent to state that this thesis will focus on the decision making processes involved in women and partners' participation in screening and not the outcomes of the decision. It is not important whether women accepted or declined screening, but the focus is on the service contextual factors that influenced their decision-making process.

#### **1.5 Participation in prenatal Down's syndrome screening**

Participation in Down's syndrome screening is based on the ethical principles of autonomy, beneficence, justice and informed consent. Respect for autonomy is a norm respecting the

decision-making capacities of individual women and it is a professional obligation. It directs the healthcare provider to identify and implement clinical management that is expected in the judgement of the pregnant woman, to result in greater benefits that are important to her over risks she wants to avoid. It maintains her right to refuse or choose prescribed tests. Autonomous choice is a right, not a duty of the pregnant woman (Beauchamp and Childress, 2001; Sharma et al., 2007). In the context of Down's syndrome screening this means competent pregnant women and partners are expected to make decisions based on their beliefs, values, and personal circumstances without influence to determine their participation (Beauchamp and Childress, 2001; Sharma et al., 2007; Ahmed et al., 2012c).

### **1.5.1 Beneficence**

Beneficence is a group of norms for providing benefits and balancing benefits, risks and costs fairly. It guides the healthcare provider to identify and implement clinical management that is expected on the basis of the best available evidence to result in a greater balance of good over harm for the pregnant woman. In order to offer screening tests to patients, the first ethical question to ask is whether it is medically reasonable (reliably expected to improve outcome). If it is not, then there is no ethical basis to offer it. In prenatal screening for Down's syndrome, it has been proven, from international studies, that it is medically reasonable, as it reduces the incidence of invasive tests and unnecessary pregnancy loss (Wald et al., 2003; Nicolaides, 2004; Malone et al., 2005; Sharma et al., 2007).

The reasonable person standard requires disclosure of it to pregnant women in the informed consent process for the management of their pregnancies. First trimester screening has obvious benefits to second trimester screening: early disclosure of results, gestational age, dating and detection of multiple gestations, chorionicity and malformations. The risks are poor techniques and lack of quality control. In addition, the offering of invasive diagnostic procedure to women is ethically justifiable, as some women have made the informed decision not to accept the birth of a child with any detectable chromosomal abnormality (Beauchamp and Childress, 2001; Sharma et al., 2007).

### **1.5.2 Justice**

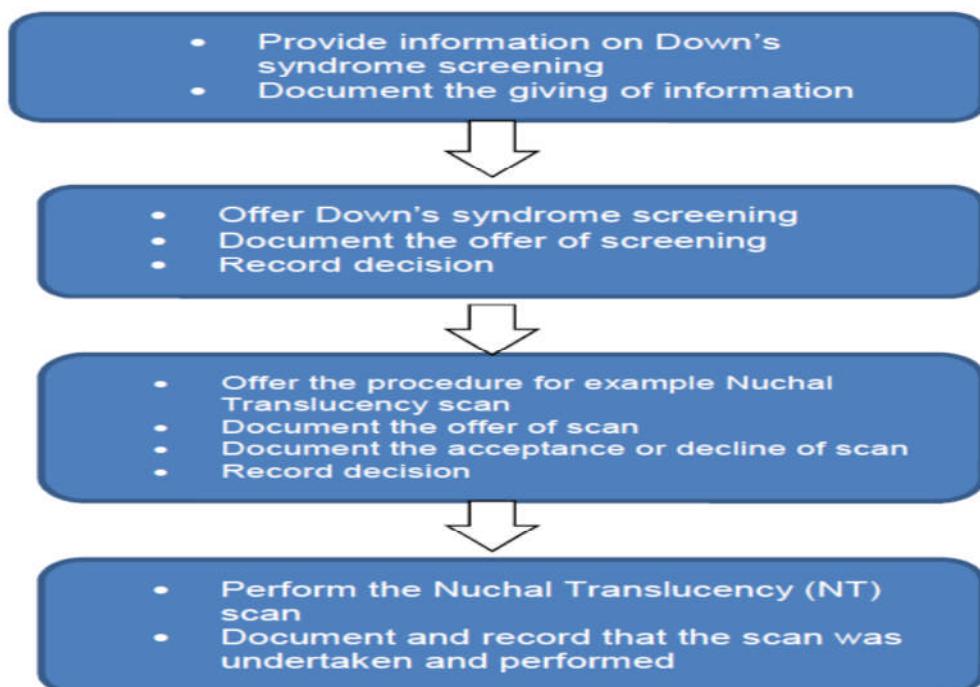
Justice is a group of norms for distributing benefits, risks and costs fairly. It directs healthcare providers to prevent exploitation, in which only a few have access to significant benefits and many experience only burden. The obligation to avoid exploitation shapes the

process of the informed consent and provision of information. The principle of justice requires nondirectional counselling about alternatives to screening and invasive testing and women should be allowed to select the best choice for their needs. Withholding this choice exploits women. It is a population-based ethical principle, in contrast to the individual woman focus of respect for autonomy and beneficence (Beauchamp and Childress, 2001).

### 1.5.3 Informed choice or consent

In autonomous informed consent the role of the midwife is to give information and then allow the pregnant woman to decide whether to accept or decline the offer of screening (McKenzie, 2009). The concept of informed choice is based on the doctrine of informed consent. Informed consent is the clinical tool that implements the ethical principles of autonomy and beneficence in the decision-making process with women about the clinical management of the pregnancy (Hodge, 2004; Sharma et al., 2007; Chervenak and McCullough, 2010). There is a consensus that undergoing screening should be the result of informed choice (See figure 1). This is particularly the case for prenatal Down's syndrome screening where one of the possible outcomes of the screening process is termination of pregnancy (TOP) (Dormandy et al., 2006).

**Figure 1: Consent pathway for Down's syndrome screening** (NHS FASP, 2011)



Informed choice, or consent, is recognised worldwide and accepted as a crucial part of ethical biomedicine (World Health Organization, 2006; Ahmed et al., 2012b). In the UK, the national screening policy for Down's syndrome states that the primary purpose is to facilitate informed choice (NICE, 2008; NHS FASP, 2011). In accordance with the policy on Down's syndrome screening (NICE, 2008), health professionals are required to offer screening in a nondirective way to enable pregnant women and their partners to make an informed choice about screening. Informed choice is similar to informed consent, as in both cases the pregnant woman has the final decision-making right.

Nonetheless, there are differences. First, informed consent is a form of legal requirement for which certain criteria need to be met before the woman's consent is valid, such as her competence, understanding of information and nondirective counselling by healthcare professionals (Faden and Beauchamp, 1986; Wirtz et al., 2006; Ahmed et al., 2012c). Second, in informed choice, the pregnant woman is presented with all screening options by the healthcare professional and then the woman decides which option to choose. Informed consent does not require that a woman selects one screening method out of a number of other methods. Usually, the pregnant woman is expected to consent to the standard screening method for her gestation (first or second trimester screening), whilst she is informed about others such as the foetal anomaly ultrasound scan (Wirtz et al., 2006).

Informed choice involves not only the provision of high quality, complete, up-to-date information about prenatal Down's syndrome screening, but is also about making sure that the decision-making of pregnant women and partners is autonomous and free from influence or coercion. The role of community midwives in Down's syndrome screening is to promote informed choice by providing relevant information about the advantages and disadvantages of screening or implications of participating in screening to pregnant women (McNeill and Alderdice, 2009; Ahmed et al., 2012c). This role is consistent with both the legal and moral requirements of informed consent (Wirtz et al., 2006). Therefore, ensuring that pregnant women and their partners' decision-making are free from external influences or coercion is central to enabling informed choice.

When providing information on Down's syndrome screening at booking, the midwife needs to recognise pregnancy as a state of health and respect the choices of the pregnant woman even when it is contrary to clinical evidence (Wirtz et al., 2006). Additionally, the information-giving should be based on updated evidence and the standard guidelines on

screening, whilst taking into consideration local clinical practice. Deviation from any of these concerns could result in a departure from nondirectiveness.

Goldberg (2009) stated that the reasons for adopting informed consent were, first, to demonstrate proof that the rights of women and partners have been incorporated into the process of offering screening. Secondly, it provided legal documentation against litigation and thirdly, it distances screening from eugenics and paternalistic decision-making involving health professionals. Informed consent or choice, therefore, placed the decision-making strictly on the woman.

The association between informed choice and patient autonomy appears based on the bioethical version of autonomous decision-making. As such, the standard bioethical concept of autonomous decision-making is based on rational choice models of health behaviour which view individuals as autonomous, rational and free from external influences. It does not take into account the diverse contextual factors where women make choices in clinical settings. It certainly does not acknowledge that any attempt to facilitate informed choice necessarily engages midwives and pregnant women in a dynamic relationship (Heritage and Maynard, 2006; McKenzie, 2010).

Interaction between midwives and pregnant women operates at the interface of workplace and everyday systems in the antenatal clinics and when women visit for antenatal screening tests. Importantly, the guidelines and service processes in place for the routine offering of screening and the smooth running of the programme may be experienced by pregnant women and partners as constraining on their decision making (Burkell and McKenzie, 2005; Spoel, 2007; McKenzie, 2009). Also, the interaction of the midwife - pregnant woman communication and the institutional contextual factors may be perceived by pregnant women as directive.

## **1.6 Variation in uptake of Down's syndrome screening**

Uptake of screening is defined as accepting either the first trimester combined test or second trimester quadruple test as recorded in the woman's hand-held notes. The UK National Screening Committee 2011-2012 annual report for England stated that more than 540,000 pregnant women were screened for Down's syndrome with 17,314 screen positive for the condition (UK NSC, 2012). Tringham et al. (2011) reported based on laboratory and booking data that there has been an increase in the uptake rates of screening across all

maternal age groups by a mean of 12.6% since the introduction of the combined screening test in an area that has been experiencing a decline in uptake rates (Gidiri et al., 2010) in the North of England. The researchers posited that the increase may be as a result of women's preference for earlier timing of the combined test or because of the improved detection rate.

Nevertheless, variation in uptake rates of 20% and 33% in three hospitals with an overall uptake rate of 28% has been reported in a study conducted in England using questionnaire surveys (Shantha et al., 2009). Participation rates have also been shown to vary widely between 22.7% and 73.9% across and within regions in the North of England (Walker and Tarn, 2012). Other countries such as the United States, France and the Netherlands which have a universal screening policy for Down's syndrome, have marked variation in uptake rates (McNeill et al., 2009). Variation in uptake rates persists within and between countries with similar socio-cultural factors and screening policy in terms of content and implementation based on autonomous and informed choice. For example, variation in uptake rates between countries in Northern Europe, such as the Netherlands 27%, UK 61% and 90% in Denmark have been reported (Hall et al., 2007; Boyd et al., 2008; van den Heuvel et al., 2008; van den Heuvel et al., 2009; Crombag et al., 2013).

Existing studies have attributed the variation in uptake rates of Down's syndrome screening to psychosocial differences among women (Khoshnood et al., 2003b; Bryant et al., 2006; Kuppermann et al., 2006; Fransen et al., 2007; Li et al., 2008; McNeill et al., 2009; Maxwell et al., 2011). A recent study by Bakker et al. (2012) using cross sectional survey questionnaires evaluated factors affecting take up of screening and variation in uptake rates in the Netherlands. The authors reported that the low and variable uptake rates of screening were as a result of the inter-regional differences in attitudes (positive attitude towards the Down's syndrome and a negative attitude towards TOP). The availability of family support, special education and specialised medical care reinforced the positive attitudes of women to children with the condition. Another factor was the perception of age-related risk with younger women declining screening over their perceived low risk. Additional factors reported were the influence of midwives on women's decision-making and the policy of reimbursement. The significance of the findings is that the reasons for the low and variable uptake rates are a combination of socio-demographic and organisational factors.

However, other studies have not found any associations between psychosocial differences and uptake rates (Press and Browner, 1997; Paul, 1998; Rowe, 2007; Alderdice et al., 2008; Rowe et al., 2008; Seavilleklein, 2009; Usta et al., 2010; Prathapan et al., 2012). Drawing a conclusion that psychosocial differences among women account for variation in uptake rates is therefore difficult. This has led to an assumption that service organisation and delivery may explain the variation in uptake rates of Down's syndrome screening.

Chiang et al. (2006) using observations and interviews explored informed choice of a convenience sample of 26 screen positive pregnant women who have accepted to undergo amniocentesis for Down's syndrome in Taiwan. The findings suggest that the variation in uptake may be influenced by the information given about the test. In addition, the manner of presentation and the knowledge and attitudes of the health professional who communicated the offer may account for the differences. However, the study involved a convenience sample of screen positive, fee paying women who agreed to amniocentesis which limits the trustworthiness and relevance of the findings. Similar findings have previously been reported by Baillie and Hewison (1999) in an editorial.

Existing work by Lynn et al. (2010) using questionnaire surveys completed by directors and managers of maternity units found variations in service provision and offers of Down's syndrome screening across most maternity services in a region in Northern Ireland (NI). The authors reported that the variations were due to factors such as the lack of agreement about the provision of screening even among health professionals. In addition, midwives experienced time pressure, inadequate training and termination of pregnancy was illegal by law except in the interest of the pregnant woman's mental or physical wellbeing. Hence, women cannot be offered abortion for foetal anomaly, owing to the social, moral or religious contexts unless in limited circumstances. Midwives therefore, experienced professional and personal conflict arising from the structure and organisation of the maternity services and when offering screening to women. The implication was that midwives offered screening to women from a negative perspective, which may account for the low uptake rates of Down's syndrome screening in NI as previously reported (Alderdice et al., 2008; McNeill and Alderdice, 2009; McNeill et al., 2009). The ramification of these studies is that contextual factors may influence women's participation in screening but this has not been fully investigated. The lack of a universal screening policy and limited TOP services in NI limit the generalizability of the study to the NHS context in England.

## **1.7 Influence of contextual factors on decision-making in healthcare programmes**

Contextual factors can be defined as any situations, forces or circumstances that may exist within or outside a healthcare setting and has the possibility of influencing the healthcare settings and the different programmes. Interestingly, existing studies have demonstrated that various contextual factors contribute to the decision-making process during information delivery in clinical practice (Stivers, 2002; Collins et al., 2005; Stivers, 2005; Koenig, 2011; Toerien et al., 2011).

Smith et al. (2008) in their book chapter reported that the broader context of health professional's clinical decision-making can be seen to consist of different types of factors that become relevant to particular decisions: social, professional, organizational, physical and environmental. The interaction between these contextual factors and decision-making is reciprocal, complex and dynamic. Importantly, the influence of specific contextual factors is based on the unique features of the decision being undertaken. The study found that physiotherapists used chatting with colleagues to validate their decisions and to create new ideas. Further, physiotherapists based their current decisions on decisions that have been made in the past. The author suggests that the collaborative nature of decision-making implied factors influencing healthcare professionals' decisions could also be considered factors influencing the decision-making processes of service users in clinical settings. Based on the hierarchy of evidence adapted for this study this is a low quality literature.

Existing research by Hedberg and Larsson (2004) that involved observation of nurses in clinical settings and Smith et al. (2008) suggest that the quality of decision making outcomes is determined by the decision maker, the undertaking and the setting where the decision-making process is conducted. Therefore, healthcare professionals and patients' decision-making cannot be separated from the context in which they occur.

Moreover, research has shown that Down's syndrome screening may be offered in a routine context with an assumption of acceptance, and that unless the pregnant woman clearly declines the offer, it is assumed she has agreed to screen (Pilnick, 2008; Tsouroufli, 2011; Pilnick and Zayts, 2012). The issue of how service contextual factors might affect participation in screening has not been comprehensively explored. The emphasis of this

thesis is on seeking an understanding of service contextual factors that influence women and their partners' participation in screening as they make choices in the maternity services.

Previous studies have mentioned how decision-making by service providers and users in clinical practice are sensitive to contextual influences, but most of the research has been limited in scope and not focused on prenatal Down's syndrome screening. The focus of some of the studies has been on the biomedical, rational choice model of healthcare professionals and patients' decision-making in clinical environments. However, Lippman (1991) cited in Seavilleklein (2009, p.74) "*argues that claims that women themselves need or choose prenatal screening is something constructed by the context of testing*".

Existing work by Charise et al. (2011), using a range of multidisciplinary insights from authors with previous or current interest in healthcare and in the field of public health, social sciences and engineering, produced a tool that could be used to systematically consider factors affecting health decisions. In developing the tool, the researchers adopted four themes: Bodies, Technologies, Place and Work (BTWP) from a framework developed by Health Care Technologies at the University of Toronto, Canada (McKeever and Coyte, 1999; HCTP, 2009 cited in Charise et al., 2011). These conceptual themes, together with the findings from the discussions between the multidisciplinary authors, yielded topics on the role of non-medical factors on an individual's decision-making.

The authors conducted a review of literature to identify health decision frameworks focused on non-medical factors. These existing frameworks were used to develop an iterative cycle of vignettes and questions which were reviewed by experts to ensure the vignettes had face validity. The vignettes generated a list of questions that were related to specific contextual factors relevant to health decisions that were not well represented in existing frameworks. The researchers identified the role of the human body and the relationship that existed between bodies as fundamental in health decision-making. This is similar to the findings by McKenzie (2010) using conversational analysis of tape recorded consultations in maternity units between midwives and pregnant women in Canada.

Technology was highlighted as a contextual factor in health decision-making. Women deciding whether to participate in prenatal Down's syndrome screening may be conflicted by information received on the internet (technology). The influence of place on health decisions was highlighted in the tool, as health decisions occur in different contexts which may affect an individual's decision-making processes. This was also noted in McCormack and McCance

(2006) framework that was developed through an iterative process that mapped and combined existing conceptual frameworks involving critical dialogue and focus groups with practitioners and co-researchers in person-centred nursing development. Charise et al. (2011, p.10) stated “*the symbolic, geographical, historical, economic, social, physical and formal connotations of place articulate a range of contextual factors that may influence individual preferences in health decision*”.

The concept of work was also recognised as critical in health decision-making by Charise et al. (2011). This was based on the responsibility placed on patients or service users to be well-informed and be involved in the health decision-making process. This has also been reported in existing work (Coulter and Ellins, 2007; McKenzie, 2009). Hunter (2006) used an ethnographic approach that involved observations, interviews and focus groups to explore emotion work experience of a purposive sample of 19 community midwives in the UK. The author found that how midwives experienced and managed emotions with pregnant women in the workplace is equivalent to physical labour or work. The involvement of student midwives in the study limits the transferability of the findings. The potential implications or relevance of the findings in prenatal screening is that informed choice placed cognitive labour on community midwives to inform pregnant women and partners about Down’s syndrome screening and on women to decide. This involvement placed a burden on women and partners which could be referred to as work, especially when combined with managing their health.

Charise et al. (2011) concluded that health decision-making is multi-factorial and iterative reflecting the complexity of health decisions. The authors contended that by carefully considering non-medical contextual factors within health services, the decision-making process may be improved. The improvement may come as a result of suggestions or innovations to improve decision-making or expose barriers that need to be addressed to enable the best conditions for informed decision-making processes.

### **1.7.1 Social contextual factors**

Eisenberg (1979) cited in Clark et al. (1991), Heyman and Henriksen (2001) and Lawson and Pierson (2007) pointed out that decision-making in clinical settings is subject to ‘sociologic influences’, such as social characteristics of medical providers and users, social interaction and differential power structure in clinical settings (expert authority) and the structure of health care organizations in which clinical encounters occur. These social influences could

have a positive or negative effect on health professionals' and patients' decision-making. However, Lawson and Pierson (2007) work was based a literature review that lack details of the method used. In addition, Lerrick (2008) in his book chapter suggests that when there is a prevailing culture or norm in a clinical setting, service providers and users could be constrained from exhibiting or adopting a different perspective.

The social context also highlights the status or power differential between the expert authority of health professionals (information giver) and service users (information receiver) (Clark et al., 1991; Spoel, 2007). The work by Spoel (2007) involved analysis of Ontario's policy documents for midwives through a selection of values and criteria for feminist rhetoric. The study lacked details of the method used in the review. Therefore, the interplay between the midwife-pregnant woman interaction in the maternity services and the exercise of authority depends on the basic processes of information-giving and the negotiation of meaning, as well as on the use of technical language in controlling the terms of the consultation. The implication of the findings in Down's syndrome screening is that this interplay could constrain women and their partners' participation.

In addition, McKenzie (2010) using conversation analysis of 40 tape recorded consultations between midwives and pregnant women in Canada reported that the interaction between that takes place at antenatal appointments contributes to the building of a relationship between them. Therefore, the transition of a pregnant woman to motherhood involved relationships with the foetus, health professionals and partner. The meaning of this finding is that the relationships formed in the prenatal context may influence women and partners' participation in antenatal screening programmes.

Thachuk (2007) reported in a discussion paper on how a pregnant woman's social context influenced her decision-making and how the desire to 'please the midwife' may influence participation in healthcare programmes. Existing research using qualitative methods such as interviews and focus groups revealed the quality of the midwife-pregnant woman relationship determines the woman's care experience in the maternity services (Berg et al., 1996; Walsh, 1999; Hunter, 2006; Aston et al., 2010). In particular, the research highlighted the active presence of the midwife as crucial to the woman's maternity care experiences. The midwife's presence created a friendly personal relationship, a sense of trust, safety and care that is woman-centred, above their professional based role. For others, the most important experience was feeling satisfied with the skills and expertise of the midwife. Yet

researching women's experiences of maternity care is becoming difficult and is complicated by women's reluctance to criticise midwives or healthcare staff (van Teijlingen et al., 2003; Hunter, 2006; McLachlan et al., 2009). There is also the notion of having been offered the best care possible and an expected attitude of gratitude.

The limitations of these studies were that they draw on the experiences of a small number of women and health professionals of childbirth and therefore may not be representative of midwifery users. The studies also focused on the perspective of either health care professionals or women without seeking the views of women, partners and midwives in a midwifery encounter.

Bandura (1986) observed that the social status and observable characteristics of healthcare professionals' could influence their social environment, including service users, before they say or do anything. In addition, existing research indicated that the social context has an influence on clinical decision-making (Lerrick, 2008; Smith et al., 2008). The literature is not clear on how social or professional contexts, such as the presence of a community midwife in the consulting room where pregnant women consent to Down's syndrome screening, influence their decision-making process.

Furthermore, there is a chronic shortage of midwives and there are concerns about falling maternity care standards in the UK (Hunter, 2006; Royal College of Midwives, 2011). There is clearly scope for further research into midwives', women's and partners' perspectives as evidence has shown that midwife-pregnant woman interactions are central to women's experiences in maternity care.

### **1.7.2 Organisational contextual factors**

A study by Jones et al. (2004) employed interviews and focus groups with a purposive sample of 24 GPs in England to explore involvement of patients in care when decisions involved scarce resources. The authors reported that organisational factors such as the volume and type of caseload and institutional policies and procedures may influence decision-making. The volume, type and distribution of caseload could influence decision-making processes by making less time available to make decisions and provide intervention. This could result in less effective interventions, scaling back assessments, less time for offering patients' information to enable them make informed decisions. Time, as an organisational resource, has also been found to influence decision-making processes by affecting healthcare professionals' involvement with patients. This tends to affect the

development of rapport with patients and create an asymmetrical relationship between patients and health professionals during consultations. Similar findings were reported among physiotherapists by Smith et al. (2008) in their book chapter

A qualitative study by Hedberg and Larsson (2004) used participant observations of nurses in three settings in Sweden to investigate how the environment affected health professionals' decision-making processes in nursing practice. Unstructured observations of nurses ( $n = 6$ ) throughout their shifts focused on their interactions, activities, events, time and the atmosphere in the clinical settings. The content analysis revealed the predominant themes influencing decision-making were environmental.

The two main environmental themes were interruptions and work procedures. Interruptions consist of what was done by people including patients, staff members requesting assistance and technical interruptions such as noises, telephone or emergency alarms. Interruptions occur when nurses provided direct personal care such as during check-up and ward rounds. The work procedures were elements that disturbed the continuity of nurses' decision-making such as time pressure to provide routine care, allocation of work and being available to patients and staff. The authors concluded that interruptions and work-related procedures are present all the time in nurses decision-making activity and could affect the quality of their decisions and the quality of medical care provided.

What is important to this thesis was that the study provided evidence that environmental factors within the healthcare settings influence nurses' decision making processes and outcomes in practice. However, the study involved observation of nurses and the data collection method was prone to observer bias and influence. In addition, the method does not allow participants to tell their stories which limit the findings. It was not possible to know from this study how the environmental factors in the prenatal settings where women consent to be screened could influence their decision making. Similarly, studies by McCabe (2004) and McCormack and McCance (2006) using qualitative methods such as interviews and focus groups, revealed that nurses can communicate well with patients when they use a person-centred approach, but that the ability to do so is heavily influenced by the care environment.

### **1.7.3 Physical environmental context**

Bandura (1986, p.18) offered a theory in which the environment acts in a dynamic and reciprocal way with the knowledge, attitudes, beliefs, values and other personal attributes of

individual decision makers. He suggested that "*human functioning is explained in terms of a model of triadic reciprocity in which behaviour, cognitive, and other personal factors and environmental events all operate as interacting determinants of each other*".

Smith et al. (2008) reported in their book chapter that the physical environment influences decision making by physiotherapists in healthcare settings. The physiotherapist needed to make decisions about the room layout, room décor, and location of equipment, type of equipment to be supplied and used within the environment. The literature by Davis and Walker (2010) explored how discourse operates through place based on interviews with midwives in New Zealand. The authors suggest that the space and layout of the obstetric hospital setting may shape the behaviour of women and the practices of midwives. Moreover, a literature review conducted by Kaplan and Kaplan (2008) reported that the Reasonable Person Model (RPM) suggests that people are more thoughtful, helpful, reasonable, co-operative and constructive when the environment is conducive. Howbeit, the influence of the prenatal environment on women and their partners' participation in screening has not been investigated.

Donabedian (2005) reviewed reviews which evaluated methods for assessing the quality of medical care and found that the outcomes of medical care such as quality of care could be assessed by studying the process of care and the healthcare setting in which care takes place. The assessment of the setting was roughly designated as the assessment of structure such as the physical facilities of the institutions providing care, policy, the qualifications and role of the medical staff. The assessment of process involved judgements based on considerations of information provision, continuity and acceptability of care and the involvement of patients in decision-making. Therefore, based on Donabedian quality framework, healthcare contextual factors were grouped into structure and process with the linear interaction between structure and process, as outcome. The Donabedian conceptual framework was adapted in this thesis to explore the influence of service organisation and delivery on women and partners' participation in screening as an outcome.

A study of the contextual factors and the process of information-giving takes the focus away from the woman and her partner's characteristics and allows for a study of the concrete contextual factors and processes through which informed choice is accomplished. This shift in focus is potentially valuable for policy makers and health professionals. Understanding the contextual factors at work during the consenting process of women for Down's syndrome

screening could alert healthcare professionals and policy makers to the dilemmas inherent in obtaining autonomous informed consent in practice. It would also provide strategies for recognising and overcoming these dilemmas. The dilemmas may include the different competing sources of information, women's understanding and knowledge of screening (McKenzie and Oliphant, 2006), the translation of the antenatal screening policy and care pathways in practice (Burkell and McKenzie, 2005; Spoel, 2007), the navigation of mandated woman-centredness (Salmon and Hall, 2003; NICE, 2008; Midwifery 2020 Programme, 2010) and nondirective counselling when these may be neither possible nor desirable (Williams et al., 2002b; McKenzie, 2009; Ahmed et al., 2012b; Ahmed et al., 2012c). However, the influence of service organisational and delivery contextual factors involved in prenatal Down's syndrome screening programmes on women's choices and how the national screening policies and guidelines are implemented in practice in maternity services in England have not been fully explored.

## **1.8 Overview of review**

The studies reviewed suggest that decision-making processes about screening involve complex contextual factors. These contextual factors challenge health behaviour models that assume rational patterns of individualistic, mechanistic behaviour in individuals (Lawson and Pierson, 2007; Charise et al., 2011). The findings of this study may challenge the current emphasis on nondirective informed choice in prenatal Down's syndrome screening and shed more light on the issue of informed consent generally.

## **1.9 What we know**

Pregnant women are offered Down's syndrome screening nondirectively and their decision-making is without influence and coercion such as the midwife's view regarding a particular option. Yet there are marked variations in uptake rates of Down's syndrome screening within and between regions in the UK. The reasons for variability of screening uptake are not well known, but several studies have suggested that differences in women's demographic and psychosocial status are responsible for the variation. The findings have been largely inconclusive. However, the literature review indicates some studies have reported on the influence of health service organisational and delivery contextual factors on women's decision making. This has led to the suggestion that service organisation and delivery could

account for the variation in uptake rates. Furthermore, variation in uptake rates may indicate ineffectiveness of the screening programmes. No matter the predisposition, a critical starting point for any programmes exploring ways to improve the service is recognition and understanding of variation in participation rates.

### **1.10 Gaps in the literature**

Some of the studies that reported on the influence of health service contextual factors on decision making were based on expert and anecdotal evidence (Lippman, 1999; Lerrick, 2008; Smith et al., 2008). A few studies have reported on the influence of service organisation and delivery on women's participation in prenatal Down's screening programme, but have been limited methodologically in scope. For example, studies that employed quantitative methods such as RCT and questionnaire surveys did not find any associations between the attitudes of midwives and uptake rates (Dormandy and Marteau, 2004; Bakker et al., 2012). The reason may be that the processes of decision making are complex, multifaceted and cannot be fully explored numerically. Studies that adopted observation and conversational analysis of tape recordings were focused on the content of the information and strategies used by midwives in the UK to impart information (Pilnick, 2004; Pilnick, 2008).

Many of the studies that explored the influence of service contextual factors using qualitative methods such as interviews and focus groups predate the introduction of universal screening in the UK (Williams et al., 2002a; Williams et al., 2002b). Furthermore, existing studies have reported on second trimester screening for Down's syndrome (McNeill and Alderdice, 2009; McNeill et al., 2009). Studies that have reported on first trimester combined screening have been limited in scope with regards to the findings given by women without the views of partners and midwives or vice versa. For example, Tsouroufli (2011) research that explored routinisation and the constrained contexts of Down's syndrome screening in the UK was conducted in a research context. It is also limited by the involvement of healthcare assistants offering screening to pregnant women.

Moreover, the little research available has been conducted with GPs in countries such as the USA, Netherlands, Australia and Northern Ireland, Sweden, Norway, Iceland, Greece and Canada. The findings may not necessarily be relevant to the NHS service within England.

Nevertheless, none of the studies have comprehensively explored the influence of service organisation and delivery on women and partners' decision-making process from the perspectives of midwives, women and their partners using a health service organisation and delivery model. Moreover, the relationship between service organisation and delivery and variation in uptake rates has not been fully investigated. In addition, how the national policies and guidelines are operationalised in practice in maternity services in England has not been fully explored. Therefore, we know little about how pregnant women and their partners' decision-making process are affected by the real life contexts of clinical practice. These are gaps in the literature.

This study is not meant to draw a cause and effect relationship between service organisation and delivery, participation in screening and variation in uptake rates. It is rather to provide a better understanding of the relationship between service contexts, participation in screening and uptake rates, thus revealing potential influences and pressures faced by pregnant women and their partners.

### **1.11 Research question**

Do the structures, processes and people in the prenatal settings influence women and partners' participation in screening for Down's syndrome?

### **1.12 About the researcher and his PhD Journey.**

I was born into a large family that lived on less than 50 pence a day. I worked hard to get through college and, against many odds, was admitted into a medical school in a national university. I graduated from the medical school and worked as a general practitioner (GP) for some years before joining my state (regional) university as a senior medical officer. Being in a university environment and interacting with staff and students brought up a desire to become a lecturer. I enrolled into a part-time Master in Public Health programme (MPH) and requested deployment to the College of Health Sciences as a lecturer in Public Health. The request was eventually granted. Whilst in the part-time programme, I accepted a scholarship to Denmark for an MSc programme in Public Health (Health Promotion). Towards the end of the programme in Denmark, I saw the School of Healthcare advertisement for PhD studentship positions. The position that caught and held my attention was the research on uptake of prenatal screening for Down's syndrome. As a GP, I have attended to children with

Down's syndrome and have seen children with the condition as a medical student. I was also aware that screening for prenatal Down's syndrome is not widely available in my home country, only in few teaching hospitals. I applied for the position and was subsequently awarded a studentship. The PhD journey has been tough, especially with the data collection methods which I nearly gave up on, due to the technical difficulties and initial participants' apathy. On the family side, it has not been easy either, with my wife and I having to endure a series of obstetrics/medical challenges.

However, my supervisors have been wonderful and magnanimous in their support, which in no small way helped to overcome some of the difficulties. The PhD research has been an opportunity to acquire research skills and knowledge which I hope to pass on to my students. It will also enable me to conduct further research and contribute to the policy debate on women and partners' participation in prenatal Down's syndrome screening. Hopefully, this PhD will enable me to climb up the academic ladder, especially with the publications that will come out of it.

### **1.13 Structure of the thesis**

This thesis is presented in seven chapters.

#### **Chapter 1**

Introduces the issues involved in prenatal Down's syndrome screening to provide background and context to the thesis. The first section will define participation in healthcare programmes. Participation in Down's syndrome programmes and variation in uptake rates are thereafter discussed. The second section will present a review of interdisciplinary and healthcare studies that have examined how contextual factors influence service providers' and users' participation in healthcare programmes.

#### **Chapter 2**

This chapter continues with the literature review that was initiated in chapter 1 and critically reviews contemporary literature on studies which have investigated factors influencing pregnant women and their partners' participation in prenatal Down's syndrome screening.

#### **Chapter 3**

The Donabedian Healthcare Organisation and Delivery Model as the conceptual framework adopted for this study is described. The logic for adopting and adapting it to the aim and objectives of this study is also explained.

#### **Chapter 4**

A discussion of the document review conducted as part of this study to provide supplementary data on how the national guidelines are operationalized in the maternity services and the context for the online interviews with community midwives, pregnant women and their partners is presented.

#### **Chapter 5**

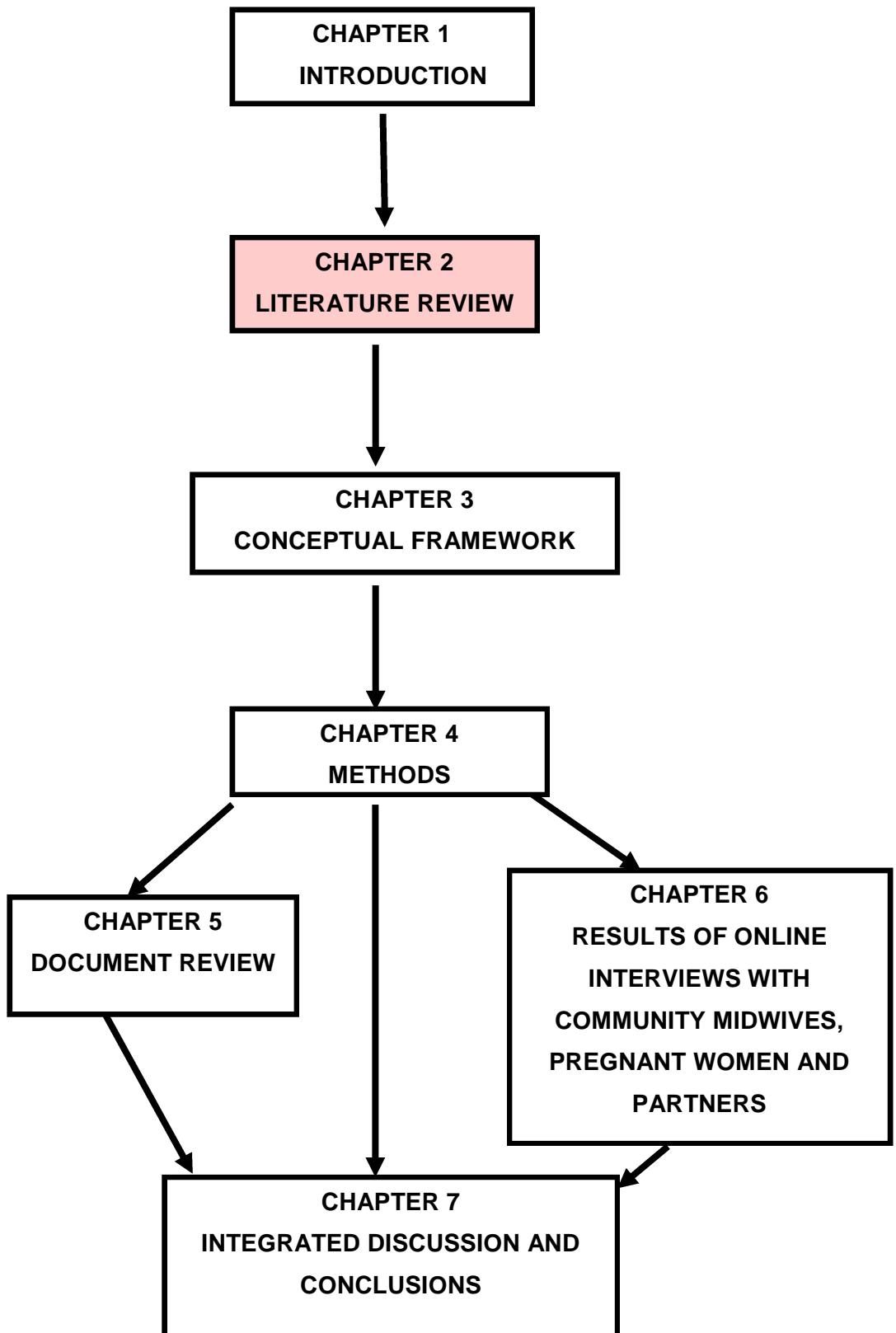
A detailed explanation of the methodology and the methods employed in this study is presented in this chapter. The pilot study and its findings are also presented. The main study, trustworthiness of the findings, data management and analytic procedure are described.

#### **Chapter 6**

The results of the online interview of community midwives, pregnant women and partners are presented in this chapter. An overview of the findings including a comparison of the findings is also provided.

#### **Chapter 7**

This chapter provides an integration of the findings from the document review and the online interviews. A model depicting the influence of service organisation and delivery is developed and presented here. A social model of shared decision-making process is proposed to mitigate the influence of service organisation and delivery. The thesis will then end with the conclusions drawn and plans for dissemination of the findings



## CHAPTER 2: LITERATURE REVIEW

### 2.1 Introduction

This chapter is a continuation of the review of the contemporary literature in chapter 1 on contextual influences on decision-making in the healthcare settings including prenatal Down's syndrome testing programmes. Although a systematic review was not conducted, systematic methods were nevertheless employed to ensure relevant studies were included as explained in the first section. This is followed by the review of literature on the experiences of women, partners and midwives in prenatal screening. Subsequently, factors influencing women's participation in prenatal Down's syndrome screening are presented. This is followed by a section on decision-making, the purpose of the study and a summary of the chapter with the research aim and objectives.

### 2.2 Method of the literature review

In order to explore the influence of service organisation and delivery on women and partners' participation in screening, it is important to understand the experiences of health professionals, pregnant women and their partners. In addition, contemporary studies on factors influencing decision-making processes and uptake of screening in the prenatal context of Down's syndrome screening were also reviewed. The literature search was conducted from October 2010 to September 2013. The electronic databases searched from 1990 – 2013 were EMBASE, Global Health, HMIC Health Management Information Consortium, Maternity and Infant Care, Ovid MEDLINE(R), PsycINFO, CINAHL, Cochrane Central Register of Controlled Trials (CENTRAL), Cochrane Database of Systematic Reviews (CDSR), NHSEED (NHS Economic Evaluation Database), EBM Reviews - ACP Journal Club, EBM Reviews - Database of Abstracts of Reviews of Effects, EBM Reviews - Health Technology Assessment, Open SIGLE (System for Information on Grey Literature in Europe), Web of Knowledge (Web of Science). The date limit of 1990 was chosen, as prenatal Down's syndrome screening was introduced in the late 1980s (McCormick, 2011).

The search was limited to publications in English, because of the cost involved in translation and also the time required. Hand searching the reference lists of retrieved articles, the Journal of Medical Screening and the Journal of Intellectual Disability for relevant studies

was also conducted. The hand searching of retrieved articles yielded valuable resources especially on contextual factors influencing decision-making in healthcare settings mostly reported in Chapter 1.

The search strategy generated 746 hits after excluding papers in other languages, 64 papers remained following the review of titles, abstracts and full text including those derived from the retrieved articles' reference lists. They consist of qualitative (31), quantitative (20), review (6) and mixed methods studies (3), books (3) and conference abstract (1). Therefore, most of the literature based on participation in Down's syndrome testing was read to obtain potential insights into the influence of service organisation and delivery on women and partners' decision-making process. A limitation of the selection process is that some studies may have been missed. It is not possible to identify all articles or papers that reported or mentioned aspects of the influence of service organisation and delivery at the phase of appraisal using titles and abstracts. See appendix 1 for search terms, databases and number of hits.

### **2.2.1 Quality appraisal**

Each article was read for its relevance to the inclusion criteria and the quality of the studies was evaluated using a critique-led approach that took the whole body of the literature as the object of inquiry. Existing research suggests that quality should be defined by the extent to which the article answers the research question, instead of the quality of reporting. Hence, no study was excluded based on the quality of the research (Greenhalgh and Peacock, 2005; Dixon-Woods et al., 2006; Dixon-Woods et al., 2007; Hannes, 2011). Sandelowski et al. (1997) reported that there are no gold standard criteria for identifying, including or excluding a high quality qualitative research paper. Nevertheless, the included studies were appraised using the CASP tools for trial and review (see [www.caspinternational.org/?o=1012](http://www.caspinternational.org/?o=1012) for more information), the Health Evidence Bulletins-Wales for observational studies (see <http://hebw.cf.ac.uk/projectmethod/appendix8.htm>) and EBN tool for qualitative research (Russell and Gregory, 2003). The quality appraisal process was used to identify weaknesses in study methodologies and to better interpret and assess their findings. To overcome the potential problems associated with methodological incomparability of studies, where different methods might have been used to explore the research aim, the inclusion criteria were based on the aim of the thesis rather than on study design or method (Sandelowski et al., 1997; Sandelowski et al., 2007). In this sense, all studies, regardless of study design,

whose aim was to explore and report on health professionals', pregnant women and their partners perspectives or experiences of, or attitudes towards Down's syndrome testing, antenatal testing or decision making in healthcare programmes were eligible.

Therefore, an unstructured approach as described by Dixon-Woods et al. (2007) was also adopted in this study. The approach required researchers to use their expertise and judgement to include articles in their study. This was also supported by the fact that no single appraisal tool can identify all the flaws that occur in different types of studies (Dixon-Woods et al., 2004; Dixon-Woods et al., 2007). This approach was also crucial to this thesis, as the influence of service organisation and delivery in the prenatal context is subtle, complex, multifaceted and most often not fully explored in studies that have reported on participation in Down's syndrome screening. Hence, the research question for this study determined the hierarchy of evidence used in this study and not the traditional hierarchy of evidence as noted in Sackett et al. (1996) work based on effectiveness of interventions. The hierarchy of research developed for this literature review has qualitative studies as the highest type of evidence with expert and anecdotal opinion papers as the lowest type of evidence. Qualitative studies, meta-synthesis of qualitative research and systematic review (highest level of evidence), observational studies and RCT (medium level of evidence), expert and anecdotal opinion (lowest level of evidence). There are emerging hierarchies of evidence for qualitative studies (Daly et al., 2007) but there is little agreement about their value.

Several of the included studies failed to report piloting of questionnaires and testing for reliability and validity. The quality of the studies were variable with some of low quality based on the hierarchy of evidence (Lippman, 1999; Rapp, 2000; Lerrick, 2008; Smith et al., 2008; Ghosh et al., 2011). Some lack details regarding methods (Dudgeon and Inhorn, 2004; Lawson and Pierson, 2007; Spoel, 2007; Thachuk, 2007). Others based on the critical appraisal revealed weaknesses in the study design, sample, size, response rate and techniques to ensure validity or trustworthiness and relevance of the study (Chiang et al., 2006; Winquist et al., 2008; Aune and Möller, 2012). Most of the quantitative studies that employed cross sectional survey questionnaires relied on a convenience sample of participants and may be limited by selection bias. A flaw of many of the qualitative literature was that reflexivity and how trustworthiness was ensured were not always discussed. In addition, the methods and sample size were not always justified and the theoretical framework adopted not stated. Data were sometimes collected postpartum in some of the studies with recall bias as a possible limitation. Therefore, generalising the findings to the

whole target population or transferability of the findings to other settings required careful consideration. However, the overall quality of most of the included studies was fairly good. The quality appraisal of peer reviewed Journals may be affected by the word limit given by publishers and the articles may not include all the necessary information for quality reporting.

### **2.2.2 Selection of studies**

#### **2.2.2.1 Inclusion criteria**

The retrieved articles were based on the following criteria:

1. Refereed journal articles and grey literature
2. The paper is published in English
3. Studies on the perspectives, attitudes, experiences of women, partners and midwives' participation or decision making process in prenatal Down's syndrome testing and antenatal testing.
4. Studies on factors that may influence participation or decision-making in healthcare programmes.

#### **2.2.2.2 Exclusion criteria;**

1. Articles reporting findings from book reviews.
2. Editorials
3. Articles reporting on pregnant women with twin pregnancies or below the age of 16 years.
4. Non-English language studies.

### **2.2.3 Literature synthesis**

The synthesis of this literature review was based on a subjective narrative overview of previous studies as noted in Weed (2005) and (Dixon-Woods et al., 2005). It is essentially a scoping exercise that summarises existing studies to establish the research need. The significant results of quantitative studies and the authors' findings in qualitative studies were collected, compared and categorised into major topics and subtopics.

## **2.3 Health professionals' experiences of prenatal Down's syndrome screening**

Most of the literature about participation in screening has been based on the views and perspectives of women. The perspectives of midwives offering Down's syndrome screening have seldom been explored (McNeill and Alderdice, 2009; Ahmed et al., 2013). This may be as a result of the difficulties encountered recruiting midwives for research in the constrained context of maternity services as noted in Reed (2009a).

Existing research conducted in Australia by Nagle et al. (2008) involving four focus groups with 27 GPs suggests six themes have an impact on health professionals' experiences. Data was analysed using constant comparison methods. The themes were categorised as intrinsic factors which were elements within the context of the consultation and extrinsic factors which were elements outside the context of the consultation.

The intrinsic factors were the limitations of screening (a risk, not definitive) which the GPs found difficult to communicate to women either because of lack of skills or confidence and to avoid making women anxious. Variation in the amount and quality of information given to patients was also highlighted. Selective information-giving was adopted by GPs either intentionally or unintentionally based on the information needs of women and was usually based on age or parity. Some GPs claimed to be directive during information-giving, whilst others indicated that they involved the women in decision-making. Time constraint was a source of stress within GPs owing to the multiple issues during the prenatal encounter.

The extrinsic factors were the negative emotional impact of screening information on the GPs largely due to the implications of screening. This generated conflict within GPs mostly from the potential of screening to do 'more harm than good'. The lack of control with the process of informing women especially over the demands on them such as workload, training needs, empowering and advocating for women were issues raised by the GPs. Additionally, they felt frustrated with offering screening to women with poor understanding and from poor communication with staff. The finding that the contextual factors in the prenatal setting may affect women's decision making process is highly relevant to this thesis. However, the study was conducted in a setting without universal screening policy and some services were on fee basis which limit its transferability to the NHS UK context. Further, the

views of midwives, women and their partners were not sought. This is a high quality study with a research question that was explored using an appropriate study design. It has few flaws that may affect its trustworthiness. Similar findings have been reported among GPs offering Down's syndrome screening to women in Canada who were interviewed in the study (Park and Mathews, 2009).

Existing studies in the UK using qualitative approach have reported midwives experience difficulties with some of the intrinsic and extrinsic factors during consultations with pregnant women (Williams et al., 2002a; Williams et al., 2002b; Pilnick, 2004; Williams et al., 2005; Pilnick, 2008; McNeill and Alderdice, 2009; Tsouroufli, 2011). Though, most of the studies were limited in scope as it involved other health professionals not necessarily midwives and conducted before the implementation of the universal screening policy in the UK. However, how the intrinsic and extrinsic factors present in the prenatal context of Down's syndrome screening has not been comprehensively studied in the NHS context in England.

Winquist et al. (2008) used postal questionnaire surveys to investigate the ethical and social dilemmas associated with Down's syndrome screening from the perspectives of 191 GPs and family physicians in one Canadian province. The study found that most healthcare practitioners offer Down's syndrome screening to all pregnant women, but many were of the opinion that it should be offered to high risk women only. Seventy-three percent of the responders believed that screening creates anxiety for women, with two thirds believing it is useful as it enabled women to decide. About 54% reported difficulties in explaining the limitations of screening to women and 51% felt they needed more training. Sixty percent of the responders (obstetricians more than physicians) felt they were comfortable referring women with a positive diagnostic test for TOP. About 23% felt conflicted with the referral based on their moral values and beliefs. This may indicate a lack of support for serum screening and women's acceptance of it. In addition, the study found that female professionals were more concerned about the ethical, social and clinical implications of screening than male professionals. The authors surmised that the views of physicians about screening were diverse. It is within this opposing diversity and biases that clinical interactions and dilemmas faced by physicians should be understood and the variation in take-up rates of serum screening explained. The low response rates (39%) and the study involved physicians and obstetricians limit its reliability and validity. It is not clear if confounding and bias was considered at the validity assessment stage. In addition, the

questionnaire surveys may not have been the ideal method to meet the research aim. The conclusions reached may therefore not be generalisable to the target population.

The diversity of views and opinions about testing information among health professionals was demonstrated in a study by Durand et al. (2010). The study involved interviews with 17 health professionals and 17 pregnant women and sought to explore the information and decision support needs of parents considering amniocentesis. There was no agreement among health professionals about the nature and quantity of information to provide about the risk of possible chromosomal disorders, risk of miscarriage, TOP and probability associated with the tests. Yet most professionals agreed on the need for updated information in different formats and that the information should be optional, easy to understand, given gradually and tailored to women's individual needs. Interestingly, few professionals responded to the question that women should be given time to think about the test, decide with a partner and also be reminded that they could opt out at any point. The authors surmised that some of the diversity of opinions among health professionals may be as a result of the limitations of current policies and guidelines. Whilst Durand et al's work has some limitations in that it is focused on diagnostic testing, the findings are relevant. The implication of the study is that the various guidelines and policies in Down's syndrome screening may influence how information is presented by health professionals to women. However, we know little about how the various national guidelines and policies are implemented in practice in maternity services and the effect on women's decision making processes.

The differing agendas between health professionals and women have been reported in the literature as dilemmas experienced by health professionals involved in offering Down's syndrome screening (Williams et al., 2002a; Williams et al., 2005). Women wanted to keep the pregnancy, in addition to wanting reassurance about the pregnancy and an opportunity to see the baby during dating and NT screening scans. Still, healthcare professionals were interested in screening for congenital disorders. The studies are well conducted but the findings are limited by the absence of a universal screening policy for prenatal Down's syndrome when they were conducted.

Existing research using questionnaire surveys involving obstetricians, midwives and women conducted in Australia and the UK suggests they have different preferences towards the timing of different screening tests and detection rates (Bishop et al., 2004; Lewis et al.,

2006a; Lewis et al., 2006b). The attitudes exhibited by the health professionals showed between and within group differences in preferences. Further, women have greater preference for the safety of screening tests compared to health professional who placed greater emphasis on earlier testing. The implication is that the different perspectives may influence the way Down's syndrome screening is presented to pregnant women and their partners thereby influencing their decision making. However, the survey methodology adopted in the studies limits the exploration of the views of participants.

In addition, midwives in the UK surveyed by Skirton and Barr (2010) online or through hard copies of questionnaires sent by post felt they had the skills to offer Down's syndrome screening to women. Though, a minority of midwives felt they needed training updates. Interestingly, among the midwives who felt they had the skills to offer screening to women, some could not distinguish screening from diagnostic testing and the conditions currently being screened for in the UK. The majority of the midwives knew that Down's syndrome was one of the conditions screened for, but few midwives did not know which test was used to diagnose it. Most midwives in the study reported providing screening information to male partners when present. Additionally, some midwives reported being comfortable when women declined the offer of screening. Yet, many of the midwives did not respond to the question about being comfortable when women declined screening. In spite of the low response to the question, the majority of the midwives reported not feeling vulnerable to litigation when women declined screening. This is in contrast to the findings by Ahmed et al. (2013) where midwives expressed concern about the fear of litigation. The reason for the contrasting findings may be the different study design adopted in the studies. The study by Ahmed et al. (2013) used a qualitative approach (interviews) whilst Skirton and Barr (2010) administered questionnaire surveys to 78 midwives.

In a recent study conducted by Ahmed et al. (2013) in the North of England that involved semi-structured face to face interviews with 15 community midwives reported midwives experienced challenges such as time constraints and language problems with women from ethnic minority groups. Time pressure was exacerbated when midwives engaged interpreters to inform women with limited English language skills. Midwives often resorted to the use of photographs of children with Down's syndrome. The midwives also reported having difficulties offering the combined screening test, on account of the short window of opportunity. Further, facilitating informed choice with teenagers and women with complex social needs was a challenge to the midwives. Another challenge encountered at booking,

was when women asked for advice. The issue of women asking for advice has been extensively reported in research by Ahmed et al. (2012b) and Ahmed et al. (2012c) using Q methodology . The difficulty with knowing what advice to give to women in the prenatal context was highlighted as a concern by midwives, to avoid influencing women's decision-making processes. Thus, advice could range from information-giving to being directive by recommending a particular course of action. The studies are high quality with appropriate study designs, analysis of data and the findings are relevant. The potential ramifications of these findings are that the context of Down's syndrome screening may be confusing and conflicting.

McNeill and Alderdice (2009) conducted research involving qualitative interviews with 15 midwives in Northern Ireland (NI) using an ethnographic approach. The findings suggest that the dilemmas experienced by midwives offering Down's syndrome screening to women include the ethical issues of TOP, limited availability of TOP services and lack of professional control. The negative experiences of midwives revealed in the study have the potential to interfere with the midwives' role in objectively facilitating women's understanding of screening information. The negative experiences of midwives' offering Down's syndrome screening to women have also been reported in other research conducted in England (Tsuroufli, 2011; Ahmed et al., 2013).

In addition, existing research using a combination of 70 semi-structured interviews and some small group discussions with a broad range of health professionals revealed that health professionals were concerned about the challenges encountered when facilitating informed choice in the prenatal context (Williams et al., 2002a; Williams et al., 2002b; Williams et al., 2002c). The concerns arose from the experiences of women having difficulties opting out of screening, understanding the concept of risk and the offer of screening as a constraint on women's choices. The experiences of midwives revealed in these studies have the potential to interfere with the midwives role in objectively facilitating women's understanding of screening information. The research has well defined aims and appropriate study designs but with few flaws such as involving health professionals with indirect involvement in perinatal care. The findings are relevant to this study. However, the findings provided little discussion of the influence of service organisation and delivery on women's decision making processes and predate universal prenatal screening for Down's syndrome in the UK.

Winquist et al. (2008) reported that the linking of screening to TOP was psychologically challenging to health professionals and may influence how screening was offered to women. The researchers suggested the routine offer of screening would overcome some of the emotional issues involved in screening. However, this suggestion is not supported by the findings of the study.

## **2.4 Partners' experiences of prenatal Down's syndrome screening**

It has been a national policy in the UK for all pregnant women to be offered Down's syndrome screening since 2003. Where possible, the policy recommends the involvement of partners, but the experiences of male partners in the screening process has not been well studied (Skirton and Barr, 2010). The importance of female partners of pregnant women in same-sex partnership is appropriately acknowledged. However, the focus of this thesis is on male partners' participation in screening.

Most existing research about participation in screening has been on the experiences of pregnant women (Green et al., 2004; García et al., 2008b; Skirton and Barr, 2010). The literature suggests the reason may be owing to men's reluctance to take part in research on pregnancy as it is perceived to be a woman issue (Rapp, 2000; Reed, 2011). This indicates that male partners' participation in prenatal Down's syndrome screening has not been fully explored. In Europe, there has been a call by the World Health Organisation for recognition of the role of male partners in healthcare (WHO, 2007). In the prenatal context, the Council of Europe (1990) and NICE guidelines (NICE, 2008) were made with regards to women and their partners. For example, they state men should be involved in screening and that women and their partners' views, values and beliefs should be explored and respected.

However, busy schedules such as work commitments prevented some partners from attending prenatal appointments, and sometimes women and their partners may decide it is not necessary for the male partners to take time off work to attend what is seen as a routine appointment (Locock and Alexander, 2006). Furthermore, partners do not have a legal right to paid time off work to attend prenatal appointments, as this only applies to pregnant women (Maternity Action, 2013).

The trend in the UK is the first trimester combined screening, which may pose difficulties for women and their partners, because of the risk of participating in screening without being able to fully discuss the implications of screening and make informed decisions, considering

the short window of opportunity (Heyman et al., 2006; Locock and Alexander, 2006). Moreover, partners' participation in screening is important as women preferred to make decisions about screening with their partners (Ahmed et al., 2012a; Ahmed et al., 2012b). This enables women to make informed choices, howbeit, the majority of women perceived their partners to have a strong influence on their decisions (Jaques et al., 2004b; Ahmed et al., 2006a; Korenromp et al., 2007).

Importantly, as early screening such as the era of non-invasive prenatal diagnosis (NIPD) draws near, partners may be left out of screening as a result of the short window of opportunity. This may be further exacerbated by partners' inability to attend prenatal screening appointments owing to work commitments and limited engagement with midwives (Skirton and Barr, 2010; Williams et al., 2011). There is, therefore, a need to explore the experiences of partners in first trimester Down's syndrome screening as noted in previous research (Green et al., 2004; García et al., 2008a; Skirton and Barr, 2010).

Research using questionnaire surveys administered to 737 pregnant women 37 years or over opportunistically sampled in Australia has shown that women are eager for their partners to participate in screening (Jaques et al., 2004b). However, partners experience some conflicts when attending prenatal screening. This may be as a result of some partners' believing that prenatal care is the responsibility of women. This belief is reinforced by partners' views of maternity services as being dominated by pregnant women and health professionals who are mainly women as noted in other studies using a qualitative approach (Plantin et al., 2003; Ekelin et al., 2004; Locock and Alexander, 2006; Reed, 2009b; Theuring et al., 2009).

Research conducted in the UK by Skirton and Barr (2010) using questionnaire surveys reported that 79.3% of the 111 responders ( 100 women and 11 men), were involved in the decision-making about screening. Yet, only 31.5% believed they discussed the choice of screening with their partners, with 19.8% reporting some discussion with their partners. Therefore, almost a third of those who reported joint decision-making did not actually discussed it with their partners.

Furthermore, research by Gottfredsdóttir et al. (2009b) involving 40 semi-structured interviews conducted at 7–11 weeks and at 20–24 weeks of gestation with 10 couples in Iceland. The findings revealed inconsistencies between women and partners' reports on joint decision-making about screening. Women reported making the decisions with their partners. Whilst some of the partners reported that women made the decisions alone. These studies

clearly demonstrated the need to involve partners in research to obtain a complete perspective of their participation in screening. However, previous research using qualitative approach suggests that male partners would have valued being in control of decision making concerning the pregnancy, but felt it is the woman's body that is involved (Gottfredsdóttir, 2005; Gottfredsdóttir et al., 2009b). Supporting women in screening decision-making processes may be a way to meet this need and thereby decrease uncertainty.

A recent metasynthesis of qualitative research on men's experiences of antenatal screening by Dheensa et al. (2013a) reported that partners' experiences of prenatal care were dependent on the support they received. Partners who attended prenatal appointments, but felt ignored by midwives, reported feeling unsupported and hence perceived the appointments as unnecessary (Bronte-Tinkew et al., 2007; Cabrera et al., 2008; Hildingsson and Sjöling, 2011; Williams et al., 2011). This finding demonstrates that the level of engagement of partners in screening was most important to their experiences of screening. The meta-synthesis was a well conducted study with well-defined search questions that was supported by appropriate inclusion and exclusion criteria, included papers were appraised using the CASP tool, the data extraction and synthesis were done under the guidance of independent supervisors. Given the evidence presented the authors' conclusions appear appropriate.

Importantly, previous work suggests partners' attendance at prenatal appointments reflects their responsibility as good fathers which they expressed by supporting their female partners during antenatal testing to ensure the birth of a healthy baby (Browner, 1999; Markens et al., 2003; Locock and Alexander, 2006; Reed, 2009a; Williams et al., 2011). The studies used traditional qualitative methods to explore male partners' experiences with prenatal screening. However, Williams et al. (2011) involved online email interviews with 8 male partners longitudinally at 16 and 28 weeks and postpartum to explore their experiences of antenatal testing. Recall bias and only middle class participants with access to the internet may have agreed to participate, which are limitations of the study. However, these flaws have little to no effect on the trustworthiness or relevance of the findings.

Research suggests that whilst women were primarily focused on the emotional aspect and the effect of screening on their foetus, partners were more interested in technical and statistical information about screening especially from healthcare professionals, friends, family and searches made on internet (Browner, 1999; Markens et al., 2003; Ekelin et al.,

2004; Locock and Alexander, 2006; Gottfredsdóttir et al., 2009b; Reed, 2009b; Skirton and Barr, 2010; Williams et al., 2011). Obtaining such information empowered partners to be involved in joint decision-making with their pregnant partners. The information obtained was used to support their female partners during the decision-making process.

In spite of being empowered by the information and their participation in decision-making, partners' participation in screening remained limited (Draper, 2002; Locock and Alexander, 2006). The meta-synthesis by Dheensa et al. (2013a) explained that only midwives could provide the information necessary to guide women's decision making about screening.

Furthermore, midwives had a duty to accept women's, and not partners, final decisions. This may be a source of tension between pregnant women and their partners if they have different views about the options for screening.

Previous work suggests that male partners felt that their participation in screening such as information dissemination and formulation of options and integration of information was important to support their pregnant partners' decision making (Markens et al., 2003; Locock and Alexander, 2006; Williams et al., 2011). They were also concerned about their unborn baby. This concern is reflected in pregnant women and their partners having strong emotional feelings about accepting or declining screening. In some instances, partners declined screening over the risk of false-positive results that unfairly reveal an abnormality for a healthy foetus. Additionally, some women and partners would not consider TOP in case of the condition and in other instances, felt capable of coping with any child (Ivry and Teman, 2008; Williams et al., 2011).

A few studies on the influence of partners on women's take up of screening were found, but none was found for how service organisation and delivery influenced their participation in screening for Down's syndrome. A book written by Rapp (2000) reported that women's acceptance or decline of amniocentesis is influenced by the beliefs of the partners as they were less likely to undergo the test if the male partner is supportive of raising a child with disability. Furthermore, partners may directly affect women's participation in TOP as noted in a discussion paper based on the analysis of literature (Dudgeon and Inhorn, 2004). Partners may provide or withhold economic and emotional support either for a TOP or for parenting, or they may actively or passively impose their desires for or against TOP on their partners. The potential significance of these findings is that partners can influence women's

decisions to accept or decline screening. However, the low quality of the work based on the hierarchy of evidence limits the trustworthiness and relevance of the findings.

Factors such as absence of male partners at counselling and the woman's own assessment of risk have been found to be strong determinants of refusal or acceptance of prenatal diagnostic testing (Browner, 1999; Browner et al., 1999; Gidiri et al., 2007). An existing study using longitudinal questionnaire surveys explored the acceptability of screening for cystic fibrosis in primary care settings. The study revealed twice as many women as male partners agreed to be screened (Bekker et al., 1993). This may be owing to male partners' perception of carrier testing as associated with reproduction and childbirth and hence, more of the woman's responsibility. However, the study design adopted may not be ideal to explore the reasons for the higher acceptability of carrier screening among women compared to their male partners.

The association between partners' participation and uptake of interventions was strongest when male partners who came to the clinic agreed to be counselled together with their female partners. The prospective cohort study by Farquhar et al. (2004) using questionnaires revealed that partners' participation in voluntary counselling and testing (VCT) for HIV can improve acceptance and utilisation of preventive strategies. The authors suggest that counselling women and their partners together in the antenatal setting may have additional benefits to individual VCT. However, the quality of the relationships between the women and their partners may have independently led to improved uptake of interventions rather than couple counselling, which limits the validity of the finding.

A few studies that involved interviews with women and their partners to explore their decision making have been conducted. Browner (1999) reported on diagnostic testing and indicate disagreement on the issue, with more men in favour of accepting amniocentesis. Ekelin et al. (2004) reported that partners were weakly involved in decision-making, have little knowledge of prenatal diagnosis and minimal or no attachment to the developing foetus. However Gottfredsdóttir et al. (2009b) suggest most pregnant women reported making joint decisions with their partners about screening, but some male partners claimed they left the decision to the women. In addition, some male partners claimed information about screening was given to them by their female partners. None of the male partners sought information on their own nor discussed it with others.

Previous research conducted by Reed (2009a) involved interviews and focus groups with 15 midwives, 22 pregnant women and 16 male partners, explored fathers involvement in general antenatal screening. The study reported that male partners' participation in screening varied from clinic to clinic but perhaps was greater in clinics where the dating and anomaly scan were both performed. Ethnicity and level of education were issues in partners' participation in screening, particularly regarding the decision-making process. Language was also a concern with partners' participation in screening with midwives reporting that male partners do not pass the correct information to their female partners when acting as interpreters (Bowler, 1993). This failure of male partners to pass on information to their pregnant partners restricted midwives' ability to fully inform pregnant women about their screening choices. Additionally, it takes away women's reproductive autonomy, thus undermining their ability to make informed, autonomous decisions about screening. This is a well conducted study that explored the perspectives of midwives, women and their partners and there are few flaws that affect the trustworthiness or the relevance of the findings.

Nevertheless, in situations where screening identified potential problems, some of the men saw themselves as bystanders and their individual feelings overlooked. In other cases, partners became the main channel of communication between the pregnant woman and health professional, acting as an advocate in a difficult situation. Though, other studies have argued that the emphasis on the partner's role as the main support person for the woman during pregnancy ignores their psychosocial aspects and emotions (Beardshaw, 2001; Schott, 2002; Green et al., 2004; Gottfredsdóttir, 2005; Gottfredsdóttir et al., 2009b).

The findings of these studies do not indicate the influence of service organisation and delivery on partners' participation in screening, but provided some insight into their experiences of the prenatal Down's syndrome programmes.

## **2.5 Pregnant women's experiences of prenatal Down's syndrome screening**

Existing research using qualitative approach suggests women are initially anxious about screening, especially first time pregnant mothers, but found the first trimester screening test reassuring when the result came back lower risk (Heyman et al., 2006; Hundt et al., 2008). In addition, the pregnancy was made more real from the visualisation of a healthy foetus. However, a few women experienced anxiety which was not alleviated by a negative diagnostic test result.

Engaging in the screening test enabled women to move from a tentative state to a more definitive pregnancy (Williams et al., 2005; Favre et al., 2008; Hundt et al., 2008; Gottfredsdóttir et al., 2009b; Aune and Möller, 2012). It allowed women to publicise the pregnancy, especially through social networks like Facebook and obtain scan photos for family members. The researchers concluded that the NT scan in the first trimester screening and the 20<sup>th</sup> week scan were perceived as routine tests and may be difficult to decline, as they were understood to be part of the antenatal care package.

Skirton and Barr (2010) conducted a questionnaire survey with 100 pregnant women, 11 male partners and 78 midwives in the UK. Some women reported not remembering if Down's syndrome screening was discussed or offered at booking. Interestingly, most of the women reported not having other sources of information about screening, making health professionals the most important source of information. In addition, the women felt they had enough support to make a decision about screening and found information from midwives the most helpful in making a decision, with other sources the least helpful. Women who wanted more support reported wanting more information from the midwives and would have preferred it at an earlier stage in pregnancy. There is lack of ethnic diversity and less educated women were not sufficiently represented in the sample survey. This should be taken into account during interpretations of the findings.

Nevertheless, the qualitative study by Aune and Möller (2012) conducted in Norway with a sample of 10 women aged 38 to 42 years who were interviewed twice. The authors revealed that many women reported being more comfortable with finding out information from sources such as the internet rather than from healthcare professionals. The relevance of the study is limited by the exclusion of women with fertility problems or history of miscarriages. The small sample size of 10 older women (only one primigravida) and the fact that the study was conducted in Norway, limits its transferability to the NHS UK context.

## **2.6 Factors influencing women's participation in prenatal Down's syndrome testing**

Existing studies have generally suggested that factors which influence women's participation in screening are multifactorial, but could be grouped into demographic and psychosocial factors (Skirton and Barr, 2007; McNeill and Alderdice, 2009; Choi et al., 2012). Yet, other studies have reported on the influence of organisational context such as the

availability of a universal screening policy and health professionals' influence on women's decision-making. These are reviewed separately.

### **2.6.1 Demographic factors**

#### **2.6.1.1 Ethnicity**

Existing research using quantitative approach has shown women from ethnic minority backgrounds were less likely to participate in Down's syndrome screening compared to white women (Dormandy et al., 2005; Fransen et al., 2009; Fransen et al., 2010a).

Nevertheless, other studies using quantitative approach have not found any associations between ethnicity and uptake rates (Gidiri et al., 2007; Rowe, 2007; Alderdice et al., 2008; Rowe et al., 2008; Asongu et al., 2010).

Ahmed et al. (2012b) using Q methodology (multimethods) with 98 postpartum women recruited from 21 practices in England reported on the diversity of views expressed by women in general and within ethnic groups. The authors recommended that healthcare professionals should not have stereotypical beliefs about women from ethnic minority backgrounds. For example the assumption that, women from ethnic minority backgrounds are likely to decline screening based on religious or cultural beliefs.

#### **2.6.1.2 Socioeconomic factors**

Research using quantitative approach has shown that uptake rates of Down's syndrome screening is high among women that are socioeconomically advantaged compared to those with low socioeconomic status (Khoshnood et al., 2003a; Dormandy et al., 2005; Kuppermann et al., 2006; McNeill et al., 2009; Fransen et al., 2010b). Nevertheless, other research found that the socioeconomic status of women was not related to their attitudes to screening (Seror and Ville, 2009). However, the study involved questionnaire surveys of postpartum women in the Netherlands. The findings may be limited by recall bias.

Existing studies that involved questionnaire surveys conducted in the UK have been inconclusive about the association of screening uptake and socioeconomic status (Rowe and Garcia, 2003; Alderdice et al., 2008; Rowe et al., 2008). Prathapan et al. (2012) reported in a study conducted using population-based register of congenital anomalies in the North of England. The study found no evidence of socio-economic differences in uptake of Down syndrome screening in the UK. However, most of the previous studies were based on self-report from pregnant women and may be subject to socio-economic bias.

### **2.6.1.3 Educational level**

Women with a higher educational level have been reported to decline screening more often than women with less education in a study conducted in the Netherlands using survey questionnaires administered to 593 pregnant women with a response rate of 79% (Müller et al., 2006). Yet, a study by Stefansdottir et al. (2010) that adopted a quasi-experimental design with 237 (control) and 142 (intervention) pregnant women completing the questionnaires (95% response rate). The study found that women with higher education in Iceland were more likely to accept screening compared to those with less education. Similar finding was also reported by McNeill et al. (2009) using a survey methodology.

### **2.6.1.4 Religion**

Reasons for declining screening included opposition to TOP or a more fatalistic attitude of accepting an affected pregnancy as God's will, considering religious or moral beliefs (van den Berg et al., 2005a; Williams et al., 2005; Alderdice et al., 2008; Fransen et al., 2009; Reid et al., 2009; Fransen et al., 2010a). Women who believed children are gifts from God or nature were more likely to decline screening compared to women who saw children as the woman's right. Such women are more likely to consider the use of screening technology (García et al., 2008a). Howbeit, other studies have not found any strong associations between religion and uptake of Down's syndrome screening (Ahmed et al., 2006a; Ahmed et al., 2006b; Li et al., 2008; Usta et al., 2010; Ahmed et al., 2012a). The research by McNeill et al. (2009) conducted with 666 pregnant women in the UK (79% response rate) using survey methodology communicated that religious denomination was not significantly associated with screening uptake. Alsulaiman et al. (2012) research conducted using survey questionnaires administered to 100 women in Saudi Arabia, 222 UK white and 198 UK-Pakistani (response rate not stated). The study suggests that religious affiliation was not a strong indicator of attitudes towards diagnostic testing and TOP. The limitation of the study is that it explored non-pregnant women's hypothetical decision-making following prenatal diagnosis.

### **2.6.1.5 Maternal age**

Research has shown that different factors influence women's participation in screening (van den Berg et al., 2005a; Kuppermann et al., 2006; Crombag et al., 2013). For women below 35 years old, the factors were willingness to terminate an affected pregnancy and the influence

of healthcare professionals. Women who resent the interference of medical technology on pregnancy may reject the offer of screening. Older women (35 years or over) were influenced by the importance placed on information about the foetus. This may suggest that older women were more worried about their increased risk of having a child with the condition and are more likely to accept screening for decision-making as noted in previous work (Muggli et al., 2006; Tringham et al., 2011). In addition, women with a family history of genetic disorder may accept screening, because of their awareness of an increased risk of having a baby with genetic disorder (Skirton and Barr, 2007; Alderdice et al., 2008; McNeill et al., 2009; Morris and Alberman, 2010).

Recent research reputed that pregnant women expressed concern that age was an influence on their decision-making to accept screening (Aune and Möller, 2012). Furthermore, some of the women felt there was much negative focus on age. However, the findings are limited by the small sample of older women. Nonetheless, other studies have shown that older women, over problems with getting pregnant decline screening (Liamputpong et al., 2003). The meta-synthesis by Reid et al. (2009) posits that the reasons were mostly based on the possibility of diagnostic testing and the associated risk of miscarriage. The study by Reid et al. (2009) is a high quality meta-synthesis of studies on women's decision making about Down's syndrome screening.

#### **2.6.1.6 Parity**

Research using questionnaire surveys completed by 744 pregnant women (97% response rate) in the suggests multiparous women were less likely to accept screening than primiparous women (Shantha et al., 2009). The reasons given for declining were; no specific reasons, some women just did not want screening, some believed they were low risk, whilst others felt it was unnecessary. Yet McNeill et al. (2009) revealed in their study that involved survey methodology that more multiparous women compared to primigravida accepted screening in both hospitals studied. The association was, however, not statistically significant. Other studies using quantitative approach have found no association between gravidity, parity and uptake rates of screening (Dormandy et al., 2005; Nawaz et al., 2011; Tringham et al., 2011). All the studies were conducted in the UK, however, McNeill et al. (2009) was conducted in a culturally constrained context with limited availability of TOP services.

## 2.6.2 Psychosocial factors

### 2.6.2.1 Perceived parenting burden/reward

García et al. (2008a) study that conducted face to face interviews with 59 pregnant women who were randomly selected in a larger RCT that explored women's decision making about Down's syndrome screening. The author's suggest that women accepted screening, as they wanted to know and possibly prepare for the birth of a baby with Down's syndrome. This may be in view of their perceived ability to cope. Likewise, some women accepted screening, owing to their inability to cope with the physical, social, emotional and financial burdens of having a baby with Down's syndrome. Women who declined screening, felt society has a place for children with learning difficulties and can cope with the burden of taking care of the child as noted in previous work (Liamputpong et al., 2003; Chiang et al., 2006; Remennick, 2006; García et al., 2008a). Women with experience of disability did not seem to have different attitudes or perceptions of screening. The women felt that experiential knowledge of the condition did not influence their decisions about screening (García et al., 2008a). However, some women felt that the burden of a child with learning difficulties and society's negative attitude to disability had an influence on their decisions to screen (Gottfredsdóttir et al., 2009b)

Previous work suggests some women accepted screening to find out if the foetus has Down's syndrome in order to prepare for diagnostic testing and possible TOP (García et al., 2008a), whilst others accepted screening to prepare for the birth of a baby with Down's syndrome. Notwithstanding, some women declined screening as a result of their perceived ability to take care of a child with Down's syndrome and therefore, resigned to fate (Bryant et al., 2005; van den Berg et al., 2005a; Korenromp et al., 2007; Etchegary et al., 2008; Choi et al., 2012). The integrative review by Choi et al 2012 that describes the actual and hypothetical decision making by women following a prenatal diagnosis of Down's syndrome was well conducted with a well-defined search question and inclusion criteria, and attempts were made to identify all relevant published literature. However, how the included peer reviewed literature was critically appraised was not explained.

In addition, personal reward for taking care of a child with Down's syndrome has been reported as an influence on women's decisions to decline screening (Lawson, 2006; Gottfredsdóttir et al., 2009a; Lawson and Walls-Ingram, 2010). Nonetheless, the absence of

reward may cause women to accept screening for possible diagnostic testing and TOP if screen positive for Down's syndrome.

#### **2.6.2.2 Quality of life for a child with Down's syndrome**

Gottfredsdóttir et al. (2009a) reported in their study that involved pre and post screening interviews with 10 pregnant women and their partners that most women who declined screening did so on the basis of their perception that Down's syndrome was not a severe enough condition to warrant TOP. The limitation of the study is that the disability law in Iceland provides services for independent living of disabled individuals, which may have influenced the views expressed by the women. However, Hewison et al. (2007) employed questionnaire surveys in the UK, Ahmed et al. (2008) research involved 19 women in the UK using questionnaire surveys and interviews and Kelly (2009) interviews with 40 women in the USA suggest that women considered the severity of the condition when making testing and termination decisions. Therefore, women's perception of the quality of the life of a child with the condition was a factor in the decision-making process.

#### **2.6.2.3 Attitudes towards individuals with disabilities**

Personal experiences with individuals that have Down's syndrome have been associated with either declining or accepting screening for the condition (Etchegary et al., 2008; Gottfredsdóttir et al., 2009a; Crombag et al., 2013). On the other hand, research has shown a weak association between attitudes towards people with Down's syndrome and screening intentions (Bryant et al., 2010). The research involved 197 pregnant women administered survey questionnaires. The authors surmised that other normative variables such as external factors might have a strong influence on women's decision making. The assumption is that service organisation and delivery could influence women's decision-making processes. The low response rate (24%) limits the validity of the finding. However, the finding is supported by Potter et al. (2008) research conducted with 38 pregnant women in Canada who were interviewed post screening. The study showed that different factors in women's values and knowledge influence the process of decision-making producing different outcomes. Women may feel uncomfortable expressing their intention to terminate but in practice when faced with the diagnosis, most women choose to terminate (Mutton et al., 1998; Müller et al., 2006). Existing qualitative work suggests women have more positive attitudes about children with learning difficulties compared to male partners (García et al., 2008a). Anyhow, the majority of women in the quantitative study conducted by Skirton and Barr (2010) believed

that experiential knowledge of the condition did not influence their decisions about screening. Previous quantitative studies suggest attitudes may be a poorer indicator to behaviour (Hewison et al., 2007; Alsulaiman et al., 2012).

#### **2.6.2.4 Risk and anxiety**

Women accepted screening to overcome anxiety, in the best interest of the child and to gain reassurance about the well-being of their baby (Chiang et al., 2006; Etchegary et al., 2008; García et al., 2008b; Gottfredsdóttir et al., 2009b; Bryant et al., 2010; Crombag et al., 2013). Yet, other studies revealed that women decline screening over the anticipated stress and anxiety that may occur from screening (Markens, 1999; Remennick, 2006; García et al., 2008a; Reid et al., 2009).

The perception of being classified as low risk in terms of organisation of care, self-perception of being healthy, especially with no complications in earlier pregnancies and no family history of genetic diseases may influence women's decision-making processes (Liamputpong et al., 2003; McNeill et al., 2009; Reid et al., 2009). Though, women may be classified as high risk considering age, previous history of high risk results and family history of congenital anomaly. Such high risk status may influence women to accept screening for reassurance, preparation for outcome and decision-making.

Screening may be declined if women have high perceived risk of procedure related miscarriage (Skirton and Barr, 2007; McNeill et al., 2009). Additionally, uncertainty and anxiety were reasons for women declining screening as it was closely linked to miscarriages and TOP, therefore was not a personal option. Importantly, women who accepted screening may not have excluded TOP as a possibility in diagnostic testing, but did not directly link screening with TOP (Markens, 1999; Reid et al., 2009). This is illogical from an ethical context as it may result in women facing psychological stress if the result returns high risk.

The longitudinal surveys conducted in the Netherlands by Müller et al. (2006) explored women's attitude to NT screening using questionnaires administered to pregnant women at different stages in their pregnancy. The reasons for younger women's acceptance of screening were for reassurance, to prepare for outcome and decision making. Women aged 36 years or over accepted screening with the hope of avoiding diagnostic testing if lower risk. However, their reasons for declining screening were that they would not consider TOP, diagnostic testing, and any interference with the pregnancy or be faced with difficult choices. Recall bias is a limitation of the study.

Previous work reported that women's experiences with screening were associated with either declining or accepting screening (Santalahti et al., 1998; Carroll et al., 2000; Etchegary et al., 2008; Crombag et al., 2013). For example, negative experiences of family members or friends such as excessive anxiety and fear with screening may result in women refusing the test. Crombag et al. (2013) literature review was well conducted with inclusion criteria and the Anderson model of healthcare utilisation as the framework for the review. However, the included articles were not critically appraised. This should be taken into account during the interpretation of the findings.

Aune and Möller (2012) research conducted in Norway reported that screening caused anxiety especially for women faced with a choice that differed from that of their partners, family or society or if they just do not want to participate in screening. Moreover, some screen positive pregnant women maintained an emotional distance from the foetus. Others became emotionally connected with the foetus when they saw the scan photos and subsequently declined amniocentesis. Still, some women experienced feelings of guilt from thoughts of TOP owing to their advanced age especially with society's disapproval of older pregnant women.

#### **2.6.2.5 Ability to take action**

Existing studies using qualitative approach suggest women accepted screening, owing to the perception that it is morally responsible to engage in prenatal care, for mother and foetal wellbeing (Remennick, 2006; García et al., 2008a). Moreover, women accepted screening because they felt responsible to prevent the suffering and misery of a baby with a congenital disorder (Remennick, 2006; Hewison et al., 2007; Etchegary et al., 2008). Regardless, other studies using qualitative approach have demonstrated women decline screening, to ensure the birth of a healthy baby by avoiding the risks associated with invasive diagnostic testing (Markens, 1999; Liamputpong et al., 2003; Reid et al., 2009).

#### **2.6.2.6 Support from others**

The opinions and support of partners during decision-making for Down's syndrome screening have been shown in the literature to be important to whether women accepted or declined screening (Gottfredsdóttir et al., 2009a; Åhman et al., 2010; Aune and Möller, 2012). In addition, the qualitative studies also suggest friends and family also influence women's choices. However, a survey questionnaire study in Australia by Jaques et al.

(2004b) among women aged 37 years or over that reported their partners as having the most influence on their decisions found no significant difference in the partner's role in the group of women that accepted screening and those that declined screening. This finding was also reported by Choi et al. (2012) in their review.

#### **2.6.2.7 Views about termination of pregnancy**

Women decline screening over their moral views about abortion; they were against TOP on any grounds. Some women explained that the decision to terminate a pregnancy, over foetal anomaly should not lie with human beings. They felt women should accept whatever nature or God gives, as life is made of varieties and not of perfect human beings (Williams et al., 2005; van den Berg et al., 2005a; Heyman et al., 2006; Remennick, 2006; Etchegary et al., 2008; Shantha et al., 2009). Yet research has found that women who ruled out TOP based on their personal views accepted screening for reassurance or to confirm their low risk status (Heyman et al., 2006; Müller et al., 2006).

#### **2.6.2.8 Test characteristics**

Some women declined screening over concerns that the screening results were not definitive but a risk value (Kuppermann et al., 2006; Li et al., 2008; McNeill et al., 2009). However, some women valued and accepted the first trimester screening because of the earlier result which enable decision making (Kerr and Shakespeare, 2002; Williams et al., 2005; Hundt et al., 2008). It also gave women time to discuss screening with both partners and health professionals.

#### **2.6.2.9 Trust in the medical establishment**

Patients' trust on healthcare providers has also been illustrated as an influence on decision-making with women who trust their healthcare providers likely to accept the offer of screening (Kuppermann et al., 2006; Entwistle et al., 2008; Schwab, 2008; Reid et al., 2009). Further, women who distrust their providers are likely to decline the offer of screening. In addition, some women were strongly against the medicalisation of pregnancy and hence declined screening (Williams et al., 2002a; Hundt et al., 2008). Interestingly, the majority of women in a study that explored why Israeli women seek genetic testing did not object to excessive medicalisation of pregnancy (Remennick, 2006). The cited studies used qualitative approaches except Kuppermann et al. (2006) which was a prospective cohort study using questionnaires.

#### **2.6.2.10 Women's understanding of information about screening.**

Women were infrequently aware of the possible implications of screening, therefore, their decision-making about screening may not depend on their understanding of the information about screening (Markens, 1999; Heyman et al., 2006; McNeill et al., 2009; Reid et al., 2009; Seror and Ville, 2009). For example, some women accepted screening just to have a positive experience of prenatal care. Further, some women found it difficult to distinguish screening results from diagnostic tests, with a high risk result being seen as definitive of the condition (Lewando-Hundt et al., 2001; Pilnick, 2004; Pilnick et al., 2004). Hence, women declined screening, because they did not wish to have a diagnostic test.

Previous studies using interviews and conversational analysis of tape recorded consultations between midwives and women have also found women's understanding of screening questionable (Smith et al., 1994; Pilnick, 2004; Pilnick et al., 2004). Though, existing work using cross sectional survey questionnaires suggests that a majority of women claimed they understood the information about screening and it helped them to make informed decisions (Müller et al., 2006; Shantha et al., 2009). Interestingly, a systematic review by Green et al. (2004) reported that among women who believed their choices were informed, few women deliberated about the screening information before making their decisions. The review is a high quality study with minor flaws that have no effect on the relevance of the findings.

Furthermore, research has shown women's level of knowledge was not associated with the amount of information received (Ferguson et al., 1995; Ockleford et al., 2003; Rostant et al., 2003; Gourounti and Sandall, 2008). Ironically, women who stated that they received sufficient information did not have a higher level of knowledge than women who reported that they received too little information about screening. This demonstrates that the association between an individual's perception of the adequacy of given information and their understanding of information is positive, but not perfect. Individuals tend to exaggerate the adequacy of given information. Women's lack of full understanding of screening may reinforce the notion that screening is routinely conducted in the prenatal context. The study by Gourounti et al. (2008) was a descriptive, prospective questionnaire survey conducted in one hospital in Greece and the findings is also limited by the inability of the questionnaires to explore fully women experiences.

The literature so far reviewed on factors that influence women participation in prenatal testing revealed that the evidence that psychosocial and demographic factors are

responsible for the variation in uptake rates is inconclusive. Drawing a conclusion that psychosocial and demographic differences among women account for variation in uptake rates is therefore difficult. However, the differing findings may be due to the varying study design, sample, size, response rate, methodology and location of the included studies.

Lippman (1999) drawing on and synthesising a range of literature and expert opinion explained that the focus of rational, individualistic biomedicine to locate all influences on decision-making on the internal psychosocial and demographic attributes of women has been proven to be erroneous. The author suggests that there are powerful external structural and social factors that constrain informed choice. For example, the culture prevailing in the healthcare setting as regards the importance, reliability or sophistication of the available screening test has been shown to influence women's participation in screening (Pilnick, 2004; Pilnick, 2008; Tsouroufli, 2011). However, how the literature was searched, selected, synthesised and expert opinion sought by Lippman were not stated which is a flaw that affects the trustworthiness of the study.

## **2.7 Influence of service organisation and delivery on women's Down's syndrome screening choices.**

### **2.7.1 The influence of screening policy on screening uptake rates**

Variation in detection rates of prenatal Down's syndrome persists within and between countries with similar demography, attitudes and screening policy (Boyd et al., 2008; Crombag et al., 2013). This has been attributed to differences in the implementation of screening policy, non-availability of legal TOP services for foetal abnormality and differences in screening technology. Further, uptake rates vary between countries with different screening policies. For example, in the Netherlands before the introduction of the universal screening policy the uptake rate of Down's syndrome screening offered as part of a study was low compared to the UK with a universal screening policy (Skirton and Barr, 2007). Existing research indicates that uptake rates vary between healthcare settings with different screening policies (Lewando Hundt, 2004; Skirton and Barr, 2007; Tsouroufli, 2011).

Interestingly, the introduction of the universal screening policy to the Netherlands in 2007 has not led to an increase in uptake rates. Work by Bakker et al. (2012) that involved questionnaire surveys completed by 820 pregnant women (73% response rate) in two regions in the Netherlands suggests the policy of reimbursement undermined equal access

to screening. Women 36 years or over have free access to screening whilst younger women have to pay. The authors surmised this created a perception that screening in younger women was needless. In addition, women are first presented the opportunity to receive information about screening. The women have to consent, before the actual information about screening is given. Similar findings have been reported in Schoonen et al. (2012). The policy of reimbursement and the right not-to-know limits the transferability of the findings of this study to the UK NHS context.

McNeill et al. (2009) study in NI that involved survey methodology with open ended interviews questions explored women's reasons for accepting or declining serum screening. The authors reported similar low uptake rates of Down's syndrome screening in two hospitals with different screening policies. Hospital one, had an uptake rate of 26% with Down's syndrome screening offered to all pregnant women. Hospital two, with an uptake rate of 28%, offers screening to women aged 35 years or over and women with a family history of the condition. The authors suggest that the low uptake rates in the two hospitals may be due to the cultural and societal contexts of screening. The contexts may constrain midwives to present screening from a negative perspective as noted in McNeill and Alderdice (2009) study conducted using an ethnographic approach. The inference is that women may often not ask the difficult questions about the implications of screening. Therefore, the societal and cultural contexts encouraged collusion between midwives and women to decline testing, in order to avoid the complex negotiation involved in screening decisions. However, the non-systematic consecutive recruitment of women in the settings may have introduced a bias.

The study conducted in the Netherlands by Müller et al. (2006) using a questionnaire survey reported on the relationship between local Down's syndrome screening policies and decision-making for screening. Women reported accepting policies they were familiar with and were offered by health providers, as it must be for the well-being of mother and baby. This is consistent with the research by Seror and Ville (2009) also using questionnaire surveys, reported that women in the Netherlands tend to accept the local care pathways offered to them. Therefore, the absence of a policy on screening for Down's syndrome screening may reflect it was not an important part of prenatal care with a consequent low uptake rate. The absence, therefore, reflects a considered decision which to service providers and users is 'a policy' in itself. Further, it has been suggested in existing research that the uptake rate of Down's syndrome screening is usually high in countries where it is

routinely offered to all pregnant women (van den Berg et al., 2005b; McNeill et al., 2009). The ramification of these findings is that the local care pathways have an influence on women's decision-making processes but this has not been investigated comprehensively in the NHS context in England.

Ghosh et al. (2011) reported in a conference abstract on the addition of a new service (first trimester combined test) to a site previously offering only the second trimester triple screening test. The result showed an increase in uptake rates of Down's syndrome screening during the first year of introduction. The reason for the increase was attributed to the implementation and access of the local population to the 'best screening programme'.

Further, this trend was reported in a study by Tringham et al. (2011) using hospital records in a region in the North of England. The study indicates the addition of a new service, the combined screening test, led to an increase in uptake of Down's syndrome screening during the first year, in a population that was experiencing a declining uptake. Previous research also using hospital records in the north of England has also reported on the increase among age groups that were experiencing a decline in uptake rate before the introduction of the new service (Gidiri et al., 2010). It is not clear if the increase was because the results allowed early decision-making or the enhanced reliability of the test was responsible for the increased uptake. The authors surmised that the factors responsible for the increase in uptake are unknown as the reasons for accepting or declining screening are multi-factorial. However, how confounders and bias were controlled was not stated and human error in data input may be limitations of the study.

### **2.7.2 The influence of organisational context on screening uptake rates**

Previous work by Dormandy et al. (2002b) explored variation in uptake of serum Down's syndrome screening in settings that offer screening as part of the routine visit and those that required a separate visit. The study used a prospective descriptive design with the multidimensional measure of informed choice questionnaire to describe the rate of informed choice in two hospitals, one where screening was conducted at routine visit and the other at separate visit. The authors reported that the high uptake rate seen in the routine visit hospitals may reflect convenience or a less considered decision. Whilst the low uptake rate associated with the separate visit may reflect a physical barrier to testing such as making arrangements for childcare, absence from work or the choice was considered more carefully. The implication of the study is the context in which screening options are offered

should be considered as well as the information provided but contextual factors have not been fully explored in the NHS in England. The limitations of the study are that it involved only two settings and the questionnaires were completed earlier in the separate visit setting. The attitudes and behaviour may have been assessed closer in time in the routine visit setting.

Furthermore, Dormandy et al. (2002a) that involved questionnaire surveys completed by 28 screening representatives found that the rate of informed choice to accept screening was higher when the test was conducted as part of a routine visit than as a separate visit. In the separate visit hospital, women with positive attitudes to undergo the test did not turn up for screening. This inconsistency was greater in the separate visit hospital compared to the routine visit hospital. The author surmised that the reason may be the physical barrier to screening such as competing demands at the time of the separate visit or women simply forgot about the appointment. The limitations of the study are the audit and questionnaire were not collected concurrently and the questionnaires were not validated.

Large variations in uptake rates of screening between hospitals which were not related to the influence of health professionals on women's decision-making has been noted in a study by Dormandy and Marteau (2004). The study employed survey questionnaires administered to 89 health professionals and medical records of women's uptake of Down's syndrome screening in two hospitals conducting screening at routine and separate visits. The authors hypothesised that at the organisational level, midwives with positive attitudes may conduct screening at the same routine booking visit and midwives with negative attitudes may require a separate visit. Therefore, screening may be organised in such a way to facilitate take-up of screening by women who are undecided or passively engaged with the process of screening. At the individual level, health professionals with positive attitudes may present the positive aspects of screening and those with negative attitudes may highlight the negative aspects of screening. However, the authors found that midwives in the high uptake hospital had more positive attitudes compared to midwives in the low uptake hospital, but found no significant association between attitudes of health professionals and women's uptake of screening. The limitations of the study are that health professionals may have responded differently as they would in practice, the study was design to detect a correlation of 0.5 or greater and the number of midwives seen by each woman cannot be verified.

Moreover, Dormandy et al. (2006) cluster randomised control study found that the rates of informed choice in women were similar in both the routine and separate visit groups. The authors suggested that there is no evidence to support the study hypothesis that more women make informed choice when Down's syndrome screening is conducted at routine visits compared to separate visits. The limitations of the study are that the pre-screening completion of questionnaires by women may have influenced the uptake of screening and some women completed the questionnaires after randomisation. The work by Dormandy et al was conducted before the introduction of universal screening in the UK

Studies using qualitative approaches have reported that the time available to give women information about screening is inadequate (McNeill and Alderdice, 2009; Tsouroufli, 2011). The time pressure may affect pregnant women and their partners' ability to make choices that are consistent with their values and beliefs. Further, the organisation of care within the maternity services by midwives has been shown to disempower women in a systematic manner. This is achieved by midwives' task oriented care as a result of heavy workload, concern with getting the job done and the manner of language used. The idea is often to get the job done as quickly as possible (Porter et al., 2007; McNeill and Alderdice, 2009; O'Connell and Downe, 2009). The mechanistic delivery of care in the prenatal context is untenable from an ethical context.

### **2.7.3 The influence of health professionals on women's participation in screening**

Existing studies have reported that the personal opinions and attitudes of health professionals may influence women's decision-making about screening (Williams et al., 2002b; Farsides et al., 2004; Pilnick, 2004; Lewis et al., 2006b; Pilnick, 2008; Gottfredsdóttir et al., 2009b; McNeill et al., 2009; Park and Mathews, 2009; Tsouroufli, 2011; Bakker et al., 2012; Hertig et al., 2013). Women claimed that the covert and overt directiveness of health professionals had an influence on their decision-making processes about screening. For example, health professionals have been shown to direct women and partners' decision-making, both by appearing to assume that testing will take place, recommending screening and through selective information-giving. Nevertheless, the finding by Bakker et al. (2012) using questionnaire surveys which suggests that midwives recommended combined screening test to women in the Netherlands was not statistically significant.

Further, the research of Tsouroufli (2011) using observations (taped recorded consultations) and questionnaire surveys suggests that 67% of women in the study stated that clinic staff

were in favour of the screening test, this was not surprising as the uptake rate of first trimester screening was 95%. What was surprising was that the majority of the women reported they made the decisions based on their moral values and beliefs as previously noted in Williams et al. (2005). However, research by Paul (1998) cited in Seavilleklein (2009), Dormandy et al. (2002a), Dormandy et al. (2006) and Skirton and Barr (2007) suggests that women may make decisions that are at variance with their attitudes, beliefs and values owing to the influence of health professionals. The meaning is that women's acceptance of Down's syndrome screening may not be consistent with their beliefs and values.

Previous research by McNeill et al. (2009) reported that midwives influence women who accepted or declined screening in a study conducted in a context of low uptake rate, ad hoc policies (lack of a universal screening policy) and limited availability of TOP services.

Moreover, previous work by Pilnick (2008) and McNeill and Alderdice (2009) suggest that midwives are supportive of making sure women have choices about screening but did not seem to carry through in practice.

However, other studies using quantitative approaches have found no evidence that midwives' attitudes influence women's choices and decisions (Dormandy and Marteau, 2004; van den Berg et al., 2007; Bakker et al., 2012). Nonetheless, Dormandy and Marteau (2004) maybe limited, as it was powered to detect a large effect size and van den Berg et al. (2007) was conducted in the Netherlands where screening was at the time of the study, not routinely offered to all pregnant women and younger women (<36 years) pay for the services.

#### **2.7.4 Routinization of prenatal Down's syndrome screening**

Existing studies have reported that ultrasound scans are now seen as a routine part of antenatal care. It is eagerly seen by pregnant women and their partners as an opportunity to see the baby (Pilnick et al., 2004; Williams et al., 2005; Seavilleklein, 2009; Åhman et al., 2010). The indication is that the offer of first trimester combined screening made in the context of an ultrasound scan may be accepted by women as a routine procedure and to visualise the baby without thinking through the implications of the scan.

Research conducted in Iceland reported that women felt because screening was non-invasive, they did not really think about it. In addition, they described screening as part of the routine tests in pregnancy, and explained they made a choice to have it (Gottfredsdóttir

et al., 2009b). Indeed, the offer of screening by health professionals as a choice may be done at the expense of important information about the implications of screening. Other qualitative studies conducted in the US by Markens (1999) and in Taiwan by Chiang et al. (2006) have suggested that screening is perceived by most women as a routine test, as it was part of the prenatal care package that is offered to all pregnant women in the health care system. Therefore, some women may not perceive screening as optional. Interestingly, research in the UK has reported that few women were concerned that declining screening may be a potential barrier to accessing other services offered during prenatal care (Heyman et al., 2006; Reid et al., 2009).

Conversational analysis of data obtained from tape recorded women's consultations with midwives in the UK indicates little involvement of women in the discussions which reflect that screening was not understood as a choice even though the midwives made it known that the decision lies with the woman (Pilnick, 2004; Pilnick, 2008). Expert authority was invoked to stifle participation, so that when screening was offered as a choice, women did not perceive it as such. Consent was often assumed by the midwife rather than allowing an independent choice. Further, the offer of Down's syndrome screening is made with other routine blood tests at the booking visit, with some midwives recommending screening to women as 'the best' test. The significance for practice is that some women may bypass the decision-making process, as screening may be perceived as a routine test. The limitation of the studies by Pilnick is that they were conducted before the introduction of universal Down's syndrome screening policy in the UK.

Existing research conducted in Israel using qualitative approach suggests the offer of screening in the prenatal setting was seen by some women as an endorsement by healthcare professionals (Remennick, 2006). Additionally, many women have trust in the medical institution which may influence their decisions to accept screening (Heyman et al., 2006). Other studies conducted in the USA and Israel found that some women decline screening because of their mistrust of healthcare professionals and the medical institution (Kuppermann et al., 2006; Remennick, 2006).

Health professionals' ability to interpret technical and statistical information when informing women about screening may influence women's decision-making processes (Markens, 1999; Liamputpong et al., 2003; Williams et al., 2005; Chiang et al., 2006; Remennick, 2006; Nagle et al., 2009; Reid et al., 2009). It creates a power differential between women and health

professionals' knowledge of screening. This may result in women depending on the expert knowledge of health professionals with the consequent impairment of women's decision-making process as noted in previous work.

Tsouroufli (2011) reported on a study conducted in an innovative site offering NT screening using data from 57 tape recorded consultations involving a mix of midwives and healthcare assistants (HCAs) with women in the UK. Questionnaire surveys were also employed to explore the information-giving process and the uptake rates in the one stop clinic. The aim of the study was to explore routinisation and constraints on informed choice in the context of first trimester screening. The method of data analysis was a collaborative team approach through discussion and also to obtain inter-rater reliability. The study found midwives approached the offer of screening with optimism and presented the condition as unlikely or having a small risk of occurring in pregnancy. Time pressure was an issue at booking with midwives and HCAs providing minimum to no information about the implications of screening, thereby reinforcing the normality of pregnancy. Women found it easier to opt-in than to opt-out of screening, as it was perceived to be an integral part of care, health professionals encouraged screening and there was a workplace expectation for women to conform. The author surmised that directive information and the absence of purposeful discussion with pregnant women constrained the offer of screening for prenatal Down's syndrome.

Similar findings have been previously reported in existing studies (Pilnick, 2004; Williams et al., 2005; Pilnick, 2008; McNeill and Alderdice, 2009; Park and Mathews, 2009; Seavilleklein, 2009; Bryant et al., 2010). Tsouroufli (2011) suggests that midwives acting relationally, as well as in a less constrained environment would improve the care and quality of information received by pregnant women. The findings by Tsouroufli are relevant but transferability is limited as the combined screening was offered at this site as part of research where women received the result within an hour and HCAs were involved in offering screening to women. In addition, the method of data analysis used may not be ideal for tape recorded consultations.

## **2.8 Decision-making in the prenatal Down's syndrome screening context**

Bekker (2010) work based on the conceptual review integrating the science behind individuals' decision making with the demands of designing complex, healthcare

interventions explored whether checklists damage the quality of informed choice interventions. The study suggests patients employed heuristic and/or systematic strategies to attend to information in order to make screening or treatment decisions. Heuristic strategy is an easy way of handling information by subconsciously making decisions with little attention to details. The deliberate, conscious and comprehensive attention to details or all options available is a systematic strategy in decision-making. Factors affecting these strategies are values, beliefs, experience (internal factors) and external factors such as information, events and the views of others. The author surmised that women may have difficulties separating decisions from the process of care. The implication for screening programmes is the need to consider other factors such as the influence of service organisation and delivery on women's decision-making processes.

The study by Aune and Möller (2012) explored the experiences of ultrasound risk assessment for chromosomal anomalies of women in Norway who had accepted first trimester screening. It involved 10 women interviewed twice (19 interviews) and the transcripts were analysed using Grounded Theory. The study found that women wanted to participate in screening but experienced difficulties with decision-making which constrained their choice. Some women demanded advice and guidance from health professionals. Whilst others felt the choices available with a high risk result were complicated, with some women reporting it was best not to know and hence, declined screening. The ramifications of these findings are that women consider diagnostic testing when deciding about screening and the decision-making processes create psychological stress.

The involvement of the health professionals in women's decision-making has been shown in existing research to result in three models of decision-making. The models may be used to describe the level of involvement of women in decision-making. The models are the paternalistic model in which the midwives give information about screening, but the midwives' preferences take precedence over that of the women's. In shared decision-making there is a two way information exchange where the beliefs and values of the woman are taken into consideration with the midwife participating in all stages of the decision. In the informed choice model the information is given objectively and nondirectively by the midwives with the decision made strictly by the woman (Carroll et al., 2000; Chiang et al., 2006; Park and Mathews, 2009; Hertig et al., 2013). However, the qualitative studies focused mostly on consultations involving GPs and pregnant women and were conducted in Canada, Taiwan and in Switzerland limiting their transferability to the UK NHS context.

## **2.9 Purpose of the study**

In order to shed light on pregnant women and their partners' participation in screening and how decisions about screening are shaped by the service contexts, there was a need to explore the decision-making processes involved in the offering of screening. It is within the service contexts that decisions about screening are made or initiated by women and their partners during the prenatal experience. It is within this environment that participation in screening must be understood and explained. A multiple case study design involving document review, online interviews using vignettes, open-ended questions and prompts were used to explore the influence of service organisation and delivery on women and their partners' participation in screening. The rationale for the case study was to have an extensive understanding of the influence of service organization and delivery on the decision-making processes from the perspectives of midwives, women and their partners in two settings with different uptake rates.

### **Aim**

To explore the influence of service organisation and delivery on women and partners' participation in screening in two settings with different uptake rates which may help explain the variation in uptake rates of screening.

### **The objectives of the study were to explore:**

1. How the structures of maternity services influence women and partner's participation in prenatal screening
2. How the process of prenatal Down's syndrome screening influence women and partner's participation in the programme.
3. The influence of community midwives on women and partner's participation in prenatal screening for Down's syndrome.

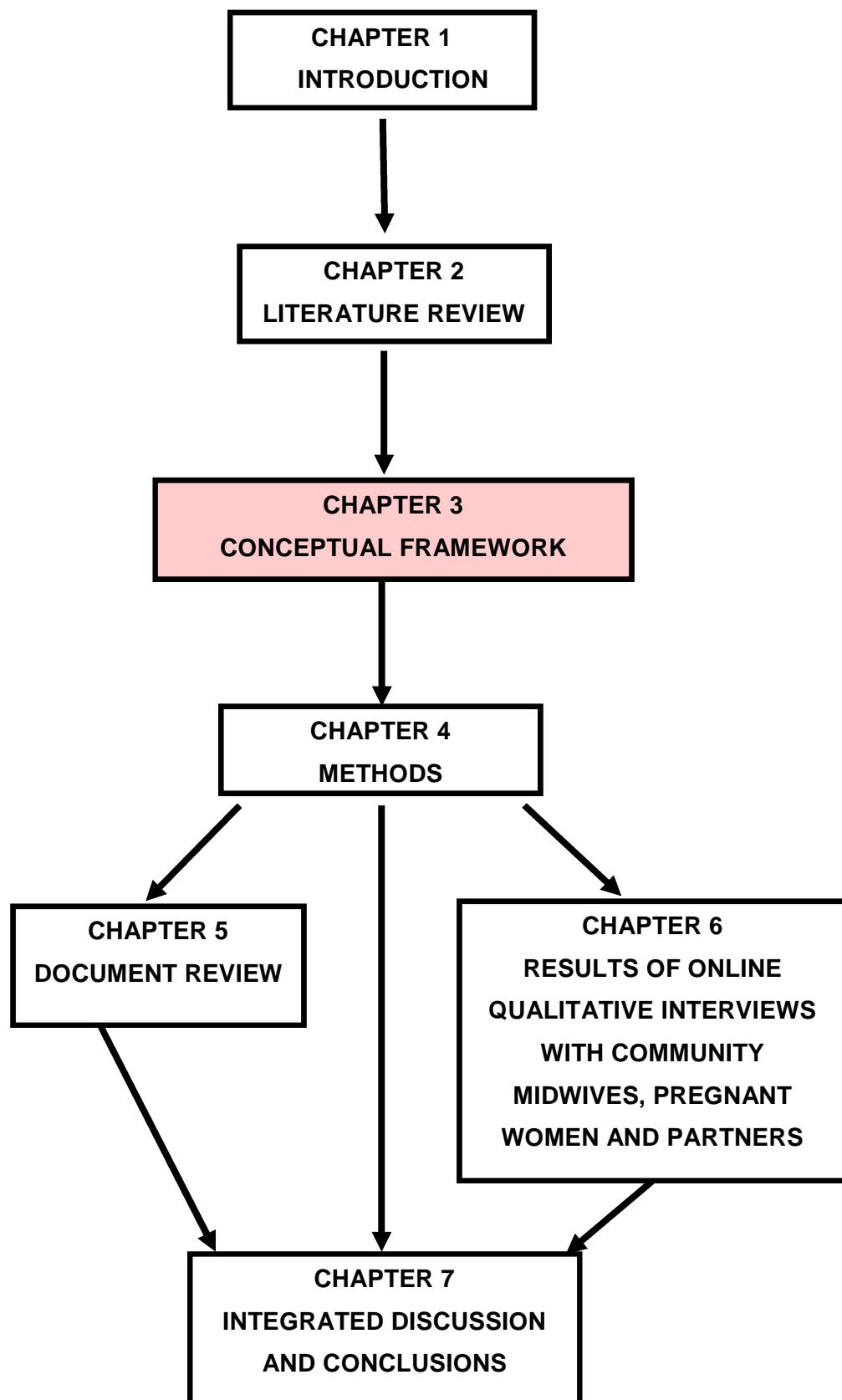
## **2.10 Chapter summary**

The review synthesised diverse methodological studies to produce a number of recurring subthemes and themes that appear to illuminate the gaps in the literature. The evidence that psychosocial differences among women may account for the variation in uptake rates is inconclusive. The review revealed that the service organisational and delivery contexts of

Down's syndrome screening may influence participation in Down's syndrome screening and account for the variation in uptake rates. However, this has not been comprehensively investigated in the NHS context in England. The review indicates that qualitative methods using an inductive approach such as interviews and focus groups were most suitable in exploring the experiences of health professionals, pregnant women and their partners including the external and internal constraints encountered in prenatal testing. The reasons may be that qualitative interviews and focus groups allowed for the expression of views of participants and the analyses are grounded in their perspectives.

van Teijlingen et al. (2003) Hunter (2006) and McLachlan et al. (2009) revealed that researching women's experiences of maternity care is becoming difficult and is complicated by women's reluctance to criticise midwives or healthcare staff. In addition, Reed (2009a) and Reed (2011) reported on the difficulties recruiting midwives and male partners for research in the maternity services in the UK. To overcome these difficulties, studies are using the internet as a data collection tool in the maternity services. Skirton and Barr (2010) survey research and Williams et al. (2011) qualitative interviews used online methods to explore midwives, women and their partners' experiences of prenatal testing in the UK.

The need for research on the influence of healthcare settings on women's participation has been identified by existing studies (Press and Browner, 1997; Dormandy et al., 2002b; McNeill and Alderdice, 2009). A national workshop on revisiting 'Choice' held in the Leeds Institute of Health Sciences, University of Leeds on 19.11.10, brought together experts in the field of prenatal Down's syndrome screening. Stakeholders spoke of the influence of service organisation and delivery on women's participation which may challenge the current emphasis on non-directional counselling. Additionally, research on the factors responsible for the differences in take-up of screening in settings with high and low uptake rates and first trimester combined screening policy was not found.



## CHAPTER 3: CONCEPTUAL FRAMEWORK

### 3.1 Introduction

Chapter Two provided a review of contemporary literature on the influence of service organisation and delivery on women and their partners' participation in prenatal Down's syndrome screening. This chapter will focus on the Donabedian Healthcare Organisation and Delivery conceptual framework adopted in this study. The literature review in chapters 1 and 2 revealed that, whilst the influence of service organisation and delivery on women's choices in clinical settings has been mentioned, the emphasis has been on women making autonomous, rational and individualistic choices. Besides, the policy guiding the offer of prenatal Down's syndrome screening is informed by the biomedical rational choice model which views women's autonomous and informed decisions as free from external influences or coercion. Little attention has been paid to the ways in which service contextual factors inherent in prenatal screening, influence women and their partners' decision-making processes. Missing from the literature are studies that investigated the influence of service organisation and delivery from the perspectives of frontline service providers and users in two settings with different uptake rates using a health service conceptual framework.

### 3.2 Conceptual framework for this study

**Figure 2: The Donabedian Healthcare Organisation and Delivery Model.**  
(Shojania et al., 2007).



The Donabedian healthcare organisation and delivery model (Donabedian, 1980; Donabedian, 1988) is one of the most frequently used models to evaluate the quality of healthcare services including nursing and midwifery services (Hulton, 2000; Morestin et al., 2010). Hulton (2000) adapted the Donabedian model to develop a new model for evaluating

the quality of maternity services in developing countries. The Donabedian model typically categorises service providers and users, services provided, service environment and outcome variables according to structure, process and outcome. This model assumes a linear relationships between structures, process and outcome variables (see figure 2). Structures denote the attributes of the settings in which service or care is provided. This includes the physical setting, such as the facilities and equipment available, and the care environment, such as the ambience of the consulting rooms where women are consented for screening. It also includes the health professional's attributes, such as professional competence, interpersonal skills, commitment to the role and personal characteristics. The others are organisational issues such as time, skill mix, staff role, the number and qualification of staff, policies and guidelines, staff attitudes and types and volume of caseload.

Process refers to activities involved in giving and receiving care. It includes a range of healthcare activities that make up caring as perceived by patients such as providing information, reassurance, showing concern and respect. The others are having a sympathetic presence, getting to know the patients and taking time to discuss issues with them. Further, it involves factors that affect the quality of interaction between midwives and women (McCabe, 2004; McCormack and McCance, 2006). For example, in the prenatal context, it refers to how healthcare providers deliver Down's syndrome screening or what is actually done in giving and receiving information about screening. This includes the offering of Down's syndrome screening and the processes of decision-making.

Outcomes in biomedical care denote the result of care on the patient's quality of life and satisfaction. However, patient satisfaction is a poor indicator of medical service quality, as some users may be satisfied with poor care. Moreover, quality outcomes such as patient attitude and satisfaction are difficult to measure, because they are not clearly defined. Factors other than medical care may influence outcomes, hence its limitation for assessing quality of medical care. Existing studies suggest both patient and physician factors influence the quality of care rendered in healthcare settings (Peterson, 1963; Moorehead, 1964) cited in Donabedian (2005). Therefore, the emphasis must shift away from the preoccupation of evaluating quality and concentrate on the distinct elements of structure and process to understand the medical care process itself (Donabedian, 2005).

The outcomes of this thesis are the influence of service organisation and delivery on women and partners' participation in screening (see figure 3, page 64). Therefore, this thesis

explored the perspectives of midwives, women and partners that are related to understanding how structures and processes in prenatal Down's syndrome screening influence participation in screening. This led to a consideration of the key drivers in structures and processes that can be expected to influence women and partners' decision-making process. The Donabedian framework is adapted in this study to classify service organisation as structures and service delivery as processes in the prenatal context. The influence of service organisation and delivery are the outcomes of the framework (see table 1, page 65 and figure 3).

### **3.2.1 Structures**

Structures refer to the service organisational context. The literature has reported on how the organisational context of the maternity services influenced pregnant women and their partners' participation in Down's syndrome screening (McNeill and Alderdice, 2009; Tsouroufli, 2011), but the influence of the physical environment (where consent is obtained) on women's choices has not been fully investigated. An objective of this study, therefore, is to explore the influence of the environment (ambience) of the prenatal context on women and their partners' participation in screening. The Donabedian framework on structures involved the attributes of the settings where services are provided. These include the physical attributes such as facilities and equipment, human resources such as the number, and qualifications of midwives (refer to table 1). Hence, the dimensions involved in structures based on the Donabedian framework may be grouped into social or professional, organisational and physical environmental factors according to Smith et al. (2008). The specific contextual factors explored are the social or professional factors, such as the presence of midwives, and the physical environmental factors, such as the room layout, décor, temperature, lighting and medical equipment in the consulting rooms of antenatal clinics where women are consented for screening. These factors are classified as the prenatal environment in which women and partners make choices about participating in Down's syndrome screening. The literature is not clear on how the social or professional context such as the presence of a community midwife in the consulting room, influences women and their partners' choices and decisions. Previous work indicate the social status and observable characteristics of midwives could influence the social environment before the midwives say or do anything (Bandura, 1986). In addition, research revealed that the social context has an influence on clinical decision-making (Lerrick, 2008; Smith., 2008).

Existing studies have shown that the attitudes of midwives influence women and partners' choices and decisions (Williams et al., 2002b; Farsides et al., 2004; Lewis et al., 2006b; McNeill and Alderdice, 2009). However, other studies have found no evidence that midwives' attitudes have any influences on women's choices and decisions (Dormandy and Marteau, 2004; van den Berg et al., 2007; McNeill and Alderdice, 2009). The attitude of the midwife, an attribute of structure in the adopted Donabedian framework, has a linear relationship with process. Based on the framework and the inconclusive evidence from previous research, the influence of midwives' attitudes on women and partners' participation in screening was investigated in this thesis (see table 1).

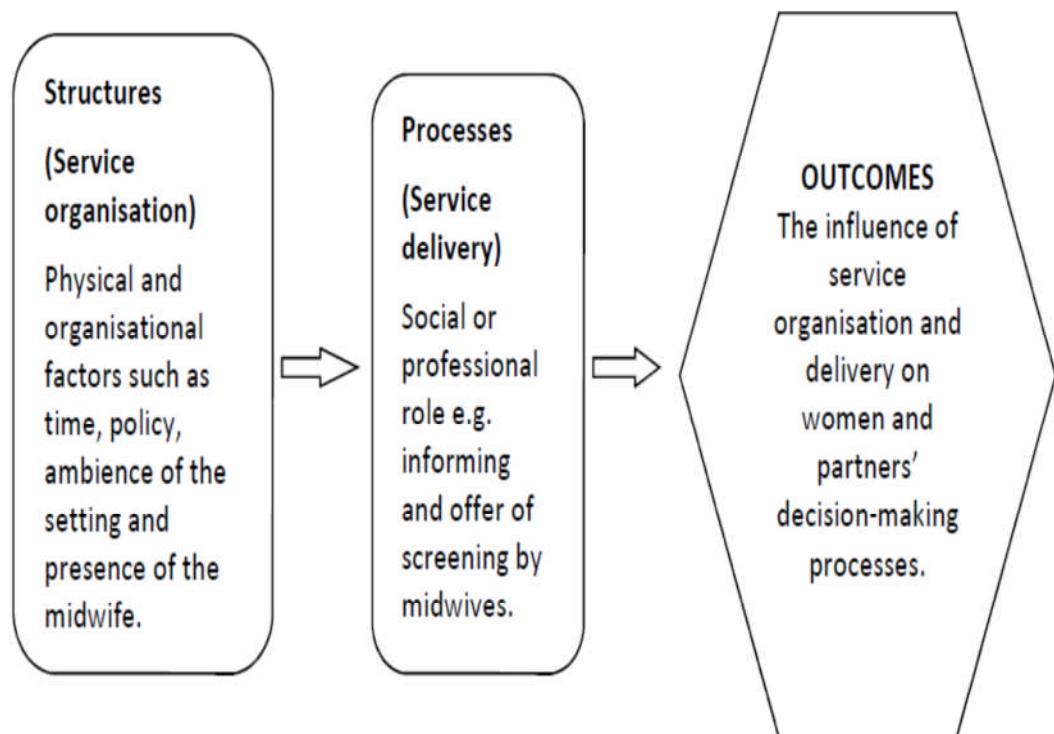
### **3.2.2 Process**

Processes refer to service delivery context, which includes the activities of providing services and the resources involved or used in the activities (Shojania et al., 2007). Therefore, process is what is done in prenatal Down's syndrome screening, by whom and how it is done. Process refers to services offered, test characteristics and quality, interpersonal relationships, information-giving and receiving, involvement of women and partners' in decision-making (see table 1 and figure 3). It includes the provision of privacy, confidentiality and the same standard of care to all women. In addition, it involves emotional support, respectful treatment and adequate contact time with midwives. This is care that meets the information, emotional and physical needs of women and partners in a way that is consistent with their values and beliefs. The role of information exchange is crucial to enabling women and their partners to make informed choices. It involves appropriate timing, understanding, quality and quantity of information. There is concern that the process of information-giving about prenatal Down's syndrome screening may affect whether or not women and partners clearly understand what is offered, why and any specific instructions. Thus, women and their partners may not be given the opportunity to discuss, ask questions, and understand that screening is optional and the implications of participating in screening. This may affect their decision-making process about the screening test. Previous studies suggest when screening is offered in a routine context, women and partners may perceive the offer as a routine test or a recommendation by health professionals to accept screening (McNeill et al., 2009; Tsouroufli, 2011).

### 3.2.3 Outcomes

The outcome in this thesis using the Donabedian Healthcare Organisation and Delivery Model is the influence of service organisation and delivery on women and partners' participation in screening. This is not an outcome of screening decisions but attributes of the process of decision-making, as it was not important in this thesis whether women accepted or declined screening. However, the exploration of the influence of service organisation and delivery on women and partners' participation in screening may give some insights into the screening decision outcomes.

**Figure 3: The adapted Donabedian Healthcare Organisation and Delivery Model.**



**Table 1: The study's adapted Donabedian conceptual framework; criteria, Indicators, Data collection and Tools.**

<b>Research Question</b>	<b>Objectives</b>	<b>criteria</b>	<b>Indicators</b>	<b>Data collection</b>
Do the structures, processes and people in prenatal settings influence women and partners' participation in screening for Down's syndrome?	1. To explore whether the structures of the maternity services influence women and partners' participation in prenatal screening for Down's syndrome.	Surroundings (ambience), facilities, equipment, staff, training, provider role, supervision, amenities, policies and guidelines.	Layout of clinical environment, ventilation or poor air quality, lighting, temperature, availability and accessibility of services and facilities, number and qualifications (staff), quantity, content and attendance (training), knowledge, attitudes and skills, information leaflets for women, care plan, antenatal screening policy/protocol (Down's syndrome screening policy), waiting time for services	Policies/guidelines and care pathways review. Pilot of web-based interviews and discussion forum with coordinators (interviews only), community midwives, pregnant women and partners.  Main online data collection method using open-ended interviews with vignettes and prompts involving community midwives, pregnant women and partners.

	2. To explore whether the process of offering prenatal Down's syndrome screening influences women and partners' participation in the programme.	Services offered Technical quality of services Promotion of continuity of care  Information exchange Good interpersonal relations	Quality of screening tests, involvement of women and partners' in decision-making, information-giving and receiving, emotional support, provision of privacy and confidentiality, same standard of care to all women, respectful treatment and adequate contact time with midwives.	Policy/guidelines and care pathways review  Pilot web based group discussion and interviews with midwives, pregnant women and partners  Main online data collection method using open-ended interviews with vignettes and prompts involving community midwives, pregnant women and partners.
	3. To explore whether community midwives influence women and partners' participation in prenatal screening for Down's syndrome.	Midwife's attitude	Attitude to screening tests and role.	Pilot web based group discussions and interviews with community midwives, pregnant women and partners.  Main online data collection method using open-ended interviews with vignettes and prompts involving community midwives, pregnant women and partners.

### **3.3 Other conceptual frameworks considered**

The health belief model (HBM) (Rosenstock, 1974; Lawson and Pierson, 2007), theory of reasoned action (TRA) (Fishbein and Ajzen, 1980) and the theory of planned behaviour (TPB) (Ajzen, 1991) have been shown to be good predictors of a range of health-related behaviours (Armitage and Conner, 2000; Ajzen, 2001; Bryant, 2003). Nevertheless, the sense of ambivalence associated with women's attitudes and behaviour on their screening choices and decisions highlighted in the literature review points towards the inappropriateness of the TRA/TBP for examining women's decision making process. The TRA/TBP models have been criticised for not incorporating factors such as prior behaviour and experience known to affect an individual's actions. Prior behaviour or experience is relevant in the prenatal testing context as women who accepted Down's syndrome screening in an earlier pregnancy may likely accept it again in a subsequent pregnancy (Rausch et al., 2000; Bryant, 2003). Moreover, the TRA/TBP theories also do not take into account the influence of the social, organisational, cultural and physical environmental contexts in which individuals and their behaviours are located in the prenatal testing situation (Conner and Sparks, 1995; Bryant, 2003; Lawson and Pierson, 2007).

The alternative models also assumed that a conscious, rational intention to act in a certain way precedes actual behaviour. For example, the models assume that women and partners will carefully consider the various options based on their beliefs and values and decide on an option that will maximise benefits and minimise the risks of screening. Thus, the models are based on the premise that women are able to make decisions or complete mental tasks autonomously in a logical manner. Research has shown that women are more vulnerable in pregnancy than when they are not pregnant (Farrell et al., 2011). This is because previously memorised coping strategies are often not effective which may affect their decision-making process during antenatal screening and testing as noted in Brudal (2000) cited in Aune and Möller (2012). In addition, women do not make screening decisions in isolation. Women make decisions within a service context which may influence their decision-making processes and constrain informed and autonomous choice. The TRA/TPB and HBM theories also tend to focus more on the outcomes of decision-making, rather than the process of decision-making (Reid et al., 2009). It remains doubtful if women can fully engage in the biomedical model of informed choice. Therefore, the use of the biomedical models of

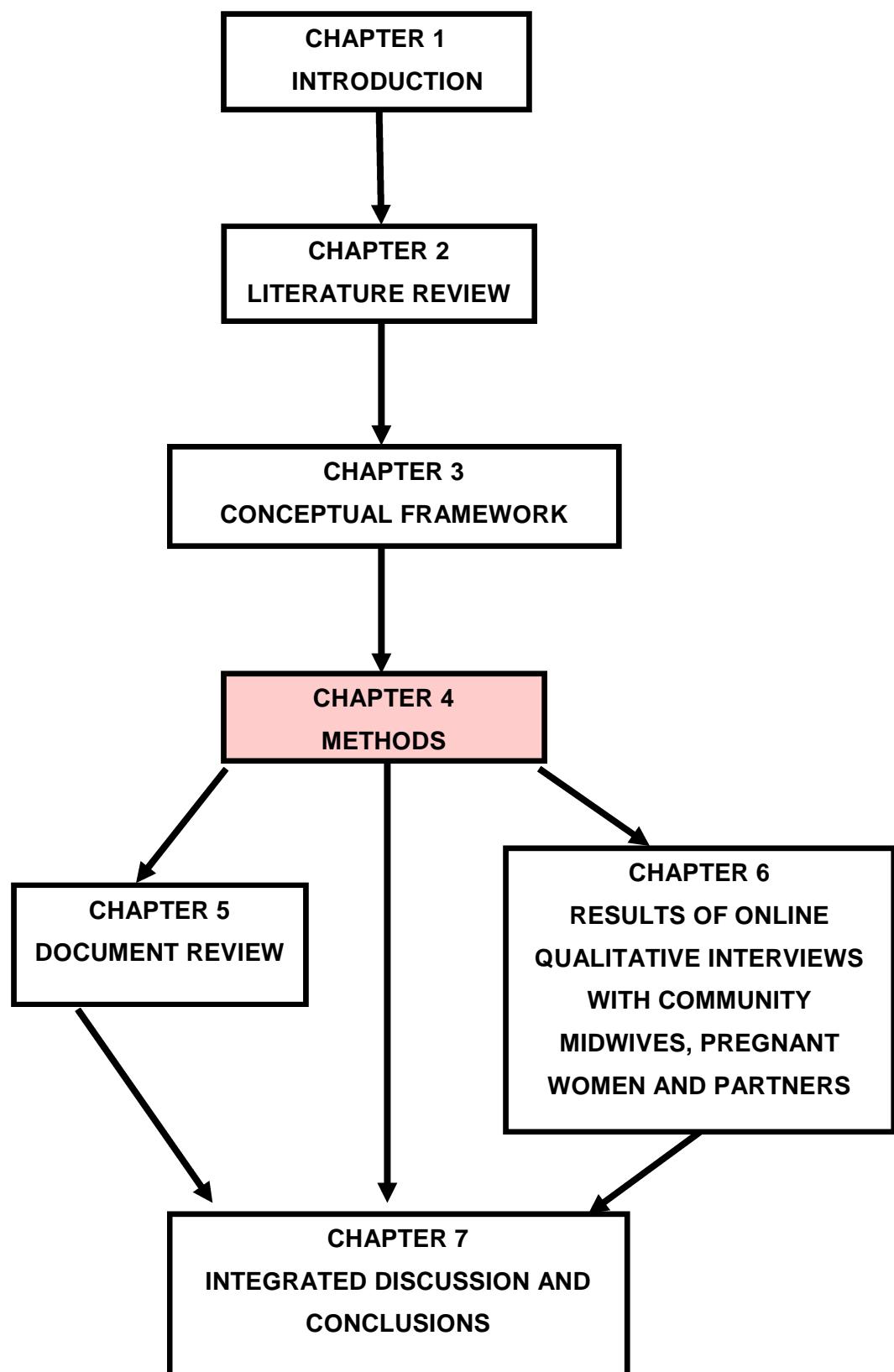
TRB/TRA and HBM cannot fully explore and explain women and partners' participation in Down's syndrome screening.

Indeed, a model that includes both the autonomous and rational decision-making efforts of midwives, women, partners and the service contextual factors that may affect screening uptake was needed. The Donabedian Healthcare Organisation and Delivery Model fulfilled this requirement. The framework can therefore explore the relationships between women, partners, midwives and the service context. This interconnectedness can then be fully explored to explain or identify service factors that influence participation in screening.

### **3.4 Summary**

The Donabedian framework provides a basis for the research question and objectives by grouping attributes in service organisation as structures, process and people (midwife-woman and partner interactions) as service delivery. The outcome of the framework is the influence of service organisation and delivery on women and partners' participation in screening. As such its relevance is in the selection of an appropriate research design and present findings within a structured framework. The framework is intended to highlight the essential care or service attributes identified in the prenatal context of screening and qualitatively explore their influences on participation in screening. Further, the Donabedian framework will provide some insight into the experiences of community midwives, pregnant women and partners in the programmes. The findings might help change practice in ways that will benefit future users (Janssen and Wiegers, 2006; Kobayashi et al., 2011).

The limitation of the adapted Donabedian framework is that it excludes, processes primarily related to the effective delivery of medical care at the community level, for example, home visits for antenatal booking appointments (Donabedian, 2005). However, it is still relevant for prenatal Down's syndrome screening, as consent is an on-going process and consent is obtained when women present at the antenatal clinic or ultrasound department for Down's syndrome screening tests. In addition, the midwife is seen as a proxy of the maternity services in the NHS. Another weakness of the Donabedian framework is that it does not give insight into the dynamic relationship between attributes or factors in structure and process as suggested in existing studies. Therefore, the linear relationship between structure and process is not well established (Donabedian, 2005; Shojania et al., 2007).



## CHAPTER 4: METHODS

### 4.1 Introduction

Methodology describes the overall framework used to investigate reality based on a philosophical position. The philosophical positions adopted in this thesis and the rationales are described in the first section of this chapter. Next, the research design which links the research question to data collection and the conclusions to be drawn is presented. The research design provides coherence; a pathway for proceeding from questions to conclusions. This is followed by the methods of data collection underpinned by the conceptual frame work (see table 1). The pilot study conducted in the course of this research and the methodological issues encountered are discussed. Subsequently, the use of scenarios or vignettes as a methodological tool in the main online qualitative interviews to address some of the issues raised in the pilot study was described. This is followed by a description of the use of the vignettes, open-ended questions, written and photo prompts to obtain qualitative data. Lastly, steps to ensure trustworthiness of findings, data management and analytic procedures are explained.

### 4.2 Methodological Framework

Gill and Johnson (2010, p.64) reported that research "*requires the use of clear methodological procedures and protocols*" to establish trustworthiness. Saunders et al. (2009) propose the research 'onion' design methodological framework. The framework consists of layers each representing a key design decision needed to conduct the study. Previous work has proposed some of the key drivers that affect the methodology used (Bryman and Burgess, 2002; Saunders et al., 2009). These are:

- (1) the philosophical position of the researcher
- (2) the objectives of the study
- (3) the nature of the research problem
- (4) theoretical framework(s) supporting the study.

The research objectives, the research problem and the Donabedian quality framework underpinning the research have been discussed in the preceding chapters of this thesis.

#### **4.2.1 Research Philosophy**

Existing works suggest research philosophy affects the conduct and quality of research (Tashakkori and Creswell, 2007; Saunders et al., 2009). It is described as the way one thinks about the development of knowledge and is associated with the concepts of ontology and epistemology. Ontology and epistemology have different assumptions which have led to what is known as the research paradigm or the worldview stance (Guba and Lincoln, 2004; Tashakkori and Creswell, 2007). Research paradigms have been associated with different traditions, but two commonly used are the positivism/ post-positivism paradigm (realism), and the interpretivist/ phenomenological paradigm (widely known as constructivism).

A quantitative method (positivism/ post-positivism paradigm) of inquiry used alone was considered unsuitable for this thesis, because little is known about the topic and decision making processes are complex. Moreover, the purpose was not to generalise the findings to the population, but to provide detailed descriptions and interpretations of the findings that may be transferable to similar settings. As a result an exploratory qualitative approach was considered most appropriate.

The descriptions of the interpretive paradigm best fit the nature of this study and it was the approach adopted. Limited research on the topic has used conversational analysis of tape-recorded consultations between healthcare professionals and women to examine the topic, but this was considered limited in scope, as it did not allow participants to tell their story or describe their experiences.

A number of traditional face-to-face qualitative methods were considered, but an online qualitative interviews method was selected. The rationale is the problems with recruiting busy healthcare professionals and partners of pregnant women, necessary for traditional face-to-face qualitative research. The difficulty with recruiting healthcare professionals and male partners of pregnant women owing to work commitments have been reported in previous studies (Hamilton and Bowers, 2006; Curry et al., 2009; Reed, 2009b).

The initial design adopted for the pilot study was the sequential exploratory mixed-methods approach (QUAL→quan) (Patton, 1990; Tashakkori and Teddlie, 1998; Creswell and Clark, 2007; Morgan, 2007; Creswell, 2009). This approach was replaced by a qualitative case study design, as a result of the technical problems encountered during the pilot study, participants' lack of interest in the study, poor response rates and ethical issues

encountered. The rationale for replacing the mixed methods design is explained in section 4.10.

A qualitative descriptive study was undertaken in the main study to obtain a detailed source of descriptive data regarding the influence of service organisation and delivery on women and their partners' participation in screening. Sandelowski (2000, p.336) stated that a qualitative descriptive study provides "*a comprehensive summary of an event in the everyday terms of those events*". It aimed to explore the meaning participants or individuals hold about the issue under investigation or their social world. The social world is created through interactions of people with the world around them. In qualitative research the participants are the experts and the researcher seeks to interpret their views of the social world or the issue under investigation. Participants' views are assumed to be subjective and multiple, as there is not just one perspective of the social world or reality.

The philosophical underpinning of this qualitative research is based on constructivist realism which encompasses interpretive and positivism traditions as described in Cupchik (2001). The two traditions were viewed as complementary and in parallel, but the ontology of constructivist realism presented a challenge. The interpretive tradition assumes a subjectively experienced reality that is made up of multiple meanings of the world in which we live and work. The researcher is interested in understanding human actions and experience as constructed by the actors in a given context. In addition, the tradition believes meanings are varied and multiple hence the qualitative researcher is interested in the complexity or multiplicity of perspectives rather than a narrow or 'average' perspective.

The researcher employed vignettes, broad open-ended questions and prompts to elicit the meaning of the issues under investigation underpinned by the adopted Donabedian framework. The focus was to gain useful insights into the structures and processes through which decisions are adjudicated in the prenatal context where the participants interact.

Thus, the theoretical orientation of this research is grounded in the perspective of the participants and is inductive (Creswell, 2007; Hesse-Biber, 2010). The interpretivist approach is a philosophy used to unravel thoughts and feelings of people rather than numbers.

Previous research by Walliman (2006) and Saunders et al. (2009) reported that subjectivity should be recognised between humans and that subjective meanings play a crucial role in everyday life and aim to reveal interpretations and meanings. Therefore, this thesis adopted the 'inductive' approach to enable the development of descriptions and theories about

what has been reported (Crotty, 1998; Gill and Johnson, 2010). The constructivist philosophy is also based on the assumption that the researcher's bias, experience, expertise and insight are all part of the subjective meanings constructed and described (Denzin and Lincoln, 2003).

In contrast, the positivistic tradition believes that there is an objective social reality in the real world. The strength and usefulness of this tradition are that it allowed the adopted Donabedian framework to be interposed as a lens between the researcher and the study phenomenon, to obtain a clear view especially during the data analysis. The deductive approach yielded insights into the patterns of socially meaningful events in this study. It clarified patterns that lie within the data and these patterns were then formally described. Therefore, the two traditions in constructivist realism brought richness and precision which complemented each other.

#### **4.2.2 Rationale for the qualitative approach**

The rationale for using a qualitative approach was based on the literature review which indicated that qualitative interviews and focus groups are the most suitable methods for this study. In addition, qualitative research is particularly well suited for an exploratory study for which previous research is limited. Qualitative research provides a useful method for describing the multifaceted nature of midwives, pregnant women and partners' experiences and the complex process of decision-making in the context of the maternity services (Bradley Eh, 2001; Curry et al., 2009). However, the limitation of qualitative research is that it usually involved a small sample size because of the intensive and time consuming nature of data collection. The findings cannot be generalised to the target population.

### **4.3 Research design for the main study**

The research design is a multiple case study involving online qualitative interviews with vignettes and document analysis of the screening policies and guidelines in two maternity settings. The case study explored the influence of service organisation and delivery on participation in screening from the perspectives of midwives, pregnant women and their partners. The document review is presented in chapter 5. It is imperative to state that the case study design adopted in this thesis is different from clinical case studies in the field of biomedicine where the focus is on describing patients or cases. Case study design has been used successfully to investigate processes and outcomes in dynamic clinical settings in order to gain multiple perspectives (Payne et al., 2007; Curry et al., 2009; Yin, 2009).

The qualitative case study is the preferred design, as the aim of this study was to describe and understand a contemporary event (prenatal Down's syndrome screening) and relevant behaviours (influence of service context) over which the researcher has no control. The case study is an empirical approach that investigates a contemporary complex event (the offer of screening and decision-making processes) within a real world context (maternity services) when the boundaries between the event and context are not clearly visible. The logic underlying the use of the qualitative multiple case study design is to describe similar and contrasting results from the two maternity services studied (Yin, 2009). This is to obtain a description of the frontline providers and users' perspectives on the influence of service organisation and delivery on participation in screening and help explain the variation in uptake rates.

#### **4.4 Research Methods**

The qualitative data collection methods employed in the pilot study were online interviews and focus groups. For the main study, online interviews using vignettes with open-ended questions and prompts were adopted. Interviews have been particularly useful in cases where confidentiality and anonymity may remove fear of reprisal from the authorities or health professionals for unfavourable statements during programme evaluation or patient satisfaction studies (Curry et al., 2009).

Although interviews and focus groups were traditionally conducted either in person or by telephone, the internet or web-based (online) method was adopted. The mode of the online interviews conducted was asynchronous and the open-ended questions were semi-structured with prompts. In asynchronous mode, an individual participant may choose to respond to the questions at any time that is convenient. The same vignettes, open-ended questions and prompts were presented to all participants in a group in the same online format.

##### **4.4.1 Rationale for use of online methods**

The literature review in chapter 2 revealed difficulties with recruiting midwives and male partners in the maternity services and women find it difficult to criticise health professionals in traditional research. In addition, over the years, it has become increasingly difficult to recruit participants for traditional interviews and focus groups, considering the problems with scheduling meetings (Tates et al., 2009).

The internet has been used in the maternity services to explore the perceptions and experiences of service users and providers in prenatal Down's syndrome programmes as demonstrated in the literature review. It has also been used in the field of health research to explore the experiences of women on bed rest (Adler and Zarchin, 2002), behavioural data collection (Rhodes et al., 2003), research on hidden or hard to reach populations (Ahern, 2007; Matthews and Cramer, 2008), in paediatrics and adolescents (Gray et al., 2005; Fox et al., 2007; Zwaanswijk et al., 2007; Tates et al., 2009; Nicholas, 2010) and in surveys of health professionals (Burls et al., 2010).

The goals of internet and traditional qualitative health research involved understanding a phenomenon from the view point of participants that have the experience of the health related condition being investigated (Curry et al., 2009; Nicholas, 2010). The advantages of using the internet over traditional methods in this thesis includes its ability to reach participants that are difficult to recruit because of hectic work schedules, family commitments, cost and geographic location (Denzin and Lincoln, 2005; Kenny, 2005; Tates et al., 2009). It also provides an environment for disclosure of sensitive information due to the anonymity afforded participants by the internet and offers more time for reflection, thus producing a range of data especially from shy participants.

The barriers to the use of the internet in healthcare data collection relate to factors such as, participants and researchers' poor computer literacy and lack of access to internet services which have been reported in previous work (Mann and Stewart, 2000; Kenny, 2005). The drawback of adopting the online methods is that only literate, middle to upper class individuals may agree to participate in the research. However, the Ofcom Communications Market Report indicates that 80% of UK households have access to the internet and text messages are the most used methods of communicating with family and friends (Ofcom, 2012). This has opened up opportunities for data collection using the internet and World Wide Web in healthcare research.

#### **4.5 The main research question for the study**

Do the structures, processes and people in the maternity services influence women and partners' participation in prenatal screening for Down's syndrome?

#### **4.5.1 The broad objectives of the study**

1. To explore whether the structures of the maternity services influence women and partners' participation in prenatal Down's syndrome screening
2. To explore whether the process of offering prenatal Down's syndrome screening influence women and partners' participation in the programme.
3. To explore whether community midwives influence women and partners' participation in prenatal screening for Down's syndrome.

#### **4.6 Settings**

A purposive sample of maternity services in the North of England were identified from routine reporting of regional data (Walker and Tarn, 2012). Two NHS Trusts at the high and low end of the range of prenatal Down's syndrome screening uptake rates for the region were invited to participate in the study. Routine data for the past three years have shown wide variation in uptake rates in this region (see figure 4). Though, the data for the past 2 years have been primarily second trimester screening (Walker and Tarn, 2012). The two locations were chosen, as the first trimester combined screening test is the preferred screening test in both maternity services. For the purpose of this thesis, the pseudonym Terrace maternity service was used for the low uptake Trust and Landscape maternity service for the high uptake Trust.

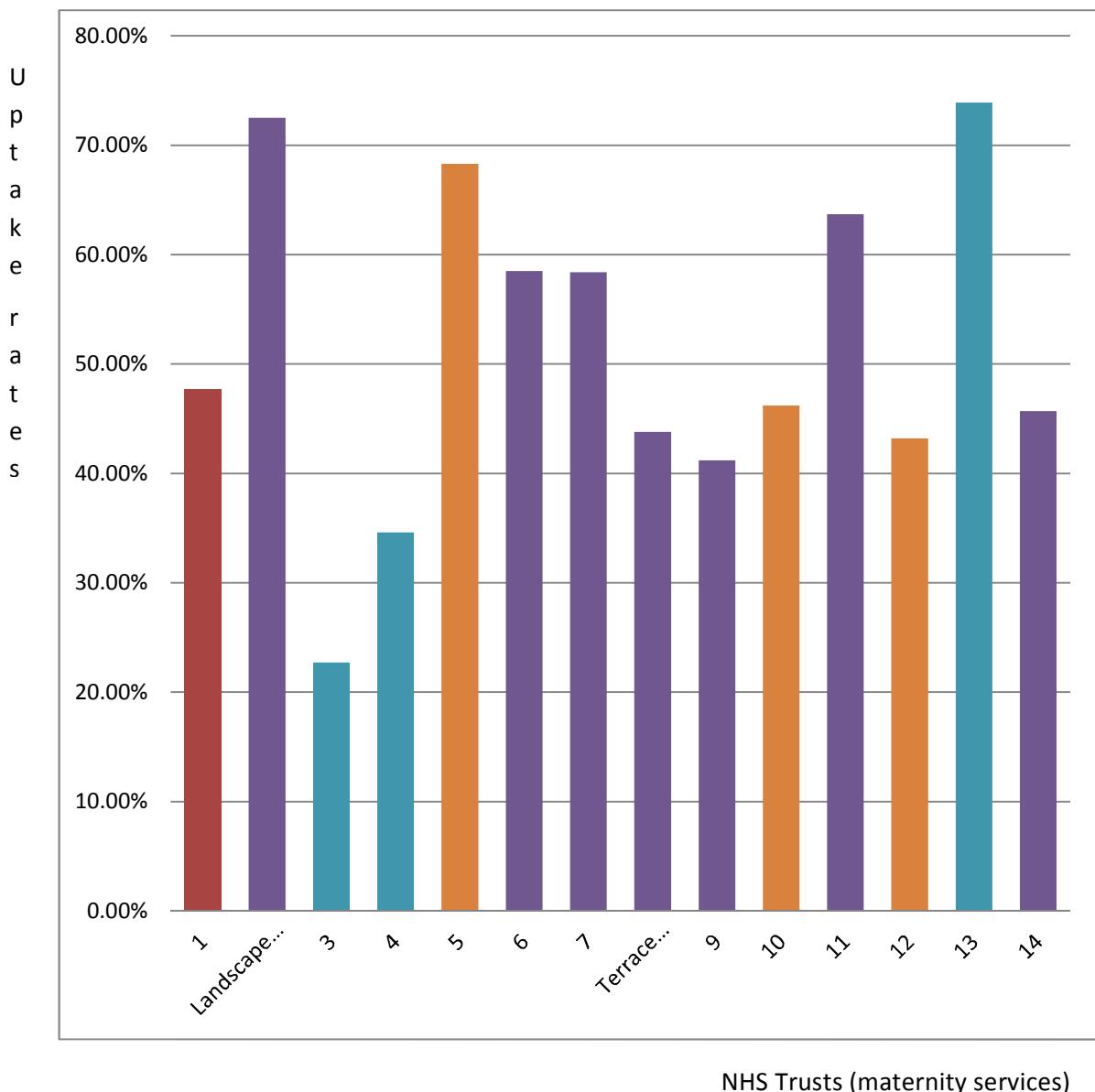
##### **4.6.1 Terrace maternity service (Low uptake Trust)**

The NHS Trust is a large multi-site setting located in an urban area in the North of England. The maternity service provides coverage for a multi-ethnic population. The population is heterogeneous, but the white British ethnic group make up the majority of the population. The Trust is a designated teaching hospital that teaches undergraduate and post-graduate students. See chapter 5, subsection 5.9.2 page 120 for more details.

##### **4.6.2 Landscape maternity services (High uptake Trust)**

This is a smaller single site setting, located in a semi-urban area in the North of England. The NHS trust is not a designated teaching hospital. See chapter 5, subsection 5.9.3 page 120 for more details.

**Figure 4: Uptake rates for 1<sup>st</sup> and 2<sup>nd</sup> trimester prenatal Down's syndrome screening for the North of England 2011-2012.**



#### **4.7 Sample and recruitment of community midwives**

The aim is to identify information-rich participants who have certain characteristics, detailed knowledge, or direct experiences relevant to the phenomenon of interest. In this thesis, it is a role or participation in prenatal Down's syndrome screening. The sampling strategy adopted was purposive sampling, which seeks to include community midwives who provide information about prenatal Down's syndrome screening to pregnant women at booking

appointments. Community midwives were therefore expected to provide a range of responses to the online interviews. It was therefore important to recruit midwives who were willing to take part in the study, so that they can honestly and openly share their viewpoints and experiences.

In qualitative research the sample size varies according to the type of study. However, the sample size is usually smaller than those used in quantitative research. The interest in a qualitative sample is on depth, richness of data and its analysis as previously reported (Curry et al., 2009; Carlsen and Glenton, 2011). Purposive sampling was employed to recruit 18-24 community midwives. To achieve this sample size, it was necessary to over recruit as suggested in the literature for online studies (Bloor et al., 2001; Fox et al., 2007).

This sample size was considered adequate for the study, based on the recommended sample size by the literature for conducting qualitative interviews to achieve data adequacy and diversity of views (Patton, 1990; Kuzel, 1992; Baum, 2008; Curry et al., 2009). It was also considered sufficient to allow for attrition and gather sufficient data. In addition, participants tend to respond sparsely to online interviews compared to face-to-face interviews. The sample size enabled data to be collected from a range of community midwives with varied experiences in their role of delivering Down's syndrome screening. Further, it is the sample size for conducting four focus groups with a minimum of six participants. This is important as the trustworthiness of the data from focus groups depends on the credibility of comments from the participants and also the number of focus groups held as noted in Duffy (2002).

The Local Collaborators in the participating NHS trusts were requested to distribute invitation letters and the participant information sheets (PIS) (see appendices 5 and 7) to all community midwives with a request to join and contribute to the study. Midwives who agreed to participate in the study were informed to log in online to the study by using the link provided in the invitation letters. This was to enable them to read the PIS and consent forms. They were also told they could contact the researcher for further information.

Participants who registered or signed in for the study could print copies of the consent form to keep. Nevertheless, by registering and clicking on the submit button that read "*I agree to participate*" or responded to the questions, participants were giving their consents to participate in the study (see appendix 6). To increase the response rates, the community midwives were assured of their confidentiality and anonymity. They were also told a

summary of the findings of the research will be sent to them if required. This strategy has been used in previous studies (Partridge and Winer, 2002; Fernandez et al., 2003; Holmes, 2009).

Furthermore, prior to data collection, the researcher attended the monthly meeting of community midwifery team leaders in Landscape maternity services in order to promote the study among the midwives. At the meeting, the objectives of the study and how to navigate the study's website were explained, but there was no computer with internet access in the room to demonstrate it visually. The issue of requiring a password for registration was also explained. They were also assured of the security of the website as it was being hosted, secured and backed-up by the University of Leeds.

#### **4.8 Sample and recruitment of pregnant women and partners**

The aim was to identify information-rich participants who have certain characteristics or direct experiences relevant to the research topic such as age, ethnicity, gender and education thereby reflecting a wide range of perspectives and experiences. The sampling strategy employed was purposive sampling which seeks to include pregnant women and partners who were offered prenatal Down's syndrome screening and it was not important whether they accepted or declined screening. Based on the research questions, pregnant women and partners were expected to provide the most credible responses to the study. It was therefore important to recruit pregnant women and partners who were willing to take part in the study, so that they can honestly and openly share their viewpoints and experiences. Purposive sampling was employed to recruit 18-24 pregnant women and 18-24 partners to the study. To achieve this sample size, it was necessary to over recruit as noted in the sampling and recruitment of community midwives. The rationale for the sample size is as explained for community midwives in section 4.7 page 77.

The head of the ultrasound department in both NHS trusts were informed about the research through the Local Collaborators and they were willing to assist in the recruitment of pregnant women and their partners. The line managers and the sonographers were subsequently informed about the study. The researcher usually reported to the sonographers on duty in the antenatal ultrasound department on each day of recruitment, so that they will be aware of his presence and assist in the recruitment.

The sonographers briefly introduced the study to the pregnant women and partners on completion of the 20 week foetal anomaly scan and obtained consent for them to be introduced to the researcher. The purpose of the study was briefly explained to pregnant women and their partners who agreed to be introduced to the researcher. Those who met the inclusion criteria were given the PIS and invited to participate in the study.

The period of the 20<sup>th</sup> week foetal anomaly scan was chosen for recruitment, as many male partners accompany their pregnant partners to the foetal anatomy scan (Reed, 2009a). To increase the response rates, pregnant women and their partners were assured of their confidentiality and anonymity. They were informed a summary of the findings would be sent to them, if required, on completion of the research. Further, pregnant women whose partners were not present at the foetal anomaly scan were requested to introduce the research to partners who had been present at the booking appointment. Women, who agreed, were given information sheets for partners to take home.

#### **4.8.1 Inclusion criteria for pregnant women and partners**

- 1) The study included pregnant women and partners who were offered prenatal Down's syndrome screening by a midwife and accepted or declined the offer.
- 2) The study required pregnant women and partners to have sufficient competency in written and spoken English to understand the PIS, consent forms and navigate the online study web page. Though, as the data collection for this group was online, languages other than English could be translated using free web translation services like Google translate.
- 3) Women and partners were 16 years or over, owing to the care pathway for women below 16 years being different. They need specialist care throughout pregnancy.
- 4) Access to the internet and an email account which may be created for this study.
- 5) Willingness to participate in the study.
- 6) Capacity to consent. Nonetheless, this was assumed as the sonographer must have assessed and obtained consent for the foetal anomaly scan. Additionally, the sonographer must have obtained consent to be introduced to the researcher.

#### **4.8.2 Exclusion criteria for pregnant women and partners**

- 1) Women less than 16 years of age, since their care pathway is different, as they require specialist care.

- 2) Partners less than 16 years of age, because the researcher could not account for their ability to comprehend or understand the information for the research and give informed consent to participate in the study.
- 3) Women with multiple pregnancies were excluded, as they have a different prenatal Down's syndrome screening programme.
- 4) Women who were not offered prenatal Down's syndrome screening by midwives or were offered screening or booked in a different maternity service.

#### **4.9 Ethical considerations**

To facilitate the ethical approval process, contact was made with health professionals in managerial roles in the maternity services to act as Local Collaborators, as it was a requirement for ethics approval. They functioned as the local contact throughout the study by facilitating access, information and making sure the research caused as little disruption as possible in the maternity services.

Using NHS premises for recruiting community midwives, pregnant women and their partners required ethics, research and development (R&D) approvals and obtaining research passports from the respective Trusts. The approvals and research passports were obtained prior to recruitment and data collection. The reference number for the ethics approval for this study is 11/YH/045. Copies of the approval letters, consent forms, PIS, and invitation letters to midwives are in appendices 2, 3, 4, 5, 6 and 7.

The ethics, R&D approvals, research passports and letters of access took quite a while to obtain. Other researchers have had similar or worse delays (Hunter, 2005; Ahmed et al., 2006a; Galbraith et al., 2006). Preparation for ethical approval started in September 2011 and the final approval (letter of access) was obtained by the end of March 2012. Nevertheless, clear consideration was given to the potential ethical issues arising in undertaking this study. The issues were;

##### **4.9.1 Informed consent**

Verbal consent was obtained from pregnant women and partners by the sonographers to be introduced to the researcher at the end of the foetal anatomy scan. The study was explained verbally to pregnant women and partners who agreed, for about 2- 5 minutes. They were given PIS by the researcher. Community midwives were sent letters of invitation and PIS by

post via the Local Collaborators in the participating NHS Trusts. The information for the research clearly identified the purpose and sponsor of the study, how participants had been selected and the nature of their commitment. Further, participants were told they could contact the chief investigator for more information.

Those who agreed to take part in the study were asked to register for the study online. The PIS and consent forms were placed on the website for participants to read and agreed to by registering or signing-in for the study. They were advised to save or print a copy of the consent form for their records. The study's website was designed to allow access only to participants who registered or signed-in and hence, consented to participate in the study.

#### **4.9.2 Confidentiality and anonymity**

The confidentiality and anonymity of participants' and organisational data were ensured by replacing names with pseudonyms and identifier numbers. Prospective participants were assured of the confidentiality of their responses. Data were stored in accordance with the Data Protection Act (1998). Only the chief investigator and academic supervisors had access to personal data. The personal data was kept separately from the anonymised data in the password secured, firewall protected University of Leeds researcher's computer. To further strengthen the confidentiality and anonymity, participants were informed they could create a new email account for the study.

#### **4.9.3 Harm or risk**

The researcher was aware that in any online study there are two potential sources of harm. First, the research questions may provoke emotional reactions. Second, a breach of anonymity or confidentiality may cause harm especially in research with an under-studied population for example, undocumented migrants. In general, evidence has shown that online studies are no more risky than traditional methods (Kraut et al., 2004). Indeed, in practice, they may be less risky since the online environment may make it easier for participants to withdraw if they wish to do so. The concern was that it is not possible in an online study to know when a participant suffers an adverse psychological reaction to the research topic or questions and needs support. This has also been reported in previous research (Kraut et al., 2004; Holmes, 2009). Therefore, participants were informed in the PIS and on the study's website that if they felt uncomfortable or distressed whilst responding to the questions they should stop and contact the chief investigator, their GP or midwife. The

researcher was also careful to offer support to participants by providing a link on the study's website to contact details of sources of support such as the Antenatal Results and Choices group. Rules about appropriate online behaviour, where they could report abuse or make complaints were provided on the website. The website was safeguarded, backed up and monitored by University of Leeds Information Systems Services (ISS).

#### **4.9.4 Autonomy of participants**

Community midwives who agreed to take part in the study were requested to register for the study using the web link provided. Those who agreed to the consent form online by registering for the study were recruited. Pregnant women and partners who registered online were also recruited into the study. Three individual web pages were created for community midwives, pregnant women and partners to ensure confidentiality and anonymity. Throughout the study, community midwives, pregnant women and partners completed the study separately when they logged-in to their specific web pages. In addition, they were informed that they could withdraw from the study at any time without informing the researcher and that their medical care or legal rights would not be affected.

### **4.10 Pilot Study**

The initial study design was sequential exploratory mixed-methods involving online interviews and discussion forum with antenatal screening coordinators (online interviews only), pregnant women, partners of pregnant women and community midwives. Hence, the pilot study was conducted primarily for the phrasing, sequencing and construction of the final topic guides for the main study. Piloting also explored the feasibility of using the web-based methods in the study. The pilot study was therefore to test the technical aspects of the data collection tools for flaws, limitations or weakness in the study and web designs. It was also to indicate the approximate duration required to conduct the main study. This was to enable flaws to be addressed before the commencement of the main study as suggested in previous research (van Teijlingen and Hundley, 2002; Kvale, 2007; Turner, 2010).

#### **4.10.1 Topic guides for pilot study.**

Topic guides were designed and prepared to capture the experiences and perspectives of antenatal screening coordinators, community midwives, pregnant women and partners. Careful attempt was made by the researcher to generate the topic guide around research

questions (moving from topics to questions). To do this, a list of potential questions was provided to represent the research question. These questions were generated from the literature review. The initial drafts of the topic guide were then discussed in several supervision meetings to ensure that all potential interview questions matched the research question and objectives. The topic guides were initially pre-piloted among colleagues and the feedback together with that of my supervisors were used to restructure the topic guides before the piloting.

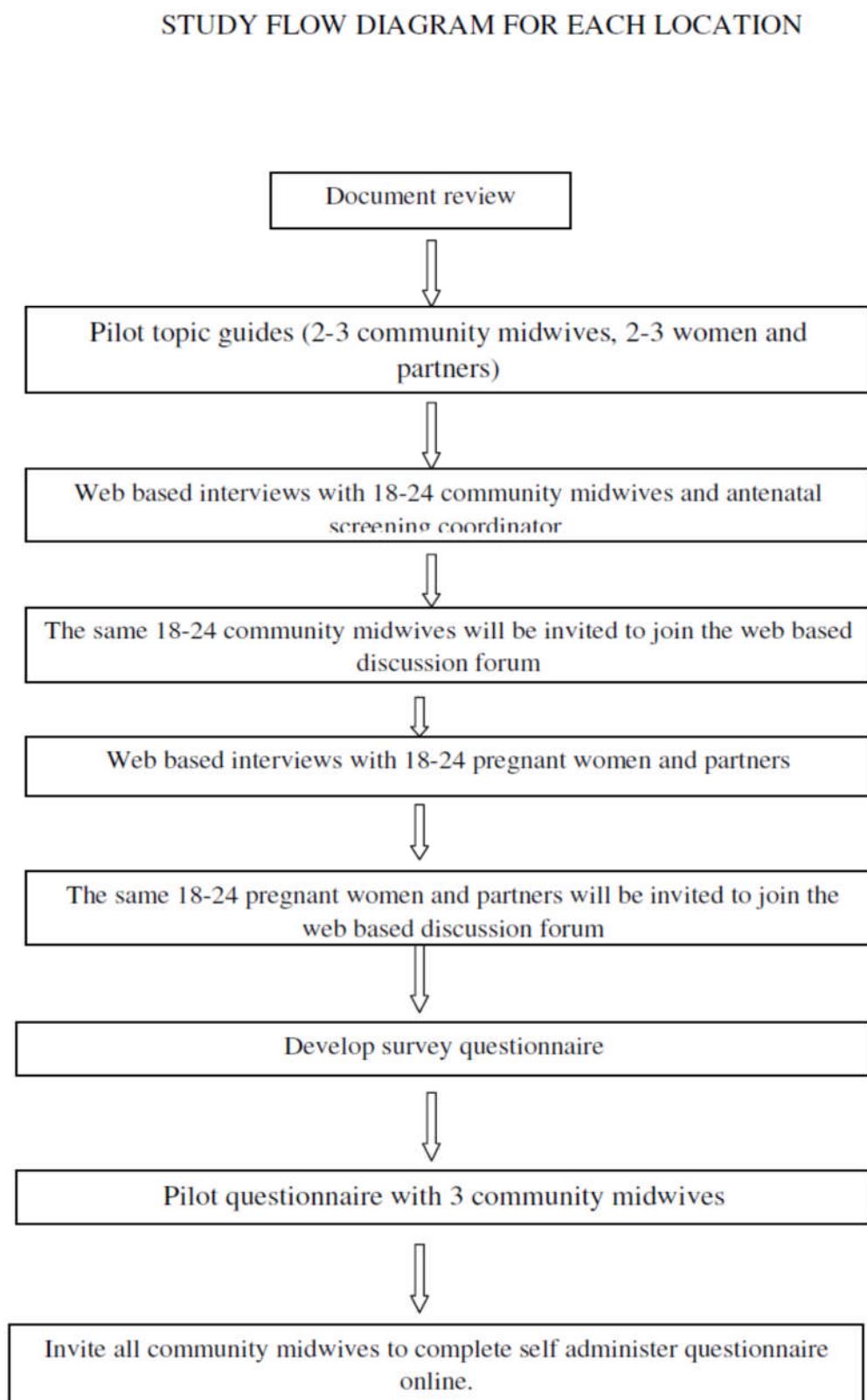
#### **4.10.2 Pilot study data collection**

The secure web based interview and discussion forum were set up by Professor William Montelpare, a visiting Professor in the School of Healthcare, University of Leeds who provided the necessary expertise. The web based interviews and discussion forum were hosted in the firewall protected, secure server of the University of Leeds ISS. This was a novel, contemporary, exploratory part of data collection in the study design.

The online interviews took about 30minutes to complete. Participants who completed the interviews were invited to the discussion forum. The web based discussion forum required community midwives, pregnant women and partners to log in to their specific discussion boards with their unique passwords and usernames. The usernames and passwords were self-generated when participants registered for the study.

Participants who responded to the questions or posted comments on the discussion boards were required to identify themselves to the group by a self-chosen pseudonym throughout the duration of the study. Pregnant women are regarded as vulnerable people by the nature of their condition hence community midwives, pregnant women and partners had different (separate) discussion forum during the pilot study. The rationale was to provide a safe environment for pregnant women to discuss freely with other pregnant women of similar gestational age. Further, partners' presence in the same online forum might have influenced women's responses. Participants were advised to keep their username and password in a safe place and to take no more than 30 minutes to respond to each question that was posted on the discussion forum weekly, to avoid the research becoming a burden.

**Figure 5: Pilot study flow diagram**



#### **4.10.3 Findings from the pilot study**

The pilot study raised numerous methodological issues. For example, at the start of the pilot study the website was taken down for maintenance by the University's ISS for a period of two days which could have adversely affected the response rates. Additionally, in both maternity services, the community midwives could not log on to the discussion board after completing the online interviews. Some reported a blank screen when they logged on to the study;

Email sent on 30.04.12:

"Hi

*My name is RM (pseudonym) and I am a community midwife for NHS (pseudonym), I am trying to register on your research but the link does not take me to your research??*

*Please help,*

*Thanks RM".*

The University ISS was contacted immediately about the technical problems experienced by participants accessing the website. The ISS initially felt there was no problem, since the website was working fine after diagnostic checks were done. In the course of trying to find out what the possible cause or technical issues were, the researcher decided to investigate using his home computer. By typing in the study's web address into several browsers, the researcher noticed that the study's website goes blank and reappears when the compatibility mode was switched on. This finding was relayed to the ISS. Thereafter, a description of how to turn on and off the compatibility mode was sent to the local collaborators who in turn informed the community midwives. However, many of the midwives had difficulties locating the compatibility mode:

Thu, 31 May 2012

*Hi Hyacinth,*

*Just to inform you that I am still having problems trying to access the study. Tried again several times today and am still getting a blank page when I log in and I have tried through the word document but I am unable to find the compatibility view after using both methods you gave. I will keep trying and am waiting for IT dept to help me access the site. RM.*

As shown by the emails above the complaints kept coming and the researcher was left with no option but to forward the anonymised emails to the ISS. The response from the ISS few days later may be seen in the email below:

22.05.2012

*Hi,*

*There are some design issues with the site and these are probably causing later versions of IE to throw it out. You at least need a correct doctype, add this line to the very top and see if it cures it:*

*<!DOCTYPE HTML PUBLIC "-//W3C//DTD HTML 4.01 Transitional//EN"*

*"<http://www.w3.org/TR/html4/loose.dtd>">*

*ISS.*

The link was added, as suggested by the ISS, but the problem was not cured. This was easy for the researcher to diagnose using the compatibility view in his home computer. The information that the blank screen was not cured by the doctype was relayed to ISS.

The researcher had to be reflexive to understand that the problem may have occurred when the website was formatted to fit the requirements of the University's ISS. This new insight by the researcher was relayed to the technical team and the website was subsequently reformatted. The reformatting of the website resolved the issues of the blank page using the researcher's home computer. Hopefully, the technical problems were finally resolved, but a new and menacing issue emerged; the midwives were no longer interested in the study.

Other methodological issues encountered were poor response rates especially to the discussion forum. This may be due to design issues with the website. The forum required participants to log on to the website frequently to either respond to the questions on the discussion board or post comments. With the busy nature of midwives, this was difficult as many did not remember to do so at work. Moreover, they were required to log on using passwords and usernames. Some midwives complained through the gatekeeper that the password retrieval was problematic. Additionally, the requirement for registration with a username and password might have made some participants suspicious, owing to fears over hacking into their accounts or computers and identity theft. The discussion forum also required participants to post comments on responses from other participants at least twice for each topic or question, which the midwives may have found undesirable, owing to their

busy schedules. Importantly, having 18-24 participants in one forum meant participants having to read all or skip some of the responses before posting a comment. This was a methodological flaw in the study and by hindsight; each cohort should have been allocated into four groups with a group size of four to six to allow for easy participation. The study was also designed without a backup plan in the event of a failure or breakdown in the web-based data collection tool, as reported in existing literature on conducting online interviews (Salmons, 2011).

The sensitive nature of the research topic may have been a barrier to participation in the discussion forum. Further, the fear of employers accessing the website and reading participants' responses and the risk of identification may have been a deterrent. Finally, the midwives refused to take part in the study outside working hours, because they were not going to be paid for their time.

## **4.11 Main Study**

A decision was reached by the researcher in agreement with the supervisors who are skilled in qualitative research and online methods to replace the pilot study with an online qualitative interview using vignettes with open-ended questions and prompts. This decision came as a result of reflecting on the data collected during the pilot study. It was noticed that participants who responded, completed the online interviews. However, the discussion forum had few responses and most of the responses were to the first question with progressively fewer responses to the prompts and other topics.

The reason may be due to the fact that the online interviews took about 30 minutes to complete in one sitting, whilst the discussion forum meant going back and forth over a period of one to two months. The pilot data from the online interviews indicate that participants were providing socially desirable answers and responses were limited to a few lines or words. The few responses to the discussion forum were more detailed and revealed some of the contextual factors that might influence participation in screening compared to the online interviews.

### **4.11.1 The main online interviews using vignettes**

At the end of several monthly supervision meetings, a decision was reached to combine features of the online interviews with that of the discussion forum. This was to enable the

development of an asynchronous online qualitative interview using vignettes with open-ended questions and written prompts. The idea was to develop a data collection tool that combines methodologies from traditional and online data collection tools. This was to provide an approach that was innovative, accessible, captures participants' attention and capable of eliciting detailed qualitative data. Importantly, it has to be an online qualitative interview that was efficient and also quick to complete.

The rationale for continuing with the online methods in the main study was the availability of the study's website in the University of Leeds server for the next few months. In addition, most of the technical problems were resolved with the reformatting of the website. The website became widely accessible and functional with most internet browsers able to access the study's web page without producing a blank page or screen.

The busy nature of midwives, partners of pregnant women and lack of full access to the internet by midwives, necessitated the use of asynchronous online interviews with vignettes to be completed at the participant's convenience. Furthermore, the online qualitative interview is a useful method for gathering information from people who were willing to share their stories, but frightened or hesitant to do so in a face-to-face interview as noted in McCoyd and Kerson (2006). To allay concerns regarding confidentiality and anonymity, the researcher built trust with participants by assuring them that their responses would be treated as confidential. Hence, this bond of trust was reinforced in the instructions and participant information sheets as well as in the overall conduct of the online interviews.

#### **4.11.2 Vignettes**

The terms vignette and scenario are used interchangeably within this thesis. The main online interviews used the term scenario to describe the short stories. The reason is scenarios are commonly used in midwifery educational training.

Vignettes are short stories or scenarios in textual, video or pictorial format or delivered through computers that describe hypothetical situations to which participants are asked to respond as noted in previous research (Hughes, 1998; Johnson, 2000; Taylor, 2006; Jenkins et al., 2010). Since vignettes portrayed hypothetical characters, existing work suggests they offer a less threatening way to investigate sensitive topics (Barter and Renold, 1999; Styles et al., 2011).

Vignettes have been used to elicit participants' perceptions, attitudes, and beliefs about specific situations, to investigate topics that may be sensitive to participants and to compare perceptions between groups (Finch, 1987; Barter and Renold, 1999; Martin, 2004). For example, it has been used to explore the quality of physicians' practice and decision making (Peabody et al., 2004; Berney et al., 2005), to explore midwives' intrapartum decision making in relation to their attitudes towards risk (Styles et al., 2011) and to survey health-related attitudes of the general population (Denk et al., 1997; Link et al., 1999; Hughes and Huby, 2002). It has also been used to investigate attributions and reported communication by healthcare professionals to the diagnosis of Down's syndrome (Elwy et al., 2007) and in a study that explored the views of health care professionals regarding informed choice and prenatal testing (van den Heuvel et al., 2010).

The rationale for adopting scenarios in the online interviews was firstly, the ethical, moral and safety dilemmas presented by direct observation of a booking visit that involved the community midwife, pregnant woman and partner where Down's syndrome screening test is offered. These dilemmas have been previously reported in existing research (Hughes and Huby, 2002; Styles et al., 2011). Secondly, the influence of service organisation and delivery on users' participation in screening is a sensitive topic among healthcare professionals as noted in previous work (Williams et al., 2002b; Dormandy and Marteau, 2004). Thirdly, with vignettes, participants do not necessarily need to have in-depth knowledge of the topic under study as suggested in Hughes (1998). This is particularly important as women and partners may not have in-depth knowledge of prenatal Down's syndrome screening or may have forgotten the information given as reported in previous research (Jaques et al., 2004a; Khoshnood et al., 2004; Williams et al., 2011). The final rationale is time constraints. Vignettes represent a simple tool that can be used to explore midwives', women and their partners' perspectives and experiences quickly and efficiently. Similar findings on the use of vignettes to quickly explore participants experiences have been reported in previous research (Veloski et al., 2005).

#### **4.11.3 The use of open ended questions, written and photo prompts**

Studies have shown that the use of vignettes with open-ended questions allowed participants to describe their experiences in detail, as it encourages participants to write in their own words about their experiences and opinions (Kalafat and Gagliano, 1996; Hughes, 1998; Sheppard and Ryan, 2003; Hughes and Huby, 2004; Williams et al., 2005). A study by

Veloski et al. (2005) stated that the most effective vignette-based surveys used open-ended questions. In this study, each participant group had identical open-ended questions, but the questions were worded to elicit detailed responses from participants as much as they were comfortable with.

The use of prompts has also been advocated in traditional qualitative research with open ended questions to keep participants focused with their responses and elicit as much detailed information as they are comfortable with. Previous work suggests prompts ensured that participants provided optimal responses of their viewpoints and experiences by reducing misunderstanding of the main open-ended questions (Creswell, 2007; Turner, 2010). In the online interviews with community midwives, women and their partners, the written and photo prompts employed with the vignettes reduced non-response to the main open-ended questions.

Photo prompts were employed with scenario 4 for community midwives, pregnant women and partners to stimulate the memory of participants (see appendix 8). This was necessary as the influence of the social and physical prenatal environment may not be easily isolated by participants as an influence among other influences at booking. Moreover, responses to vignette 4 may not be accurate because of time-lagged recall bias. In addition, existing work suggests photo prompt was effective among participants with low literacy as it enable them to understand and focus on the purpose of the study thereby facilitating in-depth responses (Bender et al., 2001). The written and photo prompts were useful tools in the main online interviews and made it easier to elicit a range of responses from participants.

#### **4.11.4 Constructing the scenarios for the main online qualitative interviews**

Two criteria are important in constructing scenarios. The first is the constructed scenario(s) should appear realistic to participants. A realistic scenario creates interest and engenders greater involvement by the participants as reported in Nicholas and Yavas (2006). The second criterion is that the vignettes should be clear about the problem being portrayed, while being vague enough to elicit multiple responses from the participants (Hughes, 1998).

To portray realistic situations in this study, the vignettes were created from data obtained from the pilot online interviews and discussion groups, literature review and the NHS choices website (an internet resource for pregnant women). Each vignette was carefully written and edited by the researcher during several supervision meetings from June to October 2012. This afforded the opportunity for input from the researcher's supervisors who are highly

skilled in qualitative research and online studies. The aim was to achieve clarity and brevity and avoid clutter which may have given participants reasons not to respond appropriately to the online interviews. The vignettes were presented in an orderly manner and provided enough realistic information to reflect how actual pregnant women and partners were seen by midwives at booking.

Existing research has shown that open-ended questions in studies that use vignette elicit responses that are close to real experiences by promoting creativity among participants as in a focus group (Kalafat and Gagliano, 1996; Hughes, 1998; Sheppard and Ryan, 2003; Hughes and Huby, 2004). However, the design of the qualitative online interviews using vignettes and prompts did not facilitate in-depth responses as in face-to-face in-depth interviews but provided the opportunity for a considerable range of responses to be obtained from participants.

There are several types of vignettes such as a snapshot, portrait and composite vignettes. The snapshot represents a story about an aspect of the problem under investigation. The portrait is a vignette that describes the participants' characteristics and experiences whilst the composite describes a vignette that is a combination of snapshot, portrait and some analysis of the problem as noted in Spalding and Phillips (2007). The type of vignette adopted in this study was the content specific vignette. It contained a brief description of the setting, participants, the problem, the interacting dimensions, the dialogue between participants and the event(s) participants should pay attention to as noted in Veal (2002). The content specific vignette is crucial in eliciting different perspectives among participants, the subtleties of the interactions at booking and factors that may have influenced the decision making process. Besides, it provided participants with the feeling of being in a discussion group and helped capture the context in which screening was offered as participants responded to the questions.

#### **4.11.5 Pilot of main study**

The main study was piloted with two participants in both settings. They reported the vignettes and questions were comprehensible and encountered no problem accessing the study online.

#### **4.12 Sample and recruitment of community midwives**

This was conducted as described in section 4.7 (Sample and recruitment of community midwives). Although, before the commencement of the main study in both maternity services, the researcher held meetings with the Local Collaborators to explain that the website had been reformatted and was now easily accessible. Modalities for facilitating access and promoting interest in the study within the maternity services were also discussed. Both Local Collaborators promised to facilitate the study in the Trusts and were particularly satisfied the data collection method was an online interview that took about 30 minutes to complete.

#### **4.13 Sample and recruitment of pregnant women and partners**

This was conducted as described in section 4.8 (Sample and recruitment of pregnant women and their partners).

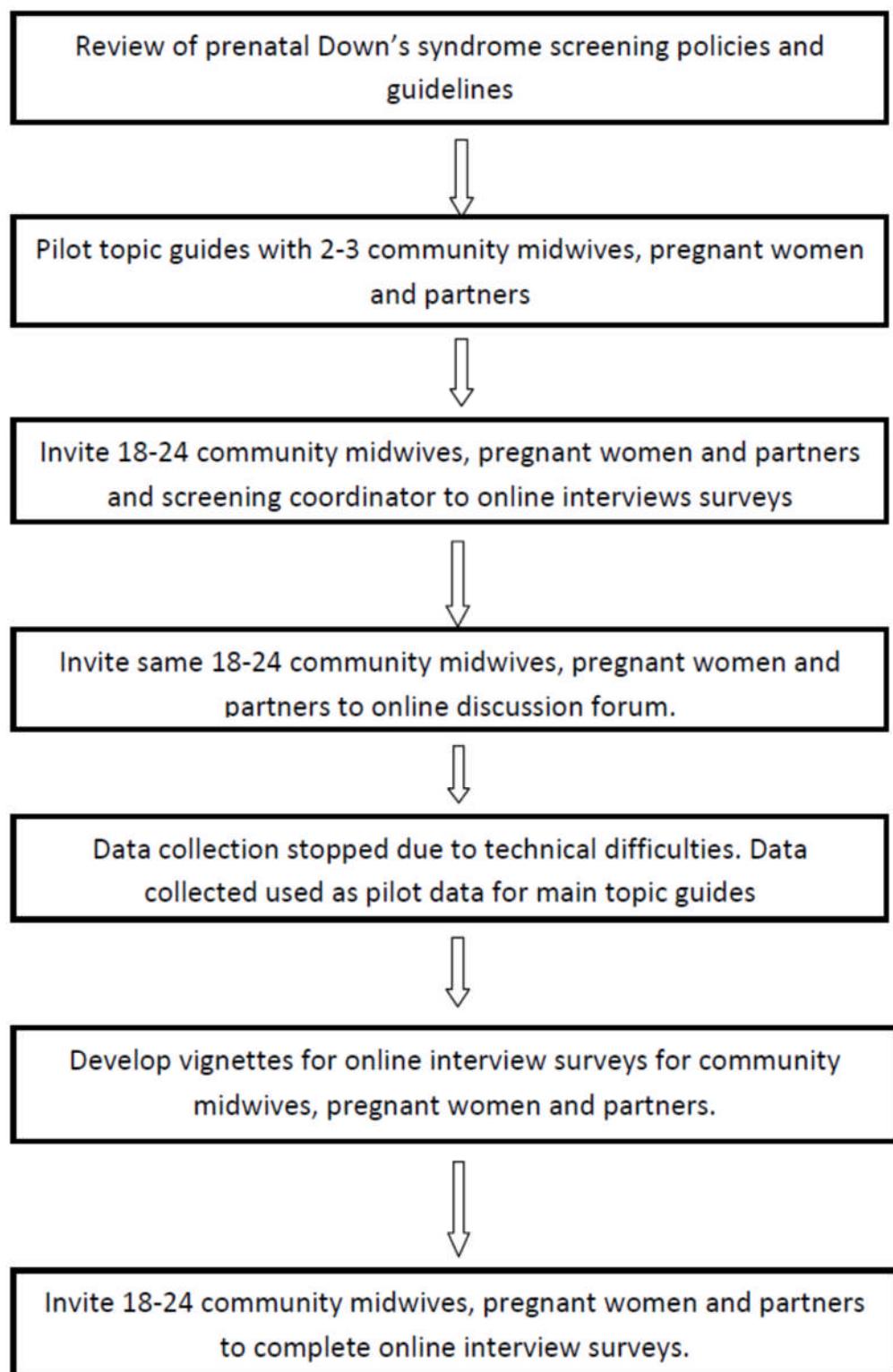
#### **4.14 Data collection for the main study**

Recruitment and data collection were conducted between November 2012 and February 2013 in both NHS Trusts. However, before the start of recruitment the researcher contacted the ISS to be granted access to the Bristol Online Survey (BOS) which is free to staff and students of the University of Leeds. Access was granted and the BOS was kept as a backup for the main online study.

##### **4.14.1 Main online interview with community midwives**

Community midwives who agreed to take part in the study were requested to sign-in at the website: <http://matserv.leeds.ac.uk/index.php>. This requires access to the internet and an email account. Email address and demographic details were requested during registration. By signing-in, midwives provided consent to participate in the study. The online interviews took about 30 minutes to complete and the midwives were expected to have an email account and access to the internet. They could log in at any time and place to respond to the online interviews. Questions were related to their experiences informing women and on contextual factors that may influence women and partners' participation in Down's syndrome screening (see appendix 8). Importantly, a web-based interview may be

Figure 6: Flow diagram for main data collection in each location



convenient for participants and removes the possibility of coercion, but it requires self-motivated individuals with access to online services.

It provided participants with a sense of privacy and confidentiality to disclose sensitive information about their experiences that may not be obtained in a group discussion (Lewis, 2003; Lambert and Loiselle, 2008). Responses were saved automatically to a web report data base accessible only to the investigator and academic supervisors in secure, firewall protected server of the University of Leeds.

#### **4.14.2 Main online interview with pregnant women and partners**

Pregnant women and partners who decided to take part in the study were requested to sign-in at the website: <http://matserv.leeds.ac.uk/index.php>. This required access to the internet, but they were told the researcher would be glad to introduce them to library internet services if they have no access at home to the internet. Demographic details and email addresses were requested during sign-in. The email address ensured the follow-up of participants who entered their demographic details without responding to the questions. This enhanced the integrity of the data obtained. Another reason is to have a summary of the study sent to participants on completion of the research. By signing in, women and their partners gave consent to access and complete the open-ended individual interviews lasting approximately 30 minutes depending on their views. Questions were related to the way in which Down's syndrome screening was offered to them, the information they received and on factors in the prenatal context that may have influenced their participation (see appendix 8).

#### **4.14.3 Field notes**

Field notes from the researcher's experience during recruitment, thoughts on the research and discussions with local screening coordinators were collected. The rationale is that it offered useful insights during document and interview data analyses and enhance the credibility of the data obtained as noted in previous literature (Emerson, 1995; Kahn, 1999).

#### **4.14.4 Trustworthiness of the main online study**

The basic issue in relation to trustworthiness is how the researcher can persuade the reader that the findings are credible and worth taking into account. Specific steps were employed to establish the trustworthiness of the data by ensuring credibility, dependability, confirmability and transferability as suggested in previous work (Lincoln and Guba, 1985; Baum, 2008).

#### **4.14.4.1 Credibility**

Credibility means confidence in the truth of the data. The inability to identify responders in online studies may affect the credibility of the data. This was avoided by the researcher recruiting the pregnant women and partners personally from the antenatal ultrasound department and the community midwives via the Local Collaborators. Additionally, participants' responses were text-based resulting in error free transcripts which were automatically generated. From a researcher perspective, the major advantage of the online interview emerged during the analysis process. The arduous and costly task of error prone transcription which is a major disadvantage in any traditional qualitative research was avoided in this study.

The anonymity offered by the online interview, reduces inhibitions among participants. Lower inhibitions decrease socially biased responses, facilitating open and honest responses which have previously been reported in online studies (Rezabek, 2000; Oringderff, 2004; Kenny, 2005; Reid and Reid, 2005; Cantrell and Lupinacci, 2007; Fox et al., 2007; Nicholas, 2010; Seale et al., 2010). Furthermore, the online interviews afforded participants anonymity, resulting in a sense of protection from the negative consequences of employers' and healthcare professionals' knowledge of their participation. The fact that participants could withdraw from the online study without giving reasons, adds to the credibility of the data obtained in this study. In addition, participants may choose to respond to the questions as much as they were comfortable with.

The semi-structured nature of the online interviews allowed the variations seen in the data to result from the variations in the concepts being explored (Holstein and Gubrium, 2001). The use of different methods and sources of evidence to triangulate the data enhanced the credibility of this thesis. Furthermore, the researcher looked for evidence from the data to support claims or conclusions made.

#### **4.14.4.2 Dependability**

A study by Fairweather et al. (2012) found that the themes derived from face to face interviews and online interviews were the same even though face-to-face interviews generated more words. The online interviews using vignettes with open-ended questions did not produce detailed in-depth responses as obtained in face-to-face interviews, but

provided the opportunity for a considerable range of responses to be obtained from participants.

Participants log on to individual web pages for the open-ended interviews at their convenience to give well considered responses. Besides, participants were informed not to discuss the interview questions with their colleagues to enhance diversity of views and the dependability of the findings. This was stated in the consent form (see appendix 6).

The social desirability effect that results from proximity to and potential pressure from interviewer or researcher was reduced in this study, owing to the anonymity afforded by the online interview enabling honest views of participant to be obtained (Montoya-Weiss et al., 1998; Griffiths, 2005; Tates et al., 2009; Nicholas, 2010). The study website required participants to sign in with an email address and provide demographic details, enhancing the dependability of the study. The findings, interpretation and recommendations supported by the data, enhanced the dependability of the study. The researcher also had prolonged engagement with the study sites and the data over long periods of time.

#### **4.14.4.3 Confirmability**

Confirmability refers to the neutrality of the data so that two or more independent individuals can reach an agreement about the data's relevance or meaning. The researcher's academic supervisors agreed with the identified themes and all stages of the research were assessed by them. The inability to achieve theoretical saturation with asynchronous online studies has been cited as one of the reasons for its lack of confirmability (Im and Chee, 2006). However, in traditional face-to-face interviews obtaining theoretical saturation is almost impossible owing to difficulties in establishing the point at which saturation is reached, as new data will always add something new (Mason, 2010). Providing an audit trail which involves thick description of the audit process such as sampling decisions and data collection procedures, field notes, analysis and interpretation enhanced the confirmability of the finding of this study. In addition, the audit trail included the experiences of the researcher during the study.

#### **4.14.4.4 Transferability**

Transferability was achieved in this study through the provision of sufficiently detailed information (thick, rich description) to allow readers to determine whether the findings are applicable to their settings. Transferability was promoted by paying careful attention to

describing both typical and atypical views expressed by the participants. In addition, there was a reasonably wide range concerning ethnicity, age, educational status and experiences among participants (Tables 3, 4, 5). It is also notable that the results of the online interviews be expected to vary depending on the prevailing routines of the particular maternity service. It is important to reiterate that the purpose of the qualitative multiple case study is not to generalise findings to the population, but of theoretical generalisation (Kohlbacher, 2006; Yin, 2009). Readers may have to judge its transferability to similar settings.

#### **4.14.4.5      Reflexivity**

The trustworthiness of the data obtained in this research was enhanced by the researcher being sufficiently reflexive about the study. The researcher was aware of his position at the centre of the online interview and understood his active role in the process of data generation and analysis. The potential influence of the researcher based on his background, gender, ethnicity, interest in the topic, on the data collection and analysis can be countered by an active process of reflexivity, which the researcher actively engaged in.

The study was relevant to the previous experience of the researcher as a medical practitioner and a lecturer in community health, but who has not practised in the United Kingdom. This background was helpful for the researcher to form an unbiased understanding of the context of prenatal Down's syndrome screening programmes and in the development and use of the online qualitative interview using vignettes.

Personal biases and values cannot always be bracketed when engaging in qualitative data analysis, hence, the researcher was a co-constructor of the meaning of the values and biases contained in the responses from participants as noted in Creswell (2009). Although, the researcher attempted to see himself as an investigator of midwives, pregnant women and partners' views and perspectives. This approach helped the researcher to reduce his influence on the data to a minimum and kept out his personal values and biases as much as possible during the data analysis.

The impact of the researcher's ethnicity on the response rates is not known, but it may have contributed to the diversity of ethnicity in the sample of women and partners that responded to the online interviews. Likewise, in an attempt to promote acceptance of this study and create rapport during recruitment, the researcher usually dressed smartly, with the NHS Trust ID badge displayed with the researcher's name and status as a temporary researcher boldly written on it.

#### **4.15 Data management**

The transcripts were printed directly from the study's web report database and anonymised by erasing email addresses and names of the participating NHS Trusts. Each anonymised transcript was given a pseudonym and identifier ID number. Moreover, the transcripts were copied from the web report database into a Word document and saved separately in the firewall, password protected, backed up University of Leeds computer server (PhD student M drive). Two sets of transcripts were saved. One set had the personal details of the participants which were saved separately from the second set of anonymised transcripts.

The anonymised transcripts were printed out for reading, re-reading and manual coding. The reason for working with the anonymised paper based method was the researcher's preference and the data obtained were not very extensive, so it was easy to use the manual methods. The manual method used was a combination of cutting and pasting into initial categories using a Word document, then printing out each category and colour coding to search for subcategories. A journal record that included the researcher's thoughts and emerging theories was maintained. The journal memos offered useful insights for data analysis during the writing up of the study and enhanced the credibility of the findings of the study (Emerson, 1995; Kahn, 1999; Lacey and Luff, 2007).

#### **4.16 Analysis of transcripts**

This involved the reading and re-reading of the complete transcripts enabling the researcher to gain an overview of the richness, depth and diversity of the data. It essentially involved the researcher's immersion in the data. Question and content analyses were conducted according to Morse and Field (1996), which has been used in previous research on exploring women's reasons for accepting or declining Down's syndrome screening in NI (McNeill et al., 2009). Qualitative content analysis is a method of analysis that may be used either in an inductive or deductive way. It has also been used in existing research to build models which closely fit the data and to describe a study's phenomenon in a conceptual form (Kohlbacher, 2006; Elo and Kyngäs, 2008). The unit of analysis of the interviews in both maternity services was the transcript of an individual interview (Graneheim and Lundman, 2004).

#### **4.16.1 Question analysis**

The semi-structured interview format with scenarios used in this study enabled participants to be asked the same questions throughout the course of the research. In this instance, all of the responses under each scenario were sorted into one category (theme). These initial four categories (experiences of participants, the influence of process, the influence of midwives and the influence of the environment on participation in screening) and the responses were then read and content analysis of this initial sort was conducted.

#### **4.16.2 Content analysis**

The analysis of data from both Trust sites (cases) entailed comparisons both within and between cases (Miles and Huberman, 1994; Strauss and Corbin, 1998; Green and Thorogood, 2004). The unit of analysis used was line-by-line coding with single words, phrases and sentences closely examined to give them labels known as meaning units as described by Graneheim and Lundman (2004) and Lacey and Luff (2007).

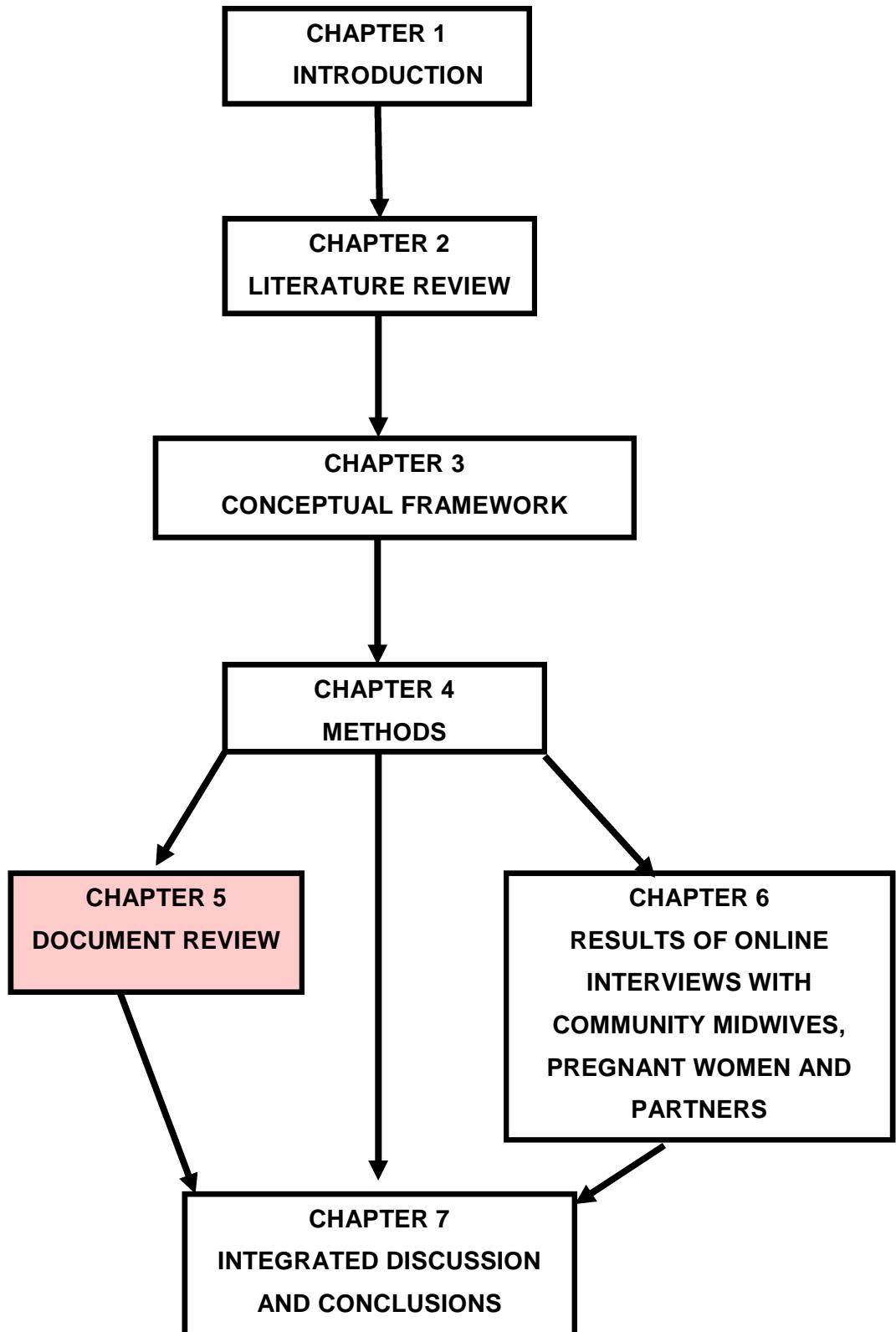
- 1) The entire interview was read and divided by topics into categories based on the conceptual framework for this study (analysis by topic). The topics derived based on the framework were among others the influence of structure, process and people in the prenatal context. Others were the experiences of participants, decision-making and reasons for decision. These topics then became broad primary categories (themes)
- 2) Large amounts of data (meaning units) were sorted into each category.
- 3) Categorises with few meaning units were combined until the categories had ample data.
- 4) The meaning units in the entire categories in the transcripts were grouped, re-grouped, labelled and re-labelled into subcategories or subthemes.
- 5) This resulted in a tree diagram with ‘types of’ the main category. For example;  
Category or theme: Influence of the structure of the screening programme on partner’s participation.  
Subcategory or subtheme: Consent in the context of dating scan.  
Meaning unit:  
*“I was at the 12 weeks scan when I was asked if I wanted the scan” ID105L*

*"I wasn't aware of the part where they measure fluid round the baby's head. I just thought it was a blood test. But it was a safe test and caused no harm to my partner and unborn child so I let it go ahead" ID106L*

- 6) After a few days the original transcripts were read without looking at the subcategories and categories.
- 7) The categories, subcategories and meaning units, were reconsidered; whether they were too small, large or too many. They were collapsed or subdivided and re-labelled as appropriate.
- 8) Again, after a few days, the subcategories and categories for all the transcripts were re-read to consider whether there were too many or too few subthemes and themes to make overall sense of the transcripts for the research question.
- 9) Previous instructions were revisited until satisfactory themes and subthemes were reached. The subthemes were quantitatively counted to identify patterns in the data as reported in previous work (Morgan, 1993; Babbie, 1998).
- 10) The relationships between themes including subthemes were explored and connections between them were made. Strauss and Corbin (1998) referred to categories (themes) as having 'analytic power', due to their potential to explain and predict. 'Constant comparisons' between collected data, subthemes, themes and initial findings help to crystallise ideas to become part of the emerging theory.

#### **4.17 Chapter summary**

The role of different internet browsers in accessing the research's web page should be taken into consideration when planning an online study. In addition, it is important that researchers have a back-up plan when considering using an online data collection tool. The challenge in conducting online interviews using vignettes is in getting participants to answer the questions in a manner that corresponds as closely as possible to their actual behaviour. For example, when informing or consenting to screening in the prenatal setting and not respond hypothetically to the scenarios or what they think to be the correct answers. The use of written and photo prompts in this study was invaluable in keeping participants focused on the context of prenatal screening when responding to the main open-ended questions.



## CHAPTER 5: DOCUMENT REVIEW

### 5.1 Introduction

Document review is a data collection method based on a systematic means for appraising documents. The documents may be hard copies or electronic (computer-based and Internet-transmitted) and may include reports, programme policies and guidelines and minutes of meetings. Like other qualitative research methods, document review requires that existing documents are evaluated and interpreted in order to elicit meaning, gain understanding and develop empirical knowledge (Corbin and Strauss, 2008; Rapley, 2008b; Bowen, 2009).

The rationale for the document review is presented in the first section; followed by the methods of document retrieval, data management and analysis. The findings were subsequently grouped into either those related to the national guidelines or those related to local screening guidelines. This is followed by a description of the structures and processes common to both maternity services and structures and processes specific to each maternity service. Subsequently, this is followed by a discussion of the findings. Lastly, the chapter summary is presented.

### 5.2 Rationale for conducting the Document review

We know little about how the national guidelines on screening for foetal anomaly including Down's syndrome screening are implemented in practice. This is a gap in the literature. The justifications for this research approach are that the researcher has not worked in the NHS, the concept of the maternity services is structurally complex and to illuminate the gap in the literature. The review

Adopting an ontological position rooted in constructivist realism allowed the reading or construction of aspects of the social reality of the programme through the guidelines and care pathways based on the Donabedian framework. These documents were rich in portraying the context of the study settings. Epistemologically, the documents are formal statements of the purpose of Down's syndrome screening programmes, but the review afforded a means to interpret them critically and explored how they are operationalised in practice. The ontological and epistemologically positions in qualitative research have been noted in Mason (2002).

The advantages of the use of documents are the availability of the documents prior to the start of the study and that they were written without the researcher's intervention, thereby eliminating 'researcher bias' (Silverman, 2011). The documents provided access to the procedures involved in offering Down's syndrome screening to pregnant women and their partners at booking, which may be otherwise difficult to directly observe. This is as a result of the difficulties with gaining access for direct observation due to the context, confidentiality and sensitive nature of the consultations. Document review may illuminate issues not reported by other research methods

Nonetheless, it is not possible to know from documents alone how the screening programme operates in practice. Firstly, the documents are not transparent representations of the routines and decision-making processes in the programme. Secondly, policies and guidelines do not take into consideration the practical issues encountered by service providers and users in the prenatal context. Thirdly, documents provided access to information/framework about offering screening not about how it actually happened. Importantly the documents reviewed may not have been written for the same purposes as this thesis and the data would be restricted to what already exists which may be incomplete, inaccurate or not current. Therefore, drawing conclusions from document review alone may not be possible. A limitation of document review is that it may be intensive and time consuming to retrieve and review several documents.

The findings from the document review were used as a means to verify or corroborate findings from the online interviews with midwives, pregnant women and partners. This is known as 'triangulation of data' which was defined by Denzin (2009, p.301) as "*the use of multiple methods in the study of the same object*". Triangulation entailed drawing upon multiple (at least two) sources of data to seek convergence and corroboration through the use of different data sources and/ or methods. Thus, the researcher corroborated findings from the online interviews and document review to reduce the effect of potential biases that can exist in a single study. Patton (1990) reported that triangulation helps to guard against the accusation that a study's findings were simply an artefact of a single method, a single source or a single researcher's bias. Indeed, triangulation increased the trustworthiness of the study's findings. Furthermore, Merriam (1988, p.118) stated: "*Documents of all types can help the researcher uncover meaning, develop understanding, and discover insights relevant to the research problem*" .

The overall aim of the document review is to explore how the national policies/ guidelines and care pathways are interpreted and operationalised in two maternity services with different uptake rates.

### **5.3 Methods**

Documents such as screening policies, guidelines and care pathways which aimed to inform the offer of prenatal Down's syndrome screening were identified, retrieved using internet searches and during field visits. They provided information about national and local guidelines, prenatal care and screening pathways. The socio-demographic, economic and epidemiological profiles of the population covered by the maternity services were also obtained from internet searches. The screening and care pathway documents were requested from the screening coordinators in two maternity services. However, the researcher could only obtain hard copies of the local care pathways for Down's syndrome screening from the coordinators. The researcher was directed to the UK National Screening Committee (UK, NSC) website. The UK NSC, including the Yorkshire and Humber UK NSC, the local NHS websites and Google were searched for reports, training resources, policies, guidelines and care pathways on prenatal Down's syndrome screening. A few documents were obtained from a hand search of retrieved documents. The field notes used as part of the document review were generated during the field visits through discussions with the antenatal screening coordinators regarding how the national screening policy was operationalised locally.

### **5.4 Data management**

The retrieved documents were sorted and coded manually as described in chapter 4 section 4.15. Hard copies of documents which identify the maternity services involved in this study were anonymised and kept in the researcher's locked drawer, in the secured University of Leeds PhD suite.

### **5.5 Settings**

For confidentiality, the first maternity service was given the pseudonym 'Terrace' maternity service (low uptake service) and the second maternity service studied was given the pseudonym, 'Landscape' maternity services (high uptake service). See subsections 5.9.2,

5.9.3 and chapter 4, section 4.6, on page 76 for a more detailed description of the two settings.

## **5.6 Analytic procedure**

The unit of analysis was written text based on census data, three reports, eight policies and guidelines, two care pathways (appendix 11) and the field notes (appendix 9) obtained during field visits, discussions and pilot interviews with the local screening coordinators (LCO) in both maternity services (see Table 2). The method of analysis was content analysis as described by Morse and Field (1996). The contents were analysed deductively into the broad categories of structure, process and people based on the adopted Donabedian framework. The documents were analysed inductively for meaning units which were added to the categories. The categories were further analysed inductively into subthemes or subcategories by combining the meaning units. The meaning units were derived from line-by-line coding of the documents (Graneheim and Lundman, 2004; Lacey and Luff, 2007). Comparisons, both within and between cases, were also undertaken (Miles and Huberman, 1994; Strauss and Corbin, 1998; Green and Thorogood, 2004). The analysis was conducted in steps as described in chapter 4, subsection 4.16.2, and page 100, but in step 9 the subthemes were not counted quantitatively in the document review.

## **5.7 Findings**

### **5.8 National screening policies and guidelines for antenatal screening**

#### **5.8.1 Structures of the antenatal screening programmes**

##### **5.8.1.1 Aims and objectives**

The national guidelines reported that the UK NSC screening programmes aim to ensure the establishment of firm quality assurance mechanisms to improve the quality of the results of the screening tests in use. The guidelines also ensured that the recommendations were implemented in all maternity services.

*"A detection rate (DR) of more than 90%, for a screen positive rate (SPR) of less than 2% (of affected pregnancies) for England for those undergoing combined screening. ....A detection rate (DR) of more than 75%, for a screen positive rate (SPR) of less than 3% (of affected pregnancies) for England for those undergoing quadruple screening"* (UK NSC, 2011, p.6)

**Table 2: Documents reviewed**

Types of document	Title of document	References
Field notes	Field notes	Field notes
Report	Annual report: Screening in England 2011-12.	(UK NSC, 2012)
Guideline	Maternity Matters: Choice, Access and Continuity of Care in a Safe Service	(Department of Health, 2007a)
Policy	British Medical Association: Consent tool kit	(BMA, 2009)
Policy	Fetal Anomaly Screening Programme: Consent Standard and Guidance	(NHS FASP, 2011)
Guideline	Antenatal care: Routine care for the healthy pregnant woman. Full guideline. National Institute for Health and Clinical Evidence.	(NICE, 2008)
Guideline	Prenatal genetic screening, prenatal genetic diagnosis and associated genetic counselling.	The Council of Europe (1990)
Policy	Clinical Negligence Scheme for Trusts: Maternity Clinical Risk Management Standards	(NHSLA, 2012)
Report	Antenatal and Child Health Screening in Yorkshire and the Humber. Status Report 2011/2012.	(Walker and Tarn, 2012)
Report	Health Profile 2010. Yorkshire and the Humber	(Department of Health, 2010)
Guideline	Reference guide to consent for examination or treatment. 2 <sup>nd</sup> edn.	(Department of Health, 2009)
Census	Ward Population Estimates, Mid-2011	(Office for National Statistics, 2013)
Policy	Screening for Down's syndrome: UK NSC Policy recommendations 2011–2014 Model of Best Practice	(UK NSC, 2011)

### 5.8.2 Access to antenatal care

Women could book directly with the midwives (direct access service) or with their GPs.

Women with uncomplicated pregnancies are offered care in midwifery-led service. Women who have experienced complications with previous pregnancies may choose to have care

delivered by a maternity team consisting of midwives, obstetricians and other specialists. For women with multidimensional social needs, care is provided in collaboration with other care services such as children's services, domestic abuse teams and mental health services. Information on self-referral and the need to see a healthcare professional early in pregnancy is publicly available. The national guideline recommends that women 'book' into prenatal care by 12 weeks and 6 days.

*"Enabling women and their partners to access midwifery services directly should mean that they enter the maternity care system in places and at times that suit them and at an earlier and more advantageous stage of their pregnancy"* (Department of Health, 2007b, p.13)

*"Developing maternity services in easily accessible and visible community facilities such as Sure Start Children's Centres is one way to engage with the most vulnerable families especially in disadvantaged areas"* (Department of Health, 2007b, p.14).

#### **5.8.2.1 Clinical environment**

The guidelines advise that the ambience of the clinical environment should be conducive to encourage open discussions between the midwives, women and their partners.

*The environment in which antenatal appointments take place should enable women to discuss sensitive issues such as domestic violence, sexual abuse, psychiatric illness and illicit drug use* (NICE, 2008, p.14).

#### **5.8.2.2 Frequency of antenatal appointments**

The number of visits for antenatal care for pregnant women is determined by their parity and previous obstetric and medical history. The guidelines identify an appropriate schedule of appointments for women with a healthy pregnancy. The appointments are more for women with a first pregnancy than women who have given birth previously. At first contact with a health professional, pregnant women are referred to or given an initial booking appointment with the midwife for comprehensive assessment and a subsequent care plan.

*"A schedule of antenatal appointments should be determined by the function of the appointments. For a woman who is nulliparous with an uncomplicated pregnancy, a schedule of ten appointments should be adequate. For a woman who is parous with an uncomplicated pregnancy, a schedule of seven appointments should be adequate"* (NICE, 2008, p.15).

### **5.8.2.3 Documentation of care**

Women are provided with a hand held note at booking. Any information given is documented in these notes. Verbal consent is obtained for any test conducted. The consent is recorded by the midwife in the hand-held notes and/ or IT system. If the test is invasive such as chorionic villus sampling (CVS) and amniocentesis, written consent may be required. If the woman declines screening or diagnostic testing, it is documented by the midwife in the hand-held notes and/ or IT system along with any discussion and advice given. The hand-held record provides complete, accurate and timely records of treatment and events in the management of the pregnant woman. The records are also central to any claims and complaints against the NHS Trust, for evaluating practice, demonstrating good care has been given and that the service has been proactive.

*"Structured maternity records should be used for antenatal care. Maternity services should have a system in place whereby women carry their own case notes...A standardised, national maternity record with an agreed minimum data set should be developed and used. This will help carers to provide the recommended evidence-based care to pregnant women"* (NICE, 2008, p.70).

### **5.8.2.4 Local leadership in screening programme**

The NSC guidelines advise that Trusts should have an LCO that has an integrated prenatal and new-born screening role for appropriate continuity and to fully exploit all resources for the risk management of the screening programmes. The LCO functions as expert adviser and monitors performance and take-up rates of screening. The screening coordinator is usually supported by a screening midwife. In addition, all midwives must have a supervisor of midwives (SoMs). The SoMs superintend the role of midwives to ensure that the appropriate standard of care is provided to all women. SoMs also meet regularly with midwives to guide and support them in developing their skills and expertise.

*"It is imperative that organisations have good leadership, within an open and supportive culture which will provide the foundation for good maternity services that can fulfil the needs and expectations of women and their families. Organisations will need to consider the level of investment required to build and enhance leadership that will also support job satisfaction and staff morale"* (Department of Health, 2007b, p.24).

### **5.8.2.5 Midwife's role**

The role of midwives described in the screening programme guidelines is to provide information about the potential benefits and risks of screening to women and their partners and ascertain that they understand they have a choice. The midwives are responsible for ensuring that women know what tests are available, when they are conducted and how results will be communicated and acted upon to avoid confusion. Screening for Down's syndrome is usually undertaken by the community midwife at the appropriate time following discussion with the women and their partners (when present). However, the test can only be conducted with consent from the woman.

*"At each antenatal appointment, midwives and doctors should offer consistent information and clear explanations and should provide pregnant women with an opportunity to discuss issues and ask questions"* (NICE, 2008, p.339).

## **5.8.3 Processes involved in offering antenatal screening**

### **5.8.3.1 Booking visit**

This is the first antenatal appointment with the community midwife usually at 10 weeks of pregnancy. This provides enough time to arrange ultrasound scans and screening tests to take place at the appropriate stage of pregnancy. The booking appointment is usually of a longer duration than routine appointments, due to the comprehensive information and assessment needs in early pregnancy. The booking appointment usually takes place in the community. 'Booking' also offers women and partners the opportunity to discuss any concerns, such as if they need extra support.

Women are offered verbal information such as on nutrition, diet and exercises in pregnancy. The pregnant woman's height, weight (BMI calculated) and blood pressure are measured. Routine screening tests, such as blood group and rhesus D status are undertaken at the visit. Women are also offered screening for Down's syndrome (combined test for first trimester screening and the quadruple test for second trimester screening) and ultrasound scans for dating. The foetal anomaly scan is arranged following the dating scan.

*"Wherever possible, appointments should incorporate routine tests and investigations to minimise inconvenience to women"* (NICE, 2008, p.15)

*"The gestational age window for combined test starts from 10 weeks + 0 days to 14 weeks + 1 day in pregnancy. The quadruple test window starts from 14 weeks + 2 days to 20 weeks + 0 days. A maternal blood sample is required for the analysis of human chorionic gonadotrophin (hCG), alpha-fetoprotein (aFP), unconjugated oestriol (uE3) and inhibin-A. This test has been retained in this policy because there will always be women who book too late in pregnancy for combined testing (about 15% of the pregnant population) and wish to have screening" (UK NSC, 2011, p.6).*

#### **5.8.3.2 Informing women about Down's syndrome screening**

The guidelines state that all maternity services must have a care pathway to provide evidence that the approved booklet and leaflets by NSC and NHS FASP are being used. All pregnant women must be offered up-to-date information on screening based on current evidence in addition to the approved screening booklet and leaflet. Pregnant women should be offered testing with the offer documented in the hand held note and/ or IT system. Healthcare professionals involved in prenatal Down's syndrome screening should have the expertise and adequate knowledge to communicate information to women about screening tests for foetal anomaly including Down's syndrome and their implications. In general, the information is expected to be objective and adapted to the women and their partners' circumstances. Written information, such as the 'Screening tests for you and your baby' booklet (National Screening Committee, 2012) that supports the relevant verbal information, is given to women to take home and read.

*"Give information (supported by written information and antenatal classes), with an opportunity to discuss issues and ask questions" (NICE, 2008, p.30).*

*"Good communication between healthcare professionals and women is essential. It should be supported by evidence-based, written information tailored to the woman's needs" (NICE, 2008, p.37).*

*"Women and their partners must have access to unbiased information which includes benefits, risks and alternatives (as appropriate) in order to make an informed choice regarding their care and treatment in line with national guidance" (NHS LA, 2012, p.116).*

### **5.8.3.3 Procedures and content of information about Down's syndrome screening**

The Council of Europe (1990) recommends that antenatal screening for congenital abnormality should include the following topics or procedures: (1) counselling before and after screening and individuals should be told the test is optional (2) the healthcare professional must adopt nondirective counselling (3) autonomous and informed consent must be obtained (4) if the decision-making capacity of the individual is impaired, consent should be obtained from the legal representative (5) information about the purpose, nature and risks of the test should be given (6) in the case of a serious genetic inherited disorder, preconception information should be available.

The topics to be covered at booking and in the antenatal classes should include the following: (1) the rationale, advantages and disadvantages of foetal anomaly screening including Down's syndrome screening and diagnostic tests for higher risk results (2) information about the ultrasound scans for dating and for foetal anatomy (3) information about the choice of continuing or terminating a pregnancy in the event of a diagnosis of a genetic condition (4) information about the screening test offered locally, procedures and timing involved and that the tests are optional (5) the psychological and clinical impact of the test results and the choices that have to be made (6) the screening pathways for women who accepted or declined screening (7) information about the various conditions that could be detected by screening including Down's syndrome and the limitations of screening (8) balanced and accurate information about diagnostic tests, such as what they are, procedures and associated risks, other conditions detected, and that the tests might be repeated and associated risk, care after tests, results and options available.

The local contents of the information provided to women as seen in the checklists or aide memoir for midwives in the two maternity services were: (1) that they have received and understood the 'Screening for Down's syndrome' information booklet (2) scan is normally trans-abdominal and takes 15-20 minutes to perform. Occasionally, it may be necessary to perform a transvaginal scan in 5% of cases (3) the gestational age is calculated using the crown rump length (CRL), from the top of baby's head to the tail of its bottom (4) the Down's syndrome risk is calculated using maternal age, CRL, nuchal translucency (NT) measurement and blood test taken in the first trimester (5) a screen positive result is a numerical result of 1:150 or greater and a screen negative result if less than 1:150 (6) the combined test

provides a detection rate of 85% and false positive rate of 3%. The detection rate and false positive rate will alter with maternal age (7) CVS and amniocentesis are diagnostics tests. If a screen positive result is received and the couple are considering diagnostic testing, the couple will be seen by a midwife experienced in counselling for diagnostic testing prior to referral for the test (8) Foetuses with a Nuchal translucency >3.5mm and with normal chromosomes, have an increased risk of a structural anomaly such heart defects at a later stage in pregnancy. An appointment will be made for a scan of the baby's heart (9) an early scan could detect 40% of major structural abnormalities such as anencephaly. Any detected abnormality will trigger a referral to the Foetal Medicine Unit.

*"Specific information should include: • the screening pathway for both screen-positive and screen-negative results • the decisions that need to be made at each point along the pathway and their consequences • the fact that screening does not provide a definitive diagnosis and a full explanation of the risk score obtained following testing • information about chorionic villus sampling and amniocentesis • balanced and accurate information about Down's syndrome. If a woman receives a screen-positive result for Down's syndrome, she should have rapid access to appropriate counselling by trained staff" (NICE, 2008, p.178).*

#### **5.8.3.4        Women's understanding of information on Down's syndrome screening**

The NICE guidelines advise that the information provided to all pregnant women about Down's syndrome screening should be clear about the tests and the options available. Health professionals must use simple and consistent language when informing women about screening and should find out if the woman understands. Women with additional needs, such as the physically and mentally challenged, should be given information in a format that is accessible and easy to understand. Pregnant women, who cannot speak, read or understand English language should be provided with an interpreter.

*"Give information, with an opportunity to discuss issues and ask questions; offer verbal information supported by written information (on topics such as diet and lifestyle considerations, pregnancy care services available, maternity benefits and sufficient information to enable informed decision-making about screening tests)" (NICE, 2008, p.78).*

### **5.8.3.5 Good interpersonal relations**

In the guidelines, psychological support and an empathic relationship between the midwives, women and partners were seen as an important part of prenatal care. The models of care adopted in the guidelines and care pathways were meant to promote and enhance relationships through continuity of care.

*“Having the time to talk, engage and build a relationship with women and their partners to understand and help meet their needs throughout pregnancy and afterwards”* (Department of Health, 2007b, p.16).

*“Antenatal care should be provided by a small group of carers with whom the woman feels comfortable. There should be continuity of care throughout the antenatal period”* (NICE, 2008, p.69).

### **5.8.3.6 Support for women and their partners**

Providing support to pregnant women and their partners was seen as important in the guidelines and care pathways to enable women to make informed choices and feel comfortable with antenatal screening. The relationships between the midwives and the women formed in the prenatal context form the basis of the support for women and their families during the prenatal period.

*“Every opportunity should be taken to provide the woman and her partner or other relevant family members with the information and support they need”* (NICE, 2008, p.37).

### **5.8.3.7 Autonomy of women**

The NICE guidelines state that in antenatal screening, all pregnant women should be given information that is clear, consistent, balanced and easily understood to enable informed decisions. To protect the autonomy of pregnant women and partners, the guidelines further advise that couples should be given the opportunity to discuss concerns about screening and ask questions. Their choices and decisions should be respected even when contrary to those of the health professionals. Therefore, in the context of Down's syndrome screening, competent pregnant women are expected to make choices and decisions based on their beliefs, values, religion, attitudes and interests without influences that determine their participation.

*“Women, their partners and their families should always be treated with kindness, respect and dignity. The views, beliefs and values of the woman, her partner and her family in relation to her care and that of her baby should be sought and respected at all times. Women should have the opportunity to make informed decisions about their care and treatment”* (NICE, 2008, p.37).

#### **5.8.3.8 Informed choice and consent for screening for Down’s syndrome**

Informed choice is a process that includes information-giving, discussion and decision-making. Making a choice and giving consent are not done only during the booking appointment, but throughout the process of Down’s syndrome screening testing in England. If a pregnant woman declines first trimester screening at the booking appointment, the dating and foetal anomaly ultrasound scans are still offered and the woman has the option of accepting second trimester screening if she changes her mind. However, the NICE guidelines advise that the 20 week foetal anomaly scan should not be used to routinely screen for Down’s syndrome. The NSC guidelines state that only the pregnant women have the right to consent to screening. A partner or other family members cannot consent on her behalf. The pregnant woman has the right to withdraw her consent at any time and consent is obtained by a range of health professionals at every stage of the Down’s syndrome screening process. At every stage, the health professionals confirm that pre-screening information has been received and documented. If information has not been given, the woman is referred to the appropriate healthcare professional for pre-screening information. The pregnant woman’s decision is recorded in her hand-held notes and/ IT system. In England, consent is valid when the decision-making capacity of the pregnant woman is not impaired and the pregnant woman is 16 years old or over. Provided there is no impairment in her capacity to understand fully what is involved in screening and is able to retain information, a pregnant woman under the age of 16 years may be able to give consent.

*“Informed decision making involves making reasoned choice based on relevant information about the advantages and disadvantages of all the possible courses of action (including taking no action). It requires that the individual has understood both the information provided and the full implications of all the alternative courses of action available. In providing information for women antenatally it is important that healthcare professionals are aware of what informed choice entails and that they provide information in order to facilitate this. The provision of clear information, and time for women to consider decisions*

*and seek additional information, as well as the need for care to be provided in an individualised, woman-focused way” (NICE, 2008, p.38)*

#### **5.8.3.9 Validity of written or verbal consent for prenatal screening**

The national guidelines reported that the validity of consent does not depend on whether consent was given verbally or in written format. Written consent will not demonstrate informed consent if the criterion of choice, being voluntary without pressure or undue influence, was not met. Such pressure can come from partners or family members, as well as healthcare professionals. Midwives are advised to be alert to this possibility and, where appropriate, should arrange to see the pregnant women on their own in order to establish that the decision was truly their own. In addition, a signature on a form does not make the consent valid if the criteria of adequate information and understanding of information by the pregnant woman have not been met.

*“Consent may be expressed verbally or non-verbally: an example of non-verbal consent would be where a person, after receiving appropriate information, holds out an arm for their blood ... to be taken. However, the person must have understood what examination or treatment is intended, and why, for such consent to be valid. It is good practice to obtain written consent for any significant procedure, such as a surgical operation (even if only minor procedures are involved)” (Department of Health, 2009, p.17).*

#### **5.8.4 Decision-making and consent for pregnant women with learning disability**

The informed decision-making capacity of a pregnant women is impaired and valid consent cannot be given if she does not meet one or more of the following criteria: (1) understand relevant information about screening (2) remember the information long enough to decide (3) process or apply value judgement on the information as part of the decision-making process (4) convey the decision reached by speaking or using sign language. This also includes the use of simple muscle movements such as blinking an eye or squeezing a hand. To give informed consent, the women need to understand the nature and purpose of screening. Hence, women with learning disability may not be able to consent to screening.

The UK NSC national guidelines state that midwives who are concerned about the competency of a pregnant woman with learning disability, to consent, should seek advice from more experienced midwives. In the UK, no one can give consent to screening for a pregnant woman 16 years or over who has learning disability or lacks the capacity to give

consent, unless they have the legal authority under a Lasting Power of Attorney or as a court appointed deputy. Indeed, parents, relatives or healthcare professionals cannot consent on behalf of an adult who lacks capacity for decision-making in most cases. The Mental Capacity Act sets out the criteria for health professionals to lawfully conduct screening on women that have learning disabilities (BMA, 2009; NHS FASP, 2011).

The Mental Capacity Act (2005) provides healthcare professionals with immunity from civil and criminal legal liability for acts or decisions made in the best interests of service users who lack capacity. The Mental Capacity Act explicitly states that a woman's best interest should not be judged by healthcare professionals on the basis of the woman's age, appearance, condition or any aspect of the woman's behaviour. Furthermore, the health professional should consider the following: (1) the possibility of the pregnant women regaining capacity and thus the decision being delayed (2) the importance of involving the pregnant woman as much as possible in the decision-making process (3) any stated or written past, present wishes and feelings of the woman (4) her religious or moral values and beliefs in addition to any special circumstances that may sway the decision if she were able to decide (5) significant others, such as court appointed individuals and carers who should be consulted and their views taken into consideration in the decision-making. The decision-making process should be documented in the hand-held notes/ IT system to demonstrate that all evidence and conflicting views have been taken into account.

*"The Mental Capacity Act 2005 came fully into force in October 2007 and applies in England and Wales to everyone who works in health and social care and is involved in the care, treatment or support of people over 16 years of age who may lack capacity to make decisions for themselves"* (Department of Health, 2009, p.23).

### **5.8.5 Service user's involvement in Down's syndrome screening**

#### **5.8.5.1 Involvement of partners**

The various policies and guidelines, advise that male partners of pregnant women should be encouraged to be involved in prenatal screening.

*Encourage partners to be involved in counselling sessions* (Council of Europe, 1990, p.1).

## **5.9 Local Down's syndrome screening policies and guidelines**

### **5.9.1 Common structures and processes for screening in both maternity services**

The national policies, guidelines and care pathways are implemented locally in both maternity services. Information about Down's syndrome screening is offered by midwives at booking to women and their partners (when present) usually in the community. Women who present on or before 13 weeks gestation are offered the combined screening test and those who booked late, usually after 13 weeks, are offered the quadruple test. The NT and dating scans are conducted in the hospital's antenatal ultrasound department by sonographers whilst, blood for the serum biochemistry is collected after the NT and dating scan. Women and partners who present for the Down's syndrome screening test and need more information or lack understanding of the purpose of Down's syndrome screening are seen and given information by the hospital midwives. Consent is obtained before screening. All decisions are recorded in the woman's hand-held notes/ hospitals IT system (see appendix 10 for care pathways for Terrace and Landscape maternity services).

#### **5.9.1.1 Structure of the screening programmes**

Each setting has community midwives who offer screening to pregnant women, usually at booking in the community. There are antenatal screening coordinators in both settings responsible for the smooth running of the antenatal screening programmes. The regional screening coordinator oversees all prenatal screening services in the region and a regional training coordinator who is responsible for prenatal screening staff training. The maternity services in both settings operate a caseload system and team midwifery care models. In caseload midwifery, women are allocated a midwife who provides care throughout pregnancy, labour, birth and postnatally until care is transferred to the health visitor. In the team midwifery model, women are cared for by a group of midwives, but with each pregnant woman having a named midwife. However, during the course of her pregnancy, care may be provided by other members of the midwifery team.

*"Maternity services should ensure optimum skill mix is in place and staff at suitable levels, with relevant skill sets, to undertake appropriate tasks" (Department of Health, 2007b, p.43).*

### **5.9.1.2 Staffing levels**

The field notes revealed that Terrace maternity service has 84 community midwives and Landscape maternity service has 32 community midwives. The national guidelines recommended appropriate staffing levels to ensure that pregnant women have continuity of care with midwives that they know and trust. Further, the guidelines advise that maternity services should have arrangements in place for coordination of on-going midwifery support, should the known midwife not be available.

*“Sufficient numbers of midwives and support staff working flexibly across community and settings”* (Department of Health, 2007b, p.35).

### **5.9.1.3 Staff education and training**

The findings from the field visits and interviews with the antenatal screening coordinators in both settings revealed community midwives participate in yearly mandatory training and educational days. Midwives have telephone access to the screening coordinators for support and team leaders in the community via mobile and landlines and they are sent regular bulletin updates. There are monthly meetings with one representative from the community whose role is to act as a communication conduit between coordinator and community midwives. However, the community midwives could make an appointment with the LCO whenever necessary. Each month the prenatal Down’s syndrome laboratory request forms are audited to check if midwives are completing the laboratory request forms properly.

Educational updates may then be given based on the results of the monthly audit and audit or surveys of pregnant women seen by the midwives to ensure accurate information is being given. All midwives complete the UKNSC continuing professional development e-learning package annually as part of their mandatory training. The online training resources include topics in informed choice, parent perspective on screening, getting the best from the consultation, understanding and communicating risk and understanding genetics. Midwives are also expected to complete a questionnaire at the end of their mandatory week to provide feedback on the training and their needs. Midwives are kept fully updated and changes to service are cascaded to all midwives by the LCO. Midwives are advised to pass on the contact details of the LCO to pregnant women and their partners when necessary for further information.

*"The UK NSC looks for cost effective ways of facilitating training, for example by providing screening information for use in general training for midwives, nurses and doctors, getting recognised bodies to deliver accredited courses at a variety of levels, working with professional bodies and developing eLearning courses and information to enable more people to access suitable training in a flexible way"* (UK NSC, 2012, p.19).

### **5.9.2 Structures and processes specific to Terrace maternity services**

Terrace maternity service is located in an urban setting in the North of England. It is a large maternity service in a designated NHS teaching hospital Trust. The population is heterogeneous, with the white British ethnic group making up 89.2% of the total population. It is an area of low to moderate social disadvantage, with deprivation not significantly worse from the regional average of 27.2%. The level of deprivation is 28.6% (95%CI 28.5-28.7). There were 10,357 bookings in the maternity services between 2011 and 2012. The introduction of first trimester screening into the maternity services had brought about a 54% increase in the uptake rates for Down's syndrome screening.

There are subtle differences in the operationalisation of the national screening guidelines in Terrace maternity service compared to Landscape maternity service. Information and the offer of Down's syndrome screening are presented by midwives at booking. Consent is obtained from women who agree to screening at booking and an appointment is made for the screening test. For women who are undecided, screening is recommended by the midwives and an appointment is made for the screening test with their 'consent'. Nonetheless, if the women decide to decline screening later, then they have to inform the sonographers of their decisions at the dating scan.

### **5.9.3 Structures and processes specific to Landscape maternity service**

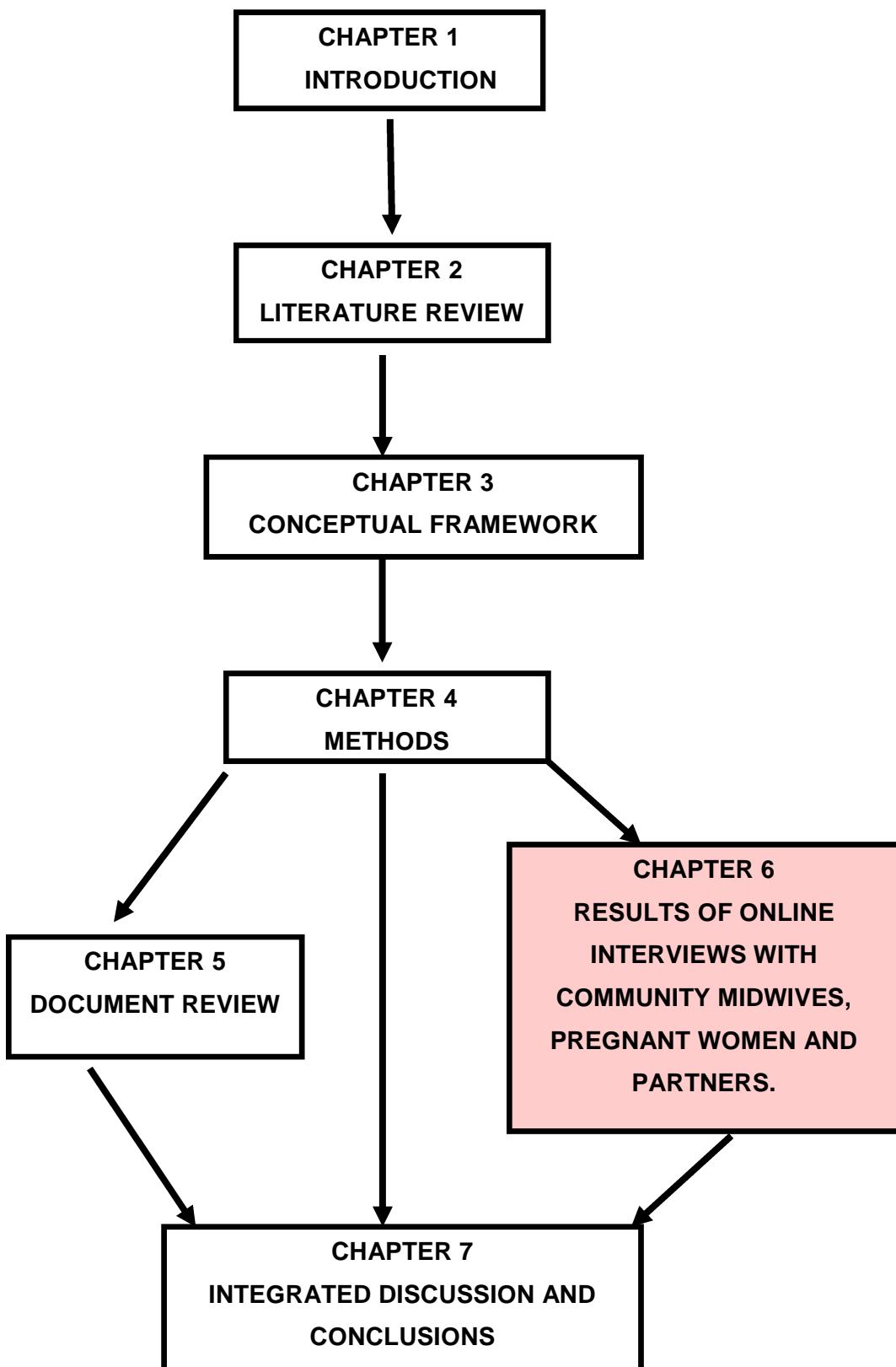
The maternity service is located in a peri-urban setting in the North of England. It is designated as a Foundation hospital. The setting is an area of social disadvantage with deprivation significantly worse than the regional average of 27.2%. The level of deprivation is 32.4% (95%CI 32.2 - 32.6). There were 3225 bookings in the maternity service between 2011 and 2012. The introduction of first trimester screening into the maternity service brought about an 85% increase in the uptake rates for Down's syndrome screening.

There are subtle differences in the implementation of the screening guidelines when compared to the Terrace maternity service. Information about the combined screening test

is presented to women at booking. The midwife does not request a decision on whether or not the woman wants to screen until her antenatal clinic appointment for the dating scan. Consent for Down's syndrome screening is obtained from women at the dating scan in the hospital antenatal ultrasound unit by the sonographers. Furthermore, women who decline the combined screening test are offered the quadruple test when they present for the 15 to 16 weeks appointment where results of routine tests are discussed.

## **5.10 Chapter summary**

This chapter described the data obtained from the review of national and local policies, guidelines and care pathways in two maternity services with different participation patterns. It provided insight into an array of service contextual factors involved in the Down's syndrome screening programme. Overall, the review of the policies and guidelines revealed influences and contradictions within these official documents which could undermine women's, partners' and midwives' autonomy and affect participation in screening. The analyses of the field notes, local guidelines and care pathways revealed nuances in the operationalisation of the national guidelines at the level of the maternity service. The nuances in operationalisation of the national guidelines may account for the difference in the uptake rates of screening seen between the two maternity services. In addition, the various programme policies, guidelines and care pathways may be made up of rhetoric of informed choice, support and relationships. These findings are summarised in table 6, page 155 and will also provide context for the final discussion in chapter 7.



## **CHAPTER 6: RESULTS OF ONLINE INTERVIEWS**

### **6.1 Introduction**

This chapter presents results of the online interviews using vignettes with open-ended questions and prompts that explored the influence of service organisation and delivery on participation in screening from the perspectives of midwives, women and their partners. The findings are important as they may advance knowledge and inform policy. The findings are reported in three sections; the first section is the midwives' online interviews, the second section is the pregnant women's interviews and the third section is on the partners' online interviews. This is followed by an overview of the findings that includes a comparison of the findings from the three groups. Finally, the overall patterns on the findings of the influence of service organisation and delivery obtained from the document review and the online interviews are presented in tables 6 and 7.

### **6.2 Online interviews with midwives**

#### **6.2.1 Findings**

Thirty-four community midwives participated in the study, 15 midwives in Terrace maternity service (T) and 19 midwives in Landscape maternity service (L). The age groups, ethnicity, length of time qualified and length of time working as community midwives are presented in Table 3.

#### **6.2.2 Response to online interviews**

All community midwives in both maternity services were invited to participate in the study via invitation letters. Fifteen midwives responded in Terrace maternity service giving a response rate of approximately 18%. In Landscape maternity service, 19 community midwives responded with a response rate of 59%. The reason for the marked difference in response rate is unknown. All communication with the midwives was through the Local Collaborators. Although, a gatekeeper reported that midwives were often very busy, the research was secondary to their roles and midwives have limited access to the internet in the community.

**Table 3: Midwives' demographic profile**

Age (years)	Terrace (n=15) n (%)	Landscape (n=19) n (%)
21-25	2 (13.3%)	1 (5.3%)
26-34	1 (6.7%)	5 (26.3%)
35-44	10 (66.7%)	13 (68.4%)
45-54	2 (13.3%)	-
Total	15 (100%)	19 (100%)
Ethnicity:		
White British	15 (100%)	17 (89.4%)
White Irish	-	1 (5.3%)
Other White background	-	1 (5.3%)
Total	15 (100%)	19 (100%)
Length of time qualified as midwife*		
0 -10 years	2 (13.3%)	5 (26.3%)
11 - 20	1 (6.7%)	5 (26.3%)
21 - 30	8 (53.3%)	6 (31.6%)
31 - 40	3 (20%)	2 (10.5%)
Missing values* ID174T &175L		
Total	14 (93.3%)	14 (94.7%)
Length of time working as community midwife:		
6 months – 2 years	4 (26.6%)	7 (36.8%)
2 years - 5 years	10 (66.7%)	11 (57.9%)
5 years - 10 years	1 (6.7%)	1 (5.3%)
Total	15 (100%)	19 (100%)

### **6.2.3 Midwives' experiences with offering Down's syndrome screening**

#### **6.2.3.1 Midwives struggle to inform women**

Midwives in both maternity services reported struggling to inform women about screening due to the volume and type of caseload and depth of information provided.

*"I have a very large caseload (160) but I try hard to ensure that all relevant info is given"*  
*ID174T.*

*"I.... strive to give all women the same depth of information," ID181T.*

*"Difficult to give a thorough explanation mainly due ...caseload of 90-110. Large numbers of caseload are women in a low socio-economic situation" ID195L*

#### **6.2.3.2 Time constraints**

Most midwives in Terrace maternity service, 80% (n=12) and 21% of midwives (n=4) in Landscape maternity service reported time constraints at booking as the main barrier to adequately informing women about screening. The time constraint was exacerbated by women from minority ethnic groups who required interpreters. In many cases the midwives were meeting the women and partners for the first time.

*"Time restraints are always a problem I feel there is not enough time to give all the information necessary to help them make an informed choice at the first appointment where they have to decide whether they want Down's screening" ID211T*

*"Sometimes feel that due to time restrictions and depth of information the clients do not get the chance to discuss their feelings" ID 188L*

*"It is not easy to fully explain the test in the given amount of time" ID194L.*

*"I am concerned that in a busy clinic there may be a temptation to give written info but not then explaining it to the women" ID206T*

Some midwives 13% (n=2) in Terrace maternity service and 32% (n=6) in Landscape maternity service reported having adequate time to inform and offer Down's syndrome screening to women. They explained there was no restriction on the duration to inform women about screening.

*"I work as part of a case loading team with the specific aim of reducing infant mortality. One benefit of this is that we conduct the booking appointment (most commonly) in the woman's home and we can take as little or as much time as the woman likes/needs. Therefore, I do not feel under any pressure to rush my discussion around screening". ID170T*

*"I give the latest screening information as per national screening guidelines. I discuss the NT scan and blood test for 1st Trimester and the blood test for 2nd Trimester. There is enough time allocated to provide the relevant information at booking". ID184L*

A midwife in Landscape maternity service felt that the time factor was irrelevant during the visit, as midwives should be able to offer Down's syndrome screening within the allocated duration of a booking appointment.

*"The time allocated is irrelevant it still has to be discussed and a leaflet given". ID193L*

#### **6.2.3.3 Coping with time pressure**

A midwife in Landscape maternity service aptly described how midwives cope with time pressure by keeping the information to the barest minimum and giving the same information to all women.

*"I discuss choice and timescales of 11-14 weeks. I point out the section to read in the NSC leaflet, explain about high risk and low risk and how they will be contacted if they are high risk. I keep the information basic and give them the leaflet as there can be information overload at booking...I give the same information to all women and explain and outline the procedure" ID197L*

#### **6.2.3.4 Information related anxiety**

One midwife in Terrace maternity service reported that some women look terrified when informed about the possibility of their baby having the condition.

*"The women look a little shocked to be given this information". ID181T*

#### **6.2.3.5 Women's lack of understanding of information**

Midwives in both maternity services, Terrace 40% (n=6) and Landscape 37% (n=7) believed that women lack understanding regarding the information about screening.

*"I think screening is an important subject but I am not always sure if all the women totally understand what they are saying yes or no to". ID175L*

*"It is my experience that it is at my first meeting with the couple that this occurs and it is a very complex full appointment. Consequently I do wonder if all the information I try to portray has been understood fully". ID210T*

However, 20% (n=3) of midwives in Terrace and 16% (n=3) of midwives in Landscape maternity services, felt many women seen at booking have a good understanding of the information given about screening.

*"I think most of the women understand the information given and are able to make an informed choice". ID212T*

#### **6.2.3.6 Educational/training needs**

A midwife in Terrace maternity service reported concerns about unmet training needs for midwives taking part in the programme.

*"Midwives also need the skills to get women to repeat back information in order to check understanding, this is not taught". ID181T*

#### **6.2.3.7 Fear of Litigation**

A midwife in Terrace maternity service stated that when women decline information about Down's syndrome screening at booking, they were made to document their refusal by signing the consent form.

*"If they do not wish me to impart the information I get them to sign they have declined in case later they said I did not offer them the information!" ID204T*

#### **6.2.3.8 Stereotypical beliefs among midwives**

Five midwives in Terrace maternity service believed that women, particularly those from ethnic minority groups decline screening for cultural reasons

*"Many of the women I look after are migrants, who don't speak English or struggle with complexities of the English language...many women decline the test because of cultural preferences" ID 191T*

*"The majority of my caseload decline screening on the basis of their cultural beliefs" ID 171T*

*"Many women decline the test because of cultural preferences". ID 191T*

### **6.2.4 Influence of the structures of screening programme on women's participation in Down's syndrome screening**

#### **6.2.4.1 Screening in the context of a scan**

A midwife reported that since the combined screening test was conducted with the dating scan, take up of screening has increased.

*"As it is offered with the dating scan I have seen a massive increase in uptake". ID207T*

#### **6.2.4.2 Consent required at booking**

Midwives reported obtaining consent for screening at booking. Yet, midwives in Terrace maternity service explained that women who are undecided during the visit, had screening recommended to them, due to the short window of opportunity. If the women changed their minds, they have to either inform the sonographer during the dating scan or destroy the request form.

*"Due to the implementation process a decision is required immediately/at referral therefore if there is some indecision it is more common to recommend screening and decline later than to postpone decision consequently the majority would probably just continue with the screening". ID210T*

*"They can be quite disconcerted by the request to make a decision as it may not have entered their thinking as being a possibility that they could be at risk of having a baby with Down's syndrome". ID211T*

*"Some are not sure at the first interview, so I complete the paperwork for the test, and advise them to let the sonographer know if they do or do not want to do the nuchal fold measurement." ID205T*

#### **6.2.4.3 Influence of the clinical environment**

Several midwives in Terrace maternity service, 40% (n=6) and 63% (n=12) in Landscape maternity service, believed that the clinical environment has no influence on women's decision-making processes. Some of the midwives believed the attitude of the midwife and how the information was given, had the most influence on women and partners' decision-making processes.

*"No, they are used to this environment, it is the NHS not BUPA!" ID204T*

*"No. I think it's the midwife and her manner that matters more than the environment. I don't wear uniform, and a number of other community midwives don't wear it". ID205T*

*"Clinical environment in my opinion will not affect decisions concerning participating in Down's screening. It depends on the quality of information given and the quality of the person giving it" ID196L*

However, some midwives in Terrace maternity service (n=4) and (n=5) in Landscape maternity service agreed the clinical environment may have an effect on women's decision-

making. A midwife vividly described the influence of clinical settings on women's decision-making processes.

*"Clinical setting= tests= blood= scans= being a good patient= saying yes to everything. Hard to say no, hard to ask questions. Uniforms are a barrier in my opinion. They say 'nurse who does some tests on me'. Uniform says I would like a barrier between us. Uniforms = 'I know best' this environment is scary and I think people will take any tests that may even be adhered to" ID182T*

Further, two midwives in each site believed the clinical environment could make some women anxious and in combination with the large amount of information given at booking may result in women being overwhelmed at booking. The midwives used various medical terms for the effects of the environment on women such as "medicalised" and "white coat syndrome".

*"Although we try to make the environment conducive I am sure many clients get "white coat syndrome" and feel obliged to agree to any screening. Booking clients in their own homes was in my opinion a much better and preferable environment. In a home setting time pressures are reduced for the client as they are in control and I think they used to consider the information more". ID173L*

*"I think influences of the environment vary depending on the individual couple. If a woman has a particular aversion to medical environments then she may not feel comfortable and as such may not fully absorb information about screening". ID170T*

In addition, one midwife in Terrace maternity service believed that information posters about Down's syndrome on the walls of the waiting or consulting rooms may cause women to be more aware of the condition which may influence their decision-making.

*"I don't really see how the clinical environment would affect a woman's decision re screening, unless there was information posters regarding it in the room". ID171T*

A midwife reported the seating arrangement during the consultation may heighten or exacerbate power asymmetry, where the midwife is seen as the expert and the women and partners as lay persons.

*"Yes I think the rooms are set so that the woman feels as comfortable as possible for example the desk not being in between the midwife and the woman. Therefore the woman is more likely to make the right decision for her if she feels comfortable. The midwife should talk to*

*the woman at her level; sat down and without the desk in between them so the midwife seems approachable because if the room is set out so that the woman is far away from the midwife or not at the same level; sat down, the woman may not feel as comfortable" ID192L*

Although, one midwife in each site felt the influence of the clinical environment was minimised or neutralised when there is a trusting relationship between the midwife, pregnant woman and partner especially when unbiased information is given.

*"I think the midwife's interpersonal skills should be so good that she builds up a rapport with the woman/couple and when unbiased information is given, the setting cannot have any bearing at the personal decisions. I think people in Britain (generally) are used to this sort of clinical environment and are not intimidated by it". ID208L*

*"If the midwife can develop a rapport with the couple, the environment is less significant. Women need to be able to form trusting relationships". ID181T*

## **6.2.5 Influence of the process of offering screening on women's participation in Down's syndrome screening**

### **6.2.5.1 Information is rushed**

Some midwives in Landscape maternity service (n=4) 21% and in Terrace maternity service (n=2) 13% reported information about Down's syndrome screening was presented in a hurry. A midwife felt the rushed offer of screening definitely influenced women's decision-making processes.

*"Due to time constraints I feel I can sometimes speed through the delivery of the information and then clients just agree to participate" ID 173L*

*"Always rushed, aware to give full information but anxious we get it right" ID204T.*

*"It is rushed due to everything else discussed but I always ensure they understand it is a screening test not diagnostic test" ID199L*

*"Time pressures means you sometimes have to rush especially if more than one patient is waiting" ID202T*

#### **6.2.5.2 Information overload**

Several midwives in Terrace maternity service, 33% (n=5) and 47% (n=9) in Landscape maternity service believed women are given too much information at booking which may result in women not understanding the information about screening.

*"After all that info piled upon a newly pregnant woman in 50 mins, when you get to the end and asking about Down's screening, of course the answer will be an uninformed yes as she will be tired/hot/stuffy room and brain dead. And remember we do need to ask for a decision if possible and if not we request one and tell them they can change their minds. If she is a non-English speaker the appointment may have been going on for ages so she will be even more brain dead" ID182T*

*"The amount of information given at booking dilutes the importance of the information you need them to absorb and information is not recalled adequately.....I feel clients are influenced to participate because we deliver the information "en masse" at booking" ID173L*

#### **6.2.5.3 Use of photographs and scenarios**

A midwife reported using photographs of children with Down's syndrome to facilitate women's understanding of screening, especially with women from ethnic minority groups. In addition, a midwife in Landscape maternity service reported using scenarios to explain or illustrate the implications of screening to women and their partners.

*"I usually discuss the implications of participating in the screening programme and do through possible scenarios to highlight the dilemmas that may arise so I have to say that I make more than a minimal reference" ID195L*

*"I often use pictures of people with Down's syndrome, to assist" ID181T*

#### **6.2.5.4 Screening perceived as a routine test**

Few midwives (n=1) in Terrace and (n=2) in Landscape maternity services believed that not all women are told screening is optional. Even when women were told it was optional, women may still perceive screening as a routine test, as information about screening was given with other routine blood tests at booking.

*"If a midwife seems to make out that they are all routine tests they will say yes to everything. Are they told that all tests are optional?? or are they" ID182T*

*"Combining the info for screening tests with routine blood tests info appears to put them at the same level of importance for the woman. It could also influence some women to say take the blood test while you are taking the others without really thinking about it" ID175L*

*"The amount of information given in one allocated appointment I feel trivialises the importance and significance of the screening test and relegates it to routine and therefore can be perceived as not needing special thought or consideration... Yes. I feel clients just go along with everything and may feel pressured to accept all tests as routine". ID173L*

One midwife from each site reported on their perception of Down's syndrome screening as it relates to other routine tests. They seemed to attach equal importance or weight in terms of uptake of Down's syndrome screening and other routine tests, instead of making a clear distinction between them.

*"Most people have Down's screening but a minority decline. Not many decline other screening for example HIV". ID189L*

*"I discuss blood tests (maternal screening) and Down's Syndrome screening at the same time with the intention of giving the importance of all the tests equal weight. I would hope this would encourage the woman to give all tests equal consideration and that she would feel able to ask questions at this point" ID170T*

#### **6.2.6 Influence of midwives on women's participation in Down's syndrome screening**

Most midwives in Terrace maternity services, 80% (n=12) and Landscape maternity service (n=10) 53%, believed midwives can influence pregnant women and their partners' participation in screening. In some instances, in this study, information-giving by the midwives was directive. The midwives imposed their preferences over that of the women's.

*"A number of women say they would like screening so they can prepare to care for their baby with Down's syndrome. I explain that research was done into bonding with their baby that concluded that parents bond better with their child if they were unaware of its health problems. I say that making the decision to test hinges on their view of terminating a baby with Down's syndrome and I would discourage them from testing if they would keep the baby anyway" ID214T*

*"The unreliability of the triple test made it more likely for the midwife to impose her own views on the test and warn of false positives" ID191T*

In other instances, the midwives believed that women and their partners need support in the decision-making process. They shared their knowledge and experience, while taking the beliefs and values of the women into consideration and helping them to decide on their own.

*"The women see the community midwife as the main support during their pregnancy, the first contact who they rely on to provide safe and accurate information" ID188L*

*"Women need the information in simple terms and occasionally more than once as it is a lot of information to take in... my concerns are that women who accept the Down's screening have not thought about what they would do about a high risk result therefore I ask women if they have thought about this before they accept it" ID192L*

Few midwives in Terrace maternity service, 20% (n=3) and Landscape maternity services 16% (n=3) do not believe midwives influenced women's decision-making processes, but that women's decisions are based on the information provided, their beliefs and values. In these instances, the midwives offered one-way information-giving. They reported staying completely out of the decision-making process. The midwives refused to give their opinions, as they assumed decision-making was personal to women and their partners. Yet, several midwives in both maternity services recognised their influence on women's decision-making, but felt that they were professionals and well trained to inform nondirectively.

*"I will do as much as I can in giving information and I do not influence women even when they ask me if they should proceed, sometimes they ask "what do other women tend to do?", if they are unsure and they need time to consider I advise them that I will put them forward for the test so that they can have it but that when they go for the scan to refuse when they get there and to rip up the consent form" ID174T*

*"I would hope the information given is impartial. Women do ask what you would do but I say I couldn't comment" ID202T*

*"I think it would be possible to influence their decision but as midwives I feel we are very well practised at providing neutral information to facilitate informed choice" ID189L*

Furthermore, two midwives in Landscape maternity service reported midwives' influence on women's participation in screening was related to their level of education, age and experience. They believed the offer of screening to first time mothers, young less educated mothers or women who are undecided may influence their participation in screening.

*"The woman may feel she (midwife) is a person of great experience whose views are right. This could influence a woman's choice.... the younger less educated women often accept the tests without question because the midwife has told them about it so it must be right"*

*ID175L*

*"For those who have never heard of it until they are booked by the midwife, they seem to opt to have the screening done" ID184L*

#### **6.2.6.1 Midwives appear to increase uptake rates**

A midwife in Terrace maternity services reported that some midwives influence women's decision-making processes to increase the uptake rates of screening in the maternity services.

*"Also I have felt that the hospital has almost seen pro doing screening to get the figures up!!"*

*ID182T*

#### **6.2.7 The potential influence of interpreters on women's participation in Down's syndrome screening**

Some midwives (n=3) in Terrace maternity service revealed that interpreters sometimes lack understanding or grappled with the complexities of the concept of Down's syndrome screening, in addition to slowing down the process of informing women and their partners.

*"Also difficult when there are language barriers because even with interpreters who themselves are not sure what Down's syndrome is" ID182T*

*"Interpreters slow the process down" ID204T*

*"I find that even with an interpreter it may take some time before women show signs of understanding of Down's syndrome, I often use pictures of people with Down's syndrome, to assist" ID181T.*

### **6.3 Results of the online interviews with pregnant women**

#### **6.3.1 Findings**

Thirty-five pregnant women participated in the study, 16 in Terrace maternity services and 19 in Landscape maternity services. The age groups, ethnicity, age on leaving full time education and highest level of educational qualifications are presented in Table 4.

### 6.3.2 Response to interviews

A total of 592 participant's information sheets were given to pregnant women in Terrace maternity service who agreed to participate in the study. This is equivalent to a response rate of 2.7%. The response rate in Landscape maternity services was 6.1% with 311

**Table 4: Pregnant women's demographic profile**

Age:	Terrace maternity services (N=16) n (%)	Landscape maternity services (N=19) n (%)
16-20	-	2 (10.5%)
21-25	3 (18.7%)	5 (26.3%)
26-34	12 (75%)	7 (36.8%)
35-44	1 (6.3%)	5 (26.3%)
Total	16 (100%)	19 (99.9%)
Ethnicity:		
White British	6 (37.5%)	18 (94.7%)
Mixed White and Black African	1 (6.2%)	1 (5.3%)
Black African	5 (31.3%)	-
Other Black background	1 (6.2%)	-
Any other Ethnic group	1 (6.2%)	-
Asian Pakistani	2 (12.5%)	-
Total	16 (100%)	19 (100%)
Age (years) left full time education:		
16 - 20	4 (25%)	12 (63.1%)
21 - 25	9 (56.3%)	5 (26.3%)
26 - 30	3 (18.7%)	1 (5.3%)
31 – 35	-	1 (5.3%)
Total	16 (100%)	19 (100%)
Highest level of educational qualification:		
No formal qualifications	-	2 (10.5%)
GCSE	2 (12.5%)	3 (15.8%)
A levels/SCE Higher/AS	1 (6.2%)	4 (21%)
NVQ; GNVQ or Diploma	4 (25%)	4 (21%)
First degree (e.g BA, BSc)	6 (37.5%)	2 (10.5%)
Higher degree (e.g MA, PhD, PGCE)	3 (18.7%)	3 (15.8%)
Other qualifications (e.g. City and Guilds, RSA/OCR, BTEC/Edexcel)	-	1 (5.3%)
Total	16 (100%)	19 (99.9%)

information sheets given to those who agreed to participate in the study. Poor response to online studies has been reported in the literature (Petchenik and Watermolen, 2011).

Nevertheless, some of the women who declined to participate in the study explained that they were given inadequate verbal information about Down's syndrome screening at booking. Some were only asked if they wanted to screen or not. Others reported declining information about screening. A few women explained that they were not interested in participating in a study about Down's syndrome screening.

### **6.3.3 Women's experience with information about prenatal Down's syndrome screening**

#### **6.3.3.1 Prior knowledge/ experience**

Several women in both maternity services, 75% (n=12) in Terrace maternity service and 58% (n=11) Landscape maternity service, had prior knowledge or information about screening before the booking visit.

*"I got the information about Down's syndrome prior to visiting the midwife. This was part of my biochemistry course in the University" ID124T*

*"Told by friends. I didn't really read up on the subject much" ID115L*

*"I have a Down's syndrome aunt so this is something that I am aware of" ID105T*

*"I had researched the various methods of screening for Down's syndrome prior to my booking visit so I felt I already had a good understanding. Conversations with family and friends who had also undertaken Down's screening gave me further information" ID96L*

### **6.3.4 Influence of the structure of the screening programme on women's participation in Down's syndrome screening**

#### **6.3.4.1 Consent obtained at dating scan**

A small number of women in Landscape maternity service indicated that consent was obtained for Down's syndrome screening when they presented for the dating scan. A pregnant woman reported receiving inadequate information before being asked for consent, whilst another was still undecided about screening.

*"We were asked if we would like the test, I was not sure either way....not really decided. I decided on the day to have the test" ID115L*

*"We were told on the same day of the first scan if we would like a test. We didn't receive any information about afterwards" ID145L*

#### **6.3.4.2 Influence of the prenatal environment on women's decision-making process.**

The majority of the women in both maternity services reported that the prenatal setting had no influence on their participation in screening.

*"No I don't think the room decor, lighting or any clinical setting could influence my decision. This is because it is more personal than clinical to me" ID119T.*

Though, several women in both maternity services explained that the prenatal environment might influence them to consider screening if there are photos/posters of baby's with Down's syndrome on the walls of the waiting or consulting rooms. In addition, a pregnant woman reported that because screening was routinely offered by midwives to all women mostly in a clinical environment, it was perceived as a routine test.

*"I don't think the decor would influence a decision about screening. If there were posters about the screening on the walls or waiting room then that might influence others' decisions to have the screening as it would highlight the screening to them before-hand and could possibly have frequently asked questions on" ID107T*

*"I feel the environment may influence someone if the test is considered routine because it's a clinical setting and then it may be made to feel like a normal test??" ID95L*

*"I personally did not feel the surroundings influenced my decision, however, as this is unfamiliar territory people may feel that they have to make a decision quickly as the midwife is busy. It is a clinical setting and people may feel that they are pressured into screening as this is for the best. With the push for women to accept Down's screening they may feel like they have no choice but to do so" ID96L*

The presence of the midwife in a uniform in the prenatal environment may highlight the power asymmetry between the midwife who may be seen as the expert and the woman as the lay person.

*"It's a very formal office type room, and when you are being addressed by someone in uniform in an environment where you perhaps feel that power is taken away from you,*

*you're more likely to feel that you've been told to do something rather than discussing something for you to make your own mind up on" ID94L*

Two women in Landscape maternity services reported that the ambience of the clinical setting had a definite influence on their participation in screening.

*"Yes, thought it was best to have it done". ID145L*

*"It was warm and relaxed in the room we had, it gave me a sense of trust in the person we were talking to; if I had been in a dingy room with someone who hadn't a clue what they were telling us about we would have been inclined to move to another care provider and would probably have looked further into this ourselves" ID112L*

### **6.3.5 Influence of processes involved in screening on women's participation in Down's syndrome screening**

#### **6.3.5.1 Information is rushed**

Two women in Landscape maternity service reported that the verbal information about Down's syndrome screening was rushed.

*"Felt that explanation was a bit rushed as if I was expected to partake in the test" ID110L*

*"I would have liked more info from the midwife at the time of my booking as I feel they try and hurry things along and leave you to research things yourself. I feel more info should be given at the booking visit" ID92L*

#### **6.3.5.2 Inadequate verbal information about implications of screening**

Some women in Terrace maternity service 13% (n=2) and 21% (n=4) women in Landscape maternity service explained they were given insufficient or no verbal information about the implications of screening. In some instances, the midwives just asked if they wanted screening or not.

*"I was told about the need to have the baby screened for Down's syndrome and basically given a leaflet and booklet about the condition. In all my pregnancies blood samples were taken for screening without detailed verbal information from the midwife" ID118T*

*"I was given a booklet to read in my notes rather than tests actually explained to me" ID105T*

*"No explanation just briefly mentioned it; still don't know what the blood tests I have had done are for" ID92L*

Several women in Landscape maternity service reported that the implications of screening were not explained verbally by the midwife at booking.

*"Termination was not an option; either by us and the midwife did not offer that option either" ID120L*

*"What I don't feel is stressed enough is that the test does involve you making a decision to terminate the baby" ID101L*

#### **6.3.5.3 Women's understanding of information**

About 50% (n=8) of participants in Terrace maternity service and 47% (n=9) in Landscape maternity service explained that they understood the verbal information given by the midwife.

*"I didn't have any difficulties in understanding the midwife and was happy to consent to the test. The midwife took time to explain what the process entails and that further tests may be required if necessary" ID132T*

*"The midwife made the talk about Downs very easy. I thought the test would be helpful to know if my baby has Downs and the midwife made it that easy I didn't really know he had done it. It was that simple" ID116L*

#### **6.3.5.4 Information overload**

Several pregnant women (n=5) 21% in Landscape maternity service found the information given at booking overwhelming.

*"Felt a little overwhelmed by all the advice I was given, however I understood that the mw needed to explain it all to me" ID121L.*

*"There is a lot of information to take in and I can imagine that it may be difficult for those people who are unsure as to whether they would like screening or not" ID96L*

#### **6.3.5.5 Emotional impact of screening information**

Several pregnant women in Terrace maternity service (n=4) 25% and 58% (n=11) of women in Landscape maternity service reported feeling either scared, terrified, anxious or worried by the information about screening and the possibility of having a baby with Down's

syndrome. Women also reported that their anxiety was relieved when the screening results came back low risk.

*"Terrified, the thought of having a baby with special/additional needs was scary. Before I was given advice from my midwife I understood what Down's syndrome was but never though it could be a possibility for my baby to have it. I thought you would know somehow before pregnancy if you have the missing gene or was vulnerable to having a baby with Down's syndrome" ID103T*

*"Scared, on top of all the information you are given at your first midwife appointment it can be a lot to take in. I think this was the first time that it occurred to me that I may have a child with a disability" ID101L*

*"The screening result was a relief when it came negative" ID124T*

However, 56% (n=9) of women in Terrace and 21% (n=4) in Landscape maternity service felt indifferent or confident about the information given. A pregnant woman in Terrace maternity service reported it was ridiculous to screen for Down's syndrome based on her experiential knowledge.

*"It didn't really concern me as I have a Down's syndrome cousin so if I had a baby with Down's syndrome it wouldn't make me feel any different about having it... I didn't feel any different as I told the midwife I didn't want the screening. I think the screening for Down's syndrome is quite ridiculous to be quite honest with you. Out of all the disabilities they can only screen for Down's syndrome and have the option to terminate your pregnancy if it comes out positive" ID138T*

A pregnant woman expressed concerns about the psychological impact of a high risk screen result on women who accepted screening without thinking through the implications of screening.

*"I personally feel that the problem lies where the results come back high risk as some women may not have thought about this. The prospect of having an invasive test is daunting and for those who do not wish to have this, their pregnancy may be filled with anxiety and uncertainty as a result". ID96L*

#### **6.3.5.6 Perception of risk**

The classification of women based on age or family history into higher or lower risk for the likelihood of having a baby with Down's syndrome generated anxiety in some women at both sites and indifference in others.

*"I was aware that there could be a possibility due to my age; however it did not make any difference to our baby... of course a little stress was experienced as we found out I was high risk for carrying a DS child, however this would not change anything for us" ID120L*

*"Due to my partner having Down's in the family we were not bothered about the outcome" ID116L*

#### **6.3.5.7 Reassured by the midwife**

Two women in Terrace maternity service and a woman in Landscape maternity service reported being reassured by the professionalism and concern demonstrated by the midwives at booking.

*"Yes, I feel the midwife was very professional and comforting, I didn't feel as though I couldn't approach her and ask, I feel I was able to freely express any concern I had and she answered with information helping me understand more" ID103T*

Yet, the reassurance given to a pregnant woman in Terrace maternity service may easily be perceived as directive.

*"The midwife discussed issues arising from the screening but she reassured one that the chances of results being positive were very low and that a 2nd screening was only required very rarely. I was comfortable asking questions to my midwife who was very happy to answer my queries" ID124T*

#### **6.3.5.8 Screening offered as a routine test**

Some women in Terrace maternity service 16% (n=3) and 63% (n=12) in Landscape maternity service perceived Down's syndrome screening as a routine test in prenatal care for a healthy pregnancy and for the wellbeing of the baby. In most instances, women reported that screening was offered to them by the midwives as a routine test.

*"Yes, midwife explained that it was commonly done, a routine test". ID115L*

*"Yes, I understood that the test was as part of normal pregnancy care and I should take it. And she told me it is like a normal routine test" ID144T*

*"Yes, other blood tests are routine throughout pregnancy and also must be done in any event. Yes, screening came across as an initial test carried out at the beginning of all pregnancies" ID143L*

A pregnant woman in Terrace maternity service reported accepting screening because it was offered to her. The woman felt screening was part of prenatal care. Another participant in Terrace maternity service expressed surprise that the combined test was now offered free of charge in the NHS, as in her last pregnancy she paid to have it done in a private hospital.

*"I just decided to go for the screening based on its availability" ID102T*

*"To be honest as I had this done on previous pregnancy, I didn't really think too much into things. I have a very supportive husband and we would deal with the results once they were given either way ...I was told this screening was part of a package (Gold package) offered to all pregnant women...I paid £170 for this in my previous pregnancy privately. So it was a surprise and a welcomed relief that now all pregnant women have this offered to them as part of the care offered to all pregnant ladies in the NHS care!" ID137T*

### **6.3.6 Midwives' influence on women's participation in Down's syndrome**

#### **screening**

Several women, 25% (n=4) in Terrace maternity service and 37% (n=7) of women in Landscape maternity service reported that the attitudes of the midwives particularly the implicit or explicit manner of presenting information about screening left them feeling that screening was recommended in prenatal care.

*"Yes, I felt that it was highly recommended for me to take the test. I knew that it was optional for the screening but did feel it was recommended. I had already made up my mind that I was going to have the screening done anyway but it just helped to clarify one or two things" ID94L*

*"I did not feel influenced in any way as I have already come to a conclusion, however I did feel that there was a push to consent to the screening process as if it was the right thing to do. To "just have it anyway" seemed to be the views" ID96L*

A small number of women 6% (n=1) in Terrace maternity service and 26% (n=5) in Landscape maternity services reported that they were told by the midwives to have the screening tests done. In these instances, women specifically implied information-giving by the midwife was largely directive. The midwife played an active role in the decision making process while the woman was passive. The woman gave consent to the recommended screening preferences of the midwives.

*"I had already undergone the screening test in my first pregnancy and the technique and approach used by the midwife made it easy for me to decide on taking the screening test in this pregnancy as I felt it wasn't really necessary" ID132T*

*"My midwife did push me into the decision but after I decided to do the test myself" ID146L*

*"I was told to have it by midwife and family... I decided on the day to have the test" ID115L.*

In other instances, several women reported information-giving by the midwife as a one-way process. The midwife gave information that was 'objective' to enable autonomous and informed decision making. Beyond information-giving the midwife had no further role in decision-making.

*"I understood it was her place to tell me about it and for me to make an informed decision. So no, her explanation did not influence my decision" ID 119T*

*"I was told about the need to have the baby screened for Down's syndrome and basically given a leaflet and booklet about the condition. In all my pregnancies blood samples were taken for screening without detailed verbal information from the midwife... No the midwife did not influence me in making this decision" ID118T*

*"I wasn't influenced by anyone or anything, only our choice as a couple" ID104L*

In some instances, some pregnant women reported developing trust in the midwives and acting in a relational way. It involved discussion of information and screening options available, based on the women's beliefs and values. The women sought support and advice from the midwife. Decision making was negotiated, but the women took responsibility for the final decisions.

*"I think what she said made me feel sure about my decision. She gave me enough information to make me realise what I already knew was correct and yes I was comfortable*

*asking questions and voicing my opinions.... she made me realise what I knew was correct and that I had made the right choice for me" ID95L*

*"Yes, I feel the midwife was very professional and comforting, I didn't feel as though I couldn't approach her and ask. I feel I was able to freely express any concern I had and she answered with information helping me understand more". ID103T*

Nevertheless, many of the women, 63% (n=10) Terrace maternity service and 68% (n=13) in Landscape maternity service reported they have decided before the booking visit.

*"Before the screening visit to my midwife I was determined to undertake any screening available to me in order to ensure the wellbeing of my baby... since I was determined to take any routine tests available to pregnant women, I cannot say that my decision was based on the midwife explanation. However, I can say that discussing with my midwife gave me an opportunity to request being tested" ID124T*

*"I had already made up my mind that I was going to have the screening done any way but it just helped to clarify one or two things" ID94L*

### **6.3.7 Partners' influence on women's decision-making about Down's syndrome screening.**

A pregnant woman in Landscape maternity service explained her partner's insistence on screening had precedence over her preference not to undergo screening.

*"Personally with my first child I was happy not to have the screening done since I believe that if I was carrying a Down's syndrome child then this was the child I was meant to have, I had the screening done since it was my partner's wish, this second time around I had it done not only again for my partner but because of the implications for my oldest child and how a sibling for him who needed a lot more care etc. could impact him, and so we could all prepare-however my belief was still the same" ID 112L*

### **6.3.8 Reasons for women's decisions about Down's syndrome screening**

Most women in both maternity services reported that their reasons for participating in screening were for reassurance, preparation for outcome, decision-making and to avoid harm to the foetus such as miscarriage and TOP.

*"In my culture children with Down's syndrome are stigmatised and generally looked down upon and sometimes even killed. These kids are often neglected, physically abused and sexually assaulted. Parent and families with children with this condition are often blamed for their child's condition. This made me to consider having the screening as I will be able to make a decision on whether to or not to go ahead with the pregnancy" ID118T*

*"I wanted to know the possible risk factor so I was aware of what to expect at the birth. Being aware of my baby's health was more important than my feelings towards the screening process" ID111L*

*"I was ok with not having the test so I listened but still decided to decline. The thought of a needle going in around my baby was upsetting and unnecessary for me" ID110L*

## **6.4 Results of online interviews with partners of pregnant women**

### **6.4.1 Findings**

Fifteen male partners participated in the study, 7 in Terrace maternity services and 8 in Landscape maternity services. The age group, ethnicity, age on leaving fulltime education, highest educational qualification and involvement in decision-making are shown in Table 5.

### **6.4.2 Response to interviews**

In Terrace maternity service, 218 participant information sheets were given to partners who agreed to participate in the study. This is equivalent to a response rate of 3.2%. In Landscape maternity service the response rate was 4.3% with 186 information sheets given to partners who agreed to participate in the study. No reason was given by partners who declined to participate in the study. There were fewer men than women involved in this study, despite the best efforts of the researcher and may be due to men's reluctance to be involved in research on pregnancy as it is believed to be a woman issue (Rapp, 2000; Reed, 2011).

### **6.4.3 Partners' experiences with information about Down's syndrome screening**

#### **6.4.3.1 Prior knowledge about screening**

Most partners in Terrace maternity service 86% (n=6) and 38% (n=3) in Landscape maternity service reported having prior knowledge of screening before the booking visit from sources such as the internet, friends, family members and previous pregnancies. Partners still found the information from the midwives useful.

*"I was previously aware of what Down's screening was and the implications of having a positive result. This was from general knowledge and learning about conditions in pregnancy" ID104T*

*"I knew about the screening from my sister-in-law as she had it done with her children but I had also read about it on the internet. The info we got from the mw was useful too" ID99L*

**Table 5: Partners' demographic profile**

Age (years):	Terrace maternity services (N=7) n (%)	Landscape maternity services (N=8) n (%)
21-25	-	1 (12.5%)
26-34	1 (14.3%)	5 (62.5%)
35-44	5 (71.4%)	2 (25%)
45-54	1 (14.3%)	-
Total	7 (100%)	8(100%)
Ethnicity:		
White British	1 (14.3%)	7 (87.5%)
Other White background	-	-
Mixed White and Black Caribbean	-	1 (12.5%)
Black African	4 (57.1%)	-
Other Black background	-	-
Any other Asian background	1 (14.3%)	-
Asian Pakistani	1 (14.3%)	-
Total	7 (100%)	8(100%)
Age (years) left full time education:		
16-20 years	1 (14.3%)	7 (87.5%)
21 - 25	1 (14.3%)	1 (12.5%)
26 - 30	3 (44.8%)	-
31 - 35	-	-
36-40	2 (28.6%)	-
Total	7 (100%)	8(100%)
Highest level of educational qualification:		
No formal qualifications	-	-
GCSE	1(14.3%)	1 (12.5%)
A levels/SCE Higher/AS	1 (14.3%)	1 (12.5%)
NVQ; GNVQ or Diploma	-	4 (50%)
First degree (e.g BA, BSc)	-	1 (12.5%)
Higher degree (e.g MA, PhD, PGCE)	5 (71.4%)	1 (12.5%)
Other qualifications (e.g. City and Guilds, RSA/OCR, BTEC/Edexcel)	-	-
Total	7 (100%)	8(100%)
Involvement in decision-making	6 (85.7%)	7 (87.5%)

#### **6.4.4 Influence of the structure of the Down's syndrome screening programmes on partners' participation**

##### **6.4.4.1 Consent and screening in the context of the dating scan**

Two male partners in Landscape maternity service reported that consent for Down's syndrome screening was obtained at the dating scan

*"I wasn't aware of the part where they measure fluid round the baby's head. I just thought it was a blood test. But it was a safe test and caused no harm to my partner and unborn child so I let it go ahead" ID106L*

*"I was at the 12 weeks scan when I was asked if I wanted the scan" ID105L*

##### **6.4.4.2 Influence of the clinical environment on partners' participation in screening**

Male partners in both maternity services stated that the clinical setting had no influence on their decisions to participate in screening. However, a few partners at both sites had different views about how the clinical environment may influence decision-making processes in the prenatal context.

*"For me we could have been sat on a building site cafe having the same conversation this would have no influence on the outcome. The information clearly explained is the most important thing" ID98L*

*"I think that the clincal environment is adequate. It doesn't really influence decision making (specifically regarding the test). It is better to have the midwife sitting closer to the pregnant lady rather than over a desk as it helps relax her. Too much medical equipment sometimes distracts from the consultation" ID104T*

#### **6.4.5 Influence of the process of offering Down's syndrome screening on partners' participation.**

##### **6.4.5.1 Lack of verbal information about screening**

A partner in Landscape maternity service reported the verbal information given by the midwife was inadequate and was given too quickly.

*She didn't really say that much just that the test gives a percentage chance and the booklets say a bit more about it. ID97L*

*"Bit quick but probably realised we did not want it" ID94L*

#### **6.4.5.2 Information overload**

Three partners in Landscape maternity service reported that the quantity of information provided at booking was too much.

*"In the beginning of the information that was given, too much information to handle, but by the end of the meeting, what was going to be was going to be. So testing was not needed" ID98L*

*"Plenty of information" ID94L*

*"My midwife gave me loads of information" ID105L*

#### **6.4.5.3 Partners' understanding of information about screening**

Several partners, 57% (n=4) in Terrace maternity service and 25% (n=2) of partners in Landscape maternity service reported understanding the verbal information about screening. However, the responses of some of the partners suggested difficulty in distinguishing screening from diagnostic test. In addition, one of the partners reported having some initial difficulties understanding the information.

*"I did the blood test and the outcome of the blood test did not require me to do the screening. So I had no decision to make for Down's syndrome screening" ID86T*

*"Yes, after a while of the midwife explaining but not straight away" ID106L*

*"I had no difficulty in understanding the midwife's explanation but I had to re-iterate the information to my wife so that we could both decide together" ID104T*

#### **6.4.5.4 Information related anxiety**

Several partners in Terrace maternity service, 28% (n=2) and 50% (n=4) of partners in Landscape maternity service felt the information about Down's syndrome screening made them nervous due to the possibility of having a baby with Down's syndrome or the results returning higher risk. For some partners, the period of waiting for the results was full of anxiety. However, many of the partners in both locations reported that their anxiety disappeared when the screening result came back lower risk.

*"The screening process was indeed stressful because of all the possibilities that the results would bring up and all the decisions we would have to make about the future of our family"*  
*ID100T*

*"It made me nervous to think my baby could have Down's syndrome. But I was grateful for the advice. I was nervous about hearing about the possibilities of Down's syndrome. But when the results came back low I felt much better"* *ID106L*

*"I was not around during screening due to work but waiting for the result is like waiting for an examination result"* *ID90T*

Yet, some partners were indifferent, confident about the screening either due to perceived partner's low risk as a result of age, no family history, based on their religious beliefs and / or their ability to cope with a baby that has the condition.

*"I was indifferent about it when I was told"* *ID96T*

*"I was happy to be given the information but I would be happy with any child Down's syndrome or not"* *ID95L*

A partner in Terrace maternity services was surprised to be offered Down's syndrome screening, as it was not offered routinely in his home country. This partner also believed the name Down's syndrome has a negative connotation to it.

*"Surprised as my country does not perform Down's syndrome test as a routine test...I think the word Down's syndrome test itself brings a negative ring to it not the environment. I honestly think the name of the test should be changed to a more medical name or abbreviated name"* *ID90T*

Some of the partners in Landscape maternity services were using the information about the screening pathway (diagnostic testing and its associated risk) to decline the offer of screening. It is clear they were considering the offer of screening based on a high risk result and the complications associated with a diagnostic test.

*"The risk of the testing to the baby was too high in my opinion. If in the end, baby was fit and healthy to start with...once we had the information given the 1.7% chance of damage to baby against 1% chance of the baby having Down's syndrome, the risk to the baby was too high on the baby"* *ID98L*

*"No did not want a test which would mean my wife would have to consider a further test to see if baby was Down's and could kill the baby by doing so. Prefer not to know" ID94L*

*"I think our thoughts were more about the diagnostic procedures. How it would not be very nice. We did not like the thought of just because it may have Down's that we would terminate the pregnancy" ID89L*

#### **6.4.5.5 Reassured by midwives**

A partner in Terrace maternity services reported being reassured about screening for Down's syndrome by the midwife.

*"I was reassured by the midwife about the test" ID104T*

#### **6.4.5.6 Screening perceived as a routine test**

Several partners 57% (n=4) in Terrace maternity service and 38% (n=3) partners in Landscape maternity service perceived screening as a routine test and reported the information about screening provided by the midwives made the offer appear as a routine test.

*"I did not think twice about it since I had information that pregnant women need to do the test. I never had the feeling we would have a baby with likelihood of Down's syndrome. So I just went along with it...I felt it was a screening I had to do and move on" ID86T*

*"I thought this was a routine test just like the others a woman should take in the process of a pregnancy" ID100T*

*"Yes understood the screening is routine and all the different types of tests" ID105L*

#### **6.4.6 Influence of midwives on partners' participation in Down's syndrome screening**

Few partners reported one-way information giving in which the midwife presented information about screening. The partners and the pregnant women were then expected to make decisions based on the nondirective information from the midwife.

*"The midwife explained all info and options that were open to us; the midwife never pushed or said what was the right thing to do ...an overall explanation help me decide" ID98L*

Several partners in both maternity services reported engaging in dialogue with the midwives. They reported being able to ask questions and also gave their opinion. They felt the midwives provided useful feedback and opinions.

*"I felt comfortable asking questions. I asked lots of questions and received valuable answers"*  
ID106L

*"As I already had a prior knowledge of the test, the midwife simply reassured us about the importance and usefulness of the test and helped us consider the implications of having a positive test" ID104T*

Few partners reported receiving minimal to no information from the midwives about the purpose of screening. Screening was either recommended or they were told by the midwife to have the test done. The partners felt the pregnant women gave consent to the preference of the midwives.

*"Not really my girlfriend decided on that day...just got told by the midwife to have it done"*  
ID95L

*"My midwife didn't pressure me but did recommend it, but so did all my family" ID105L*

*"Our baby is done via IVF and as such it was advisable for us to perform the test as he is not conceived naturally" ID90T*

*"Felt pressured with all the information that I should let my wife have the test...felt at one point that perhaps we should have the test because most people do!" ID94L*

#### **6.4.7 Reasons for partners' decisions about Down's syndrome screening**

Most partners in both maternity services reported that their reasons for participating in screening were for reassurance, preparation for outcome, further decision making and to avoid harm to the foetus such as miscarriage or TOP.

*"I would have accepted any tests to be done no matter what the tests as I want the best start in life for my child." ID111L*

*"I thought it would be useful in terms of removing uncertainties about the health of the baby". ID100T*

### **6.5 Overview of the results of this study.**

The findings of the online interviews revealed the influence of service organisation and delivery on women and their partners' participation in screening from the perspectives of midwives, women and partners. The interviews indicate important gaps in perception between midwives, women and their partners in both maternity services.

Whilst women and partners reported elevation in anxiety, due to the information about Down's syndrome screening and prior to receiving screening results, most midwives were focused on offering screening to women at booking. This is evident in the data as only one midwife in Terrace maternity service reported on women's anxiety in response to the information about screening. See page 126. Midwives' focus on offering screening and decision-making in the prenatal context may reflect the pressure on midwives to obtain 'consent' from women. It may indicate midwives felt incompetent or unable to meet women and partners' emotional needs in the constrained context of screening. Further it may be due to lack of understanding of the interview question by the midwives. However, the lack of understanding of the interview question is not supported by previous research which suggests that midwives lack of confidence, training, psychological support and time may be responsible for women's elevated anxiety to go unnoticed or not discussed (Miller et al., 2004; Horwitz et al., 2007; Browne et al.). This gap in perception may be as a result of the policy of objective information-giving (informed choice) that encouraged midwives to maintain emotional distance from women when offering Down's syndrome screening. However, the lack of understanding of women and their partners' emotional needs is a gap in midwives' perception and reflects the need to pay attention to women and partners' emotional needs. This finding is based on the online interviews and not on the caseload or team midwifery models adopted in both maternity services as discussed in the document review. The caseload model offers continuity of care and the development of relationships which may not be experienced in team midwifery model. However, the models of care are not important in this discussion because the general maternity service guidelines encourage midwives to form relationships with women to compensate for the weakness of the team midwifery model. Therefore, guidelines and policies are the important service organisational factors to consider in the brief midwife-woman encounter of Down's syndrome screening.

Moreover, midwives expressed differing views and opinions about the time available to present information about screening to pregnant women and their partners. Such inconsistencies may not only be due to the constraints of the current policies and guidelines (nondirective informed choice) that encouraged one-way information-giving. Moreover, it may indicate tensions within midwives as a result of trying to balance minimum information-giving about screening to enable 'consent' to be obtained from women and information overload at booking.

Midwives reported that women lack understanding of the information about screening. This is consistent with findings in other research that suggests women lack understanding of screening (Pilnick et al., 2004; McNeill and Alderdice, 2009; McNeill et al., 2009; Seror and Ville, 2009). However, many women in both maternity services reported having good understanding of the information about screening and made choices autonomously (see page 143). This is consistent with the findings of a research conducted in the UK which suggests women made choices based on their understanding of the information about screening (Shantha et al., 2009).

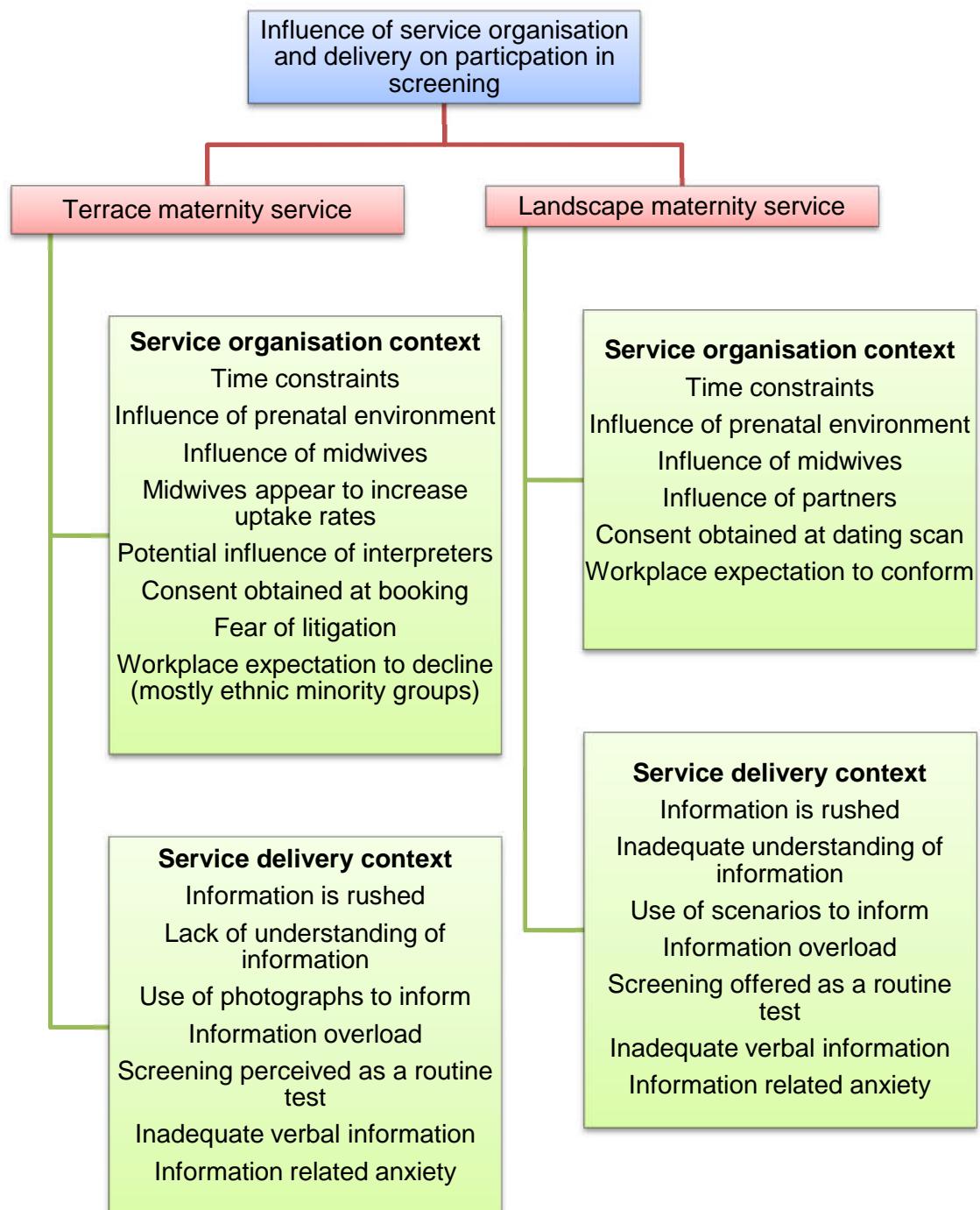
This inconsistency may reflect a genuine gap in midwives' understanding of women's information needs. The gap may be as a result of the limitations in the current policies of informed choice that encouraged objective information-giving by midwives and discouraged genuine dialogue. However, Kirkham et al. (2002) in their research suggested that the majority of midwives do not realise that women might be seeking additional information when they appear not to understand the initial information provided. This is supported by existing research that suggests that it would be misleading to assume that all pregnant women do not have knowledge about Down's syndrome screening (Lewando-Hundt, 2006). Women's need for reassurance, to accept or reject previous information and support their choice might account for this difference in perceptions. Additionally, it may be due to women not wanting information about screening and psychologically adopts avoidance (Seror and Ville, 2009). Importantly, this gap in perception points to the need for training of midwives on how to check and ensure that women understood the information about screening.

Some midwives referred to pregnant women as 'clients'. This may indicate a contractual arrangement and objectivity which supports one-way information-giving, instead of relationships between pregnant women and midwives. In addition, tensions may emerge between midwives and women in the prenatal context when midwives provide information objectively and expect immediate decisions from women. The women may be constrained by the objective nature of information-giving and discouraged from engaging in dialogue. The implication is that women may be encouraged to by-pass the complex decision making involved in screening.

Further, the involvement of partners in screening may be a source of tension in the prenatal context as demonstrated in the interviews and the document review. The findings from the

online interviews and the document review will be integrated in the discussion chapter. A conceptual model depicting the influence of service organisation and delivery on women and partners' participation in Down's syndrome screening programme is developed and described.

**Figure 7: Constraints to service provision in two maternity services (online interviews)**

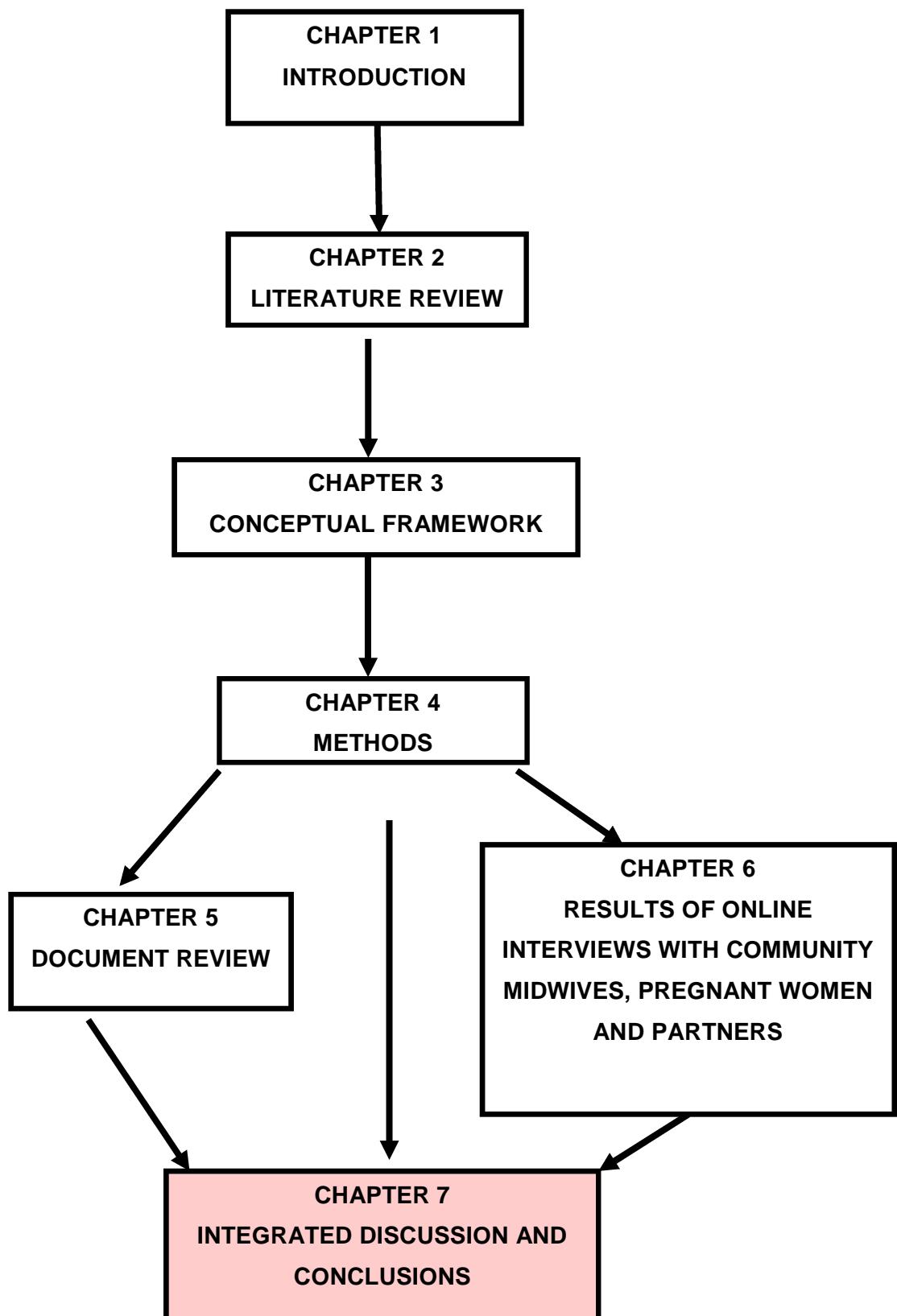


**Table 6: Comparisons of patterns of service organisation constraints in both maternity services**

	Terrace maternity services	Landscape maternity services
Policy	Informed choice/ Paternalistic policies.	Informed choice policy
Consent obtained at booking appointments	Yes	No
Consent obtained at dating scan	No	Yes
Time pressure (midwives)	80% (n=12)	21% (n=4)
Midwives' influence (women)	25% (n=4)	37% (n=7)
Potential influence of interpreters (midwives)	20% (n=3)	-
Influence of partners (women)	-	5.3% (n=1)
Quadruple test re-offered at 15 weeks appointment	No	Yes
Influence of prenatal environment (women) (midwives)	- 26.6% (n=4)	11% (n=2) 26.3% (n=5)
Workplace expectation of women to decline (especially ethnic minority groups)	Yes	No
Perceived 'push' or workplace culture to conform	No	Yes
Written information is given at 'booking'	Yes	Yes

**Table 7: Comparisons of patterns of service delivery constraints in both maternity services**

	Terrace maternity services. Uptake rate (low)	Landscape maternity services. Uptake rate (high)
Lack of verbal information (women)	13% (n=2)	21% (n=4)
Prior knowledge of screening (partners)	75% (n=12) 86% (n=6)	58%(n=11) 38% (n=3)
Information overload (midwives) (women)	33% (n=5) -	47% (n=9) 21% (n=5)
Implications of screening explained (women)	56% (n=9)	42% (n=8)
Screening offered as a routine test (women)	16% (n=3)	63% (n=12)
Information is rushed (midwives) (women)	13% (n=2) -	(n=4) 21% (n=2) 11%
Emotional impact of screening Information (midwives) (partners)	6.7% (n=1) 25% (n=4) 28% (n=2)	- 58% (n=11) 50% (n=4)
Understanding of information (partners)	50% (n=8) 57% (n=4)	47% (n=9) 25% (n=2)
Already decided (women)	63%(n=10)	68% (n=13)



## CHAPTER 7: INTEGRATED DISCUSSION AND CONCLUSIONS

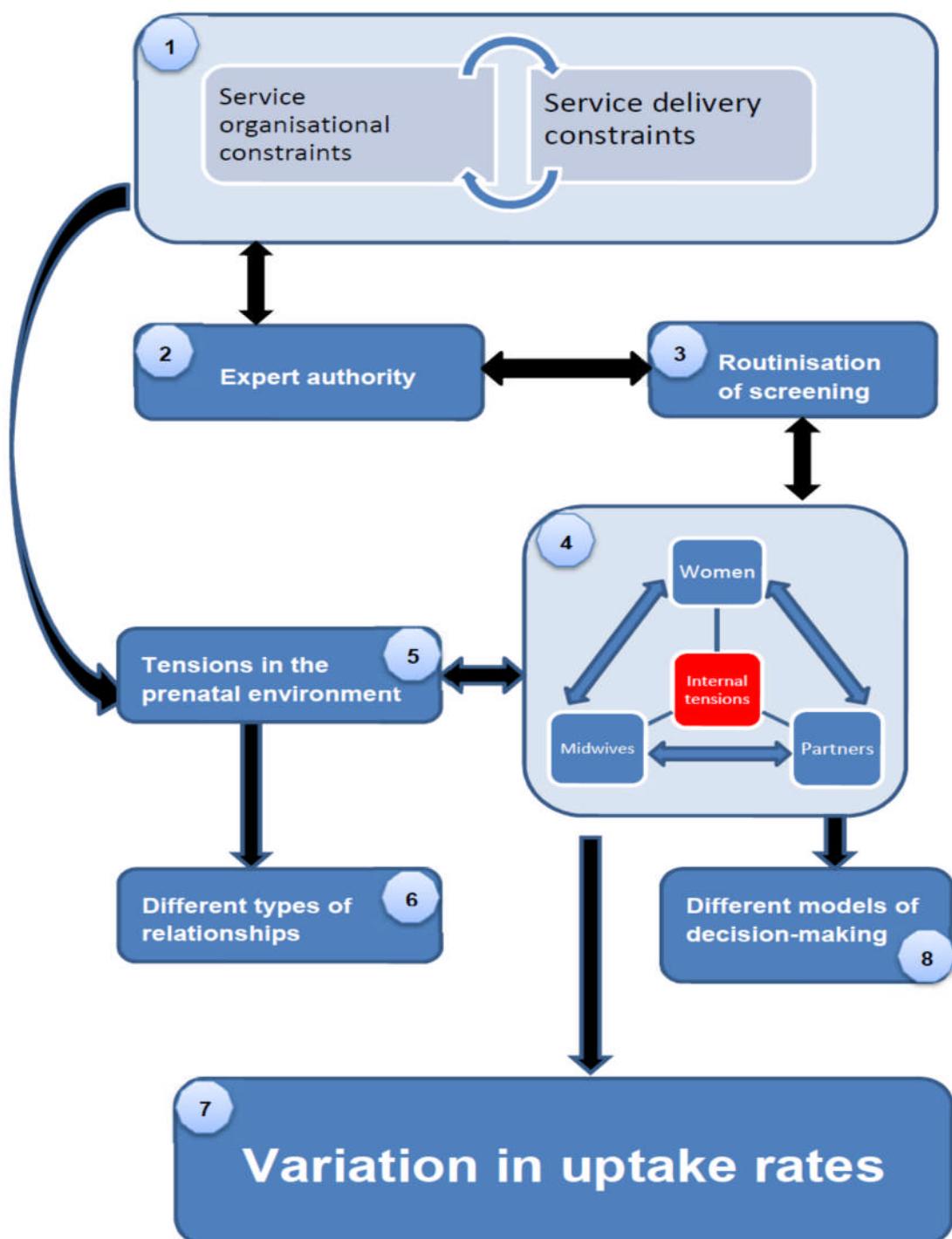
### 7.1 Introduction

The aim of this chapter is to integrate and discuss the findings of the document review and online interviews. First, the usefulness of the Donabedian conceptual framework is briefly discussed. This is followed by a description of the conceptual model depicting the influence of service organisation and delivery underpinned by the findings of this thesis. Next the variation in uptake rates is explained. The different relationships and decision-making models demonstrated in the prenatal context of Down's syndrome screening are also discussed. As a result of the constraints, tensions in the prenatal context of screening, a policy of the more social model of a shared decision-making process is proposed. The possible limitations of this thesis are considered, alongside the implications of the findings for policy, practice and education. Recommendations for research, the planned dissemination of the findings and finally the conclusions of this thesis are presented.

### 7.2 Donabedian conceptual framework

The Donabedian framework provided a structure for this thesis. It was invaluable in organising the different contextual factors in the maternity services into structures (service organisation) and processes (service delivery) as shown in fig 3 and table 1 pages 64 and 65. Moreover, the framework was useful in the construction of scenarios and questions employed in the data collection to explore the influence of service organisation and delivery. Besides, it provided a framework for the conduct of the document review and data analyses. However, the application of the Donabedian framework in this thesis to explore the influence of service organisational and delivery contexts on women and partners' participation in screening was not helpful in providing an understanding of the study findings. This may be due to the broad range of responses to the online interview questions and the findings from the document review on complex decision making processes may not be fully explained by the simple linear Donabedian model. In addition, the distinction between structure and process in the Donabedian framework still remains unclear. For example, obtaining consent from women is a process attribute but since it is the local policy to obtain consent either at booking or at the dating scan it was considered an attribute of structure.

Figure 8: A conceptual model of the influence of service organisation and delivery on the uptake of screening for Down's syndrome.



### **7.3 Conceptual model of the influence of service organisation and delivery**

Figure 8 presents the findings of this thesis as a conceptual model. At the top of this model is the concept of service organisational and delivery constraints ( see concept 1 in figure 8) developed from the findings, subthemes and themes in the online interviews and document review (see figure 7). The various constraints to service provision are described throughout this chapter and summarised in figure 7, tables 6 and 7, pages 154, 155 and 156. However, the prevailing policy of objective, nondirective informed choice in both maternity services as a constraint to service provision is explained (see subsection 7.4.1).

This is followed by the discussion about the power differential between midwives and women and how in combination with the constraints to service provision may lead to the routinisation of screening. The concepts of expert authority (concept 2 in figure 8) and routinisation (concept 3 in figure 8) were developed from the findings, themes and subthemes of midwives' influence on women's decision-making including midwives implicit or explicit directive to women to agree to screening, offering Down's syndrome screening as a routine test and women and partners' perception of screening as a routine test.

Subsections 7.4.2 and 7.4.3 provide more details about the concepts.

Next is the concept of tensions (concepts 4 and 5 in figure 8) developed from concepts 1, 2 and 3 which involved themes and subthemes such as midwives, women and their partners' experiences of Down's syndrome screening and information related anxiety. Others are midwives' unmet training needs, women and partners' lack of understanding of the concept of risk and women and partners' reasons for accepting or declining screening. In addition, these subthemes reflect differing perceptions among midwives and between midwives and women. The findings, subthemes and themes and the differing perceptions within the preceding concepts, generated tensions within and between midwives, women, and partners and in the prenatal environment. For a description of the tensions, see subsection 7.4.4.

The two-directional arrows in figure 8, indicate that the service constraints reinforced the expert authority, routinisation of screening and tensions in the prenatal environment, within and between midwives, women and partners and vice versa. Therefore, these influences set up a vicious cycle of constraints on women and partners' decision-making process in the prenatal context. The differing patterns of constraints including the nuances in the

operationalisation of the screening guidelines in both maternity services may account for the variation in uptake rates (concept 7 in figure 8). The concept of variation in uptake rates is discussed in section 7.5. The findings also revealed different types of relationships (concept 6 in figure 8) formed in the brief midwife-woman encounter in the prenatal context. These are explained in section 7.6. In addition, the different decision-making models adopted as policies or recognised in the screening programmes (concept 8 in figure 8) are described in section 7.7.

## **7.4 Integrated discussion**

The literature reviewed at the start of this thesis demonstrates the influence of service organisation and delivery on women and partners' participation in prenatal Down's syndrome screening has not been fully investigated. Existing studies have focused on the rationalistic biomedical differences in women offered second trimester serum screening to account for the variation in uptake rates of Down's syndrome. However, the evidence has been inconclusive. In addition, the review revealed that little was known about how the national screening policies and guidelines are implemented in practice. A research question, aim and broad objectives were then proposed to illuminate these gaps.

### **The main research question**

Do the structures, processes and people in the maternity services influence women and partners' participation in prenatal screening for Down's syndrome?

### **Aim**

To explore the influence of service organisation and delivery on women and partners' participation in screening in two different health districts in England with contrasting uptake rates.

### **The broad objectives**

1. To explore whether the structures of the maternity services influence women and partners' participation in prenatal Down's syndrome screening
2. To explore whether the process of offering prenatal Down's syndrome screening influence women and partners' participation in the programme.
3. To explore whether community midwives influence women and partners' participation in prenatal screening for Down's syndrome.

The findings from the document review and interviews addressed the question, the aim and objectives. This thesis has shown that the influence of the ambience of the prenatal environment and interpreters may constrain women and partners' participation in screening. In addition, the nuances in the operationalisation of the screening policy such as the repeat offer of screening and obtaining consent for screening at dating scan in Landscape maternity service may account for the difference in uptake.

The process of information-giving was often rushed, the offer of Down's syndrome screening not linked to TOP and the offer made among a battery of more routine blood tests. These constraints may influence women and partners' participation in screening. A midwife also revealed that some midwives appear to increase the uptake rates of screening. Whilst others had stereotypical beliefs that, women from ethnic minority groups decline screening for cultural reasons. These structural and process constraints influenced the midwife–woman interaction and women's decision-making processes. These findings and others are discussed under the different concepts in the model (figure 8) throughout this chapter.

#### **7.4.1 Informed choice as a service constraint in prenatal Down's syndrome screening**

The offer of Down's syndrome screening is based on the policy of autonomous and informed choice (Council of Europe, 1990; NICE, 2008; NHS FASP, 2011). The concept of autonomous and informed choice is hinged on the biomedical paradigm of nondirectiveness and autonomy to address the issue of eugenics, protect midwives from the emotional impact of screening and to guide against litigation (Clarke, 1997; Williams et al., 2002a; Williams et al., 2002b; Rantanen et al., 2008; Skirton and Barr, 2010; Hertig et al., 2013). The policy of autonomous and informed choice involves midwives presenting clear and complete information about screening, free of the influence of the midwives' value system. Women are expected to choose freely whether to accept or decline screening based on good understanding and what to do with the results of screening that is consistent with their values, beliefs, circumstance and life plans (Beauchamp and Childress, 2001; Gidiri et al., 2007; García et al., 2008b; Asongu et al., 2010).

As has been shown in this thesis, the classic instances of women capable of autonomous decision-making and midwives capable of giving full information objectively and nondirectively was not being achieved in either maternity services. Thus, midwives, pregnant women and their partners may not be in the ideal positions for the policy of informed

choice, owing to the potential interference of service constraints. A midwife in Terrace maternity service succinctly described the influence of constraints to service provision on informed choice in the prenatal context;

*"After all that info piled upon a newly pregnant women in 50 minutes, when you get to the end and asking about Down's screening, of course the answer will be an uninformed yes as she will be tired/hot/stuffy room and brain-dead. And remember we do need to ask for a decision if possible and if not we request one and tell them they can change their minds. If she is a non-English speaker the appointment may have been going on for ages so she will be even more brain-dead"* ID182T.

Indeed, in some instances, women may not want to play an active part or lack the capacity for decision-making or may want to delegate decision-making (Beauchamp and Childress, 2001; Kirklin, 2007; Hertig et al., 2013; Noseworthy et al., 2013). As such, the informed choice model may not be an ideal policy for prenatal Down's syndrome screening. Anderson (1999) challenged midwives to question the assumed ethical and moral value of nondirectiveness in autonomous decision making by exploring values and beliefs that influenced women's decision-making. The author surmised that genetic counselling based on the principle of nondirectiveness created tensions and threatened the development of a genuine midwife-woman centred relationship (Evans et al., 2004).

Further, the biomedical model failed to recognise that information presented to women and their partners is rushed considering multiple complex competing issues at booking as revealed in the online interviews. In some instances, there was little to no discussion, specific advice or support given during decision-making in the prenatal context. Thus, women were exposed and vulnerable to the overwhelming external influences and constraints to service provision. Lippman (1999) reported that there is overwhelming evidence of powerful external influences and constraints on women's decision-making processes in the prenatal context. Yet, the biomedical model of autonomous and informed choice tends to locate all influences and pressure on decision-making processes internally in women and their partners.

The individualistic, biomedical model of informed choice discourages dialogue (information exchange) between midwives, pregnant women and partners, in the interest of the principle of nondirectiveness. The policy therefore implied a one-way information transfer.

Information transfer was evident in both maternity services as midwives offer the same

minimum information to pregnant women and partners, owing to increased workload and multiple competing issues at booking. However, women and their partners have varying information needs. Therefore the policy of informed choice may in fact reinforce the midwife-woman power/ knowledge divide (see concepts 1 and 2 in figure 8). Existing research has shown that the individualistic, biomedical informed choice, is an ideal model which does not take into account the practical conditions of interactions between midwives, women and their partners in many healthcare settings (Pilnick, 2004; Pilnick, 2008; McNeill and Alderdice, 2009; McNeill et al., 2009; Reid et al., 2009; Tsouroufli, 2011). The finding that the classical situations of women capable of autonomous and informed choice and midwives capable of giving clear and complete information objectively may not coincide in the prenatal context is unique.

#### **7.4.2 Power differential between midwives, pregnant women and partners**

The information transfer implied a power and knowledge differential between midwives who were seen as experts by women, and women and their partners who see themselves as laypersons. This expert authority is achieved through the midwife's ability to interpret complex technical and probabilistic risk information that is often not fully understood by women and their partners. Expert language reinforced the midwife/ woman divide which may influence how the offer of screening is interpreted and consent negotiated. Work by Foucault (1982) suggests that the health profession is often criticised for exerting unrestrained power over the general public's decision-making about health.

This power differential may lead to emphasis being given to certain information which may be perceived as directive or influence decision-making in a certain direction, despite the best effort of the midwife to remain neutral. As shown in both maternity services, midwives exercise some control over the decision-making process by how information is presented through selective information-giving. This has been reported in previous research where health professionals influenced service users' decision-making (Hayeems et al., 2009; Park and Mathews, 2009; Hertig et al., 2013). As a result, the discussion and offer of screening is a complex interaction of several factors, some of which may not be easily addressed in the prenatal context as previously reported (Lynn et al., 2010).

Moreover, women's trust in the knowledge and beneficence of the midwives and the perceived endorsement of screening by the healthcare profession makes more visible and evident the expert/ lay divide. The result was informed 'compliance' rather than true and

informed choice as suggested in this thesis and existing research, which may serve to routinise the offer of screening (see concepts 2 and 3 in figure 8). The concepts of expert authority and routinisation have been reported in the metasynthesis by Reid et al. (2009).

#### **7.4.3 Routinisation of prenatal Down's syndrome screening**

Women and their partners in both maternity services revealed midwives explicitly directed women or influenced women and partners' choices in the prenatal context. The offer may not exclude the option of choice, but appears to undermine it. Further, this influence was exacerbated by the enthusiasms of the midwives about the reliability of the combined screening test as reported in Terrace maternity service. Additionally, the portrayal of the midwife as an expert in prenatal care or the benefits of medical technology may have reinforced the midwives' influence as noted in existing work (Anspach, 1993; Maynard and Schaeffer, 2002; Pilnick, 2008).

This finding supports the suggestion by Paul (1998) cited in Seavilleklein (2009) that the strongest determining factor in women's decision-making about Down's syndrome screening was not in the attitudes of the women, but in the manner of informing or approach by healthcare professionals. Existing research has shown that the principles of nondirectiveness and autonomy have not prevented midwives from giving their value judgments of the screening tests (Pilnick, 2008; Ghosh et al., 2011; Tsouroufli, 2011). In addition, midwives in many other aspects of their role do not practice in a nondirective way, for example, in the labour ward as noted in Hindley and Thomson (2005).

Often, decisions about screening have to be made quickly so that women do not miss out on the combined screening test, because of the short window of opportunity. Midwives may therefore spend less time explaining the test to women and more time 'pushing' screening as demonstrated in Landscape maternity service. The women may not have had the freedom for informed reflection in the decision-making process. This pressure may result in women either declining screening from the frustrations felt from lack of understanding the concept of risk or accept screening over the perception that it was a non-invasive test. Lewando-Hundt (2006) suggests that a rushed prenatal consultation where midwives are explaining procedures and soliciting 'informed consent' at the same time, does not provide the best environment for reflection. Therefore, routinsation of screening reinforces the power/knowledge or expert authority of midwives as indicated in the model (concepts 2 and 3 in figure 8).

This thesis demonstrates that the mere availability and offer of screening in the prenatal context meant it must be for the wellbeing of mother and baby, even without any intention of the midwife to be directive. Furthermore, the offer of screening may not be perceived as a choice, in view of the ‘positioning’ of the screening test among a battery of other routine tests. The positioning arose from the presentation of the offer of screening alongside the offer of routine tests at the prenatal encounter, which may serve to routinise it. Similar findings have been reported in existing work (Pilnick, 2004; Pilnick, 2008; Tsouroufli, 2011).

Moreover, midwives in both maternity services categorised Down’s syndrome screening with other routine tests in pregnancy as equal in terms of take-up in the prenatal context. Mattei, (2000) cited in Vassy (2006) suggests that health professionals and pregnant women in France categorised Down’s syndrome screening with other prenatal routine tests in pregnancy, because it is routinely offered to all women. The significance of such comparisons is that it undermined the need for active consideration of the offer and midwives may intentionally or unintentionally offer screening as a routine test.

Interestingly, women in both maternity services reported that Down’s syndrome screening was offered to them by midwives as a routine test. A pregnant woman claimed it was offered as “*part of a Gold package offered to all pregnant women*” ID137T. Thus, midwives may present screening as a non-invasive routine test to avoid complex negotiations and encourage passive engagement of women and their partners in the decision-making process. This subtheme of midwives offering the combined screening test as a routine test is a unique finding.

The interview data suggest the way information and the offer of screening was framed by the midwife influenced women and their partners’ participation in screening. The offer of screening was made with the rhetoric of choice, with midwives in some instances, not emphasising the possibility of TOP and often obscuring the issue largely by not linking it to screening. Therefore, the offer of screening was perceived by women and their partners, as requiring no active consideration of screening options undermining autonomous and informed choice. This finding is supported by previous research which reported midwives not linking screening with TOP because of organisational, cultural and religious reasons (Boyd et al., 2008; McNeill and Alderdice, 2009).

The offer of screening in a standard hospital and the screening test conducted in the context of an ultrasound scan may further the normalization or routinisation of screening in both

maternity services. The combined screening test has the incidental side benefit of an ultrasound scan. Indeed, enthusiasm for the ultrasound scan which is seen as a routine procedure for viewing and obtaining scan photos of the baby may influence pregnant women and partners' decision making processes. Most women and their partners have positive feelings about the ultrasound scan and expect it to be a time to obtain photos of the baby, to take home and display or give to family members. It is also the time to confirm the wellbeing of their baby as reported in previous research (Garcia et al., 2002; Ekelin et al., 2004; Åhman et al., 2010; Georgsson Öhman and Waldenström, 2010).

Previous research by Favre et al. (2008) and Aune and Möller (2012) reported that it may be difficult for women to exercise choice with first trimester ultrasound scanning. This is as a result of most women's beliefs that ultrasound scan is compulsory. Choice in this context may be assumed to be no longer voluntary or without influence (Beauchamp and Childress, 2001; Pilnick, 2004; Williams et al., 2005; Pilnick, 2008; Schoonen et al., 2012). Therefore, even when women understood that they have a choice about Down's syndrome screening, the context of prenatal screening not only routinise screening, but also made choice illusory (Suter, 2002; Pilnick, 2008).

In this regard, some women and their partners in both maternity services could not clearly articulate the implications of screening that they had willingly consented to. The connotation is that the normalisation or routinisation may sometimes be as a result of the collusion between midwives and women to circumvent the complex process of decision-making involved in Down's syndrome screening. In addition, women and partners may be complicit by participating passively and refusing to ask the larger questions which hindered discussion about screening resulting in uncritical and unquestioning acceptance of screening. Besides, the fact that some women paid for the screening privately in previous pregnancies, but now have it provided free in the NHS, may promote the routinisation of screening in both maternity services as indicated in this thesis. This assertion is supported by previous studies (Pilnick, 2004; Pilnick, 2008).

Furthermore, a midwife in Terrace maternity service reported that some midwives influenced women's decision-making processes in order to increase the uptake rates of prenatal screening for Down's syndrome. These constraints to service provision may result in a routine screening programme for pregnant women and their partners. Existing studies have reported that prenatal screening is institutionalised and services produced en masse

(Williams et al., 2002b; Tsouroufli, 2011). The experiences of pregnant women participating in Down's syndrome screening have also been referred to as the conveyor belt experience (Hundt et al., 2011).

#### **7.4.4 Tensions in the context of prenatal Down's syndrome screening**

##### **7.4.4.1 Tensions within midwives**

The documentary analysis revealed that the various guidelines and care pathways in the prenatal context are seen as the rules. This has the potential to create tensions from the dissonance between objective, nondirective information-giving and forming trusting relationship with women. The tensions created may result in some midwives developing coping mechanisms or professional dissociation. The ramification of coping mechanisms or dissociation may account for the different relationships seen in the prenatal context (see concept 6 in figure 8). van den Heuvel et al. (2008) noted that healthcare professionals have a duty to implement policies and guidelines wholly in practice. In addition, work by Porter et al. (2007) and Blaaka and Schauer Eri (2008) suggests that midwives may adhere to policies and protocols rather than negotiated decisions with women. The authors surmised that midwives experienced tensions when required to negotiate decisions with women.

O'Connell and Downe (2009) suggest in their metasynthesis of midwives' experiences of hospital practice, that midwives by nature of their role, desire to provide individualise care to women, but are constrained by the maternity context and organisational structure that value detachment. The reason may be the powerlessness or tensions felt by midwives from the perception that they cannot be personally responsible for the care they provide. The meaning for both maternity services is that objective information provision by midwives is incongruent with their desire to forge rapport or relationship with women, generating discord and tensions within midwives.

Tensions emerged in midwives when interpreters do not have the right words to effectively translate information about screening to women owing to lack of understanding or struggling with the complexities of the concept of risk. Health professionals have been shown to grapple with the concept of risk (Nagle et al., 2008). The difficulties experienced by interpreters in translating risk information about screening may adversely affect the communication between midwives and women. This has the potential to adversely affect women and partners' understanding of information about screening. Ineffective communication has been reported to impair women and partners' ability to understand,

retain and adjust to information about screening (Sharpe, 1996). Therefore, interpreters may influence women and partners' decision-making process about Down's syndrome screening. This subtheme of the potential influence of interpreters is a unique finding.

Reed (2009a) in a study conducted in the UK, reported that male partners acting as interpreters, did not always provide women with all the information they were requested to translate. Furthermore, Preloran et al. (2005) used observation methods to determine the influence of interpreters on women's amniocentesis decisions in the USA. The study found that interpreters appeared to have more influence over women's amniocentesis decisions than might have been anticipated.

The lack of time to inform women about screening generated tensions in midwives in both maternity services. The major impact of time pressure was on the ability of midwives to form relationships and to adequately inform women about screening at booking. The tensions experienced by midwives undermined their autonomy. Importantly, midwives may not take women's values and beliefs into consideration when informing about screening or omit certain information from consideration. This finding is supported by existing work that suggests adequate time is essential for midwives to establish trust and rapport to enable women to open up and discuss. Therefore, time constraints influence the way care is given and may lead to depersonalised care as suggested in existing literature (Browne et al.).

Existing studies have reported that time pressure affects the quantity and quality of information given to women and their partners in the prenatal context of screening (McNeill and Alderdice, 2009; Park and Mathews, 2009; Tsouroufli, 2011). McNeill and Alderdice (2009) and McNeill et al. (2009) reported that midwives were concerned with the time they had to explain screening and the actual time needed to discuss screening sufficiently. The significance is that screening was offered to women with inadequate information and understanding resulting in women participating passively in screening. This may serve to reinforce the routinisation of screening (see concepts 3 and 4 in figure 8).

To cope with the tensions and enhance women and partners' understanding of screening some midwives resort to the use of photographs of children with Down's syndrome and scenarios. Research conducted in the USA revealed that women valued the use of photographs that depict the realities of people from a different ethnicity living with the condition (Houts et al., 2006; Levis et al., 2012). The implications of using photographs or posters of children with the condition to inform women are unknown. A study reported that

it will make women more anxious about having a baby with the condition (Figueiras et al., 1999). However, the use of photographs and scenarios as illustrations when informing women and partners at ‘booking’ were not recommendations in the guidelines and policies for Down’s syndrome screening.

#### **7.4.4.2 Tensions within women**

As demonstrated in both maternity services, information about screening aroused strong emotional reactions from women. These were threatening thoughts about TOP and miscarriages from diagnostic tests, but also about having a baby with Down’s syndrome. Foetal screening for Down’s syndrome, therefore generated tensions in women as the condition has no alternative obstetric ‘intervention’ to selective abortion and not all women considered TOP to be an option. Women who wish to continue with a screen positive pregnancy are offered support and referred to the consultants in fetomedicine for care. Vassy (2006) reported that TOP is a controversial practice that is unacceptable to many women. Therefore the offer of screening is associated with some difficulties, including complex information about risk and unsure anticipation, which may lead to ethical dilemmas and psychological stress (Gates, 2004; Green et al., 2004).

The interview data revealed a pregnant woman in Terrace maternity service accepted screening in order to obtain information about her baby. The screening result was important because of her cultural or societal perception about the suffering of children with the condition. Asongu et al. (2010) and Ahmed et al. (2012b) reported on the diversity of views expressed by women in general and within ethnic groups and recommended that midwives should avoid cultural sensitivities when informing women in practice. Existing studies that explored the perspectives of women in developing countries reported that participants considered the ‘mercy killing’ of children with Down’s syndrome to be a grave sin. However, there were prevailing concerns among pregnant women about children with Down’s syndrome being vulnerable to sexual abuse within the family network (Modra, 2006; García et al., 2008b; Bryant et al., 2011). Hence, the tensions within women which arose from participation in screening must be understood within the context of familial, cultural, economic and social experiences and pressures including the stigmatisation and lack of support given to children with disability (Rozario, 2005; Ahmed et al., 2006a; Atkin et al., 2008; Tsianakas et al., 2012).

Women reported being overwhelmed with information at booking (see figure 7 on page 154). The appointment is information rich, as pregnant women are given information on a variety of routine tests, assessments and care. Therefore women face a steep learning curve about prenatal Down's syndrome screening, understanding the concept of risk and make decisions within a short window of opportunity for first trimester screening. The information overload exerts a strain on the educational and cognitive processes needed to make autonomous informed decisions. Tensions emerged within women from processing large amounts of information in the prenatal context. Existing studies have found that it may be stressful for pregnant women to deal with large amounts of information in an orderly manner, often resulting in decisions being made from the context rather than from the content of the information (Bekker et al., 1999; Green et al., 2004; Reid et al., 2009; Farrell et al., 2011; Schoonen et al., 2012; Dheensa et al., 2013b). The inference is that autonomous and informed choice which should contribute to a sense of autonomy, in reality reduces it. This does not mean the women did not act autonomously within these constraints to service provision. Most women were keen to report that their participation in screening was without coercion and based on their beliefs and value system. Interestingly, some of the women reported not thinking through their decisions. Such perceptions appeared to point towards women bypassing decision-making processes in relation to screening.

#### **7.4.4.3 Tensions within partners**

The offer of risk information about Down's syndrome screening generated tensions within partners and relief when screening results came back lower risk. This may indicate over reliance on test results and less understanding of screening, as a negative result does not mean absence of the condition. The tensions were due to the possibility of having a baby with Down's syndrome, wanting reassurance and where screen positive, miscarriage from diagnostic testing and consideration of TOP. Partners often agreed to screen for reassurance and to fully engage with prenatal care in the best interest of their baby. This may be in part, due to partner's beliefs that screening is a non-invasive routine test and/ or was compulsory in pregnancy for the well-being of mother and baby (see concepts 3 and 4 in figure 8).

A partner at Terrace maternity service believed the term "Down's syndrome" has negative connotations. This belief may generate tensions within partners. Existing work has reported that the use of terms such as abnormalities, defects and risk when informing may influence how partners interpret the information and their participation in screening (Seavilleklein,

2009). In addition, the notion of 'risk' itself is inherently negative due to its definition as the chance or probability of being exposed to hazard, damage, injury or loss as noted in Polansky (2006).

Furthermore, several partners reported they had decided before 'booking' to agree or disagree to screening. Interestingly, some partners found it difficult to distinguish screening from diagnostic testing. This generated tensions within partners, as some partners encouraged pregnant women to decline the offer of screening, due to the risk of miscarriages associated with diagnostic testing. The suggestion is that some partners may have assumed that acceptance of screening meant acceptance of diagnostic testing with miscarriage as a possible complication. Therefore, such partners may likely agree to a diagnostic test that is non-invasive and without the risk of miscarriage. With the impending implementation of early non-invasive prenatal diagnosis (NIPD) in the NHS which is now offered in some private hospitals in England, screening for Down's syndrome may be perceived as a safe procedure by partners. Partners may agree to screening based on this perception and bypass decision-making about screening and diagnostic testing. This finding emphasised the need for midwives to ensure that male partners understood screening information to facilitate informed decision-making with their pregnant partners.

#### **7.4.4.4 Tensions between midwives, pregnant women and partners.**

Research has reported that emotion is a source of difficulty for women and partners and strongly influence decision-making processes about Down's syndrome screening (St-Jacques et al., 2008; Dheensa et al., 2013a). The interview data suggest tensions emerged between the desires of women and partners to avoid harm to their baby and the focus of the midwives to detect abnormality. These tensions have also been reported in existing research which called for improvement in communication between midwives and women (Hunt et al., 2005; Li et al., 2008).

Further, asking pregnant women in Terrace maternity service for a decision at booking created tensions within and between midwives and women and in the prenatal context. The tensions within women may be responsible for women asking for advice or midwives' opinions. Women may see asking for advice as support and a means to seek the midwives' value laden interpretation of the information about risk. This assertion is supported by previous research (Novick, 2009; Ahmed et al., 2012b; Ahmed et al., 2012c).

The emotions expressed by midwives, women and partners in this thesis, demonstrated by the emotive cues seen in their responses revealed the tensions in midwives' role offering screening to women. Yet it may indicate gaps in the perceptions between midwives, women and partners regarding emotional difficulties, information overload, difficulties with understanding the concept of risk and differentiating screening from diagnostic test. The use of emotive cues in internet research and the tensions experienced by midwives in prenatal care have been reported in previous studies (Im and Chee, 2006; Pilnick, 2008; McNeill and Alderdice, 2009; McNeill et al., 2009; Tsouroufli, 2011).

Tensions within midwives and women may further encourage collusion, promoting the routinsation of screening (see concepts 3 and 4 in figure 8). This was demonstrated in Landscape maternity service when midwives offered Down's syndrome screening to women without linking the offer to the implications of screening such as TOP. Moreover, the threat of legal litigation created psychological anxiety within midwives. To cope with the anxiety, midwives presented screening as a routine test and recommended screening as a maternal responsible choice in order to collude with women. Furthermore, the fear of litigation has motivated the programme to produce a checklist for screening information and detailed informed consent form. In addition, it constrained the discussion about screening, resulting in a greater focus on the practicalities and the procedures of the screening tests as revealed in the checklist seen in the document review and in previous research (Favre et al., 2008). Seavilleklein (2009) reported that the fear of litigation, resulting from women who were not screened having a child with Down's syndrome, may cause some midwives to recommend screening to women. Further, Pilnick (2008) and McNeill and Alderdice (2009) suggest that the way midwives presented screening information to women did not invite dialogue or discussion and was perceived by women that the choice had already been made for them. Moreover, the complexity of the concept of risk made information provided by midwives difficult to understand by women and partners. The tensions generated may cause women to decline screening as a result of the frustration experienced, or accept screening from the powerlessness experienced. Existing literature reported that health professionals may 'disempower' patients by restricting their involvement in dialogue and decision making process on the basis of selective information-giving (Broom, 2005; Edwards et al., 2009).

Most male partners in this study reported midwives did not directly influence their participation in screening. One partner felt it was advisable to undergo screening, as the conception was via in vitro fertilisation (IVF). This finding is contrary to that in existing

research that suggests women with fertility problems were less likely to accept screening, particularly the diagnostic tests (Potter et al., 2008). Although, some partners reported that midwives directed or recommended screening to their pregnant partners. In addition, a partner felt pressured by a midwife in Landscape maternity services to encourage the pregnant partner to accept screening, but resisted over fears of miscarriage from diagnostic testing. The different goals, purposes and values between the midwife and the couple generated tensions, especially with the prevailing knowledge that most women accepted screening.

Draper and Ives (2013) suggested that tensions emerged when midwives tried to maintain a woman-centred focus and at the same instance ensure that the views expressed were that of the pregnant woman and not that of the partner. In addition, partners may feel ignored or excluded when midwives cater more for pregnant women than partners. However, none of the partners in either maternity service reported feeling excluded from the discussion at booking. Existing studies have reported some partners felt ignored or left as bystanders during antenatal testing (Locock and Alexander, 2006; Reed, 2009b; Reed, 2011; Williams et al., 2011; Dheensa et al., 2013a; Dheensa et al., 2013b). The reason for the difference in findings may be because most of the existing studies were not focused specifically on Down's syndrome screening.

#### **7.4.4.5 Tensions within the prenatal environment**

The document review indicates the different decision-making models demonstrated in both maternity services may be the midwives' response to the contradictions, constraints and tensions in the prenatal context. Midwives are obliged to engage in objective and nondirective informed choice which is the stated policy. However, the same guidelines encouraged midwives to develop trusting meaningful relationships with women to enhance understanding of information and support decision-making based on their beliefs, values and personal circumstances (shared decision making). Nevertheless, the online interviews revealed some midwives recommended screening or directed women down a particular pathway or they support women by being directive (paternalistic model). The shared and paternalistic decision-making models are therefore policies-in-action in the prenatal context.

The multiple complex competing issues at booking may cause midwives to prioritise some issues over others. This may be as a result of increased workload due to volume and type of caseload, language problems which may be compounded by the absence of interpreters.

Hence, information about screening may be inadequate, selective and often rushed within available time and may give the woman the impression that the midwife is busy, especially if there are other pregnant women waiting for their appointments. In addition, the fear of litigation may drive midwives to obtain consent from women with poor understanding about screening. These constraints to service provision generated tensions within the prenatal environment (see concept 5 in figure 8). Previous work by Reed (2009a) reported that the prenatal environment is pressurised which makes recruiting midwives into research difficult.

The tensions within the environment may influence women's decision-making processes and exacerbate the routinisation of screening as shown in this thesis (see concepts 3 and 4 in figure 8). A midwife believed the ambience of the consulting rooms may increase a pregnant woman's anxiety about the possibility of abnormality with the foetus which may influence them to accept screening. This could also happen to women who have an aversion for clinical environments and women who are new or not used to the NHS such as women from ethnic minority backgrounds may find the environment overwhelming. Rowe et al. (2006) and Farrell et al. (2011) suggest that anxiety generated in prenatal settings may result in women becoming less thoughtful or having impaired ability to acquire, recall and synthesize information about screening. Women's acceptance or rejection of screening would then be based on decisions made from the context rather than from the content of the information given as noted in existing work (Bekker et al., 1999; Schoonen et al., 2012). Nicol (2007) reported that it may be impossible to achieve informed choice, due to the hospital environment and culture. Nicol further suggests that when service users find themselves in a stressful situation, they cope by complying with what they perceive as the health professionals' recommendations.

Women in both maternity services perceived the prenatal setting as part of the standard NHS healthcare system where tests are done to detect any problems in pregnancy which could then be dealt with by experts within the medical system. Furthermore, participants in both maternity services demonstrated that expert authority is made more visible in the prenatal context by the ambience of the prenatal environment. The display of medical equipment in the consulting rooms, the office layout, the uniform of the midwife and the seating arrangements in the consulting room could be psychological tools that display power. A clean, tidy, relaxed seating arrangement foregrounded their perceptions of screening as a routine test. The outcome then, was some women became compliant, accepted screening and/ or all tests offered in the prenatal setting. The literature by Davis

and Walker (2010) suggests that the space and layout of the obstetric hospital setting enabled power to circulate among health professionals.

The booking appointments at women's homes are no different either, as the midwife is seen as a proxy of the standard NHS healthcare system and any tests offered must be for the wellbeing of mother and baby during pregnancy. Mander and Melender (2009) reported that women perceived midwives as extensions of the trusted healthcare system when making decisions about place of birth. The subtheme of the influence of the environment on women's participation in screening is a unique finding. Previous studies suggest that the prenatal context or environment is highly pressurised, because of the dilemmas, emotions and reactions commonly encountered (Bennett et al., 2003; Georgsson Öhman et al., 2009; Reid et al., 2009). Davis and Walker (2010) drawing on a body of literature that explored the body's chemical response to labour and birth suggests that the obstetric environment may trigger emotional responses such as fear and anxiety. Although, the emotional reactions and dilemmas encountered in the prenatal environment may cause some women and partners to reject screening and resign to fate.

The various constraints to service provision and tensions introduced a perception of lack of professional control by midwives, over their roles in screening which studies have reported may affect midwives' communication and relationships with pregnant women (Morse, 1991; McCourt, 2006; McNeill and Alderdice, 2009). Midwives attempt to relieve the tensions felt within and in the prenatal context by controlling the quality and quantity of information-giving and presented screening routinely either from a negative or positive perspective. The inference is that the tensions reinforced the expert authority and routinisation of screening within the prenatal environment maintaining the cycle of constraints on women's decision-making processes (see concepts 1, 2, 3 4 and 5 in figure 8).

## **7.5 Variation in uptake rates of screening between the two maternity services**

This thesis suggests there are powerful service organisation and delivery factors influencing and constraining women's participation in screening in both maternity services. The patterns of constraints to service provision obtained from the document review and interviews in both maternity services are summarised in tables 6 and 7, figure 7 and concept 7 in figure 8. The tables and figures will be referred to throughout this section.

The online interviews indicated that in Landscape maternity service, 21% (n=4) compared to 13% (n=2) women in Terrace maternity service reported that they were given little to no verbal information about screening, just written information to take home and read. This may result in more women in Landscape maternity services assuming that screening is a routine procedure with informed choice eroded or undermined. Previous studies suggest that women accepted screening when it was routinely presented, as it enabled midwives and women to engage with prenatal screening, while avoiding the uncomfortable issue of miscarriage and TOP (Markens, 1999; Heyman et al., 2006; Reid et al., 2009).

Additionally, 75% (n=12) of women in Terrace maternity service compared to 58% (n=11) of women in Landscape maternity service, claimed to have prior knowledge and experience of screening from sources such as previous pregnancies and the internet. This suggests more women in Terrace maternity service might have thought about screening before the booking visit. Furthermore, 63% (n=10) of women in Terrace maternity service compared to 68% (n=13) in Landscape maternity service reported having decided before the booking visit. This is similar to the findings in previous research (Pilnick, 2004; Tsouroufli, 2011). Lewando Hundt (2004) reported that 49% of women in their study had already made up their minds about screening before being offered any information.

The potential ramification is that women without prior knowledge and who are undecided about screening may be more susceptible to the influence of service organisation and delivery in the prenatal context. Pilnick (2004) reported that participation in screening is not always actively considered by women before the booking visit, making women more vulnerable to external influences. Rapley (2008a) argued that decisions can be shaped over time and influenced by knowledge or information acquired from previous experiences or sources such as the internet and family. Yet, Müller et al. (2006) argued that if prenatal screening is routinely offered to all women, more women may decline screening based on negative experiences in previous pregnancies or those of someone else.

A subtheme that is specific to Landscape maternity services was information overload reported by women. In addition, more midwives in Landscape maternity service, 47% (n=9) compared to 33% (n=5) in Terrace maternity service believed women and partners are given too much information at booking. Information overload has been shown to have a detrimental impact on an individual's ability to understand and to make informed choice. Previous studies have shown that the psychological effect could have a negative impact on

comprehension and retention of information (Polansky, 2006; Farrell et al., 2011; Schoonen et al., 2012). The indication is that women's participation in screening may be influenced by the prenatal context such as workplace culture or expectations and not the content of screening information.

More women in Landscape maternity services 26% (n=5) compared to 6% (n=1) in Terrace maternity service reported being 'pushed' (explicitly directed) by midwives to accept Down's syndrome screening. Moreover, 37% (n=7) of women in Landscape maternity service compared to 25% (n=4) of women in Terrace maternity service reported midwives' overt and covert attitudes influenced their participation in screening. These findings suggest that there may be an institutional push or culture for women to conform to screening in Landscape maternity service. The influence of midwives on women's decision-making has been reported in previous studies, but the evidence has been inconclusive as demonstrated in the literature review. This thesis adds to the knowledge base that midwives influence women's decision-making in Down's syndrome screening.

In addition, more women, 63% (n=12) in Landscape maternity service compared to 16% (n=3) in Terrace maternity service, reported that prenatal Down's syndrome screening was offered to them as a routine screening test or perceived it as a routine test in pregnancy. This suggests more women and partners in Landscape maternity service may have participated passively in screening. The differences in perception of screening as a routine test may be responsible for the high uptake rates for screening in Landscape maternity service compared to Terrace maternity service.

The implications of screening were explained to more women in Terrace maternity service, 56% (n=9) compared to 42% (n=8) women in Landscape maternity service. Thus, more women in Terrace maternity service may have decided based on the consideration of the implications of screening according to their personal values and beliefs. This is more likely to impact on women's take-up of screening. Therefore, more women in Landscape maternity service may have perceived screening as a 'no risk' procedure or for the best of mother and baby. Therefore, the uptake rate of screening is likely to be higher in Landscape maternity service compared to Terrace maternity service. Research suggests that inadequate information-giving about screening to women is equivalent to substandard care and may influence women's decision-making (Etchells et al., 1996; Winquist et al., 2008).

Likewise, the low uptake rates of screening in Terrace maternity service may be as a result of the tensions and pressures sensed by women, over the need for an immediate decision about screening at booking. This may result in some women declining the offer of screening, owing to frustration or confusion from lack of understanding and also to avoid the emotional burden of screening decision-making. Existing work suggests women who reported they were given insufficient information or the information did not meet their needs experienced frustration and were less trusting. This perception deterred participation in subsequent prenatal care visits (Novick, 2009)

Interestingly, some women in Terrace maternity service may find it easier to opt-in, but opt-out becomes difficult, especially with undecided women who may perceive the appointment for the screening test as a recommendation to screen. Whilst some midwives recommended the combined screening test to women owing to its reliability, others were dissuading women from screening by emphasising TOP, stress related conditions associated with screening and the high false positive and negative rates with serum screening.

There is also a stereotypical view among the midwives in Terrace maternity service that women from ethnic minority backgrounds decline screening for cultural reasons. This view or workplace culture may affect the way screening is presented to women and their partners. For example, ethnic minority women may have screening offered to them from a cultural perspective as noted in existing research (McNeill and Alderdice, 2009; McNeill et al., 2009). Previous research by McNeill et al. (2009) conducted in a context of low uptake rates reported that some pregnant women who accepted or declined screening were influenced by midwives.

As shown in table 7, the nuances in the operationalisation of the national screening policies and guidelines at the level of the maternity service may explain the variation in uptake rates. The document review revealed that how the service is organised may have an influence on the take-up of screening. Women who declined screening at the dating scan in Landscape maternity service were offered the quadruple test when they present for the 15-16 weeks appointment with the midwife. Repeating the offer of Down's syndrome screening at the 15-16 weeks appointment may communicate, intentionally or unintentionally, a perceived directiveness to accept screening for the wellbeing of mother and baby. Therefore, women's participation in screening may be affected by the coerciveness of the repeat offer, which may lead to increased take-up of screening.

In addition, how the information-giving and consenting processes are organized in Landscape maternity service may account for the difference in uptake rates. Information about screening is presented by midwives at booking. The request for consent to NT scan is made by the sonographers at the dating scan, which may be perceived by women and their partners as a responsible choice. This may create a perceived institutional culture, structural or organizational expectations for women to conform to screening. Research has shown that consent to Down's syndrome screening is often assumed by midwives, unless the pregnant woman actively refuses it (Pilnick, 2004; Pilnick, 2008; Tsouroufli, 2011; Pilnick and Zayts, 2012). McNeill et al. (2009) stated that the way care is organised in the maternity services may influence the midwife-woman interaction. The findings of the nuances in the operationalization of the screening policies and guidelines illuminate the gap in the literature on the implementation of the guidelines across maternity services in England.

## **7.6 The relationships formed in the prenatal context of Down's syndrome screening.**

Davis and Walker (2010) noted that the biomedical construction of the maternity services marginalised midwife's knowledge and practices which may shape the lens through which they practice. This constraint may account for the different relationships formed in the prenatal context. Work by Morse (1991) suggests that there are two categories of relationships formed in the clinical setting. These are unilateral and mutual relationships. However, there are four main types of mutual relationships. These are the clinical, therapeutic, connected and over-involved relationships. The interviews in both maternity services revealed unilateral and two types of mutual relationships; which are clinical and therapeutic relationships (see concept 6 in figure 8). In the unilateral relationship, the midwife and woman are heading in opposite directions with one of them unable or not desiring to commit to the relationship. The tensions, contradictions and pressures of constraints to service provision may result in midwives becoming burnt out and not have the emotional energy or interest to invest in a relationship. Moreover, women and their partners may resent the information or the offer of screening. This is evidenced by the midwives' copious documentation of refusal of information about screening. A pregnant woman in the Terrace maternity services reported resenting the offer of screening for Down's syndrome. She felt it was ridiculous to screen for Down's syndrome and then offer TOP, if the diagnostic test is positive for the condition.

In the clinical relationship the midwife is just doing her job which may be equal to a mechanical delivery of information within the available time. The midwife gives the impression of being busy and provides minimal to no verbal information about screening. The relationship formed is superficial. There is a distancing of emotions when screening is offered, but the woman is satisfied with the offer.

In the therapeutic relationship, the information provision and offer of screening is usually of a short duration, but there is deliberation with some emotions involved and the values and beliefs of the woman and other contextual factors taken into consideration. In the context of prenatal Down's syndrome screening, the therapeutic relationship is coveted, as it engages and takes the values and beliefs of women into the discussion. This is supported by Morse (1991) who reported that the therapeutic relationship is desired in the clinical environment.

It is difficult to demonstrate from the data if midwives and women were able to form over-involved relationships because the midwives' recommendation to some women in both maternity services to have screening may have been based on a clinical relationship and to avoid dialogue.

## **7.7 Involvement of people in the prenatal context of Down's syndrome screening on women's decision-making.**

### **7.7.1 Involvement of midwives**

The effect of expert authority, routinisation, tensions and constraints to service provision may be seen in the involvement of midwives in the decision-making process (see concept 8 in figure 8). Thus, four models of decision-making that are not in their purest forms were recognised in the online interviews conducted in two maternity services with a prevailing screening policy of informed choice.

#### **7.7.1.1 The paternalistic decision-making model**

This model usually involved minimum or one way information-giving by the midwife that does not elicit the woman's values and beliefs. The woman is passive in decision-making with her involvement limited to that of consent to the preferences of the midwife (Woods, 2007). The pregnant woman consents to the midwife's choice. Therefore, the preference of the midwife takes precedence over that of the woman. In this regard, the paternalistic model encroaches on the woman's right to be fully informed and have screening decisions

respected. This is disempowering to women. This model is no longer adopted in most healthcare settings, owing to service users becoming well informed (Woods, 2007; Noseworthy et al., 2013). Paternalistic decision-making was demonstrated in both maternity services when midwives directed pregnant women to have the screening test done without providing the opportunity for them to decide on their own.

#### **7.7.1.2 The informed decision-making model**

Informed choice is the model adopted in prenatal Down's syndrome screening programmes where clear and full disclosure of available information about screening options with their various risks and benefits is given. The pregnant woman then makes the final decision from the options, based on her preference (Dheensa et al., 2013a). The model assumes that pregnant women are provided with all the information needed to enable informed decisions. However, both maternity services demonstrated that in a busy clinic, selective to minimal verbal information is often given. Sometimes women are given written information with little to no verbal information about screening and were requested to decide. Nicol (2007) reported that many women are not aware of the potential of the early ultrasound scan to detect anomalies, even when they have received detailed written information about it. As such, women often choose to read what they want, and they interpret written information to fit into their coping strategies.

#### **7.7.1.3 The shared decision making model**

In the shared decision-making model, information is interpreted and discussed between the midwife, woman and her partner when present. The women and partners' decision-making are supported by the midwife to the degree that they want to exercise that choice and the decision is arrived at mutually (Charles et al., 1997; Murray et al., 2006; Noseworthy et al., 2013). The midwives bring expertise and the women their beliefs, values, circumstances and experiences to the decision-making process. The model has as a foundation the principles of choice and negotiation. It recognises the autonomy of the pregnant woman and her right to challenge the authority of the midwife (Charles et al., 1997). It also recognises that the final decision may lie with the pregnant woman (Elwyn et al., 2000; Murray et al., 2006).

Shared decision-making was seen in the data when midwife engage in a dialogue while taking the values and beliefs of the women into consideration. Shared decision-making requires that the discussion take place in an atmosphere conducive for deliberation and

mutual decision-making (Cooke, 2005; Murray et al., 2006; Sullivan, 2006; NICE, 2008; Edwards et al., 2009; Noseworthy et al., 2013).

#### **7.7.1.4 The dual decision-making model as stated policy in Terrace maternity service**

The interviews revealed a stated dual paternalistic and informed choice policy in Terrace maternity service. The first stated policy of the informed choice model is operationalised when women decide at booking to either accept or decline screening based on the information given by midwives. However, a different policy; the paternalistic decision-making model, kicks in for women who are undecided as screening is recommended and consent obtained for arrangement to be made for the test.

Work by Schoonen et al. (2012) and Ahmed et al. (2013) suggests that obtaining consent and arranging for the screening test changes the programme from an opt-in to an opt-out programme. Ahmed et al. (2013) further suggests that the actions may lead to more commitment to the decision compared to when women are allowed to decide without the actions. A midwife in Terrace maternity service succinctly described it as taking choice away from women, as many would continue with screening. This dual decision-making model is a unique finding.

#### **7.7.2 Involvement of partners**

Only midwives could give the right information about screening that guides women and their partners' participation in screening. Importantly, the midwives have a legal duty to accept women's final decisions or consent and not that of the male partners (Draper, 2002; Dheensa et al., 2013a). However, three types of partners' involvement in women's decision-making about Down's syndrome screening were recognised in the interview data (see concept 8).

##### **7.7.2.1 The domineering decision-making model**

In Landscape maternity services, a male partner's insistence on screening took precedence over the autonomous preference of the pregnant woman to decline screening. The interest and preferences of the partner had precedence over the pregnant woman's preference not to undergo screening. Hence, the choice to accept screening was not based on the woman's beliefs and values. This was a situation where the male partner took over decision-making, in effect placing himself in charge of the pregnancy. Nevertheless at bookings, midwives are

advised to look for signs of coercion, domestic abuse and act as women's advocate when their autonomy is threatened. In this instance, the midwife may have missed the opportunity to act as an advocate, probably as a result of the policy of objective and nondirective informed choice.

#### **7.7.2.2 The supportive or joint decision-making model**

This thesis has shown that partners have an important role in supporting pregnant women, especially in decision-making about screening. It is therefore important that male partners are well informed about the purpose of screening. An example may be seen with a partner in Terrace maternity service who explained the information given by the midwife to the female partner to enable joint decision-making. In both maternity services, male partners were taking responsibility for the wellbeing of their pregnant partners by attending the booking visits, explaining information to their partners and sharing in the decision-making. This was not related to ethnicity, level of education nor age.

Existing studies have shown that many women want to share the decision-making about prenatal screening with their partners (García et al., 2008b; van den Berg et al., 2008; Nagle et al., 2009; Åhman et al., 2010; Aune and Möller, 2012; Dheensa et al., 2013a). This may be over the psychological burden associated with decision-making and the decisions made can have a sustained effect on the women, partners, children and other family members. In addition, male partner's involvement in screening may be supportive, since both partners receive and process the information about screening together (Dheensa et al., 2013b).

#### **7.7.2.3 The detached decision-making model**

Some partners were not involved in decision-making about screening, as they left it entirely up to the pregnant women. This is consistent with findings from other studies which reported that partners believed screening involved the woman's body and therefore, the decision has to be made by the woman (Markens et al., 2003; Reed, 2009a; Skirton and Barr, 2010; Williams et al., 2011; Dheensa et al., 2013a).

### **7.8 Proposed social model of shared decision-making process**

The findings of this thesis suggest the necessity for a social model that can reduce the constraints to service provision, tensions, variation in uptake rates, different types of relationships and decision-making models in the prenatal context. The shared decision-

making (SDM) model by Elwyn et al. (2012) has been adapted below to create a shared decision-making process model (SDMP) for prenatal Down's syndrome screening (see figure 9). In SDMP, the midwives work with women and partners and encourage them to come to a mutual decision. The midwives, women and partners' autonomy is maintained, if the focus is on the nature of the decision-making process rather than on the decision itself. Therefore, the proposed SDMP model focuses primarily on the decision-making process as opposed to SDM that focuses on both the process and the decision itself.

Lawson and Pierson (2007) reported in their study that explored maternal decision-making concerning prenatal diagnosis, that women who were supported by their physicians and partners felt autonomous in decision-making. Women who reported the least support and autonomy were those, whose partners and physicians left the decision-making entirely to them. The unsupported women felt abandoned. Interestingly, women who declined testing experienced less support from physicians and partners and the lowest levels of decisional wellbeing. It is therefore important to engage and support all women in the decision-making processes, as women who choose to decline the offer of screening, based on nondirective informed choice may be a vulnerable group, such as women from ethnic minority backgrounds as demonstrated in Terrace maternity services.

A Cochrane review by Légaré et al. (2010) reported that there is evidence that shared decision making has the potential to reduce the use of less effective treatment options and increase the use of effective options. Besides, existing work suggests SDM may lead to reduced variation in practice, enhance patients' autonomy and increase the sustainability of health care systems (Bond et al., 2012).

## **7.9 Advantages of adopting shared decision-making process (SDMP) in the prenatal context of Down's syndrome screening**

- 1) A shared decision-making process model would enable midwives dispel tensions, by presenting an environment where options can be discussed together. This environment would also give midwives the opportunity to observe women and partners together and look for signs of coercion or domestic abuse (NICE, 2008; Dheensa et al., 2013a).
- 2) Time constraints may also be addressed through the shared decision-making process model as it enables midwives to determine the information required for discussion and

assist with the problem of information overload, thereby facilitating understanding. Moreover, women and their partners may be able to arrive at a decision quickly when engaged in a dialogue that involves their values and beliefs in the decision-making process as noted in Polansky (2006).

- 3) Research has shown that involving women and partners in shared decision-making may improve overall psychological and wellbeing outcomes such as reduced anxiety (Brody and Smith, 1989; Kaplan et al., 1989 cited in King and Moulton, 2006).
- 4) The UK NSC (FASP) has developed an online decision aid to support service users through screening decisions (UK NSC, 2012). The decision aid provides information at different levels and helps users to consider their values, emotions and facts. It therefore helps individuals to avoid falling into cognitive traps as noted in Schoonen et al. (2012). Further, if the midwife directs women to the decision aid as part of a shared decision-making process, the incidence of litigation may be reduced. The reason is that it may be difficult for service users to complain that the information, decision aid, and discussions were inadequate to negate consent.
- 5) Further, Huntington and Kuhn (2003) found from their review of studies that explored why patients institute litigation against physicians, that the compelling reason was a breakdown in the patient-physician relationship. This theme was often expressed as unsatisfactory patient-physician communication, which often preceded the decision to pursue litigation. The reduced medical litigations associated with the shared decision-making model has also been reported in previous research in the UK and USA (Chacko, 2009; Burkhard et al., 2011).
- 6) The shared decision-making process model may not require the pregnant woman to make the final decision. It is mandatory for the midwife to discuss the relevant information and to exchange opinions based on the woman's values and beliefs. If at the end of that process, the woman has thought through the options and prefers to allow the midwife make the decision, that option is a perfect reflection of her informed and autonomous choice. The woman may then be required to sign an informed consent form to acknowledge that choice.

## **7.10 Strengths of the study**

- 1) The strength of this thesis lies in the fact that the perspectives of frontline service providers (midwives) and service users (women and their partners) were sought to gain a

comprehensive view of the study's phenomenon. In addition, the interviews and document review were conducted to gain a better understanding of the influence of service organisation and delivery on women and partners' participation in screening.

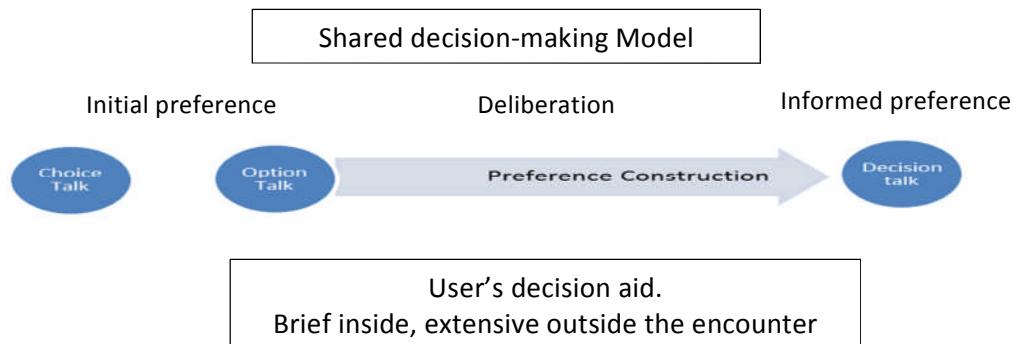
- 2) To ensure consistency between interviews, the same online interview formats were used throughout to ensure the variations seen were due to participants' responses. In order to access a wide range of views on the influence of service organisation and delivery on screening, women and partners who accepted or declined Down's syndrome screening were included in this study.
- 3) The diversity in the study samples of women and partners allowed for the exploration and identification of important issues that were common across participants. There was no limit on parity and participants had a range of ages, educational qualifications and experiences. The midwives in this study were predominantly white which is not surprising, but the ethnicity of pregnant women and partners was diverse. The diversity enhanced the credibility and transferability of the findings.

### **7.11 Limitations of the study**

- 1) Conducting the study with pregnant women at 20 weeks gestation adequately explored the screening process and may have reduced the problem of time recall and post choice biases.
- 2) The samples of midwives, pregnant women and partners may be representative of a middle class computer literate population with access to the internet. Nevertheless, effort was made to introduce those without access to the internet to public libraries. These characteristics may limit the transferability of the findings.
- 3) Selection bias may have been introduced into this study, as a purposive sampling method was employed. Those who participated in the study might have different views from those who did not. Inviting the community midwives through the Local Collaborators may have introduced a recruitment bias and a barrier to obtaining uninhibited responses. Nonetheless, there were variations in the responses from participants which may indicate the online methods minimised the bias and barriers.
- 4) As partners do not always attend routine prenatal appointments, they were recruited through the pregnant women. Recruiting partners through their pregnant partners is

**Figure 9: Elwyn's shared decision-making model**

(Elwyn et al., 2012)



**Key to figure 9 (adapted for prenatal Down's syndrome screening)**

Deliberation	A process where women and partners become aware of choice, understand their options and have the time and support to consider 'what matters most to them': It requires more than one visit and include the use of decision aid and women may discussions with others.
Choice talk	Conveys awareness that a choice exists – initiated by either a woman or a midwife. This may occur before the booking encounter (see section 7.18 (2) page 187).
Option talk	Women and partners are informed about screening in more details.
Decision talk	Women are supported to explore 'what matters most to them', having become informed.
Decision aid	Decision aid as designed in two formats: 1) brief enough to be used by midwives and women together and 2) more extensive, designed to be used by women and their partners either before or after the first booking visit (UK NSC decision aid).
Initial Preferences	Awareness of options leads to the development of initial preferences, based on existing knowledge. The goal is to arrive at informed preferences.
Informed preferences	Personal preferences based on 'what matters most to the woman, predicated on an understanding of the most relevant benefits and harms.'

perhaps not ideal and could result in a self-selecting sample. Hopefully, by adopting two different sampling strategies regarding male partner's recruitment, the bias was minimised.

5) The challenge with using scenarios is that participants may respond to the questions, based on what they know to be the correct answers and not actually how they behaved in the prenatal context.

6) Women and partners were interviewed individually using separate web pages. However, it is possible that some pregnant women and their partners may have responded to the online interviews together and might have produced different types of data. However, existing research suggests that women and their partners interviewed together may enhance the responses due to the rich dynamics of their interactions (Markens et al., 2003; Reed, 2009b). The findings in both maternity services did not indicate marked differences in the responses within each individual group.

7) This is a qualitative study based on online interviews with 34 midwives, 35 pregnant women and 15 partners in two geographical settings in the North of England. Therefore, this study can only offer transferability of findings by nature of the study design. However, the purpose of this qualitative case study was not to generalize findings but of theoretical generalization (Kohlbacher, 2006; Yin, 2009). Findings may be transferable to similar settings.

However, the low response rates reported in this thesis may be due to the researcher not having direct access to the midwives. Further, many of the participant information sheets given out to women and their partners were obtained after the brief introduction of the research by the sonographers without the researcher's input. The participants claimed to be either in a hurry or have another appointment.

To boost the response rates in future studies, researchers could explore having direct access to all participants where possible. More NHS Trusts from the low and high uptake range could be selected and included in the study. Internet enable laptops or tablets could also be made available to participants who agreed to take part in the research and are willing to complete the interviews at the point of recruitment. Another approach is to provide rewards for each participant group. Participants would then be informed that they will be entered in a draw for a general prize such as a gift voucher for an iPad or tablet.

## 7.12 Implications for policy and practice

- 1) The influence of service organisation and delivery needs to be acknowledged, if the choices of individual women are to be supported. This may enable healthcare professionals to understand the constraints to service provision and help women and partners consider the various constraints identified. This may improve communication at booking and reduce the negative effects caused by screening information, thus improving care women receive prenatally.
- 2) This thesis highlights the need for prior information preferably pre-pregnancy information about screening to all women and their partners planning on starting a family. When the information is provided again at booking, it may aid comprehension and active engagement in the decision-making processes. Information given in schools, healthcare settings and wider social networks has been advocated by Lewando-Hundt (2006). In addition, dedicated websites for teenagers about sexual health and for women and their partners desirous of starting a family may be a source of information about Down's syndrome screening prior to pregnancy.
- 3) The booking appointment should be ideally divided into two separate visits. This has also been suggested in the NICE guidelines (NICE, 2008). Adopting two separate visits may reduce the overwhelming feeling of information overload and time pressure. Information about screening may be introduced in the first booking visit. Women and their partners are then given written information and directed to the online decision aid. A shorter decision aid could then be used at the second visit to facilitate the shared decision-making process.
- 4) Shorter written information about Down's syndrome screening with illustrations may lead to better understanding than the current screening booklet. All written documents should be in simple English that could be read by an individual with a reading age that is equivalent to the average reading age of the population covered by the programme. Freer et al. (2009) reported that a shorter information leaflet is effective at improving understanding of consent.
- 5) The concept of risk in Down's syndrome screening information also presents extra challenges to pregnant women and partners. If midwives, women and their partners are to have informed discussions, midwives are likely to need more resources such as more training and time for informing women.

- 6) Midwives need to avoid the gendered, cultural, religious or morally mediated stereotype of women, as it relates to screening decisions.
- 7) The consent form for screening is too complicated and does not take into consideration the needs of women for whom English is not their first language. It should be translated into other languages to enable women and partners to understand the consenting process.

### **7.13 Implication for education**

- 1) Training to enhance midwives' communication skills using simple plain English to inform women and on how to resolve conflicts would be needed. Such skills are necessary in order to recognise and engage partners and, at the same time, ensure women's autonomy is promoted and protected. Further, low health literacy has been noted as a challenge to the SDMP model (Burkhard et al., 2011). To overcome low literacy, decision aids and the dialogue or discussion should be tailored to meet various reading and numeracy ages. The obligation is on the midwives to be aware of women's capabilities and on the women to be clear about what they understand, to enable a genuine decision-making process partnership.
- 2) Midwives require training on methods to check women's understanding to ensure that information about screening is interpreted correctly by pregnant women and their partners. This method is currently not taught probably owing to the policy of nondirective informed choice. The 'teach-back' method provides a way through which midwives can check that they have clearly communicated all the information that women and partners need and how well they understand (Bond et al., 2012). The role of written information is to provide subsequent support after the consultation. Thus, written information, despite the disadvantage that it is not tailored to an individual woman's need, can be taken away and referred to later as can referral to web-based information and a decision aid.
- 3) Midwives require training to recognise psychological implications and stress in women and their partners associated with screening information. They also need skills to support women and partners to cope with the emotional and psychological impact of participating in screening.
- 4) Interpreters should also be given mandatory training on Down's syndrome screening in their respective languages and in English. The present e-learning module for midwives could be modified and translated for interpreters.

5) Training in shared decision-making processes should be given to all midwives before its adoption as a policy. It should also be included in the annual mandatory training courses for community midwives.

#### **7.14 Recommendations for future research**

- 1) Further research is needed to verify whether the developed conceptual model reflects the influence of service organisation and delivery on women and partners' participation in screening. Focus groups and face to face interviews may be valuable qualitative methods to explore in-depth and gain a better understanding of the different concepts within the model. Based on the findings of the in-depth qualitative study the conceptual model could then be assessed quantitatively to find out its generalisability to the population.
- 2) There is a need for more research on the impact of the use of pictures of children with Down's syndrome on women's and their partners' decisions to participate in screening. The research should focus on the psychological impact and how in combination with the ambience of the environment, may influence decision-making processes.
- 3) The finding of the potential influence of interpreters on women's participation in screening needs to be fully investigated. Despite the importance of interpreters in the screening programmes little is known about their impact on women's screening decision-making in the UK context.
- 4) The findings of this thesis may be used to design and evaluate an intervention to promote the practice of the shared decision-making process model. This may include defining the characteristics and skills required by midwives, women and their partners. The findings will help to inform the training of health professionals involved in the provision of prenatal Down's syndrome screening.

#### **7.15 Dissemination strategy**

The methodological issues encountered in the pilot study were presented at the International Conference of Mixed Methods Research in 2012, University of Leeds. The title of the paper presented orally was 'Methodological dilemmas in web-based mixed methods research' (see appendix 11). The use of vignettes in health services research based on this thesis was also presented in the Postgraduate School of Healthcare, University of Leeds

conference in 2012. The title of the oral presentation was 'Using vignettes in an online qualitative study to explore participation in prenatal Down's syndrome screening' (see appendix 12). Finally, the findings from this study will be published in peer-review international journals that target policymakers and healthcare professionals involved in prenatal Down's syndrome screening and researchers involved in health services research. The objective is to contribute to the debate about the influence of service organisation and delivery on participation in healthcare programmes and the use of online methods in research.

**Table 8: Proposed publication plan**

	Proposed Titles
1	Methodological dilemmas in a web-based mixed methods research.
2	Online interviews using vignettes and prompts: An innovative approach in health services research.
3	'Power' and 'Tensions' in Down's syndrome screening: Developing a conceptual model for the influence of service organisation and delivery.
4	Variation in uptake rates of Down's syndrome screening in the North of England: A multiple case study.
5	'Collusion' in the context of Down's syndrome screening: The 'Gold package' effect.
6	Relationships and decision-making models in prenatal Down's syndrome screening programmes.

## **7.16 Conclusions**

This thesis illuminates the influence of service organisation and delivery on uptake of screening for Down's syndrome through the development of a conceptual model. The model provided new insights into the concepts involved in women and partners' decision-making process. The unique findings within these concepts were midwives offering screening as a routine test, the influence of the ambience of the environment and the influence of interpreters. Others were the dual decision-making policies, paternalistic attitudes of partners and the inability of midwives to detect coercion in the prenatal context. These influences on women and partners' decision-making process were finally expressed through women's compliance, or lack thereof, with prenatal screening. These unique findings demonstrate that the classic situation of women capable of autonomous and

informed choice and midwives capable of informing nondirectively may not exist in either maternity service.

As such, the Down's syndrome screening programmes based on the mechanistic individualistic biomedical model of autonomous and informed choice is proving surprisingly difficult to operationalise in practice. This may be as a result of the overwhelming constraints to service provision being overlooked in the current objective informed choice policy. These constraints reinforced the power/ knowledge differential between midwives and women. Therefore the informed choice policy that values emotional detachment, promotes access to screening over the need for dialogue based on the women's beliefs, values and circumstances.

Midwives in both maternity services responded to the constraints to service provision by blurring the line between nondirective informing and directiveness. The differing knowledge bases adopted by midwives, women and partners and the constraints to service provision created tensions within and between midwives, women and partners and in the prenatal environment. These constraints and tensions affected the interaction between midwives, women and partners and how screening was described to them. The interference may be responsible for the different decision-making models and relationships adopted by midwives, women and partners in the prenatal contexts.

Variation in uptake rates cannot be explained by the differences in psychosocial characteristics of women as shown in previous work. This thesis revealed that in Landscape maternity service there is a perceived organisational culture that expects women to agree to screening. However there is a mix picture in Terrace maternity service. The stereotypical beliefs among midwives that ethnic minority groups decline screening for cultural reasons, suggest a workplace culture of expectation. In addition, this thesis illuminates the nuances in the operationalisation of the national screening policies in maternity services in England which fills a gap in the literature and adds new knowledge.

The screening programmes need to be transformed, so women and their partners have the understanding needed for informed consent. Promoting informed decision-making requires a combination of information and support. Midwives play a crucial role in women and partners' participation in screening by providing the necessary support, deliberation and advice. These social interactions enable women and their partners' to make informed decisions.

As we enter into a new era of NIPD with a shorter window of opportunity, midwives need to be supported and equipped to deal with these constraints to service provision. Thus, it is essential that midwives be aware of and are positioned to meet these challenges with first trimester screening and prepare women to make informed decisions. The social model of the shared decision-making process will enable midwives to provide the extra care needed to support women and their partners' to make participation decisions in prenatal Down's syndrome screening.

The study adds new knowledge to the evidence base, providing health professionals and policy makers with new information about the influence of service organisation and delivery on women and partners' decision-making process and variation in uptake rates of screening. However, this thesis suggests that whilst women's decision-making processes are influenced by service organisation and delivery, it may not be possible to actually know all the potential influences on women and partners' participation in screening.

*"The social distance built into current ways of looking at the human body - the view of an objective scientist looking at another bodily object that is clearly separate and distinct - will be expanded to include a new type of social connectedness, where two human beings will be able to share their commonly felt experiences at their social membrane. In the new clinic, immunization from the emotional experiences of one's fellow man will no longer be seen as ...a vital necessity"* (Lynch, 1985 cited in Sharpe, 1996, p. 440).

## List of References

- Adler, C.L. and Zarchin, Y.R. 2002. The "Virtual Focus Group": Using the Internet to Reach Pregnant Women on Home Bed Rest. *Journal of Obstetric, Gynecologic, & Neonatal Nursing*. **31**(4), pp.418-427.
- Ahern, D.K. 2007. Challenges and Opportunities of eHealth Research. *American Journal of Preventive Medicine American Journal of Preventive Medicine*. **32**(5), pp.S75-S82.
- Åhman, A. et al. 2010. Did I really want to know this? Pregnant women's reaction to detection of a soft marker during ultrasound screening. *Patient Education and Counseling*. **81**(1), pp.87-93.
- Ahmed, S. et al. 2012a. Attitudes towards prenatal testing and termination of pregnancy in British Pakistani parents and relatives of children with recessive conditions in the UK. *Prenatal Diagnosis*. **32**(10), pp.954-959.
- Ahmed, S. et al. 2006a. The influence of faith and religion and the role of religious and community leaders in prenatal decisions for sickle cell disorders and thalassaemia major. *Prenatal Diagnosis*. **26**(9), pp.801-809.
- Ahmed, S. et al. 2013. Midwives' perceptions of their role as facilitators of informed choice in antenatal screening. *Midwifery*. **29**(7), pp.745-750.
- Ahmed, S. et al. 2012b. Interpretations of informed choice in antenatal screening: A cross-cultural, Q-methodology study. *Social Science & Medicine*. **74**(7), pp.997-1004.
- Ahmed, S. et al. 2012c. Is advice incompatible with autonomous informed choice? Women's perceptions of advice in the context of antenatal screening: a qualitative study. *Health Expectations*. pp.no-no.
- Ahmed, S. et al. 2006b. Attitudes towards prenatal diagnosis and termination of pregnancy for thalassaemia in pregnant Pakistani women in the North of England. *Prenatal Diagnosis*. **26**(3), pp.248-257.
- Ahmed, S. et al. 2008. Decisions About Testing and Termination of Pregnancy for Different Fetal Conditions: A Qualitative Study of European White and Pakistani Mothers of Affected Children. *Journal of Genetic Counseling*. **17**(6), pp.560-572.
- Ajzen, I. 1991. The theory of planned behavior. *Organizational behavior and human decision processes*. **50**(2), pp.179-211.
- Ajzen, I. 2001. Nature and operation of attitudes. *Annual Review of Psychology*. **52**(1), pp.27-58.

- Alderdice, F. et al. 2008. Inequalities in the reported offer and uptake of antenatal screening. *Public Health*. **122**(1), pp.42-52.
- Alsulaiman, A. et al. 2012. Attitudes to prenatal diagnosis and termination of pregnancy for 30 conditions among women in Saudi Arabia and the UK. *Prenatal Diagnosis*. **32**(11), pp.1109-1113.
- Anderson, G. 1999. Nondirectiveness in prenatal genetics: patients read between the lines. *Nursing Ethics*. **6**(2), pp.126-136.
- Anspach, R.R. 1993. *Deciding who lives: Fateful choices in the intensive-care nursery*. Univ of California Press.
- Armitage, C.J. and Conner, M. 2000. Social cognition models and health behaviour: A structured review. *Psychology and Health*. **15**(2), pp.173-189.
- Asongu, M. et al. 2010. Utilization of invasive testing following a positive triple test 2001–2009 in Hull & East Yorkshire area of the UK. *Prenatal Diagnosis*. **30**(11), pp.1107-1109.
- Aston, M. et al. 2010. Not Just Another Delivery: Women's Maternity Care Experiences of Provider Client Relationships.
- Atkin, K. et al. 2008. Decision-Making and Ante-Natal Screening for Sickle Cell and Thalassaemia Disorders: To What Extent do Faith and Religious Identity Mediate Choice? *Current Sociology*. **56**(1), pp.77-98.
- Aune, I. and Möller, A. 2012. 'I want a choice, but I don't want to decide'—A qualitative study of pregnant women's experiences regarding early ultrasound risk assessment for chromosomal anomalies. *Midwifery*. **28**(1), pp.14-23.
- Babbie, E.R. 1998. *The practice of social research*. Wadsworth Pub. Co.(Belmont, CA).
- Baillie, C. and Hewison, J. 1999. Antenatal screening: Obtaining selective consent to scanning, rather than screening, is possible. *BMJ: British Medical Journal*. **318**(7186), p805.
- Bakker, M. et al. 2012. Low uptake of the combined test in the Netherlands – which factors contribute? *Prenatal Diagnosis*. **32**(13), pp.1305-1312.
- Bandura, A. 1986. *Social foundations of thought and action : a social cognitive theory*. Englewood Cliffs, N.J.: Prentice-Hall.
- Barter and Renold, E. 1999. The Use of Vignettes in Qualitative Research. *Social Research Update*. [Online]. [Accessed 02.08.12].
- Baum, F. 2008. *The New Public Health*. Melbourne, Australia: Oxford University Press.
- Beardshaw, T. 2001. Supporting the role of fathers around the time of birth. *MIDIRS Midwifery Digest*. **11**, pp.474-477.

- Beauchamp, T.L. and Childress, J.F. 2001. *Principles of biomedical ethics*. Oxford: Oxford University Press.
- Bekker, H. et al. 1993. Uptake Of Cystic Fibrosis Testing In Primary Care: Supply Push Or Demand Pull? *BMJ: British Medical Journal*. **306**(6892), pp.1584-1586.
- Bekker, H. et al. 1999. Informed decision making: an annotated bibliography and systematic review. *Health Technol Assess*. **3**(1), pp.1-156.
- Bekker, H.L. 2010. The loss of reason in patient decision aid research: Do checklists damage the quality of informed choice interventions? *Patient Education and Counseling*. **78**(3), pp.357-364.
- Bender, D.E. et al. 2001. Tell Me What you Mean by "Si": Perceptions of Quality of Prenatal Care among Immigrant Latina Women. *Qual Health Res*. **11**(6), pp.780-794.
- Bennett, R.L. et al. 2003. Genetic counselors: translating genomic science into clinical practice. *Journal of Clinical Investigation*. **112**(9), pp.1274-1279.
- Berg, M. et al. 1996. Women's experience of the encounter with the midwife during childbirth. *Midwifery*. **12**(1), pp.11-15.
- Berney, L. et al. 2005. Ethical principles and the rationing of health care: a qualitative study in general practice. *British Journal of General Practice*. **55**(517), pp.620-625.
- Bishop, A.J. et al. 2004. Women and health care professionals' preferences for Down's Syndrome screening tests: a conjoint analysis study. *BJOG: An International Journal of Obstetrics & Gynaecology*. **111**(8), pp.775-779.
- Blaaka, G. and Schauer Eri, T. 2008. Doing midwifery between different belief systems. *Midwifery*. **24**(3), pp.344-352.
- Bloor, M. et al. 2001. *Focus groups in social research*. London; Thousand Oaks, Calif.: SAGE Publications.
- BMA. 2009. *British Medical Association: Consent tool kit*. [Online]. Available from: <http://www.southessex.nhs.uk/content/file/GP%20Zone/Safeguarding/National%20docs/consenttoolkit.pdf>.
- Bond, C. et al. 2012. Prescribing and partnership with patients. *British Journal of Clinical Pharmacology*. **74**(4), pp.581-588.
- Bowen, G.A. 2009. Document analysis as a qualitative research method. *Qualitative research journal*. **9**(2), pp.27-40.
- Bowler, I. 1993. 'They're not the same as us': midwives' stereotypes of South Asian descent maternity patients. *Sociology of Health & Illness*. **15**(2), pp.157-178.

- Boyd, P.A. et al. 2008. Survey of prenatal screening policies in Europe for structural malformations and chromosome anomalies, and their impact on detection and termination rates for neural tube defects and Down's syndrome. *BJOG: An International Journal of Obstetrics & Gynaecology*. **115**(6), pp.689-696.
- Bradley Eh, H.E.S.M.J.A.R.S.A.R.M.J.K.H.M. 2001. A qualitative study of increasing β-blocker use after myocardial infarction: Why do some hospitals succeed? *JAMA: The Journal of the American Medical Association*. **285**(20), pp.2604-2611.
- Bronte-Tinkew, J. et al. 2007. Resident Fathers' Pregnancy Intentions, Prenatal Behaviors, and Links to Involvement With Infants. *Journal of Marriage and Family*. **69**(4), pp.977-990.
- Broom, A. 2005. Virtually He@lthy: The Impact of Internet Use on Disease Experience and the Doctor-Patient Relationship. *Qualitative Health Research*. **15**(3), pp.325-345.
- Browne, J. et al. 'You've got it within you': The political act of keeping a wellness focus in the antenatal time. *Midwifery*. (0).
- Browner, C.H. et al. 1999. Ethnicity, bioethics, and prenatal diagnosis: the amniocentesis decisions of Mexican-origin women and their partners. *Am J Public Health*. **89**(11), pp.1658-1666.
- Browner, C.H., Preloran,H.M. 1999. Male partners' role in Latinas' amniocentesis decisions. . *Journal of Genetic Counselling* **8**, pp.85–108.
- Bryant, L. et al. 2005. Attitudes towards prenatal diagnosis and termination in women who have a sibling with Down's syndrome. *Journal of Reproductive and Infant Psychology*. **23**(2), pp.181-198.
- Bryant, L.D. 2003. *Understandings of Down's syndrome and their place in the prenatal testing context*. thesis, University of Leeds.
- Bryant, L.D. et al. 2011. 'All is done by Allah'. Understandings of Down syndrome and prenatal testing in Pakistan. *Social Science & Medicine*. **72**(8), pp.1393-1399.
- Bryant, L.D. et al. 2006. Understandings of Down's syndrome: A Q methodological investigation. *Social Science & Medicine*. **63**(5), pp.1188-1200.
- Bryant, L.D. et al. 2010. The role of attitudes towards the targets of behaviour in predicting and informing prenatal testing choices. *Psychology and Health*. **25**, pp.1175-1194.
- Bryman, A. and Burgess, B. 2002. *Analyzing qualitative data*. Routledge.
- Burkell, J. and McKenzie, P.J. 2005. Information Provision for Informed Prenatal Decision Making In: Vaughan, L. ed. *Data, information, and knowledge in a networked world* Canadian Association for Information Science 2005 Annual Conference.

- Burkhard, C. et al. 2011. *Shared Decision Making in Health Care Delivery*. Hanover, NH: Nelson A. Rockefeller Center, Dartmouth College
- Burls, A. et al. 2010. Oxygen use in acute myocardial infarction: An online survey of health professionals' practice and beliefs. *Emerg. Med. J. Emergency Medicine Journal*. **27**(4), pp.283-286.
- Cabrera, N.J. et al. 2008. Explaining the Long Reach of Fathers' Prenatal Involvement on Later Paternal Engagement. *Journal of Marriage and Family*. **70**(5), pp.1094-1107.
- Cantrell, M.A. and Lupinacci, P. 2007. Methodological issues in online data collection. *Journal of Advanced Nursing*. **60**(5), pp.544-549.
- Carlsen, B. and Glenton, C. 2011. What about N? A methodological study of sample-size reporting in focus group studies. *BMC medical research methodology*. **11**, p26.
- Carroll, J.C. et al. 2000. Women's experience of maternal serum screening. *Canadian Family Physician*. **46**(3), pp.614-20.
- Chacko, D. 2009. *Medical Liability Litigation: An Historical Look at the Causes for Its Growth in the United Kingdom*.
- Charise, A. et al. 2011. Questioning context: a set of interdisciplinary questions for investigating contextual factors affecting health decision making. *Health Expectations*. **14**(2), pp.115-132.
- Charles, C. et al. 1997. Shared decision-making in the medical encounter: What does it mean? (or it takes at least two to tango). *Social Science & Medicine*. **44**(5), pp.681-692.
- Chervenak, F.A. and McCullough, L.B. 2010. Ethical considerations in first-trimester Down syndrome risk assessment. *Current Opinion in Obstetrics and Gynecology*. **22**(2), pp.135-138 10.1097/GCO.0b013e3283374a9f.
- Chiang, H.H. et al. 2006. Informed choice of pregnant women in prenatal screening tests for Down's syndrome. *J Med Ethics*. **32**(5), pp.273-7.
- Choi, H. et al. 2012. Decision Making Following a Prenatal Diagnosis of Down Syndrome: An Integrative Review. *Journal of Midwifery & Women's Health*. **57**(2), pp.156-164.
- Clark, J.A. et al. 1991. Bringing social structure back into clinical decision making. *Social Science & Medicine*. **32**(8), pp.853-866.
- Clarke, A. 1997. The process of genetic counselling: beyond non-directiveness. In: Harper and Clarke AJ ed. *Genetics society and clinical practice*. Oxford: BIOS Scientific Publishers, pp.179-200.

- Collins, S. et al. 2005. 'Unilateral' and 'bilateral' practitioner approaches in decision-making about treatment. *Social Science & Medicine*. **61**(12), pp.2611-2627.
- Conner and Sparks. 1995. The theory of planned behaviour and health behaviours. In: Conner and Norman ed. *Predicting health behaviour*. Open University Press Buckingham, pp.121-162.
- Cooke, P. 2005. Helping women to make their own decisions. *Decision Making in Midwifery Practice*. pp.127-142.
- Corbin, J. and Strauss, A. 2008. *Basics of qualitative research: Techniques and procedures for developing grounded theory*. 3rd ed. Thousand oaks, CA: Sage Publications, Incorporated.
- Coulter, A. and Ellins, J. 2007. Effectiveness of strategies for informing, educating, and involving patients. *BMJ*. **335**(7609), pp.24-27.
- Council of Europe, C.o.M. 1990. Prenatal genetic screening, prenatal genetic diagnosis and associated genetic counselling. [Online]. [Accessed 31.01.11]. Available from: <http://www1.umn.edu/humanrts/instree/coerecr90-13.html>.
- Creswell, J.W. 2007. *Qualitative Inquiry And Research Design: Choosing Among Five Approaches* Author: John W. Creswell, Publisher: Sage Publica. 2nd ed. Sage Publications, Inc.
- Creswell, J.W. 2009. *Research design: Qualitative, quantitative, and mixed methods approaches* (3rd ed.). Thousand Oaks, CA: Sage Publications, Inc; US.
- Creswell, J.W. and Clark, V.L.P. 2007. *Designing and conducting mixed methods research*. Thousand Oaks, CA: Sage Publications, Inc; US.
- Crombag, N.M. et al. 2013. Determinants affecting pregnant women's utilization of prenatal screening for Down syndrome: a review of the literature. *The Journal of Maternal-Fetal & Neonatal Medicine*. (0), pp.1-50.
- Crotty, M. 1998. *The foundations of social research: Meaning and perspective in the research process*. Thousand Oaks, CA: Sage Publications.
- Cupchik, G. 2001. Constructivist realism: an ontology that encompasses positivist and constructivist approaches to the social sciences. In: *Forum Qualitative Sozialforschung/Forum: Qualitative Social Research*.
- Curry, L.A. et al. 2009. Qualitative and Mixed Methods Provide Unique Contributions to Outcomes Research. *Circulation*. **119**(10), pp.1442-1452.
- Daly, J. et al. 2007. A hierarchy of evidence for assessing qualitative health research. *Journal of clinical epidemiology*. **60**(1), pp.43-49.

- Davis, D. and Walker, K. 2010. The corporeal, the social and space/place: exploring intersections from a midwifery perspective in New Zealand. *Gender, Place & Culture*. **17**(3), pp.377-391.
- Denk, C.E. et al. 1997. How Do Americans Want to Die? A Factorial Vignette Survey of Public Attitudes about End-of-Life Medical Decision-Making. *Social Science Research*. **26**(1), pp.95-120.
- Denzin, N.K. 2009. *The research act: A theoretical introduction to sociological methods*. Transaction Publishers.
- Denzin, N.K. and Lincoln, Y.S. 2003. *Collecting and Interpreting Qualitative Materials*. 2nd ed. Sage Publications, Incorporated.
- Denzin, N.K. and Lincoln, Y.S. 2005. *The SAGE handbook of qualitative research*. Thousand Oaks: Sage Publications.
- Department of Health. 2007a. *Maternity Matters: Choice, Access and Continuity of Care in a Safe Service*. [Online]. [Accessed 22.03.13]. Available from: [http://webarchive.nationalarchives.gov.uk/20130107105354/http://www.dh.gov.uk/prod\\_consum\\_dh/groups/dh\\_digitalassets/@dh/@en/documents/digitalasset/dh\\_074199.pdf](http://webarchive.nationalarchives.gov.uk/20130107105354/http://www.dh.gov.uk/prod_consum_dh/groups/dh_digitalassets/@dh/@en/documents/digitalasset/dh_074199.pdf).
- Department of Health. 2007b. *Maternity matters: choice, access and continuity of care in a safe service*. [Online]. [Accessed 17.11]. Available from: [http://www.dh.gov.uk/prod\\_consum\\_dh/groups/dh\\_digitalassets/@dh/@en/documents/digitalasset/dh\\_074199.pdf](http://www.dh.gov.uk/prod_consum_dh/groups/dh_digitalassets/@dh/@en/documents/digitalasset/dh_074199.pdf).
- Department of Health. 2009. *Reference guide to consent for examination or treatment*. [Online]. [Accessed 08.05.13]. Available from: [https://www.gov.uk/government/uploads/system/uploads/attachment\\_data/file/138296/dh\\_103653\\_1\\_.pdf](https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/138296/dh_103653_1_.pdf).
- Department of Health. 2010. *Health Profile 2010. Yorkshire and the Humber*. [Online]. [Accessed 20.01.13]. Available from: [www.healthprofiles.info](http://www.healthprofiles.info).
- Dheensa, S. et al. 2013a. Men's experiences of antenatal screening: A metasynthesis of the qualitative research. *International Journal of Nursing Studies*. **50**(1), pp.121-133.
- Dheensa, S. et al. 2013b. Shattered Schemata and Fragmented Identities: Men's Experiences of Antenatal Genetic Screening in Great Britain. *Journal of Family Issues*.
- Dixon-Woods, M. et al. 2005. Synthesising qualitative and quantitative evidence: a review of possible methods. *Journal of Health Services Research & Policy*. **10**(1), pp.45-53B.

- Dixon-Woods, M. et al. 2007. Synthesizing qualitative research: a review of published reports. *Qualitative Research*. **7**(3), pp.375-422.
- Dixon-Woods, M. et al. 2006. Conducting a critical interpretive synthesis of the literature on access to healthcare by vulnerable groups. *BMC medical research methodology*. **6**(1), p35.
- Dixon-Woods, M. et al. 2004. The problem of appraising qualitative research. *Quality and Safety in Health Care*. **13**(3), pp.223-225.
- Donabedian, A. 1980. *Explorations in quality assessment and monitoring: The definition of quality and approaches to its assessment*. Ann Arbor, MI: Health Administration Press.
- Donabedian, A. 1988. The quality of care: How can it be assessed? *JAMA* **260**, pp.1743–1748.
- Donabedian, A. 2005. Evaluating the Quality of Medical Care. *Milbank Quarterly*. **83**(4), pp.691-729.
- Dormandy, E. et al. 2002a. Informed choice to undergo prenatal screening: a comparison of two hospitals conducting testing either as part of a routine visit or requiring a separate visit. *Journal of Medical Screening*. **9**(3), pp.109-14.
- Dormandy, E. and Marteau, T.M. 2004. Uptake of a prenatal screening test: the role of healthcare professionals' attitudes towards the test. *Prenatal Diagnosis*. **24**(11), pp.864-868.
- Dormandy, E. et al. 2005. Low uptake of prenatal screening for Down syndrome in minority ethnic groups and socially deprived groups: a reflection of women's attitudes or a failure to facilitate informed choices? *International Journal of Epidemiology*. **34**(2), pp.346-352.
- Dormandy, E. et al. 2006. Informed choice in antenatal Down syndrome screening: A cluster-randomised trial of combined versus separate visit testing. *Patient Education and Counseling*. **61**(1), pp.56-64.
- Dormandy, E. et al. 2002b. Variation in uptake of serum screening: the role of service delivery. *Prenatal Diagnosis*. **22**(1), pp.67-69.
- Draper, H. and Ives, J. 2013. Men's involvement in antenatal care and labour: Rethinking a medical model. *Midwifery*. **29**(7), pp.723-729.
- Draper, J. 2002. 'It was a real good show': the ultrasound scan, fathers and the power of visual knowledge. *Sociology of Health & Illness*. **24**(6), pp.771-795.
- Dudgeon, M.R. and Inhorn, M.C. 2004. Men's influences on women's reproductive health: medical anthropological perspectives. *Social Science & Medicine*. **59**(7), pp.1379-1395.
- Duffy, M.E. 2002. Methodological Issues In Web-based Research. *Journal of Nursing Scholarship*. **34**(1), pp.83-88.

- Durand, M.-A. et al. 2010. Information and decision support needs of parents considering amniocentesis: interviews with pregnant women and health professionals. *Health Expectations*. **13**(2), pp.125-138.
- Edwards, M. et al. 2009. What are the external influences on information exchange and shared decision-making in healthcare consultations: A meta-synthesis of the literature. *Patient Education and Counseling*. **75**(1), pp.37-52.
- Ekelin, M. et al. 2004. A qualitative study of mothers' and fathers' experiences of routine ultrasound examination in Sweden. *Midwifery*. **20**(4), pp.335-344.
- Eldh, A.C. et al. 2006. The meaning of patient participation for patients and nurses at a nurse-led clinic for chronic heart failure. *European Journal of Cardiovascular Nursing*. **5**(1), pp.45-53.
- Elo, S. and Kyngäs, H. 2008. The qualitative content analysis process. *Journal of Advanced Nursing*. **62**(1), pp.107-115.
- Elwy, A.R. et al. 2007. Attributions and Reported Communication of a Diagnosis of Down Syndrome. *Health Communication*. **22**(2), pp.115-121.
- Elwyn, G. et al. 2012. Shared decision making: a model for clinical practice. *Journal of general internal medicine*. **27**(10), pp.1361-1367.
- Elwyn, G. et al. 2000. Shared decision making and non-directiveness in genetic counselling. *Journal of Medical Genetics*. **37**(2), pp.135-138.
- Emerson, R.M., Fretz and Shaw, L.L. 1995. *Writing ethnographic field notes*. Chicago: The University of Chicago Press.
- Entwistle, V.A. et al. 2008. Communicating about screening. *BMJ*. **337**.
- Etchegary, H. et al. 2008. The influence of experiential knowledge on prenatal screening and testing decisions. *Genetic testing*. **12**(1), pp.115-124.
- Etchells, E. et al. 1996. Bioethics for clinicians: 1. Consent. *CMAJ: Canadian Medical Association Journal*. **155**(2), p177.
- Evans, M. et al. 2004. Relational Ethics and Genetic Counseling. *Nursing Ethics*. **11**(5), pp.459-471.
- Faden, R.R. and Beauchamp, T.L. 1986. *A history and theory of informed consent*. Oxford University Press, USA.
- Fairweather, J.R. et al. 2012. Computer-Assisted, Self-Interviewing (CASI) Compared to Face-to-Face Interviewing (FTFI) with Open-Ended, Non-Sensitive Questions. *International Journal of Qualitative Methods*. **11**(3), pp.280-291.

- Farquhar, C. et al. 2004. Antenatal couple counseling increases uptake of interventions to prevent HIV-1 transmission. *JAIDS Journal of Acquired Immune Deficiency Syndromes*. **37**(5), pp.1620-1626.
- Farrell, R.M. et al. 2011. Risk and uncertainty: Shifting decision making for aneuploidy screening to the first trimester of pregnancy. *Genet Med*. **13**(5), pp.429-436.
- Farsides, B. et al. 2004. Aiming towards “moral equilibrium”: health care professionals’ views on working within the morally contested field of antenatal screening. *Journal of Medical Ethics*. **30**(5), pp.505-509.
- Favre, R. et al. 2008. What about Informed Consent in First-Trimester Ultrasound Screening for Down Syndrome? *Fetal Diagnosis and Therapy*. **23**(3), pp.173-184.
- Ferguson, E. et al. 1995. A measure of knowledge and confidence in relation to HIV and AIDS: Reliability and validity. *AIDS Care*. **7**(2), pp.211-228.
- Fernandez, C.V. et al. 2003. Informing Study Participants of Research Results: An Ethical Imperative. *IRB: Ethics and Human Research*. **25**(3), pp.12-19.
- Figueiras, M. et al. 1999. Effects of Textual and Pictorial Information Upon Perceptions of Down Syndrome: An Analogue Study. *Psychology & Health*. **14**(4), pp.761-771.
- Finch, J. 1987. The Vignette Technique in Survey Research. *Sociology*. **21**(1), pp.105-114.
- Fishbein, M. and Ajzen, I. 1980. Understanding attitudes and predicting social behavior. *Prendice Hall. New York*.
- Foucault, M. 1982. The Subject and Power. *Critical Inquiry*. **8**(4), pp.777-795.
- Fox, F.E. et al. 2007. Doing Synchronous Online Focus Groups With Young People. *Qualitative Health Research*. **17**(4), pp.539-547.
- Fransen, M.P. et al. 2007. Ethnic differences in determinants of participation and non-participation in prenatal screening for Down syndrome: A theoretical framework. *Prenatal Diagnosis*. **27**(10), pp.938-950.
- Fransen, M.P. et al. 2010a. Ethnic differences in participation in prenatal screening for Down syndrome: A register-based study. *Prenatal Diagnosis*. **30**(10), pp.988-994.
- Fransen, M.P. et al. 2010b. Ethnic and socio-economic differences in uptake of prenatal diagnostic tests for Down's syndrome. *European Journal of Obstetrics & Gynecology and Reproductive Biology*. **151**(2), pp.158-162.
- Fransen, M.P. et al. 2009. Ethnic differences in considerations whether or not to participate in prenatal screening for Down syndrome. *Prenatal Diagnosis*. **29**(13), pp.1262-1269.

- Freer, Y. et al. 2009. More Information, Less Understanding: A Randomized Study on Consent Issues in Neonatal Research. *Pediatrics*. **123**(5), pp.1301-1305.
- Galbraith, N. et al. 2006. Research governance: Research governance approval is putting people off research. *BMJ*. **332**(7535), p238.
- García, E. et al. 2008a. The impact of ethical beliefs on decisions about prenatal screening tests: Searching for justification. *Social Science & Medicine*. **66**(3), pp.753-764.
- García, E. et al. 2008b. Rethinking autonomy in the context of prenatal screening decision-making. *Prenatal Diagnosis*. **28**(2), pp.115-120.
- Garcia, J. et al. 2002. Women's Views of Pregnancy Ultrasound: A Systematic Review. *Birth*. **29**(4), pp.225-250.
- Gates, E.A. 2004. Communicating risk in prenatal genetic testing. *Journal of Midwifery & Women's Health*. **49**(3), pp.220-227.
- Georgsson Öhman, S. et al. 2009. Perception of risk in relation to ultrasound screening for Down's syndrome during pregnancy. *Midwifery*. **25**(3), pp.264-276.
- Georgsson Öhman, S. and Waldenström, U. 2010. Effect of first-trimester ultrasound screening for Down syndrome on maternal-fetal attachment - A randomized controlled trial. *Sexual & Reproductive Healthcare*. **1**(3), pp.85-90.
- Ghosh, M. et al. 2011. Strategic reorganisation – impact on antenatal screening service for first trimester combined test in down's syndrome. *Archives of Disease in Childhood - Fetal and Neonatal Edition*. **96**(Suppl 1), pp.Fa62-Fa63.
- Gidiri, M. et al. 2010. Reduction in Down's syndrome screening acceptance is predominantly observed in women aged 25–35 years. *Women's Health*. **6**(4), pp.525-529.
- Gidiri, M. et al. 2007. Uptake of invasive testing following a positive triple test for Down's syndrome. Are midwives different counsellors compared with obstetricians? *Journal of Obstetrics & Gynaecology*. **27**(2), pp.148-149.
- Gill, J. and Johnson, P. 2010. *Research Methods for Managers*. 4th ed. London: SAGE Publications Ltd.
- Goldberg, H. 2009. Informed Decision Making in Maternity Care. *Journal of Perinatal Education*. **18**(1), pp.32-40.
- Gottfredsdóttir, H. 2005. Prospective first-time fathers and their views on fatherhood in the context of new policy on parental leave in Iceland. *Birth Issues*. **14**, pp.128–134.
- Gottfredsdóttir, H. et al. 2009a. How do prospective parents who decline prenatal screening account for their decision? A qualitative study. *Social Science & Medicine*. **69**(2), pp.274-277.

- Gottfredsdóttir, H. et al. 2009b. This is just what you do when you are pregnant': a qualitative study of prospective parents in Iceland who accept nuchal translucency screening. *Midwifery*. **25**(6), pp.711-720.
- Gourounti, K. et al. 2008. Women's Perception of Information and Experiences of Nuchal Translucency Screening in Greece. *Fetal Diagnosis and Therapy*. **24**(2), pp.86-91.
- Gourounti, K. and Sandall, J. 2008. Do pregnant women in Greece make informed choices about antenatal screening for Down's syndrome? A questionnaire survey. *Midwifery*. **24**(2), pp.153-162.
- Graneheim, U.H. and Lundman, B. 2004. Qualitative content analysis in nursing research: concepts, procedures and measures to achieve trustworthiness. *Nurse Education Today*. **24**(2), pp.105-112.
- Gray, N.J. et al. 2005. Health information-seeking behaviour in adolescence: the place of the internet. *Social science & medicine (1982)*. **60**(7), pp.1467-78.
- Green, J. et al. 2004. Psychosocial aspects of genetic screening of pregnant women and newborns: a systematic review. . *Health Technol Assess*. **8**(33).
- Green, J. and Thorogood, N. 2004. *Qualitative methods for health research*. Sage Publications Limited.
- Greenhalgh, T. and Peacock, R. 2005. Effectiveness and efficiency of search methods in systematic reviews of complex evidence: audit of primary sources. *BMJ*. **331**(7524), pp.1064-1065.
- Griffiths, M. 2005. Online Therapy for Addictive Behaviors. *CyberPsychology and Behavior*. **8**(6), pp.555-561.
- Guba and Lincoln. 2004. Competing Paradigms in Qualitative Research. In: Lincoln, N.K.D.Y.S. ed. *Handbook of qualitative research* London: Sage Publications, pp.105-117.
- Hall, S. et al. 2007. Undergoing prenatal screening for Down's syndrome: presentation of choice and information in Europe and Asia. *Eur J Hum Genet*. **15**(5), pp.563-569.
- Hamilton, R.J. and Bowers, B.J. 2006. Internet Recruitment and E-Mail Interviews in Qualitative Studies. *Qualitative Health Research*. **16**(6), pp.821-835.
- Hannes, K. 2011. Critical appraisal of qualitative research. In: Noyes J, B.A., Hannes K, Harden A, Harris J, Lewin S, Lockwood C ed. *Supplementary Guidance for Inclusion of Qualitative Research in Cochrane Systematic Reviews of Interventions*. Cochrane Collaboration Qualitative Methods Group.

- Hayeems, R.Z. et al. 2009. Informing parents about expanded newborn screening: influences on provider involvement. *Pediatrics*. **124**(3), pp.950-958.
- Hedberg, B. and Larsson, U.S. 2004. Environmental elements affecting the decision-making process in nursing practice. *Journal of Clinical Nursing*. **13**(3), pp.316-324.
- Heggland, L.-H. and Hausken, K. 2013. A Qualitative Identification of Categories of Patient Participation in Decision-Making by Health Care Professionals and Patients During Surgical Treatment. *Clinical Nursing Research*. **22**(2), pp.206-227.
- Heggland, L.-H. et al. 2013. Models, phases and cases of patient participation in decision-making in surgical treatment in Norway: A qualitative study. *Nursing & Health Sciences*. **15**(1), pp.39-44.
- Heritage, J. and Maynard, D.W. 2006. Problems and Prospects in the Study of Physician-Patient Interaction: 30 Years of Research. *Annual Review of Sociology*. **32**(1), pp.351-374.
- Hertig, S.G. et al. 2013. 'Doctor, what would you do in my position?' Health professionals and the decision-making process in pregnancy monitoring. *Journal of Medical Ethics*.
- Hesse-Biber, S. 2010. Qualitative Approaches to Mixed Methods Practice. *Qualitative Inquiry*. **16**(6), pp.455-468.
- Hewison, J. et al. 2007. Attitudes to prenatal testing and termination of pregnancy for fetal abnormality: a comparison of white and Pakistani women in the UK. *Prenatal Diagnosis*. **27**(5), pp.419-430.
- Heyman, B. and Henriksen, M. 2001. *Risk, Age and Pregnancy: A Case Study of Prenatal Genetic Screening and Testing*. Basingstoke: Palgrave Macmillan.
- Heyman, B. et al. 2006. On being at higher risk: A qualitative study of prenatal screening for chromosomal anomalies. *Social Science & Medicine*. **62**(10), pp.2360-2372.
- Hildingsson, I. and Sjöling, M. 2011. Fathers' experiences of support during pregnancy and the first year following childbirth – findings from a Swedish regional survey. *Journal of Men's Health*. **8**(4), pp.258-266.
- Hindley, C. and Thomson, A.M. 2005. The rhetoric of informed choice: perspectives from midwives on intrapartum fetal heart rate monitoring. *Health Expectations*. **8**(4), pp.306-314.
- Hodge, J.G. 2004. Ethical issues concerning genetic testing and screening in public health. *American Journal of Medical Genetics Part C: Seminars in Medical Genetics*. **125C**(1), pp.66-70.

- Holmes, S. 2009. Methodological and ethical considerations in designing an Internet study of quality of life: A discussion paper. *International Journal of Nursing Studies.* **46**(3), pp.394-405.
- Holstein, J.A. and Gubrium, J.F. 2001. *Handbook of interview research: Context and method.* Sage Publications, Incorporated.
- Horwitz, S.M. et al. 2007. Barriers to the Identification and Management of Psychosocial Issues in Children and Maternal Depression. *Pediatrics.* **119**(1), pp.e208-e218.
- Houts, P.S. et al. 2006. The role of pictures in improving health communication: A review of research on attention, comprehension, recall, and adherence. *Patient Education and Counseling.* **61**(2), pp.173-190.
- Hughes, R. 1998. Considering the Vignette Technique and its Application to a Study of Drug Injecting and HIV Risk and Safer Behaviour. *Sociology of Health & Illness.* **20**(3), pp.381-400.
- Hughes, R. and Huby, M. 2002. The application of vignettes in social and nursing research. *Journal of Advanced Nursing.* **37**(4), pp.382-386.
- Hughes, R. and Huby, M. 2004. The construction and interpretation of vignettes in social research. *Social Work and Social Sciences Review - An International Journal of Applied Re.* **11**(1), pp.36-51.
- Hulton, L.A., Zoe Matthews, and R. William Stones. 2000. *Framework for the Evaluation of Quality of Care in Maternity Services.* Southampton, UK.
- Hundt, G.L. et al. 2011. Inside 'Inside View': reflections on stimulating debate and engagement through a multimedia live theatre production on the dilemmas and issues of pre-natal screening policy and practice. *Health Expectations.* **14**(1), pp.1-9.
- Hundt, G.L. et al. 2008. Experiences of first trimester antenatal screening in a one-stop clinic. *British Journal of Midwifery.* **16**(3), pp.156-159.
- Hunt, L.M. et al. 2005. The routine and the traumatic in prenatal genetic diagnosis: does clinical information inform patient decision-making? *Patient Education and Counseling.* **56**(3), pp.302-312.
- Hunter, A.G.W. 2005. Is multicenter collaborative research in clinical genetics dead and, if so, what killed it? *American Journal of Medical Genetics Part A.* **134A**(3), pp.237-239.
- Hunter, B. 2006. The importance of reciprocity in relationships between community-based midwives and mothers. *Midwifery.* **22**(4), pp.308-322.
- Huntington, B. and Kuhn, N. 2003. Communication gaffes: a root cause of malpractice claims. *Proceedings (Baylor University. Medical Center).* **16**(2), p157.

- Im, E.-O. and Chee, W. 2006. An Online Forum as a Qualitative Research Method: Practical Issues. *Nursing Research*. **55**(4), pp.267-273.
- Ivry, T. and Teman, E. 2008. Expectant Israeli Fathers and the Medicalized Pregnancy: Ambivalent Compliance and Critical Pragmatism. *Culture, Medicine, and Psychiatry*. **32**(3), pp.358-385.
- Janssen, B. and Wiegers, T.A. 2006. Strengths and weaknesses of midwifery care from the perspective of women. *Evidence Based Midwifery*. **4**(2), pp.53-59.
- Jaques, A. et al. 2004a. Do women know that prenatal testing detects fetuses with Down syndrome? *Journal of Obstetrics & Gynaecology*. **24**(6), pp.647-651.
- Jaques, A.M. et al. 2004b. People who influence women's decisions and preferred sources of information about prenatal testing for birth defects. *Australian and New Zealand Journal of Obstetrics and Gynaecology*. **44**(3), pp.233-238.
- Jenkins, N. et al. 2010. Putting it in context: the use of vignettes in qualitative interviewing. *Qualitative Research*. **10**(2), pp.175-198.
- Jepson, R.G. et al. 2005. How should we measure informed choice? The case of cancer screening. *Journal of Medical Ethics*. **31**(4), pp.192-196.
- Johnson, B. 2000. Using video vignettes to evaluate children's personal safety knowledge: methodological and ethical issues. *Child Abuse & Neglect*. **24**(6), pp.811-827.
- Jones, I.R. et al. 2004. Is patient involvement possible when decisions involve scarce resources? A qualitative study of decision-making in primary care. *Social Science & Medicine*. **59**(1), pp.93-102.
- Kahn, D.L. 1999. Making the Best of It: Adapting to the Ambivalence of a Nursing Home Environment. *Qualitative Health Research*. **9**(1), pp.119-132.
- Kalafat, J. and Gagliano, C. 1996. The Use of Simulations to Assess the Impact of an Adolescent Suicide Response Curriculum. *Suicide and Life-Threatening Behavior*. **26**(4), pp.359-364.
- Kaplan, S. and Kaplan, R. 2008. Health, Supportive Environments, and the Reasonable Person Model. *Urban Ecology*. Springer, pp.557-565.
- Kelly, S.E. 2009. Choosing not to choose: reproductive responses of parents of children with genetic conditions or impairments. *Sociology of Health & Illness*. **31**(1), pp.81-97.
- Kenny, A.J. 2005. Interaction in cyberspace: an online focus group. *Journal of Advanced Nursing*. **49**(4), pp.414-422.
- Kerr, A. and Shakespeare, T. 2002. Genetic politics: From eugenics to genome.

- Khoshnood, B. et al. 2004. Socioeconomic Barriers to Informed Decisionmaking Regarding Maternal Serum Screening for Down Syndrome: Results of the French National Perinatal Survey of 1998. *Am J Public Health*. **94**(3), pp.484-491.
- Khoshnood, B. et al. 2003a. Effects of maternal age and education on the pattern of prenatal testing: Implications for the use of antenatal screening as a solution to the growing number of amniocenteses. *American Journal of Obstetrics and Gynecology*. **189**(5), pp.1336-1342.
- Khoshnood, B. et al. 2003b. Socioeconomic and state-level differences in prenatal diagnosis and live birth prevalence of Down's syndrome in the United States. *Rev Epidemiol Sante Publique*. **51**, pp.617 - 627.
- King, J.S. and Moulton, B. 2006. Rethinking informed consent: the case for shared medical decision-making. *American Journal of Law and Medicine*. **32**, pp.429-501.
- Kirkham, M. et al. 2002. Checking not listening: how midwives cope. *British Journal of Midwifery*. **10**(7), pp.447-450.
- Kirklin, D. 2007. Framing, truth telling and the problem with non-directive counselling. *Journal of Medical Ethics*. **33**(1), pp.58-62.
- Kobayashi, H. et al. 2011. Patient perception of nursing service quality; an applied model of Donabedian's structure-process-outcome approach theory. *Scandinavian journal of caring sciences*. **25**(3), pp.419-25.
- Koenig, C.J. 2011. Patient resistance as agency in treatment decisions. *Social Science & Medicine*. **72**(7), pp.1105-1114.
- Kohlbacher, F. 2006. The use of qualitative content analysis in case study research. In: *Forum Qualitative Sozialforschung/Forum: Qualitative Social Research*.
- Korenromp, M.J. et al. 2007. Maternal decision to terminate pregnancy in case of Down syndrome. *American Journal of Obstetrics and Gynecology*. **196**(2), pp.149.e1-149.e11.
- Kraut, R. et al. 2004. Psychological research online: report of Board of Scientific Affairs' Advisory Group on the Conduct of Research on the Internet. *American Psychologist*. **59**(2), p105.
- Kuppermann, M. et al. 2006. Beyond Race or Ethnicity and Socioeconomic Status: Predictors of Prenatal Testing for Down Syndrome. *Obstetrics & Gynecology*. **107**(5), pp.1087-1097 10.1097/01.AOG.0000214953.90248.db.
- Kuzel, A. 1992. Sampling in Qualitative Inquiry. In: Crabtree, B.F. and Miller, W.L. eds. *Doing Qualitative Research*. Newbury Park ; London :: Sage.
- Kvale, S. 2007. *Doing interviews*. Thousand Oaks, C.A: Sage Publications Limited.

- Lacey and Luff, D. 2007. *Qualitative Research Analysis. The NIHR RDS for the East Midlands / Yorkshire & the Humber.*
- Lambert, S.D. and Loiselle, C.G. 2008. Combining individual interviews and focus groups to enhance data richness. *Journal of Advanced Nursing*. **62**(2), pp.228-237.
- Larrick, R.P. 2008. Debiasing. *Blackwell Handbook of Judgment and Decision Making*. Blackwell Publishing Ltd, pp.316-338.
- Lawson, K.L. 2006. Expectations of the parenting experience and willingness to consider selective termination for Down Syndrome. *Journal of Reproductive and Infant Psychology*. **24**(1), pp.43-59.
- Lawson, K.L. and Pierson, R.A. 2007. Maternal decisions regarding prenatal diagnosis: rational choices or sensible decisions? *Journal of obstetrics and gynaecology Canada: JOGC=Journal d'obstetrique et gynecologie du Canada: JOGC*. **29**(3), p240.
- Lawson, K.L. and Walls-Ingram, S.A. 2010. Selective Abortion for Down Syndrome: The Relation Between the Quality of Intergroup Contact, Parenting Expectations, and Willingness to Terminate. *Journal of Applied Social Psychology*. **40**(3), pp.554-578.
- Légaré, F. et al. 2010. Interventions for improving the adoption of shared decision making by healthcare professionals. *Cochrane Database Syst Rev*. **5**.
- Levis, D.M. et al. 2012. Women's knowledge, attitudes, and beliefs about Down syndrome: A qualitative research study. *American Journal of Medical Genetics Part A*. **158A**(6), pp.1355-1362.
- Lewando-Hundt, G., Green, JM, Sandall, J, Hirst, J, Ahmed, S and Hewison, J. 2006. Navigating the troubled water of prenatal testing decisions. In: Webster, A. ed. *New Technologies in Health Care*. Basingstoke: Palgrave Macmillan, pp.25-33.
- Lewando-Hundt, G. et al. 2001. Knowledge, action and resistance: the selective use of prenatal screening among Bedouin women of the Negev, Israel. *Social Science & Medicine*. **52**(4), pp.561-569.
- Lewando Hundt, G., Sandall,J.,Spencer,K.,Williams,C and Heyman,B. 2004. *End of Award Report: Social Implications of One-Stop First Trimester Antenatal Screening*. London.: ESRC.
- Lewis, J. 2003. Design issues. In: RITCHIE AND LEWIS, J. ed. *Qualitative Research Practice. A Guide for Social Students and Researchers*. London: Sage Publications.
- Lewis, S.M. et al. 2006a. Women's and health professionals' preferences for prenatal testing for Down syndrome in Australia. *Australian and New Zealand Journal of Obstetrics and Gynaecology*. **46**(3), pp.205-211.

- Lewis, S.M. et al. 2006b. A comparison of Australian and UK obstetricians' and midwives' preferences for screening tests for Down syndrome. *Prenatal Diagnosis*. **26**(1), pp.60-66.
- Li, D.-K. et al. 2008. Factors influencing women's acceptance of prenatal screening tests. *Prenatal Diagnosis*. **28**(12), pp.1136-1143.
- Liamputpong, P. et al. 2003. Why Do Women Decline Prenatal Screening and Diagnosis? Australian Women's Perspective. *Women & Health*. **37**(2), pp.89-108.
- Lincoln and Guba, E.G. 1985. *Naturalistic Inquiry*. Beverly Hills, CA: Sage.
- Link, B.G. et al. 1999. Public conceptions of mental illness: labels, causes, dangerousness, and social distance. *American Journal of Public Health*. **89**(9), pp.1328-1333.
- Lippman, A. 1999. Choice as a risk to women's health. *Health, Risk & Society*. **1**(3), pp.281-291.
- Locock, L. and Alexander, J. 2006. 'Just a bystander'? Men's place in the process of fetal screening and diagnosis. *Social Science & Medicine*. **62**(6), pp.1349-1359.
- Lynn, F. et al. 2010. Current trends in antenatal screening services: results from a regional survey. *The Ulster medical journal*. **79**(1), pp.12-15.
- Malone, F.D. et al. 2005. First-trimester or second-trimester screening, or both, for Down's syndrome. *New England Journal of Medicine*. **353**(19), pp.2001-2011.
- Mander, R. and Melender, H.-L. 2009. Choice in maternity: rhetoric, reality and resistance. *Midwifery*. **25**(6), pp.637-648.
- Mann, C. and Stewart, F. 2000. *Internet communication and qualitative research : a handbook for researching online*. London; Thousand Oaks, Calif.: Sage Publications.
- Markens, S. et al. 2003. "I'm Not the One They're Sticking the Needle Into": Latino Couples, Fetal Diagnosis, and the Discourse of Reproductive Rights. *Gender & Society*. **17**(3), pp.462-481.
- Markens, S.B., C. H. Press, Nancy. 1999. Because of the risks: How US pregnant women account for refusing prenatal screening. *Social Science & Medicine*. **49**(3), pp.359-369.
- Martin, E. 2004. Vignettes and Respondent Debriefing for Questionnaire Design and Evaluation. *Methods for Testing and Evaluating Survey Questionnaires*. John Wiley & Sons, Inc., pp.149-171.
- Mason, J. 2002. *Qualitative researching*. SAGE Publications Limited.
- Mason, M. 2010. Sample size and saturation in PhD studies using qualitative interviews. *Forum Qual. Sozialforschung Forum Qualitative Sozialforschung*. **11**(3).

- Maternity Action. 2013. *Rights at work for fathers and partners* [Online]. [Accessed 10/09/2013]. Available from: <http://www.maternityaction.org.uk/sitebuildercontent/sitebuilderfiles/fathersandpartners.pdf>.
- Matthews, J. and Cramer, E.P. 2008. Using Technology to Enhance Qualitative Research with Hidden Populations. *Qualitative Report*. **13**(2), pp.301-315.
- Maxwell, S. et al. 2011. Socio-demographic disparities in the uptake of prenatal screening and diagnosis in Western Australia. *Australian and New Zealand Journal of Obstetrics and Gynaecology*. **51**(1), pp.9-16.
- Maynard, D.W. and Schaeffer, N.C. 2002. "Opening and Closing the Gate: the work of optimism in Recruiting Survey Respondents ". in Maynard, Douglas W et al.(red.), *Standardizations and Tacit Knowledge—Interaction and Practice in the Survey Interview*, New York: John Wiley & Sons. pp.179-204.
- McCabe, C. 2004. Nurse–patient communication: an exploration of patients' experiences. *Journal of Clinical Nursing*. **13**(1), pp.41-49.
- McCormack, B. and McCance, T.V. 2006. Development of a framework for person-centred nursing. *Journal of Advanced Nursing*. **56**(5), pp.472-479.
- McCormick, M.J. 2011. Ethical Concerns about Genetic Screening: The Down's Dilemma. *The Journal for Nurse Practitioners*. **7**(4), pp.316-320.
- McCourt, C. 2006. Supporting choice and control? Communication and interaction between midwives and women at the antenatal booking visit. *Social Science & Medicine*. **62**(6), pp.1307-1318.
- McCoyd, J.L.M. and Kerson, T.S. 2006. Conducting Intensive Interviews Using Email. *Qualitative Social Work*. **5**(3), pp.389-406.
- McKenzie and Oliphant. 2006. *The presentation of complementary and alternative medicine information in Canadian midwifery care*. [Online]. [Accessed 02.12.12]. Available from: [http://www.cais-acsi.ca/proceedings/2006/mckenzie\\_2006.pdf](http://www.cais-acsi.ca/proceedings/2006/mckenzie_2006.pdf).
- McKenzie, P.J. 2009. Informing choice: the organization of institutional interaction in clinical midwifery care. *Library & Information Science Research* **31**(3), pp.163-173.
- McKenzie, P.J. 2010. Informing relationships: small talk, informing, and relationship building in midwife-woman interaction. *Information Research* **15**(1).
- McLachlan, H.L. et al. 2009. Women's views of postnatal care in the context of the increasing pressure on postnatal beds in Australia. *Women and Birth*. **22**(4), pp.128-133.

- McNeill et al. 2009. Down's syndrome screening in Northern Ireland: women's reasons for accepting or declining serum testing. *Evidence Based Midwifery*. **7**(3), pp.76-83.
- McNeill, J. and Alderdice, F. 2009. Exploring the perspective of midwives involved in offering serum screening for Down's syndrome in Northern Ireland. *Journal of Clinical Nursing*. **18**(20), pp.2888-96.
- Merriam, S.B. 1988. *Case study research in education: A qualitative approach*. Jossey-Bass.
- Midwifery 2020 Programme. 2010. *Core Role of the Midwife Workstream Final Report*. [Online]. [Accessed 01.12.12]. Available from: [http://www.midwifery2020.org/documents/2020/Core\\_Role.pdf](http://www.midwifery2020.org/documents/2020/Core_Role.pdf).
- Miles, M.B. and Huberman, A.M. 1994. *Qualitative data analysis: An expanded sourcebook*. Sage Publications, Incorporated.
- Miller, L.S. et al. 2004. Improving the identification and treatment of postpartum depression in a managed care organization. *JCOM*. **11**(3).
- Modra, L. 2006. Prenatal genetic testing kits sold at your local pharmacy: promoting autonomy or promoting confusion? *Bioethics*. **20**(5), pp.254-263.
- Montoya-Weiss, M.M. et al. 1998. On-line focus groups: conceptual issues and a research tool. *European Journal of Marketing*. **32**(7-8), pp.713-723.
- Morestin, F. et al. 2010. Evaluating quality of obstetric care in low-resource settings: Building on the literature to design tailor-made evaluation instruments - an illustration in Burkina Faso. *BMC Health Services Research*. **10**(1), p20.
- Morgan, D.L. 1993. Qualitative Content Analysis: A Guide to Paths not Taken. *Qualitative Health Research*. **3**(1), pp.112-121.
- Morgan, D.L. 2007. Paradigms Lost and Pragmatism Regained. *Journal of Mixed Methods Research*. **1**(1), pp.48-76.
- Morris, J.K. and Alberman, E. 2010. Trends in Down's syndrome live births and antenatal diagnoses in England and Wales from 1989 to 2008: analysis of data from the National Down Syndrome Cytogenetic Register. *BMJ*. **339**.
- Morse, J.M. 1991. Negotiating commitment and involvement in the nurse-patient relationship. *Journal of Advanced Nursing*. **16**(4), pp.455-468.
- Morse, J.M. and Field, P.A. 1996. *Nursing research: The application of qualitative approaches*. Nelson Thornes.

- Muggli, E.E. et al. 2006. Mapping uptake of prenatal diagnosis for Down syndrome and other chromosome abnormalities across Victoria, Australia. *Australian and New Zealand Journal of Obstetrics and Gynaecology*. **46**(6), pp.492-500.
- Müller, M.A. et al. 2006. Women's opinions on the offer and use of nuchal translucency screening for Down syndrome. *Prenatal Diagnosis*. **26**(2), pp.105-111.
- Murray, E. et al. 2006. Shared decision-making in primary care: Tailoring the Charles et al. model to fit the context of general practice. *Patient Education and Counseling*. **62**(2), pp.205-211.
- Mutton, D. et al. 1998. Trends in prenatal screening for and diagnosis of Down's syndrome: England and Wales, 1989-97. *BMJ*. **317**(7163), pp.922-923.
- Nagle, C. et al. 2009. Reporting Down syndrome screening results: women's understanding of risk. *Prenatal Diagnosis*. **29**(3), pp.234-239.
- Nagle, C. et al. 2008. Exploring general practitioners' experience of informing women about prenatal screening tests for foetal abnormalities: A qualitative focus group study. *BMC Health Services Research*. **8**(1), p114.
- National Screening Committee, U.K. 2012. Screening tests for you and your baby [Online]. [Accessed 30.10.12]. Available from:  
<http://fetalanomaly.screening.nhs.uk/publicationsandleaflets>.
- Nawaz, T.S. et al. 2011. Direct access midwifery booking for prenatal care and its role in Down syndrome screening. *Prenatal Diagnosis*. **31**(10), pp.985-989.
- NHS FASP. 2011. *Fetal Anomaly Screening Programme: Consent Standard and Guidance*. [Online]. [Accessed 18.11.12]. Available from:  
<http://fetalanomaly.screening.nhs.uk/standardsandpolicies>.
- NHSLA. 2012. *Clinical Negligence Scheme for Trusts: Maternity Clinical Risk Management Standards*. [Online]. [Accessed 12.02.13]. Available from: [www.nhsla.com](http://www.nhsla.com).
- NICE. 2008. *Antenatal care: Routine care for the healthy pregnant woman. Full guideline*. National Institute for Health and Clinical Evidence. [Online]. [Accessed 04.02.11]. Available from: <http://www.nice.org.uk/guidance/index.jsp?action=byID&o=11947>.
- Nicholas and Yavas, U. 2006. Vignette Development: An Exposition and Illustration *Innovative Marketing*. **Volume 2**(Issue 1).
- Nicholas, D.B., Lucy Lach.,Gillian King.,Marjorie Scott.,Katherine Boydell.,Bonita J. Sawatzky.,Joe Reisman.,Erika SchippeL.,Nancy L. Young. 2010. *Contrasting Internet and Face-to-Face Focus Groups for Children with Chronic Health Conditions: Outcomes and Participant*

- Experiences.* [Online]. [Accessed 12.03.2012]. Available from:  
<http://ejournals.library.ualberta.ca/index.php/IJQM/article/view/4080>.
- Nicol, M. 2007. Vulnerability of first-time expectant mothers during ultrasound scans: an evaluation of the external pressures that influence the process of informed choice. *Health Care for Women International*. **28**(6), pp.525-533.
- Nicolaides, K.H. 2004. Nuchal translucency and other first-trimester sonographic markers of chromosomal abnormalities. *American Journal of Obstetrics and Gynecology*. **191**(1), pp.45-67.
- Noseworthy, D.A. et al. 2013. Towards a relational model of decision-making in midwifery care. *Midwifery*. **29**(7), pp.e42-e48.
- Novick, G. 2009. Women's Experience of Prenatal Care: An Integrative Review. *Journal of Midwifery & Women's Health*. **54**(3), pp.226-237.
- O'Connell, R. and Downe, S. 2009. A metasynthesis of midwives' experience of hospital practice in publicly funded settings: compliance, resistance and authenticity. *Health*. **13**(6), pp.589-609.
- Ockleford, E. et al. 2003. Do women understand prenatal screening for fetal abnormality? *British Journal of Midwifery*. **11**(7), pp.445-449.
- Ofcom. 2012. *Communications Market Report: United Kingdom*. [Online]. [Accessed 20.09.12]. Available from: <http://stakeholders.ofcom.org.uk/market-data-research/market-data/communications-market-reports/cmr12/uk/>.
- Office for National Statistics. 2013. *Ward Population Estimates, Mid-2011*
- Oringderff, J. 2004. *My Way: Piloting an Online Focus Group*. [Online]. [Accessed 12.03.2012]. Available from:  
<http://ejournals.library.ualberta.ca/index.php/IJQM/article/view/4466>.
- Park, A. and Mathews, M. 2009. Women's decisions about maternal serum screening testing: A qualitative study exploring what they learn and the role prenatal care providers play. *Women and Birth*. **22**(2), pp.73-78.
- Partridge, A. and Winer, E.P. 2002. Informing clinical trial participants about study results. *JAMA*. **288**(3), pp.363-365.
- Patton, M.Q. 1990. *Qualitative evaluation and research methods*. 2nd ed. Newbury Park, CA: Sage Publications.
- Paul, D.B. 1998. *The politics of heredity: Essays on eugenics, biomedicine, and the nature-nurture debate*. SUNY Press.

- Payne, S. et al. 2007. Case study research methods in end-of-life care: reflections on three studies. *Journal of Advanced Nursing*. **58**(3), pp.236-245.
- Peabody, J.W. et al. 2004. Measuring the Quality of Physician Practice by Using Clinical Vignettes: A Prospective Validation Study. *Annals of Internal Medicine*. **141**(10), pp.771-780.
- Petchenik, J. and Watermolen, D.J. 2011. A Cautionary Note on Using the Internet to Survey Recent Hunter Education Graduates. *Human Dimensions of Wildlife*. **16**(3), pp.216-218.
- Pilnick, A. 2004. 'It's Just One of the Best Tests that We've Got at the Moment': the Presentation of Nuchal Translucency Screening for Fetal Abnormality in Pregnancy. *Discourse & Society*. **15**(4), pp.451-465.
- Pilnick, A. 2008. 'It's something for you both to think about': choice and decision making in nuchal translucency screening for Down's syndrome. *Sociology of Health & Illness*. **30**(4), pp.511-530.
- Pilnick, A. and Zayts, O. 2012. 'Let's have it tested first': choice and circumstances in decision-making following positive antenatal screening in Hong Kong. *Sociology of Health & Illness*. **34**(2), pp.266-282.
- Pilnick, A.M. et al. 2004. Presenting and discussing nuchal translucency screening for fetal abnormality in the UK. *Midwifery*. **20**(1), pp.82-93.
- Plantin, L. et al. 2003. Talking and Doing Fatherhood: On Fatherhood and Masculinity in Sweden and England. *Fathering: A Journal of Theory, Research, and Practice about Men as Fathers*. **1**(1), pp.3-26.
- Polansky, S. 2006. Overcoming the obstacles: a collaborative approach to informed consent in prenatal genetic screening. *Health LJ*. **14**, p21.
- Porter, S. et al. 2007. New midwifery? A qualitative analysis of midwives' decision-making strategies. *Journal of Advanced Nursing*. **60**(5), pp.525-534.
- Potter, B.K. et al. 2008. Exploring informed choice in the context of prenatal testing: findings from a qualitative study. *Health Expectations*. **11**(4), pp.355-365.
- Prathapan, S. et al. 2012. Are there socio-economic inequalities in the uptake of Down syndrome screening in the UK? *Prenatal Diagnosis*. **32**(3), pp.293-295.
- Preloran, H.M. et al. 2005. Impact of Interpreters' Approach on Latinas' Use of Amniocentesis. *Health Education & Behavior*. **32**(5), pp.599-612.
- Press, N. and Browner, C.H. 1997. Why women say yes to prenatal diagnosis. *Social Science & Medicine*. **45**(7), pp.979-989.

- Rantanen, E. et al. 2008. What is ideal genetic counselling? A survey of current international guidelines. *Eur J Hum Genet.* **16**(4), pp.445-452.
- Rapley, T. 2008a. Distributed decision making: the anatomy of decisions-in-action. *Sociology of Health & Illness.* **30**(3), pp.429-444.
- Rapley, T. 2008b. *Doing conversation, discourse and document analysis.* SAGE Publications Limited.
- Rapp, R. 2000. *Testing women, testing the fetus.* New York: Routledge
- Rausch, D. et al. 2000. Participation in maternal serum screening following screen positive results in a previous pregnancy. *Journal of Medical Screening.* **7**(1), pp.4-6.
- Reed, K. 2009a. Fathers' involvement in antenatal screening: midwives' views. *British Journal of Midwifery.* **17**(4), pp.218-222.
- Reed, K. 2009b. 'It's them faulty genes again': women, men and the gendered nature of genetic responsibility in prenatal blood screening. *Sociology of Health & Illness.* **31**(3), pp.343-359.
- Reed, K. 2011. Making men matter: exploring gender roles in prenatal blood screening. *Journal of Gender Studies.* **20**(1), pp.55-66.
- Reid, B. et al. 2009. A meta-synthesis of pregnant women's decision-making processes with regard to antenatal screening for Down syndrome. *Social Science & Medicine.* **69**(11), pp.1561-1573.
- Reid, D.J. and Reid, F.J.M. 2005. An in-depth comparison of computer-mediated and conventional focus group discussions. *Journal of the Market Research Society.* **47**(2), p131.
- Remennick, L. 2006. The quest for the perfect baby: why do Israeli women seek prenatal genetic testing? *Sociology of Health & Illness.* **28**(1), pp.21-53.
- Rezabek, R. 2000. Online Focus Groups: Electronic Discussions for Research. *Forum Qualitative Sozialforschung / Forum: Qualitative Social Research.* [Online]. **1**(1). Available from: <http://www.qualitative-research.net/index.php/fqs/article/view/1128/2509>.
- Rhodes, S.D. et al. 2003. Collecting behavioural data using the world wide web: considerations for researchers. *Journal of Epidemiology and Community Health.* **57**(1), pp.68-73.
- Rosenstock, I.M. 1974. Historical origins of the health belief model. *Health Education & Behavior.* **2**(4), pp.328-335.

- Rostant, K. et al. 2003. Survey of the knowledge, attitudes and experiences of Western Australian women in relation to prenatal screening and diagnostic procedures. *Australian and New Zealand Journal of Obstetrics and Gynaecology*. **43**(2), pp.134-138.
- Rowe, H.J. et al. 2006. Are pregnant Australian women well informed about prenatal genetic screening? A systematic investigation using the Multidimensional Measure of Informed Choice. *Australian and New Zealand Journal of Obstetrics and Gynaecology*. **46** (5), pp.433-439.
- Rowe, M., H; Heron, P; Quigley, M; Askham, J and Brocklehurst, P. 2007. *Women's Experience of Antenatal Screening are there Social and Ethnic Inequalities in Access to Antenatal Screening for Down's syndrome?* Oxford: National Perinatal Epidemiology Unit; Picker Institute.
- Rowe, R. et al. 2008. Offer and uptake of prenatal screening for Down syndrome in women from different social and ethnic backgrounds. *Prenatal Diagnosis*. **28**(13), pp.1245-1250.
- Rowe, R.E. and Garcia, J. 2003. Social class, ethnicity and attendance for antenatal care in the United Kingdom: a systematic review. *Journal of Public Health Medicine*. **25**(2), pp.113-119.
- Royal College of Midwives. 2011. *State of Maternity Services Report* [Online]. [Accessed 02.12.12]. Available from: [www.rcm.org.uk/EasySiteWeb/GatewayLink.aspx?alId=185112](http://www.rcm.org.uk/EasySiteWeb/GatewayLink.aspx?alId=185112).
- Rozario, S. 2005. Genetics, religion and identity among British Bangladeshis: some initial findings. *Diversity in Health and Social Care*. **2**(3), pp.187-196.
- Russell, C.K. and Gregory, D.M. 2003. Evaluation of qualitative research studies. *Evidence Based Nursing*. **6**(2), pp.36-40.
- Sackett, D.L. et al. 1996. Evidence based medicine: what it is and what it isn't. *BMJ*. **312**(7023), pp.71-72.
- Sahlsten, M.J. et al. 2008. An analysis of the concept of patient participation. In: *Nursing forum*: Wiley Online Library, pp.2-11.
- Salmon, P. and Hall, G.M. 2003. Patient empowerment and control: a psychological discourse in the service of medicine. *Social Science & Medicine*. **57**(10), pp.1969-1980.
- Salmons, J. 2011. *Cases in Online Interview Research*. Sage Publications, Incorporated.
- Sandelowski, M. 2000. Whatever happened to qualitative description? *Research in nursing & health*. **23**(4), pp.334-340.
- Sandelowski, M. et al. 1997. Qualitative metasynthesis: Issues and techniques. *Research in nursing & health*. **20**(4), pp.365-371.

- Sandelowski, M. et al. 2007. Comparability work and the management of difference in research synthesis studies. *Social Science & Medicine*. **64**(1), pp.236-247.
- Santalahti, P. et al. 1998. Women's decision-making in prenatal screening. *Social Science & Medicine*. **46**(8), pp.1067-1076.
- Saunders, M. et al. 2009. Research methods for business students.
- Schoonen, M. et al. 2012. Informing on prenatal screening for Down syndrome prior to conception. An empirical and ethical perspective. *American Journal of Medical Genetics Part A*. **158A**(3), pp.485-497.
- Schott, J. 2002. Parent education: meeting the needs of fathers. *Practicing Midwife*. **5**(36-38).
- Schwab, A.P. 2008. Putting cognitive psychology to work: Improving decision-making in the medical encounter. *Social Science & Medicine*. **67**(11), pp.1861-1869.
- Seale, C. et al. 2010. Interviews and Internet Forums: A Comparison of Two Sources of Qualitative Data. *Qualitative Health Research*. **20**(5), pp.595-606.
- Seavilleklein, V. 2009. CHALLENGING THE RHETORIC OF CHOICE IN PRENATAL SCREENING. *Bioethics*. **23**(1), pp.68-77.
- Seror, V. and Ville, Y. 2009. Prenatal screening for Down syndrome: women's involvement in decision-making and their attitudes to screening. *Prenatal Diagnosis*. **29**(2), pp.120-128.
- Shantha, N. et al. 2009. Women's choice for Down's screening-A comparative experience in three district general hospitals. *European Journal of Obstetrics & Gynecology and Reproductive Biology*. **146**(1), pp.61-64.
- Sharma, G. et al. 2007. Ethical considerations of early (first vs. second trimester) risk assessment disclosure for trisomy 21 and patient choice in screening versus diagnostic testing. *American Journal of Medical Genetics Part C: Seminars in Medical Genetics*. **145C**(1), pp.99-104.
- Sharpe, N. 1996. Reinventing the Wheel: Informed Consent and Genetic Testing for Breast Cancer, Cystic Fibrosis, and Huntington Disease. *Queen's LJ*. **22**, p389.
- Sheppard, M. and Ryan, K. 2003. Practitioners as Rule Using Analysts: A Further Development of Process Knowledge in Social Work. *British Journal of Social Work*. **33**(2), pp.157-176.
- Shojania, K.G. et al. 2007. *Closing the quality gap : a critical analysis of quality improvement strategies volume 7, Care coordination*. Rockville, Md.: U.S. Dept. of Health and Human Services, Public Health Service, Agency for Healthcare Research and Quality.

- Silverman, D. 2011. *Interpreting qualitative data*. Sage Publications Limited.
- Skirton, H. and Barr, O. 2007. *Influences on uptake of antenatal screening for Down syndrome: a review of the literature*. Evidence Based Midwifery. vol 5, no 1, March 2007, pp 4-9.
- Skirton, H. and Barr, O. 2010. Antenatal screening and informed choice: a cross-sectional survey of parents and professionals. *Midwifery*. **26**(6), pp.596-602.
- Smith, D.K. et al. 1994. Lack of knowledge in health professionals: a barrier to providing information to patients? *Quality in Health Care*. **3**(2), pp.75-78.
- Smith, M. et al. 2008. Factors influencing clinical decision making. In: Higgs, J., Loftus & Christensen ed. *Clinical reasoning in the Health Professions* 3rd ed. Boston, Mass: Elsevier Health Sciences., Butterworth-Heinemann, pp.89-100.
- Smith., J., Higgs., Ellis E. 2008. Factors influencing clinical decision making. In: Higgs J., M., Jones ., Loftus S., Christensen, N ed. *In Clinical Reasoning in the Health Professions*. 3rd ed. Amsterdam: Butterworth-Heinemann/Elsevier, pp.89–95.
- Spalding, N.J. and Phillips, T. 2007. Exploring the Use of Vignettes: From Validity to Trustworthiness. *Qualitative Health Research*. **17**(7), pp.954-962.
- Spoel, P. 2007. A Feminist Rhetorical Perspective on Informed Choice in Midwifery. *Rhetor: Journal of the Canadian Society for the Study of Rhetoric*. **2**.
- St-Jacques, S. et al. 2008. Decisional needs assessment regarding Down syndrome prenatal testing: a systematic review of the perceptions of women, their partners and health professionals. *Prenatal Diagnosis*. **28**(13), pp.1183-1203.
- Stefansdottir, V. et al. 2010. Effects of knowledge, education, and experience on acceptance of first trimester screening for chromosomal anomalies. *Acta Obstetricia et Gynecologica Scandinavica*. **89**(7), pp.931-938.
- Stivers, T. 2002. Participating in decisions about treatment: overt parent pressure for antibiotic medication in pediatric encounters. *Social Science & Medicine*. **54**(7), pp.1111-1130.
- Stivers, T. 2005. Parent Resistance to Physicians' Treatment Recommendations: One Resource for Initiating a Negotiation of the Treatment Decision. *Health Communication*. **18**(1), pp.41-74.
- Strauss, A. and Corbin, J.M. 1998. *Basics of Qualitative Research: Techniques and Procedures for Developing Grounded Theory*. SAGE.

- Styles, M. et al. 2011. The development of research tools used in the STORK Study (the Scottish Trial of Refer or Keep) to explore midwives' intrapartum decision making. *Midwifery*. **27**(5), pp.576-581.
- Sullivan, A. 2006. Involving parents: information and informed decisions. *Midwife's Guide to Antenatal Investigations*, Elsevier, Edinburgh. pp.17-29.
- Suter, S.M. 2002. Routinization of Prenatal Testing, The. *Am. JL & Med.* **28**, p233.
- Tashakkori, A. and Creswell, J.W. 2007. The new era of mixed methods. *Journal of mixed methods research*. **1**(1), pp.3-7.
- Tashakkori and Teddlie, C. 1998. *Mixed Methodology: Combining Qualitative and Quantitative Approaches*. Thousand Oaks, CA: Sage.
- Tates, K. et al. 2009. Online focus groups as a tool to collect data in hard-to-include populations: examples from paediatric oncology. *BMC medical research methodology*. **9**(1), p15.
- Taylor, B.J. 2006. Factorial Surveys: Using Vignettes to Study Professional Judgement. *British Journal of Social Work*. **36**(7), pp.1187-1207.
- Thachuk, A. 2007. Midwifery, Informed Choice, and Reproductive Autonomy: A Relational Approach. *Feminism & Psychology*. **17**(1), pp.39-56.
- Theuring, S. et al. 2009. Male Involvement in PMTCT Services in Mbeya Region, Tanzania. *AIDS and Behavior*. **13**(0), pp.92-102.
- Toerien, M. et al. 2011. Offering patients choices: A pilot study of interactions in the seizure clinic. *Epilepsy & Behavior*. **20**(2), pp.312-320.
- Tringham, G.M. et al. 2011. Introduction of first trimester combined test increases uptake of Down's syndrome screening. *European Journal of Obstetrics & Gynecology and Reproductive Biology*. **159**(1), pp.95-98.
- Tsianakas, V. et al. 2012. Offering antenatal sickle cell and thalassaemia screening to pregnant women in primary care: a qualitative study of women's experiences and expectations of participation. *Health Expectations*. **15**(2), pp.115-125.
- Tsouroufli, M. 2011. Routinisation and constraints on informed choice in a one-stop clinic offering first trimester chromosomal antenatal screening for Down's syndrome. *Midwifery*. **27**(4), pp.431-436.
- Turner, D.W. 2010. Qualitative interview design: A practical guide for novice investigators. *The Qualitative Report*. **15**(3), pp.754-760.

- UK NSC. 2011. Model of Best Practice. Screening for Downs syndrome: UK NSC Policy recommendations 2011-2014. [Online]. [Accessed 16.11. 2012]. Available from: <http://fetalanomaly.screening.nhs.uk/standardsandpolicies>.
- UK NSC. 2012. *Annual report: Screening in England 2011-12*. National Screening Committee.
- UK Parliament. 1998. *Data Protection Act*. [Online]. [Accessed 31.01.2013]. Available from: <http://www.legislation.gov.uk/ukpga/1998/29/section/1/enacted>.
- Usta, I.M. et al. 2010. Effect of religion on the attitude of primiparous women toward genetic testing. *Prenatal Diagnosis*. **30**(3), pp.241-246.
- van den Berg, M. et al. 2005a. Accepting or declining the offer of prenatal screening for congenital defects: test uptake and women's reasons. *Prenatal Diagnosis*. **25**(1), pp.84-90.
- van den Berg, M. et al. 2007. Are counsellors' attitudes influencing pregnant women's attitudes and decisions on prenatal screening? *Prenatal Diagnosis*. **27**(6), pp.518-524.
- van den Berg, M. et al. 2008. Understanding Pregnant Women's Decision Making Concerning Prenatal Screening. *Health Psychology*. **27**(4), pp.430-437.
- van den Berg, M. et al. 2005b. Are pregnant women making informed choices about prenatal screening? *Genet Med*. **7**(5), pp.332-338.
- van den Heuvel, A. et al. 2009. Is informed choice in prenatal testing universally valued? A population-based survey in Europe and Asia. *BJOG: An International Journal of Obstetrics & Gynaecology*. **116**(7), pp.880-885.
- van den Heuvel, A. et al. 2010. Will the introduction of non-invasive prenatal diagnostic testing erode informed choices? An experimental study of health care professionals. *Patient Education and Counseling*. **78**(1), pp.24-28.
- van den Heuvel, A. et al. 2008. Informed choice in prenatal testing: a survey among obstetricians and gynaecologists in Europe and Asia. *Prenatal Diagnosis*. **28**(13), pp.1238-1244.
- van Teijlingen, E. and Hundley, V. 2002. The importance of pilot studies. *Nursing standard (Royal College of Nursing (Great Britain) : 1987)*. **16**(40), pp.19-25.
- van Teijlingen, E.R. et al. 2003. Maternity Satisfaction Studies and Their Limitations: "What Is, Must Still Be Best". *Birth*. **30**(2), pp.75-82.
- Vanstone, M. et al. 2012. Information-Sharing to Promote Informed Choice in Prenatal Screening in the Spirit of the SOGC Clinical Practice Guideline: A Proposal for an Alternative Model. *J Obstet Gynaecol Can*. **34**(3), pp.269-275.

- 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), pp.2041-51.
- Veal, W.R. 2002 Content Specific Vignettes as Tools for Research and Teaching. *Electronic Journal of Science Education*. **Volume 6**( 4 ).
- Veloski, J. et al. 2005. Clinical Vignette-Based Surveys: A Tool for Assessing Physician Practice Variation. *American Journal of Medical Quality*. **20**(3), pp.151-157.
- Wald, N. et al. 2003. First and second trimester antenatal screening for Down's syndrome: the results of the Serum, Urine and Ultrasound Screening Study (SURUSS). *Journal of medical Screening*. **10**(2), pp.56-87.
- Walker and Tarn. 2012. *Antenatal and Child Health Screening in Yorkshire and the Humber. Status Report 2011/2012*. NHS Yorkshire and the Humber.
- Walliman, N. 2006. *Social research methods*. Sage.
- Walsh, D. 1999. An ethnographic study of women's experience of partnership caseload midwifery practice: the professional as a friend. *Midwifery*. **15**(3), pp.165-176.
- Weed, M. 2005. " Meta Interpretation": A Method for the Interpretive Synthesis of Qualitative Research. In: *Forum Qualitative Sozialforschung/Forum: Qualitative Social Research*.
- WHO. 2007. *Fatherhood and health outcomes in Europe*. World Health Organization.
- Williams, C. et al. 2002a. Dilemmas encountered by health practitioners offering nuchal translucency screening: a qualitative case study. *Prenatal Diagnosis*. **22**(3), pp.216-220.
- Williams, C. et al. 2002b. Is nondirectiveness possible within the context of antenatal screening and testing? *Social Science & Medicine*. **54**(3), pp.339-347.
- Williams, C. et al. 2002c. Too many choices? Hospital and community staff reflect on the future of prenatal screening. *Social Science & Medicine*. **55**(5), pp.743-753.
- Williams, C. et al. 2005. Women as moral pioneers? Experiences of first trimester antenatal screening. *Social Science & Medicine*. **61**(9), pp.1983-1992.
- Williams, R.A. et al. 2011. Men's involvement in antenatal screening: A qualitative pilot study using e-mail. *Midwifery*. **27**(6), pp.861-866.
- Winquist, B. et al. 2008. Exploring physicians' views and values in relation to maternal serum screening. *Journal of obstetrics and gynaecology Canada : JOGC = Journal d'obstetrique et gynecologie du Canada : JOGC*. **30**(7), pp.564-572.

- Wirtz, V. et al. 2006. Patient–doctor decision-making about treatment within the consultation—A critical analysis of models. *Social Science & Medicine*. **62**(1), pp.116-124.
- Woods, M. 2007. Parental resistance: mobile and transitory discourses: a discursive analysis of parental resistance towards medical treatment for a seriously ill child: a thesis presented in fulfilment of the requirements for the degree of Doctor of Philosophy in Nursing, Massey University, Palmerston North, New Zealand.
- World Health Organization. 2006. *Medical genetic services in developing countries: The ethical, legal and social implications of genetic testing and screening*. [Online]. [Accessed 01.12.12]. Available from: <http://www.who.int/genomics/publications/GTS-MedicalGeneticServices-oct06.pdf>.
- Yin, R.K. 2009. *Case study research : design and methods*. 4th ed. Thousand oaks, CA: Sage.
- Zwaanswijk, M. et al. 2007. Young patients', parents', and survivors' communication preferences in paediatric oncology: Results of online focus groups. *BMC Pediatrics*. **7**(1), p35.

## Appendix 1: Literature review search

Search terms	Prenatal Diagnosis or antenatal screen* or antenatal test* or prenatal screen* or prenatal test* or down* syndrome or trisomy 21 or aneuploidy or uptake or up-take or take-up or "take up" or accept* or declin* or refus* or reject* or participat* or consent* or agree* or determinants or variables or emotions or factors or reasons or "decisional needs" or attitudes or "informed choice" or non-directiveness or nondirectiveness or "autonomous choice" or "external constraints" or "internal constraints" or "moral beliefs" or "moral justification" or ethics or "decision making" or utili*
<b>Database</b>	<b>Web of Science</b>
No of hits	257
Search terms	Prenatal Diagnosis or antenatal screen* or antenatal test* or prenatal screen* or prenatal test* or down* syndrome or trisomy 21 or uptake or up-take or take-up or "take up" or accept* or declin* or refus* or reject* or participat* or consent* or agree* or determinants or variables or emotions or factors or reasons or decisional needs or attitudes or informed choice or non-directiveness or nondirectiveness or autonomous choice or external constraints or internal constraints or moral beliefs or moral justification or ethics or decision making or utili#ation
<b>Databases</b>	<b>CDSR, ACP Journal Club, DARE, CCTR, CLCMR, Global Health, HMIC, MWIC</b>
No of hits	465
Search terms	(Prenatal Diagnosis or antenatal screen* or antenatal test* or prenatal screen* or prenatal test* ) AND ( down* syndrome or trisomy 21 or aneuploidy ) AND ( uptake or up-take or take-up or "take up" or accept* or declin* or refus* or reject* or participat* or consent* or agree* ) AND ( determinants or variables or emotions or factors or reasons or "decisional needs" or attitudes or "informed choice" or non-directiveness or nondirectiveness or "autonomous choice" or "external constraints" or "internal constraints" or "moral beliefs" or "moral justification" or ethics or "decision making" or utili* )
<b>Database</b>	<b>CINAHL</b>
Limiters	Limiters - English Language; Published Date: 19900101-20131231; Human; Language: English, search modes - Find all my search terms
No of hits	170. However, excluding the MEDLINE records produced a hit of 24 academic journals. The other databases searched included MEDLINE records.

Search terms	Antenatal and Screening
<b>Database</b>	<b>OpenSigle</b>
No of hits	16

## Appendix 2: Ethics approval letters



### NRES Committee Yorkshire & The Humber - Humber Bridge

Yorkshire and the Humber Research Ethics Office

First Floor  
Millside  
Mill Pond Lane  
Leeds  
LS6 4RA

Telephone: 0113 305012  
Facsimile: 0113 855619

09 January 2012

Mr Hyacinth O. Ukuhor  
School of Healthcare, Room 3.35,  
Baines Wing,  
University of Leeds, Leeds  
LS2 9JT

Dear Mr Ukuhor

**Study title:** PARTICIPATION IN PRENATAL SCREENING FOR  
DOWN'S SYNDROME: A MIXED METHODS  
MULTIPLE-CASE STUDY.

**REC reference:** 11/YH/0455

Thank you for your letter of 04 January 2012, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Vice-Chair.

#### Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

#### Ethical review of research sites

##### NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

##### Non-NHS sites

#### Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

*Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.*

*Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rforum.nhs.uk>.*

*Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.*

*For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.*

*Sponsors are not required to notify the Committee of approvals from host organisations*

**It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).**

#### **Approved documents**

The final list of documents reviewed and approved by the Committee is as follows:

Document	Version	Date
Advertisement		30 November 2011
Advertisement	1.0	30 November 2011
Advertisement	1.0	30 November 2011
Covering Letter		30 November 2011
Evidence of insurance or indemnity		30 November 2011
Investigator CV		30 November 2011
Letter from Statistician		30 November 2011
Letter of invitation to participant	1.0	30 November 2011
Other: CV - J Hirst (Supervisor)		30 November 2011
Other: CV - T Muncey (Supervisor)		30 November 2011
Other: CV - W Montelpare (Supervisor)		30 November 2011
Other: PhD Transfer Report		21 September 2011
Participant Consent Form: For partners of pregnant women	2.0	04 January 2012
Participant Consent Form: For pregnant women	2.0	04 January 2012
Participant Consent Form: For community midwives	2.0	04 January 2012
Participant Consent Form: For Antenatal screening coordinators	2.0	04 January 2012
Participant Information Sheet: For Antenatal screening coordinators	2.0	04 January 2012
Participant Information Sheet: For community midwives	2.0	04 January 2012
Participant Information Sheet: For pregnant women	2.0	04 January 2012
Participant Information Sheet: For partners of pregnant women	2.0	04 January 2012
Protocol	2.0	04 January 2012
Questionnaire: Topic guide for antenatal screening coordinators	1.0	30 November 2011
Questionnaire: Topic guide for community midwives	1.0	30 November 2011
Questionnaire: Topic guide for pregnant women and partners	1.0	30 November 2011
REC application	1.0	01 December 2011
Response to Request for Further Information		04 January 2012

#### **Statement of compliance**

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

#### **After ethical review**

##### **Reporting requirements**

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

#### **Feedback**

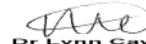
You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review

**11/YH/0455                          Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project

Yours sincerely

  
Dr Lynn Cawkwell  
Vice Chair

Email: nicola.mallender-ward@nhs.net

Enclosures: "After ethical review – guidance for researchers"

Copy to: Mrs Rachel E de Souza, University of Leeds  
Dr Michael Bramall, Barnsley Hospital NHS Foundation Trust

## Appendix 3: Research and Development approval letters



*Research and Development Directorate*  
email: michael.bramall@nhs.net

Research Governance Office  
Barnsley Hospital NHS Foundation Trust  
Tel: 01226 730000  
Extension: 2348  
Fax: 01226 208159

01 March 2012

Mr Hyacinth Ukuhor  
PhD student  
Baines Wing  
Postgraduate research suite 3.35  
School of Healthcare  
University of Leeds  
LS6 9JT

Dear Mr Ukuhor

**Study Title:** Participation in prenatal screening for Down's Syndrome: A mixed methods multiple-case study

**REC Ref:** 11/YH/0455

Thank you for submitting the above project for approval by Barnsley Hospital NHS Foundation Trust. The project was considered by the Research Governance Committee of Barnsley Health and Social Care Research and Development Alliance at a meeting and I am pleased to confirm that the committee agreed to approve the project.

The Documents received and approved were:

Document	Version	Date
Nhs R&D Form		
Protocol	2	04 January 2012
Investigator CV		30 December 2012
Participant information sheet – coordinators	2	04 January 2012
Participant information sheet – community midwives	2	04 January 2012
Participant information sheet – pregnant women	2	04 January 2012
Participant information sheet – partners of pregnant women	2	04 January 2012
Participant consent form – coordinators	2	04 January 2012
Participant consent form – community midwives	2	04 January 2012
Participant consent form – pregnant women	2	04 January 2012
Participant consent form – partners of pregnant women	2	04 January 2012
Letter of invitation – invitation to participate	1	30 November 2011
Evidence of Insurance		28 September 2011
Letter from statistician		05 December 2011
Interview Schedule – coordinators	1	30 November 2011
Interview Schedule – community midwives	1	30 November 2011
Interview Schedule – pregnant women and their partners	1	30 November 2011

Poster – coordinators	1	30 November 2011
Poster – community midwives	1	30 November 2011
Poster – pregnant women and their partners	1	30 November 2011
REC favourable opinion letter		09 January 2012
Nhs SSI Form	Barnsley	

In acting as Principal Investigator for Barnsley on this project, you must make yourself familiar with, observe and comply with:

- The informed consent and procedures approved by the Ethics Committee.
- The Department of Health Research Governance Framework and conduct your research in accordance with its principles.
- The Trust's Health and Safety policy.
- The Trust's individual reporting and reporting of adverse incident. In the event of an adverse incident the Ethics Committee and Research Governance Office must also be notified.
- The Trust's Equal Opportunities policy.
- The Trust's Information Security and Confidentiality policy.
- The Trust's Financial Regulations and procedures, if applicable.

You must also:

- Immediately notify the Ethics Committee and the Research Governance Office of any changes in protocol or new information that would raise questions about the continued conduct of the research.
- Ensure that all data and documentation is available for auditing purposes.

Basic information on the project will be entered into the Trust's research database and may be submitted to the Department of Health. The research office may seek further information from time to time in order to fulfil the information requirements of the Trust or NHS Executive.

I should be grateful if you could provide a brief annual report on the progress of the research to the Research Office, including reference to any publications that have arisen from the research. This report should be submitted during March each year, so that pertinent information can be included in the Trust's Annual Research Report.

Yours sincerely

A handwritten signature in black ink, appearing to read "Mike Vassell".

Professor S G Parker  
Director of Research & Development

Dear Mr Hyacinth O. Ukuhor

**Re: NHS Permission at LTHT for: PARTICIPATION IN PRENATAL SCREENING FOR DOWN'S SYNDROME: A MIXED METHODS MULTIPLE-CASE STUDY.**  
**LTHT R&D Number: OG11/10104**  
**EuDRCT:**  
**REC: 11/YH/0455**

I confirm that *NHS Permission for research* has been granted for this project at The Leeds Teaching Hospitals NHS Trust (LTHT). NHS Permission is granted based on the information provided in the documents listed below. All amendments (including changes to the research team) must be submitted in accordance with guidance in IRAS. Any change to the status of the project must be notified to the R&D Department.

Permission is granted on the understanding that the study is conducted in accordance with the *Research Governance Framework for Health and Social Care*, ICH GCP (if applicable) and NHS Trust policies and procedures available at [http://www.leedsth.nhs.uk/sites/research\\_and\\_development/](http://www.leedsth.nhs.uk/sites/research_and_development/).

This permission is granted only on the understanding that you comply with the requirements of the *Framework* as listed in the attached sheet "Conditions of Approval".

If you have any queries about this approval please do not hesitate to contact the R&D Department on telephone 0113 392 2878.

#### Indemnity Arrangements

The Leeds Teaching Hospitals NHS Trust participates in the NHS risk pooling scheme administered by the NHS Litigation Authority 'Clinical Negligence Scheme for NHS Trusts' for: (i) medical professional and/or medical malpractice liability; and (ii) general liability. NHS Indemnity for negligent harm is extended to researchers with an employment contract (substantive or honorary) with the Trust. The Trust only accepts liability for research activity that has been managerially approved by the R&D Department.

The Trust therefore accepts liability for the above research project and extends indemnity for negligent harm to cover you as investigator and the researchers listed on the Site Specific Information form. Should there be any changes to the research team please ensure that you inform the R&D Department and that s/he obtains an appropriate contract, or letter of access, with the Trust if required.

Yours sincerely



**Dr D R Norfolk**  
Associate Director of R&D

#### Approved documents

The documents reviewed and approved are listed as follows

Document	Version	Date of document
NHS R&D Form	3.4	02/12/2011
SSI Form	3.4	21/12/2011
Directorate Approval		12/12/2011
REC Letter confirming favourable opinion		09/01/2012
Protocol	2.0	04/01/2011
Patient information sheet: Partners of pregnant women (REC approved)	2.0	04/01/2012
Patient information sheet: Pregnant women (REC approved)	2.0	04/01/2012
Patient information sheet: Community midwives (REC approved)	2.0	04/01/2012
Patient information sheet: Antenatal screening (REC approved)	2.0	04/01/2012
Consent form: Partners of pregnant women (REC approved)	2.0	04/01/2012
Consent form: Pregnant women (REC approved)	2.0	04/01/2012
Consent form: Community midwives (REC approved)	2.0	04/01/2012
Consent form: Antenatal screening (REC approved)	2.0	04/01/2012
Advertisement: Pregnant women & partners	1.0	30/11/2011
Advertisement: Community midwives	1.0	30/11/2011
Advertisement: Antenatal screening	1.0	30/11/2011
Topic Guide: Pregnant women & partners	1.0	30/11/2011
Topic Guide: Community midwives	1.0	30/11/2011
Topic Guide: Antenatal screening	1.0	30/11/2011
Letter of invitation	1.0	30/11/2011
Letter from statistician		30/11/2011
Evidence of insurance		28/09/2011

**Conditions of NHS Permission for Research:**

- Permission from your Directorate must be obtained before starting the study.
- Favourable Opinion of the appropriate Research Ethics Committee, where necessary, must be obtained before starting the study.
- Arrangements must be made to ensure that all members of the research team, where applicable, have appropriate employment contracts or letter of agreement to carry out their work in the Trust.
- Agreements must be in place with appropriate support departments regarding the services required to undertake the project and arrangements must be in place to recompense them for the costs of their services.
- Arrangements must be in place for the management of financial and other resources provided for the study, including intellectual property arising from the work.
- Priority should be given at all times to the dignity, rights, safety and well being of participants in the study
- Healthcare staff should be suitably informed about the research their patients are taking part in and information specifically relevant to their care arising from the study should be communicated promptly.
- Each member of the research team must be qualified by education, training and experience to discharge his/her role in the study. Students and new researchers must have adequate supervision, support and training.
- The research must follow the protocol approved by the relevant research ethics committee. Any proposed amendments to or deviations from the protocol must be submitted for review by the Research Ethics Committee, the Research Sponsor, regulatory authority and any other appropriate body. The R&D Department should be informed where the amendment has resource implications within the Directorate and the Directorate research lead/clinical director notified.
- Adverse Events in clinical trials of investigational medicinal products must be reported in accordance with the Medicines for Human Use (Clinical Trials) Regulations 2004.
- Complete and return Study Status Reports, when requested, to the R&D Department within 28 days of receipt as requested. (NB Failure to comply to such request with the requirement will lead to suspension of NHS Permission.)
- Procedures should be in place to ensure collection of high quality, accurate data and the integrity and confidentiality of data during processing and storage.
- Arrangements must be made for the appropriate archiving of data when the research has finished. Records must normally be kept for 15 years.
- All data and documentation associated with the study must be available for audit at the request of the appropriate auditing authority. Projects are randomly selected for audit by the R&D Department. You will be informed by letter if your study is selected.
- Findings from the study should be disseminated promptly and fed back as agreed to research participants.
- Findings from the study should be exposed to critical review through accepted scientific and professional channels.
- All members of the research team must ensure that the process of informed consent adheres to the standards GCP outlined in the UK Clinical Trials Regulations. Investigators are directed to the R&D website for further information and training availability.
- Where applicable, this NHS Permission includes aspects of the study previously covered by the NRES Site Specific Assessment (SSA) process.
- Appropriate permissions must be in place for studies which are covered by the Human Tissue Act.
- Patient Information Sheet and Consent form must be on The Leeds Teaching Hospitals headed paper and include local contact details.

**Commercially Sponsored Trials**

If the study is commercially sponsored, NHS Permission is given subject to provision of the following documents.

- Clinical Trials Agreement - agreed and signed off by the R&D Department (on behalf of the Leeds Teaching Hospitals NHS Trust) and the Sponsor. Investigators do not have the authority to sign contract on behalf of the Trust.
- Indemnity agreement, if not included in the Clinical Trials Agreement- (standard ABPI no fault arrangements apply) signed by the R&D Department and the Sponsor

It is essential that all the responsibilities set out in the Research Governance Framework, including those outlined above are fulfilled. The Trust reserves the right to withdraw NHS Permission where the above criteria are not being met. The Trust will not accept liability for any activity where NHS Permission has not been granted.

## Appendix 4: Letters of access



### RECRUITMENT SERVICE

Ground Floor, Trust Headquarters, St James's Hospital, Beckett Street, Leeds, LS9 7TF

#### ***Private and Confidential***

Mr Onomegwonor Ukuhor  
19 Holborn Terrace  
Leeds  
LS6 2QA

Enquiries to: Jane Chim  
Direct Line: (0113) 2065982  
Our Ref: LOA/  
Date: 28th March 2012

Dear Mr O. Ukuhor

#### **Letter of Access for Research - Project Title - Participation in Prenatal Screening for Down's syndrome: A Mixed Methods Multiple Case Study**

This letter confirms your right of access to conduct research through The Leeds Teaching Hospitals NHS Trust for the purpose and on the terms and conditions set out below. This right of access commences on **28th March 2012** and ends on **31st July 2013** unless terminated earlier in accordance with the clauses below.

You have a right of access to conduct such research as confirmed in writing in the letter of permission for research from this NHS organisation. Please note that you cannot start the research until the Principal Investigator for the research project has received a letter from us giving permission to conduct the project.

The information supplied about your role in research at The Leeds Teaching Hospitals NHS Trust has been reviewed and you do not require an honorary research contract with this NHS organisation. We are satisfied that such pre-engagement checks as we consider necessary have been carried out.

You are considered to be a legal visitor to The Leeds Teaching Hospitals NHS Trust premises. You are not entitled to any form of payment or access to other benefits provided by this NHS organisation to employees and this letter does not give rise to any other relationship between you and this NHS organisation, in particular that of an employee.

While undertaking research through The Leeds Teaching Hospitals NHS Trust, you will remain accountable to your employer the University of Leeds but you are required to follow the reasonable instructions of **Colette Sarey** in this NHS organisation or those given on her/his behalf in relation to the terms of this right of access.

Where any third party claim is made, whether or not legal proceedings are issued, arising out of or in connection with your right of access, you are required to co-operate fully with any investigation by this NHS organisation in connection with any such claim and to give all such assistance as may reasonably be required regarding the conduct of any legal proceedings.

You must act in accordance with The Leeds Teaching Hospitals NHS Trust policies and procedures, which are available to you upon request, and the Research Governance Framework.

You are required to co-operate with The Leeds Teaching Hospitals NHS Trust in discharging its duties under the Health and Safety at Work etc Act 1974 and other health and safety legislation and to take reasonable care for the health and safety of yourself and others while on The Leeds Teaching Hospitals NHS Trust premises. You must observe the same standards of care and propriety in dealing with patients, staff, visitors, equipment and premises as is expected of any other contract holder and you must act appropriately, responsibly and professionally at all times.

You are required to ensure that all information regarding patients or staff remains secure and *strictly confidential* at all times. You must ensure that you understand and comply with the requirements of the NHS Confidentiality Code of Practice (<http://www.dh.gov.uk/assetRoot/04/06/92/54/04069254.pdf>) and the Data Protection Act 1998. Furthermore you should be aware that under the Act, unauthorised disclosure of information is an offence and such disclosures may lead to prosecution.

You should ensure that, where you are issued with an identity or security card, a bleep number, email or library account, keys or protective clothing, these are returned upon termination of this arrangement. Please also ensure that while on the premises you wear your ID badge at all times, or are able to prove your identity if challenged. Please note that this NHS organisation accepts no responsibility for damage to or loss of personal property.

We may terminate your right to attend at any time either by giving seven days' written notice to you or immediately without any notice if you are in breach of any of the terms or conditions described in this letter or if you commit any act that we reasonably consider to amount to serious misconduct or to be disruptive and/or prejudicial to the interests and/or business of this NHS organisation or if you are convicted of any criminal offence. Where required by law, your HEI employer will initiate your Independent Safeguarding Authority (ISA) registration, and thereafter, will continue to monitor your ISA registration status via the on-line ISA service. Should you cease to be ISA-registered, this letter of access is immediately terminated. Your employer will immediately withdraw you from undertaking this or any other regulated activity. You MUST stop undertaking any regulated activity.

Your substantive employer is responsible for your conduct during this research project and may in the circumstances described above instigate disciplinary action against you.

The Leeds Teaching Hospitals NHS Trust will not indemnify you against any liability incurred as a result of any breach of confidentiality or breach of the Data Protection Act 1998. Any breach of the Data Protection Act 1998 may result in legal action against you and/or your substantive employer.

If your current role or involvement in research changes, or any of the information provided in your Research Passport changes, you must inform your employer through their normal procedures. You must also inform your nominated manager in this NHS organisation.

Yours sincerely



Jane Chim  
Recruitment Service

cc: R&D office at Leeds Teaching Hospitals NHS Trust  
HR department of the substantive employer (and provider of honorary clinical contract, where applicable)



Gawber Road  
Barnsley  
S75 2EP

Direct line: 01226 432348

Tel: 01226 730000

Fax: 01226 202859

Minicom: 01226 321014

Our Ref: bhnft loa 073

1 March 2012

Hyacinth Ukuhor  
PhD student  
Baines Wing  
Postgraduate Research Suite 3.35  
School of Healthcare  
University of Leeds  
LS2 9JT.

Dear Hyacinth,

Letter of access for research

**Study Title: Participation in Prenatal Screening for Down's Syndrome**

This letter confirms your right of access to conduct the above research study through Barnsley Hospital NHS Foundation Trust (BHNFT) for the purpose and on the terms and conditions set out below. This right of access commences on 1 March 2012 and ends on 31 March 2013 unless terminated earlier in accordance with the clauses below.

You have a right of access to conduct such research as confirmed in writing in the letter of permission for research from this NHS organisation. Please note that you cannot start the research until the Principal Investigator for the research project has received a letter from us giving permission to conduct the project.

The information supplied about your role in research at BHNFT has been reviewed and you do not require an honorary research contract with this NHS organisation. We are satisfied that such pre-engagement checks as we consider necessary have been carried out.

You are considered to be a legal visitor to BHNFT premises. You are not entitled to any form of payment or access to other benefits provided by this NHS organisation to employees and this letter does not give rise to any other relationship between you and this NHS organisation, in particular that of an employee. While undertaking research at BHNFT you will remain accountable to the University of Leeds but you are required to follow the reasonable instructions of Sue Gibson (Head of Midwifery) in this NHS organisation or those given on their behalf in relation to the terms of this right of access.

Where any third party claim is made, whether or not legal proceedings are issued, arising out of or in connection with your right of access, you are required to co-operate fully with any investigation by this NHS organisation in connection with any such claim and to give all such assistance as may reasonably be required regarding the conduct of any legal proceedings.

You must act in accordance with BHNFT policies and procedures, which are available to you upon request, and the Research Governance Framework.

You are required to co-operate with BHNFT in discharging its duties under the Health and Safety at Work etc Act 1974 and other health and safety legislation and to take reasonable care for the health and safety of yourself and others while on BHNFT premises. You must observe the same standards of care and propriety in dealing with patients, staff, visitors, equipment and premises as is expected of any other contract holder and you must act appropriately, responsibly and professionally at all times.

You are required to ensure that all information regarding patients or staff remains secure and *strictly confidential* at all times. You must ensure that you understand and comply with the requirements of the NHS Confidentiality Code of Practice (<http://www.dh.gov.uk/assetRoot/04/06/92/54/04069254.pdf>) and the Data Protection Act 1998. Furthermore you should be aware that under the Act, unauthorised disclosure of information is an offence and such disclosures may lead to prosecution.

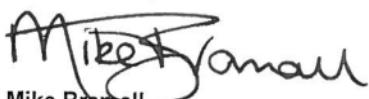
You should ensure that, where you are issued with an identity or security card, a bleep number, email or library account, keys or protective clothing, these are returned upon termination of this arrangement. Please also ensure that while on the premises you wear your ID badge at all times, or are able to prove your identity if challenged. Please note that this NHS organisation accepts no responsibility for damage to or loss of personal property.

We may terminate your right to attend at any time either by giving seven days' written notice to you or immediately without any notice if you are in breach of any of the terms or conditions described in this letter or if you commit any act that we reasonably consider to amount to serious misconduct or to be disruptive and/or prejudicial to the interests and/or business of this NHS organisation or if you are convicted of any criminal offence. Your substantive employer is responsible for your conduct during this research project and may in the circumstances described above instigate disciplinary action against you.

BHNFT will not indemnify you against any liability incurred as a result of any breach of confidentiality or breach of the Data Protection Act 1998. Any breach of the Data Protection Act 1998 may result in legal action against you and/or your substantive employer.

If your current role or involvement in research changes, or any of the information provided in your research passport changes, you must inform the University of Sheffield through their normal procedures. You must also inform the Research Governance Office in this NHS organisation.

Yours sincerely

  
Mike Bramall  
Research Governance Officer

## Appendix 5: Information about research for antenatal coordinators, community midwives, pregnant women and partners.

Version 2.0. 4th January 2012. Rec ref. 11/YH/0455

School of Healthcare



### Information about the research.

#### For antenatal screening coordinators.

#### PARTICIPATION IN PRENATAL SCREENING FOR DOWN'S SYNDROME.

You are being invited to take part in this study. Before making your decision please read the information below carefully and discuss it with others as you deem fit. You could also contact me at anytime for further information. Thank you for reading this.

**What is the research purpose?** The purpose of the study is to explore the influence of service organisation and delivery as offered by community midwives and antenatal screening coordinators on women and partners' participation in prenatal Down's syndrome screening. The study is being undertaken as part of a PhD training programme.

**Why have I been chosen?** You are being invited to participate in this study because you are an antenatal screening coordinator. You have either responded to an invitation by letter or advertisement or by word of mouth.

**Do I have to take part?** You do not have to take part as participation in this study is entirely voluntary. If you decide to take part please, register at the website: <http://matser.leeds.ac.uk/index.php>. This requires access to the internet, but we would be glad to introduce you to the library internet services if you do not have access to the internet. Demographic details will be requested during registration. This information sheet is yours to keep and is also available on the website, as is a copy of the consent form which you should read and agree to if you decides to participate in this study. You can also save or print a copy of the consent form from the website for your records. We explicitly seek your consent for the use of direct quotations which will be anonymised. If you take part and change your mind, you have the option of withdrawing from the study at any point without giving any reasons. You could withdraw from the study using the

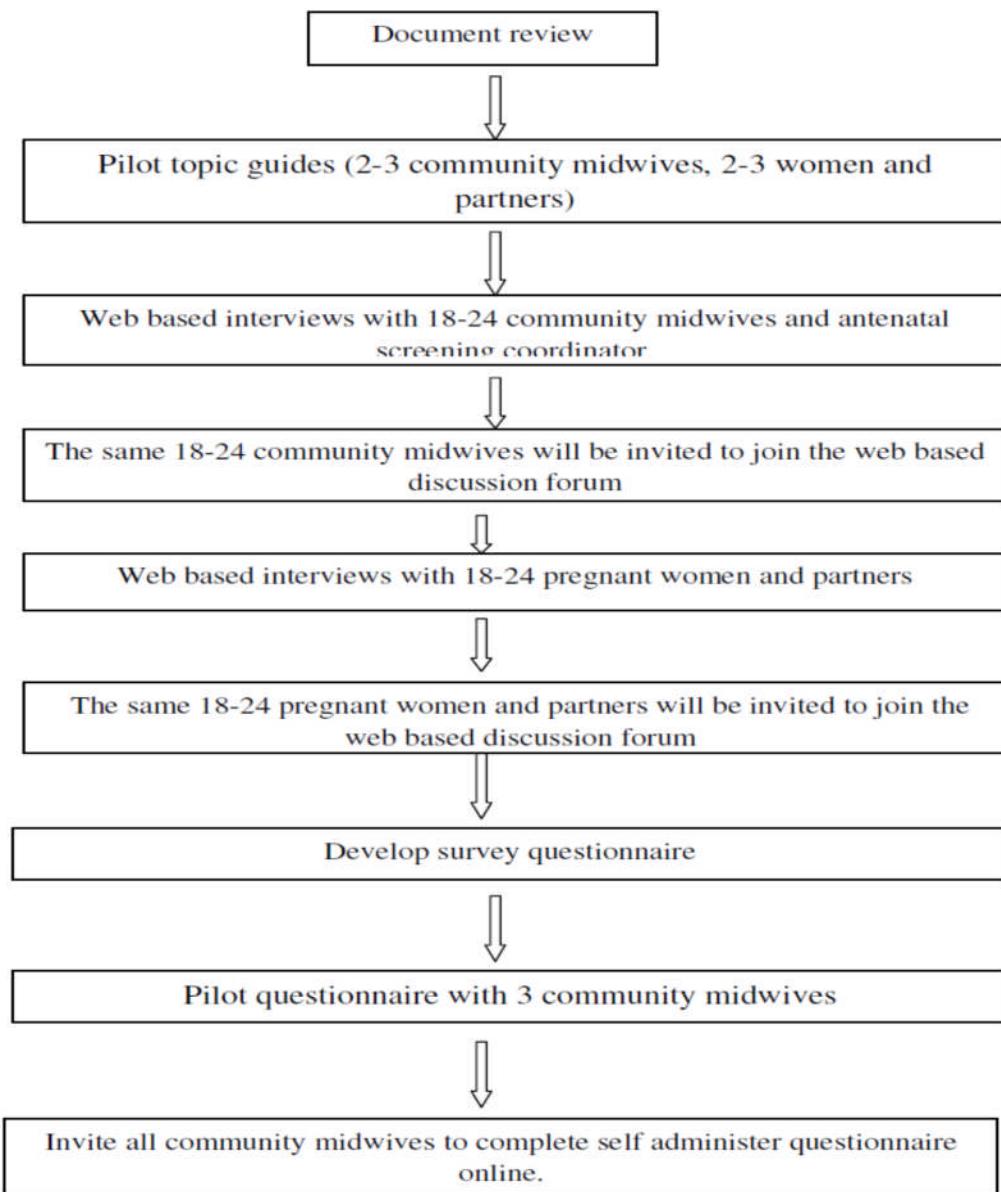
withdrawal link or button on the website. If you withdraw we seek your consent to retain and use any data (anonymously, using a personal identity number or pseudonym) which you have already given.

**What will happen to me if I take part?** You will be invited to access and complete a web based individual interview lasting 30-60 minutes depending on your views. Questions will relate to the way in which you contribute to the delivery of the prenatal Down's syndrome screening programmes in your area and your thoughts about how your influence service delivery. You would need to have access to the internet and an email account which you could create for this study. The webpage for this interview will be accessible for the duration of two weeks and you will be invited to complete the interview within this period. An email notifying you when the questions are posted and your username and password to login to the website will be sent to you once you have registered and completed the consent form.

**What are the possible disadvantages and risk of taking part?** There are no reasonable foreseeable discomforts or risks involved in participating as you could respond to the web based interview at anytime and place. If there is any unexpected discomfort, disadvantage or risk to you during the course of this research, please, bring it to my attention or use the link for support provided in the study website. The study website will be protected, monitored and back up by the University of Leeds information systems services.

**What if something goes wrong?** If you are harmed by taking part in this research, there are no special compensation arrangements. If the harm to you is due to someone's negligence, then you may have grounds for legal action but you may have to pay for it. Regardless of this, you may wish to complain or have any concerns about any aspect of the way you have been approached or treated during the course of this study, you may complain to the University secretary. The normal NHS complaints mechanisms will still be available to you. If you wish to make a complaint, then please use the link on the study website or contact the chief investigator on how to proceed.

### STUDY FLOW DIAGRAM FOR EACH LOCATION



**What are the possible benefits of taking part?** There are no immediate benefits for those participating in the study, but the study will contribute to the knowledge base and inform policy. You may gain some personal benefits from writing about your views.

**Will my participation be kept confidential?** Yes, all information collected will be kept strictly confidential. You will be assigned a personal identification number (ID) or pseudonyms to identify

your responses. Names will be removed from responses during the analysis of the data and replaced with pseudonyms. During the study the chief investigator and academic supervisors will have access to your personal data. Your ID and personal details will be stored in a password-protected computer. Your anonymised responses will not be held together with your personal details. Data will be stored in the University of Leeds (chief investigator) firewall protected secure and backed up server that is only accessible via password for security and safety. After finishing this study the data will be stored in password protected computer of the lead supervisor (Dr Janet Hirst) for 3 years and then destroyed according to the University policy on data protection.

**What type of information will be sought and why the collection of this information?** The information we need will be detailed responses to the semi-structured interview questions that will be posted on the website. This will enable the overall aim of the study to be achieved i.e. explore the influence of the service organisation and delivery as offered by antenatal screening coordinators and community midwives on women and their partners decision to participate in prenatal Down's syndrome screening.

**What will happen to the results of the research project?** A summary of the findings will be sent to all participants electronically and the Foetal Anomaly Screening Programme. Findings will be published in academic journals and presented at academic and professional conferences.

**Who is organising and funding the research?** The study is sponsored by the University of Leeds as part of a PhD training programme. No application for external funding has been or will be made.

**Contact information for further information?** If you have any further questions or if you experience difficulties accessing the website for the study, please do not hesitate to contact me:

Hyacinth Ukuhor  
PhD Student  
School of Healthcare,  
University of Leeds  
Leeds,  
LS2 9JT  
Phone: 01133433531  
E-mail: [hchou@leeds.ac.uk](mailto:hchou@leeds.ac.uk)



**Information about the research.**

**For community midwives.**

**PARTICIPATION IN PRENATAL SCREENING FOR DOWN'S SYNDROME.**

You are being invited to take part in this study. Before making your decision please read the information below carefully and discuss it with others as you deem fit. You could also contact me at anytime for further information. Thank you for reading this.

**What is the research purpose?** The purpose of the study is to explore the influence of service organisation and delivery as offered by community midwives and antenatal screening coordinators on women and partners' participation in prenatal Down's syndrome screening. The study is being undertaken as part of a PhD training programme.

**Why have I been chosen?** You have been chosen because you are a community midwife involved in prenatal Down's syndrome screening. You have either responded to an invitation by letter or advertisement or by word of mouth.

**Do I have to take part?** You do not have to take part as participation in this study is entirely voluntary. If you decide to take part please, register at the website: <http://matserv.leeds.ac.uk/index.php>. This requires access to the internet, but we would be glad to introduce you to the library internet services if you do not have access to the internet. Demographic details will be requested during registration. This information sheet is yours to keep and is also available on the website, as is a copy of the consent form which you should read and agree to if you decides to participate in this study. You can save or print a copy of the consent form from the website for your records. We explicitly seek your consent for the use of direct quotations which will be anonymised. If you take part and change your mind, you have the option of withdrawing from the study at any point without giving any reasons. You could withdraw from the study using the withdrawal link or button on the website. If you withdraw we seek your consent to retain and use any data (anonymously, using a personal identity number or pseudonym) which you have already given.

**What will happen to me if I take part?** You will be invited to participate in both phases of the study although you can take part in the first phase and or the second phase as you wish. You will need to have access to the internet and an email account.

Firstly, will invite you to access and complete a web based individual interview lasting 30-60 minutes depending on your views. Questions will relate to the way in which you contribute to the delivery of the prenatal Down's syndrome screening programmes in your area and your thoughts about how midwives influence service delivery. You would need to have access to the internet and an email account which you could create for this study. The web page for this interview will be accessible for the duration of two weeks and you will be invited to complete the interview within this period. An email notifying you when the questions are posted and your username and password to login to the website will be sent to you once you have registered and completed the consent form. This will then be followed by a web based discussion forum where the same midwives that took part in the interviews will be invited to participate. Questions from the researcher and comments from the other community midwives will be posted on the forum and you will be invited to respond to them with as much details as you think appropriate and at any time and post comments as in a discussion forum. The question and prompts from the chief investigator who will moderate the forum will be posted weekly. An email notifying you when the questions are posted will be sent to you. The web based discussion forum will be available for 1-2 months and you can access it from time to time. To avoid the study being a burden, you should spend no more than 30 minutes on each of the questions posted weekly. A survey questionnaire will be developed from the findings of the interviews and discussion forum for the purpose of exploring ways in which service organisation and delivery influence participation in prenatal screening for Down's syndrome more broadly in England.

#### Phase II.

All the community midwives in your trust will be invited to complete the survey questionnaire online as a means to access its validity. Hard copies of the questionnaire may be sent to all community midwives by post if the response to the online survey is limited.

**What are the possible disadvantages and risk of taking part?** There are no reasonable foreseeable discomforts or risks involved in participating as you could respond to the web based interview and discussion forum at any time and place. The website will be monitored and back up by the University of Leeds information systems services. If there is any unexpected discomfort, disadvantage or risk to you during the course of this research, please, bring it to my (chief investigator) attention to help you find support or use the link for support on the study website.

**What if something goes wrong?** If you are harmed by taking part in this research, there are no special compensation arrangements. If the harm to you is due to someone's negligence, then you may have grounds for legal action but you may have to pay for it. Regardless of this, you may wish to complain or have any concerns about any aspect of the way you have been approached or treated during the course of this study, you may complain to the University secretary. The normal NHS complaints mechanisms will still be available to you. If you wish to make a complaint, then please contact the chief investigator on how to proceed.

**What are the possible benefits of taking part?** There are no immediate benefits for those participating in the study, but the study will contribute to the knowledge base and inform policy. You may gain some personal benefits or support from writing or discussing your views with other participants.

**Will my participation be kept confidential?** Yes, all information collected will be kept strictly confidential. You will be assigned a personal identification number (ID) or pseudonyms to identify

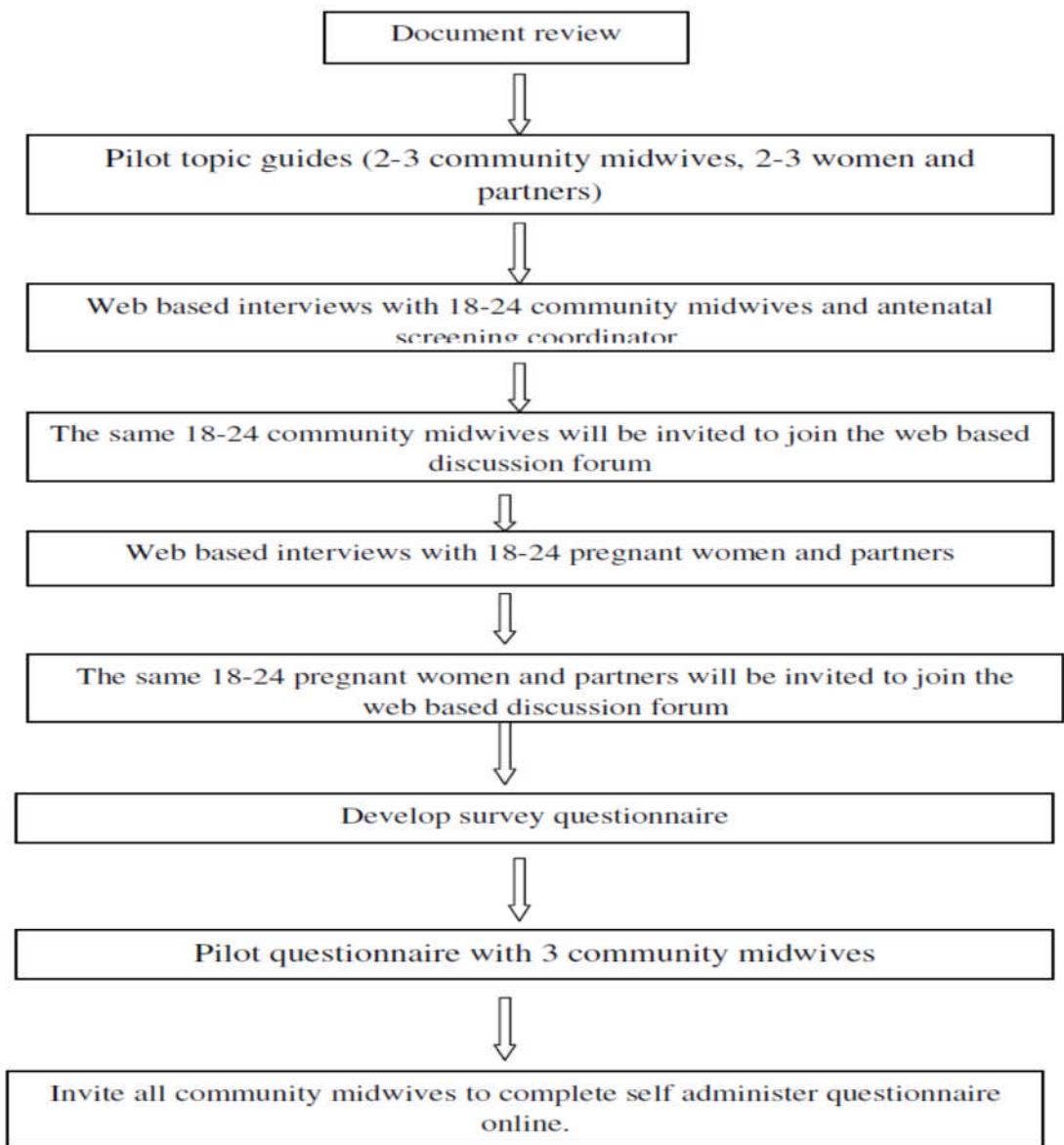
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**Will my participation be kept confidential?** Yes, all information collected will be kept strictly confidential. You will be assigned a personal identification number (ID) or pseudonyms to identify your responses. Names will be removed from responses during the analysis of the data and replaced with pseudonyms. During the study the chief investigator and academic supervisors will have access to your personal data. Your ID and personal details will be stored in a password-protected computer. Your anonymised responses will not be held together with your personal details. Data will be stored on the University of Leeds (chief investigator) firewall protected secure server that is only accessible via password for security and safety. After finishing this study the data will be stored in password protected computer

of the lead supervisor (Dr Janet Hirst) for 3 years and then destroyed according to the University policy on data protection.

#### STUDY FLOW DIAGRAM FOR EACH LOCATION



**What type of information will be sought and why the collection of this information?** The information we need will be detailed responses to the semi-structured interview questions and your comments and that of other participants that will be posted on the discussion

forum. This will enable the overall aim of the study to be achieved i.e. explore the influence of the service organisation and delivery as offered by community midwives on women and their partners decision to participate in prenatal Down's syndrome screening.

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E-mail: [hchou@leeds.ac.uk](mailto:hchou@leeds.ac.uk)

School of Healthcare

UNIVERSITY OF LEEDS



**Information about the research.**

**For pregnant women.**

**PARTICIPATION IN PRENATAL SCREENING FOR DOWN'S SYNDROME.**

You are being invited to take part in this study. Before making your decision please read the information below carefully and discuss it with others as you deem fit. You could also contact me at anytime for further information. Thank you for reading this.

**What is the research purpose?** The purpose of the study is to explore the influence of service organisation and delivery as offered by community midwives and antenatal screening coordinators on women and partners' participation in prenatal Down's syndrome screening. The study is being undertaken as part of a PhD training programme.

**Why have I been chosen?** You have been chosen because you are a pregnant woman that accepted or declined prenatal Down's syndrome screening. You are 16 years or over. You have either responded to an invitation by letter or advertisement or by word of mouth.

**Do I have to take part?** You do not have to take part as participation in this study is entirely voluntary. If you decide to take part please, register at the website: <http://matserv.leeds.ac.uk/index.php>. This requires access to the internet, but we would be glad to introduce you to the library internet services if you do not have access to the internet. Demographic details will be requested during registration. This information sheet is yours to keep and is also available on the website, as is a copy of the consent form which you should read and agree to if you decides to participate in this study. You can save or print a copy of the consent form from the website for your records. We explicitly seek your consent for the use of direct quotations which will be anonymised. If you take part and change your mind, you have the option of withdrawing from the study at any point without giving any reasons. You could withdraw from the study using the withdrawal link or button on the website. If you withdraw

we seek your consent to retain and use any data (anonymously, using a personal identity number or pseudonym) which you have already given.

**What will happen to me if I take part?** Firstly, we will invite you to access and complete a web based individual interview lasting 30-60 minutes depending on your views. Questions will relate to the way in which prenatal screening for Down's syndrome was first introduced to you and about the information you received and how this may have helped you make a decision to have the screening. You would need to have access to the internet and an email account which you could create for this study. You will be able to access the web based interview during a two week period. An email notifying you when the questions are posted and your username and password to login to the website will be sent to you once you have registered and completed the consent form. After this, you will be invited to take part in a web based discussion forum where other pregnant women that took part in the interviews will be invited to participate. Questions about if the maternity service influenced your decision about having the screening for Down's syndrome, or not, and how it happened and comments from other pregnant women will be posted on the forum and you will be invited to respond to them with as much details as you think appropriate at any time and post comments also as in a discussion forum. The question and prompts from the chief investigator who will moderate the forum will be posted weekly. An email notifying you when the questions are posted will be sent to you. The web based discussion forum will be available for 1-2 months and you can access it from time to time. To avoid the study being a burden, you should spend no more than 30 minutes on each of the questions posted weekly.

**What are the possible disadvantages and risk of taking part?** The subject area is sometimes uncomfortable for some people so it is important to remember that the information we are seeking is about how the maternity service influenced how you made your decision to have this type of screening or not. It is not important to this study whether your unborn baby was screened or not and we will not ask you. If there is any unexpected discomfort, disadvantage or risk to you during the course of this research, please, bring it to my (chief investigator) attention to help you find support or use the link for support on the study website.

**What if something goes wrong?** If you are harmed by taking part in this research, there are no special compensation arrangements. If the harm to you is due to someone's negligence, then you may have grounds for legal action but you may have to pay for it. Regardless of this, you may wish to complain or have any concerns about any aspect of the way you have been approached or treated during the course of this study, you may complain to the University

secretary. The normal NHS complaints mechanisms will still be available to you. If you wish to make a complaint, then please contact the chief investigator on how to proceed.

**What are the possible benefits of taking part?** There are no immediate benefits for those participating in the study, but the study will contribute to the knowledge base and inform policy. You may gain some personal benefits or support from writing or discussing your views with other participants.

**Will my participation be kept confidential?** Yes, all information collected will be kept strictly confidential. You will be assigned a personal identification number (ID) or pseudonyms to identify your responses. Names will be removed from responses during the analysis of the data and replaced with pseudonyms. During the study the chief investigator and academic supervisors will have access to your personal data. Your ID and personal details will be stored in a password-protected computer. Your anonymised responses will not be held together with your personal details. Data will be stored on the University of Leeds (chief investigator) firewall protected secure server that is only accessible via password for security and safety. After finishing this study the data will be stored in password protected computer of the lead supervisor (Dr Janet Hirst) for 3 years and then destroyed according to the University policy on data protection.

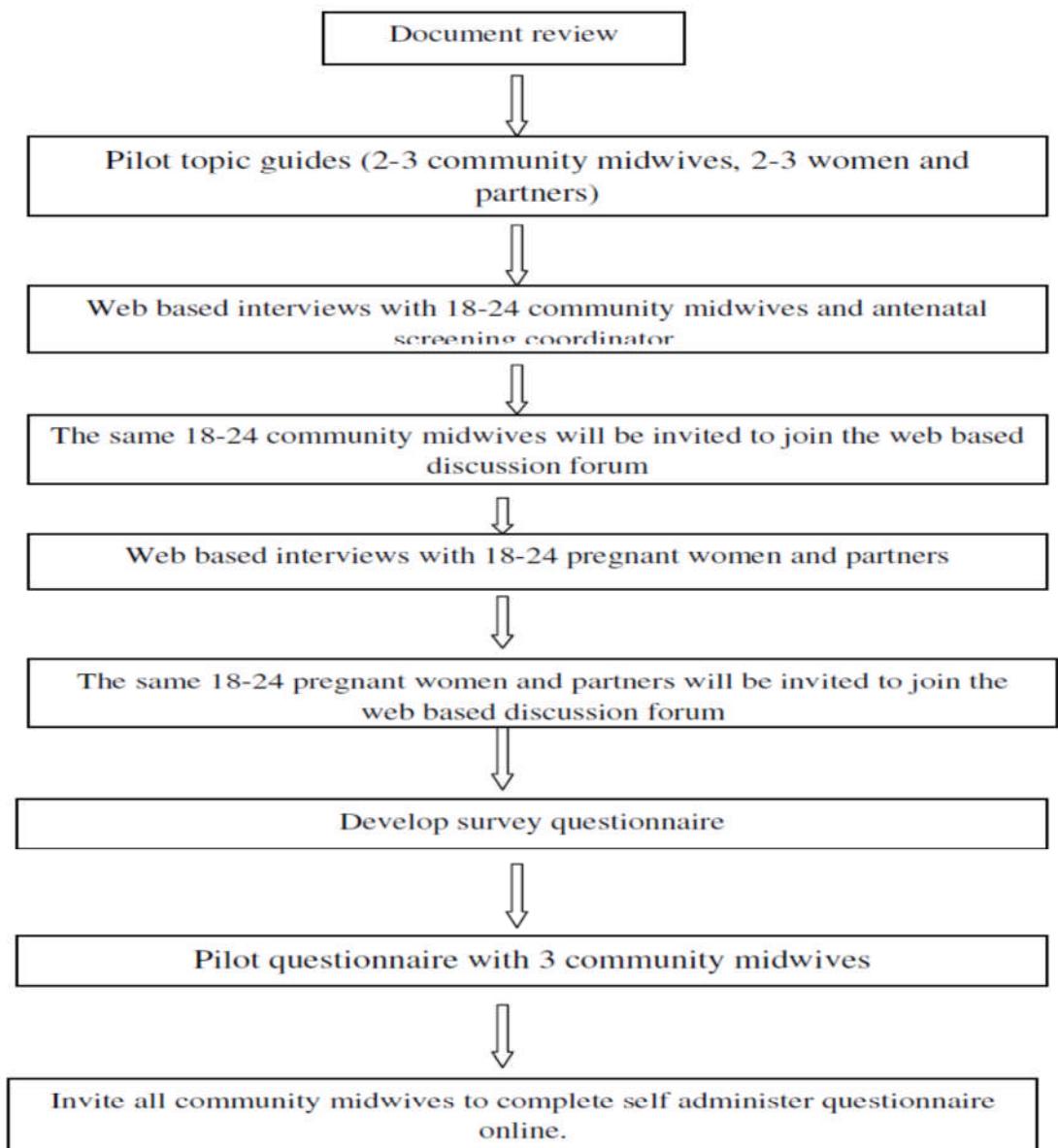
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**Information about the research.**

**For partners of pregnant women.**

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**Why have I been chosen?** You have been chosen because you are a partner of a pregnant woman that accepted or declined prenatal Down's syndrome screening. You are 16 years or over. You have either responded to an invitation by letter or advertisement or by word of mouth.

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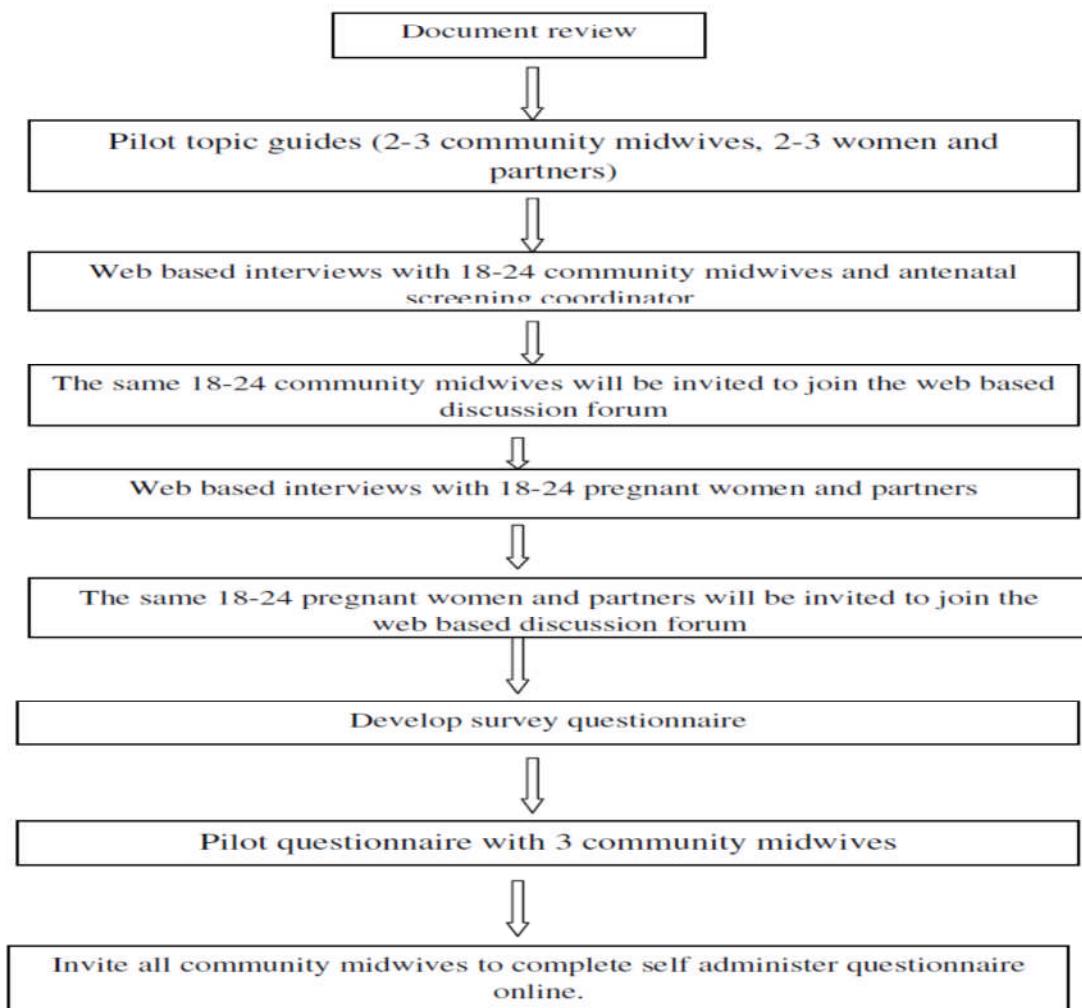
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STUDY FLOW DIAGRAM FOR EACH LOCATION



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PhD Student  
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Phone: 01133433531  
E-mail: [hchou@leeds.ac.uk](mailto:hchou@leeds.ac.uk)

## Appendix 6: Consent forms for screening coordinators midwives, pregnant women and partners

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### CONSENT FORM

Name of chief investigator: Hyacinth Ukuhor

Contact phone number: 01133433531

Rec reference number: 11/YH/0455

**Please, tick the boxes if you agree to the consent form**

**Please tick**

		Plese tick
1	I confirm that I have read, understood and kept a copy of information about the research version 2.0, dated 04.01.2012.	
2	I also confirm that I have had the opportunity to ask questions, discuss the study and when I have done so I have received satisfactory answers.	
3	I understand that I have the option of withdrawing my participation at any point and without any reason and my medical or legal rights will not be affected. I give consent for any data already given to be retained and used.	
4	I understand that my personal details and any other information I provide will be kept confidential, stored securely for 3 years.	
5	I agree not to share my answers or responses to questions on the interview or discussion forum with my friends or colleagues.	
6	I understand that any information I give may be included in published reports and will be anonymised.	
7	I give permission for my anonymised responses to be accessed by the researchers involved in this study.	
8	I understand that relevant sections of my data collected during the study, may be looked at by individuals from the University of Leeds, from regulatory authorities or the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my data.	
9	I understand that I will not benefit financially if this research leads to the development of a new service.	
10	I know how to contact the chief investigator if I need to.	

I agree to participate in this study

## **Appendix 7: Letter of invitation to community midwives**

Dear Participant,

### **Invitation to participate in research on the influence of service organisation and delivery on participation in prenatal Down's syndrome screening.**

I am a PhD student at the University of Leeds and my research aims to explore the experiences of community midwives and antenatal coordinators with first trimester Down's syndrome screening and the influence of healthcare settings on pregnant women and their partners' decision to participate.

As part of this study, I also want to explore pregnant women and partners perspectives of how service organisation and delivery has shaped their decision about prenatal Down's syndrome screening.

I understand that you are a community midwife or antenatal screening coordinator, which is why you are receiving this letter which has been distributed by the head of midwifery or supervisor of midwives on my behalf.

The enclosed information gives details of the research study and will hopefully answer any questions you have. Please, contact me if you like more information or if you are not clear with any aspect of this study. If you are interested in taking part, please register for this study at <http://matserv.leeds.ac.uk/index.php> or by email hchou@leeds.ac.uk. I will then get in touch with you to discuss things further.

Your participation remain confidential whether you decide to take part or not and no information which could identify you will be shared with your trust or be published.

Thank you for taking the time to read this letter and I look forward to hearing from you.

Yours sincerely,

Hyacinth Ukuhor.

**Please let me know if you would like this material in larger print or electronic format.**

**Appendix 8: Scenarios and open-ended questions for community midwives, pregnant women and partners**

**Scenarios for community midwives**

The following is a short research scenario concerning Down's syndrome Screening Methods. Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

**Scenario 1**

The community midwife provides information to pregnant women and their partners (when present) during booking appointments. Information sessions are usually conducted in the community antenatal clinic. A typical appointment can take between 40 - 60 minutes and includes confirming the women's medical and reproductive history, providing information about the pregnancy care pathways, dating scan, and routine blood tests for such outcomes as measuring rhesus antibodies and the detection of sexually transmitted diseases. In addition, the community midwife usually spends about 5 minutes (being the time allocated) giving information and offering Down's syndrome screening to women. The community midwife concludes the session by providing standard background information, in the form of brochures on antenatal screening tests, which she encourages the pregnant woman to take home.

a) Given the scenario described above, can you tell me what it has been like in your role as a community midwife that offers Down's syndrome screening to pregnant women and their partners, when present?

For example, can you describe your experience with the process? Consider such influences as the amount of time allocated within the visit, your volume and type of caseload, and the quantity of information that you are required to impart to the pregnant woman (and her partner).

b) Please describe your perception of the pregnant woman's responses (uptake) of the information. Do such responses, in your experience, influence what you say, and how you speak to women about Down's syndrome screening?

c) Do you have either specific or general concerns about the delivery of the Down's syndrome screening program?

Do such concerns influence what you say, and how you speak to women about Down's syndrome screening? (Please expand your responses)

---

The following is a short research scenario concerning Down's syndrome Screening Methods. Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

#### Scenario 2

During the booking visit, the Community Midwife offers the pregnant woman a range of routine blood tests which include tests for anaemia, blood grouping, and dating ultrasound scan. The Community Midwife informs the pregnant woman that the tests and information are designed to help make their pregnancy safer and assess the wellbeing of both mother and baby. In addition, the maternal blood pressure is checked, urine tested and the height and weight are measured. During this visit, the Community Midwife offers advice on diet and lifestyle. Likewise, the Community Midwife asks about previous history of physical illness or mental health issues, as well as domestic violence or sexual abuse. This round of questioning is followed by providing additional information about expected events during pregnancy and the offer to participate in Down's syndrome screening. The Community Midwife then concludes the session by providing standard background information, in the form of brochures on antenatal screening tests, which she encourages the pregnant woman to take home.

- 
- a) Considering the format that is used by the Community Midwife to deliver the various messages and information, do you think that the approach in and of itself has either a direct or an indirect influence on the pregnant mother's decision to participate in the Down's syndrome screening programme?
  - b) Do you think that by combining the presentation of information about the Down's syndrome Screening programme with routine process of blood sampling has a direct or indirect impact on the pregnant woman's interest and/or understanding of the information? In other words, does combining the presentation of routine testing with information delivery influence the decision making of the pregnant woman about the Down's syndrome screening programme?
  - c) In a typical visit of this type, the Community Midwife provides information about Down's syndrome screening to women with minimal reference to possible implications of participating in such a screening programme. To what extent do you think this approach has

either a positive or negative influence on the pregnant woman's decision about whether or not to participate in this type of screening programme?

---

The following is a short research scenario concerning Down's syndrome Screening Methods Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

**Scenario 3**

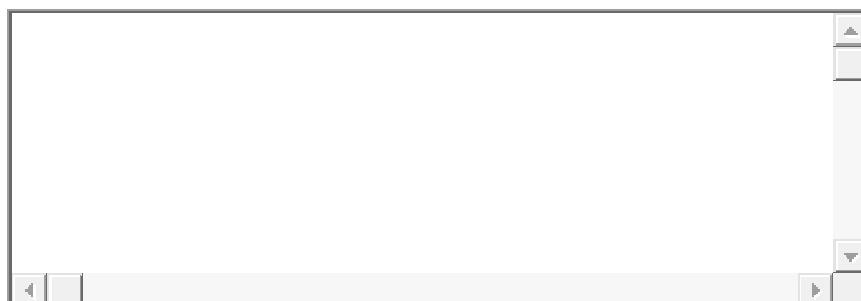
Often during the booking visit, a Community Midwife team leader will work with her team of community midwives to provide information and offer antenatal screening tests including Down's syndrome screening to pregnant women and their partners (when present). In some situations, the Community Midwife team leader will observe that the pregnant woman will hold negative views about Down's syndrome screening and subsequently decline the offer of screening.

It has been suggested that the pregnant woman's decision to decline participation in the screening programme can be attributed to the midwife using a directive approach when informing women about Down's syndrome screening.</P< font>

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Do you think that the community midwife could influence the pregnant woman's decisions about participating in Down's syndrome screening? For example by directly recommending it, by their body language, or by their tone of speech or the selective words of bias speech.

If you do, can you explain how this might happen?



Click to Submit AND ADVANCE TO NEXT PAGE

The following is a short research scenario concerning Down's syndrome Screening Methods  
Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

**Scenario 4**

The photographs below show the consulting room where the Community Midwife conducts booking appointments. During an appointment, the pregnant women and their partners are given information about prenatal Down's syndrome screening plus other information. The midwife always wears the uniform for community midwives and usually sits on a swivel chair in front of her desk. The pregnant woman sits on a chair that is placed at one end of the desk in a position where she could face the midwife. Her partner (when present) also sits on a chair besides her facing the midwife. The consulting room has a design and a décor that is found in most community antenatal clinics and is brightly lit with day light streaming through the window and a fluorescent lamp hanging from the ceiling. The room has a patient examination couch, medical examination lamps, items for measuring height and weight plus other medical equipment.

---

From this scenario and looking at the photographs below, do you think the clinical environment or any aspect of it such as the lighting in the room, room décor, medical equipment, presence of the midwife or the entire clinical setting could influence the way women and partners decide about participating in prenatal Down's syndrome screening?

If you do, can you explain in what way the clinical environment or any aspect of it could influence women's decisions to accept or decline prenatal Down's syndrome screening?



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### **Scenarios for pregnant women**

The following is a short research scenario concerning Down's syndrome Screening Methods  
Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

#### **Scenario 1**

A pregnant woman was seen by a midwife during her booking appointment at the local health centre. The midwife gave her lots of advice, such as the types of food to eat when pregnant. The woman's weight, height and blood pressure were then measured and the midwife told her about routine blood tests and the ultrasound scans. The woman agreed to have the blood tests and scans. It took another 5 minutes for the midwife to tell the woman about the screening test for Down's syndrome. At the end of the booking visit the midwife gave the woman a booklet about antenatal screening tests to take home and read.

- a) Can you explain what it was like for you when you were given information about Down's syndrome screening during the booking appointment? For example, how it made you feel about the likelihood of you having a baby with Down's syndrome.
- b) Please describe your experience with the information you received about Down's syndrome screening? For example, were you told about screening by family or friends, or had you read about the screening process prior to your visit by searching various sources like the internet? In general, were you comfortable with the amount of information that was provided to you during the booking visit?

- c) Please explain how you felt during your visit when you were talking to the midwife about Downs' syndrome screening. For example, during your visit had you any thought the screening would be helpful for you? Likewise, during the visit did you feel that the screening process would be stressful, especially when considering possible test results or the diagnostic procedures that were suggested, or the choice to terminate the pregnancy?
- d) To what extent did your experiences during the booking visit influence your decision to have Down's syndrome screening? Please explain your responses with specific examples.

The following is a short research scenario concerning Down's Syndrome Screening Methods. Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

#### Scenario 2

At the booking visit, the midwife explained a number of routine blood tests. Included in these blood tests was an explanation about the screening test for Down's syndrome.

The woman was told that the tests were designed to help make sure the mother and baby were healthy.

In addition to information about various blood tests, information about participating in daily activities, such as exercise was also presented. The visit concluded with a standard measurement of height, weight and blood pressure. Finally, as the pregnant woman was leaving, the midwife passed her an information booklet about antenatal tests and asked her to please take the booklet home to read.

- 
- a) Thinking about your booking visit, did having the midwife explain Down's syndrome screening, as well as other tests, influence your decision about screening for Down's syndrome?
  - b) Again, during your booking visit, did you feel that the information presented to you had an influence on your decision about screening? For example, did your midwife discuss issues, such as potential benefits or known risks of Down's syndrome screening? Did you feel comfortable asking questions during the interview, and if so, were the responses helpful?
  - c) In a typical visit of this type, the Midwife provides information about Down's syndrome screening. Did you have any difficulties understanding your midwife's explanation of Down's syndrome screening? If you did, please explain how this might have influenced your decision

to undergo screening?

Did you feel that you understood how screening for Down's syndrome may be different than screening for routine blood tests?

Did you feel that the screening for Down's syndrome was presented to you as a routine test?

The following is a short research scenario concerning Down's Syndrome Screening Methods

Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

#### Scenario 3

Recently a pregnant woman described how, at her booking visit with a community midwife, she was told about the screening process for Down's syndrome. The pregnant woman indicated that the way that the midwife presented the information about Down's syndrome screening, it seemed like she was suggesting that it would be a good idea for all pregnant woman to have the screening test.

As a result, after the meeting, the pregnant woman made arrangements to have the Down's syndrome screening.

Did your midwife influence your decision to have Down's syndrome screening? If so, can you explain how you felt your decision was influenced by the midwife? For example, did you feel that the discussion about the Down's syndrome screening test was part of routine care for pregnant women?

The following is a short research scenario concerning Down's Syndrome Screening Methods

Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

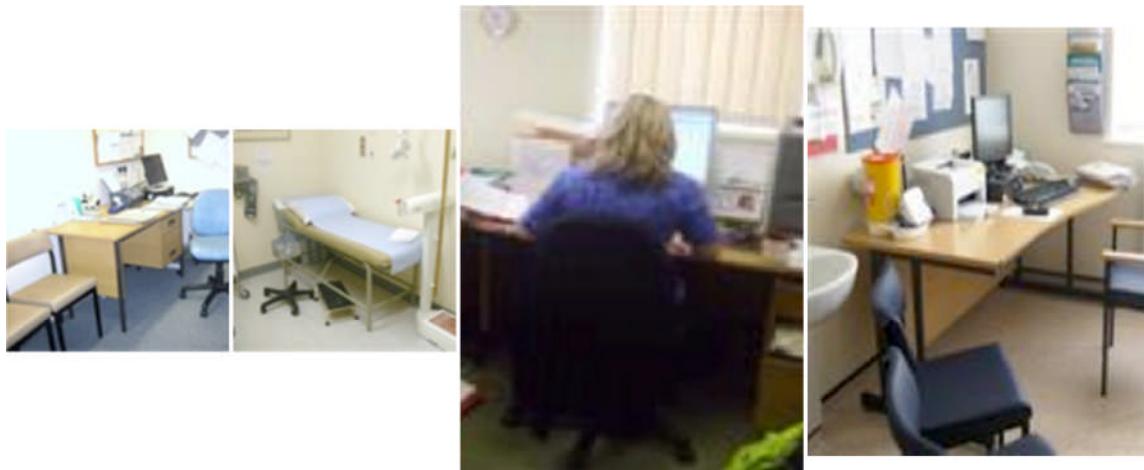
#### Scenario 4

The photographs below show the consulting room where the Community Midwife conducts booking appointments. During an appointment, the pregnant women and their partners are given information about prenatal Down's syndrome screening plus other information. The midwife always wears the uniform for community midwives and usually sits on a swivel chair in front of her desk. The pregnant woman sits on a chair that is placed at one end of the desk in a position where she could face the midwife. Her partner (when present) also sits on a chair beside her facing the midwife. The consulting room has a design and a décor that is found in most community antenatal clinics and is brightly lit with day light streaming through

the window and a fluorescent lamp hanging from the ceiling. The room has a patient examination couch, medical examination lamps, items for measuring height and weight plus other medical equipment.

From this scenario and looking at the photographs below, do you think the clinical environment or any aspect of it such as the lighting in the room, room décor, medical equipment, presence of the midwife or the entire clinical setting could influence the way you decide about participating in prenatal Down's syndrome screening?

If you do, can you explain in what way the clinical environment or any aspect of it could influence your decision to accept or decline prenatal Down's syndrome screening?



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### **Scenarios for partners of pregnant women**

The following is a short research scenario concerning Down's Syndrome Screening Methods  
Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

#### **Scenario 1**

A pregnant woman was seen by a midwife during her booking appointment at the local health centre. The midwife gave her lots of advice, such as the types of food to eat when pregnant. The woman's weight, height and blood pressure were then measured and the midwife told her about routine blood tests and the ultrasound scans. The woman agreed to have the blood tests and scans. It took another 5 minutes for the midwife to tell the woman about the screening test for Down's syndrome. At the end of the booking visit the midwife

gave the woman a booklet about antenatal screening tests to take home and read.

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- a) Can you explain what it was like for you when you were given information about Down's syndrome screening during the booking appointment? For example, how it made you feel about the likelihood of you having a baby with Down's syndrome.
- b) Please describe your experience with the information you received about Down's syndrome screening? For example, were you told about screening by family or friends, or had you read about the screening process prior to your visit by searching various sources like the internet? In general, were you comfortable with the amount of information that was provided to you during the booking visit?
- c) Please explain how you felt during your visit when you were talking to the midwife about Down's syndrome screening. For example, during your visit had you any thought the screening would be helpful for you? Likewise, during the visit did you feel that the screening process would be stressful, especially when considering possible test results or the diagnostic procedures that were suggested, or the choice to terminate the pregnancy?
- d) To what extent did your experiences during the booking visit influence your decision to have Down's syndrome screening? Please explain your responses with specific examples.

The following is a short research scenario concerning Down's Syndrome Screening Methods. Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

#### Scenario 2

At the booking visit, the midwife explained a number of routine blood tests. Included in these blood tests was an explanation about the screening test for Down's syndrome.

The woman was told that the tests were designed to help make sure the mother and baby were healthy.

In addition to information about various blood tests, information about participating in daily activities, such as exercise was also presented. The visit concluded with a standard measurement of height, weight and blood pressure. Finally, as the pregnant woman was leaving, the midwife passed her an information booklet about antenatal tests and asked her

to please take the booklet home to read.

- a) Thinking about your booking visit, did having the midwife explain Down's syndrome screening, as well as other tests, influence your decision about screening for Down's syndrome?
- b) Again, during your booking visit, did you feel that the information presented to you had an influence on your decision about screening? For example, did your midwife discuss issues, such as potential benefits or known risks of Down's syndrome screening? Did you feel comfortable asking questions during the interview, and if so, were the responses helpful?
- c) In a typical visit of this type, the midwife provides information about Down's syndrome screening. Did you have any difficulties understanding your midwife's explanation of Down's syndrome screening? If you did, please explain how this might have influenced your decision to undergo screening?

Did you feel that you understood how screening for Down's syndrome may be different than screening for routine blood tests?

Did you feel that the screening for Down's syndrome was presented to you as a routine test?



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The following is a short research scenario concerning Down's Syndrome Screening Methods  
Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

#### Scenario 3

Recently a pregnant woman described how, at her booking visit with a community midwife, she was told about the screening process for Down's syndrome. The pregnant woman

indicated that the way that the midwife presented the information about Down's syndrome screening, it seemed like she was suggesting that it would be a good idea for all pregnant woman to have the screening test.

As a result, after the meeting, the pregnant woman made arrangements to have the Down's syndrome screening.

Did your midwife influence your decision to have Down's syndrome screening? If so, can you explain how you felt your decision was influenced by the midwife? For example, did you feel that the discussion about the Down's syndrome screening test was part of routine care for pregnant women?

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The following is a short research scenario concerning Down's Syndrome Screening Methods  
Please review the scenario and respond to the questions below in as much detail as you feel comfortable.

#### Scenario 4

The photographs below show the consulting room where the Community Midwife conducts booking appointments. During an appointment, the pregnant women and their partners are given information about prenatal Down's syndrome screening plus other information. The midwife always wears the uniform for community midwives and usually sits on a swivel chair in front of her desk. The pregnant woman sits on a chair that is placed at one end of the desk in a position where she could face the midwife. Her partner (when present) also sits on a chair besides her facing the midwife. The consulting room has a design and a décor that is found in most community antenatal clinics and is brightly lit with day light streaming through

the window and a fluorescent lamp hanging from the ceiling. The room has a patient examination couch, medical examination lamps, items for measuring height and weight plus other medical equipment.

From this scenario and looking at the photographs below, do you think the clinical environment or any aspect of it such as the lighting in the room, room décor, medical equipment, presence of the midwife or the entire clinical setting could influence the way you decide about participating in prenatal Down's syndrome screening?

If you do, can you explain in what way the clinical environment or any aspect of it could influence your decision to accept or decline prenatal Down's syndrome screening?



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## Appendix 9: Field notes

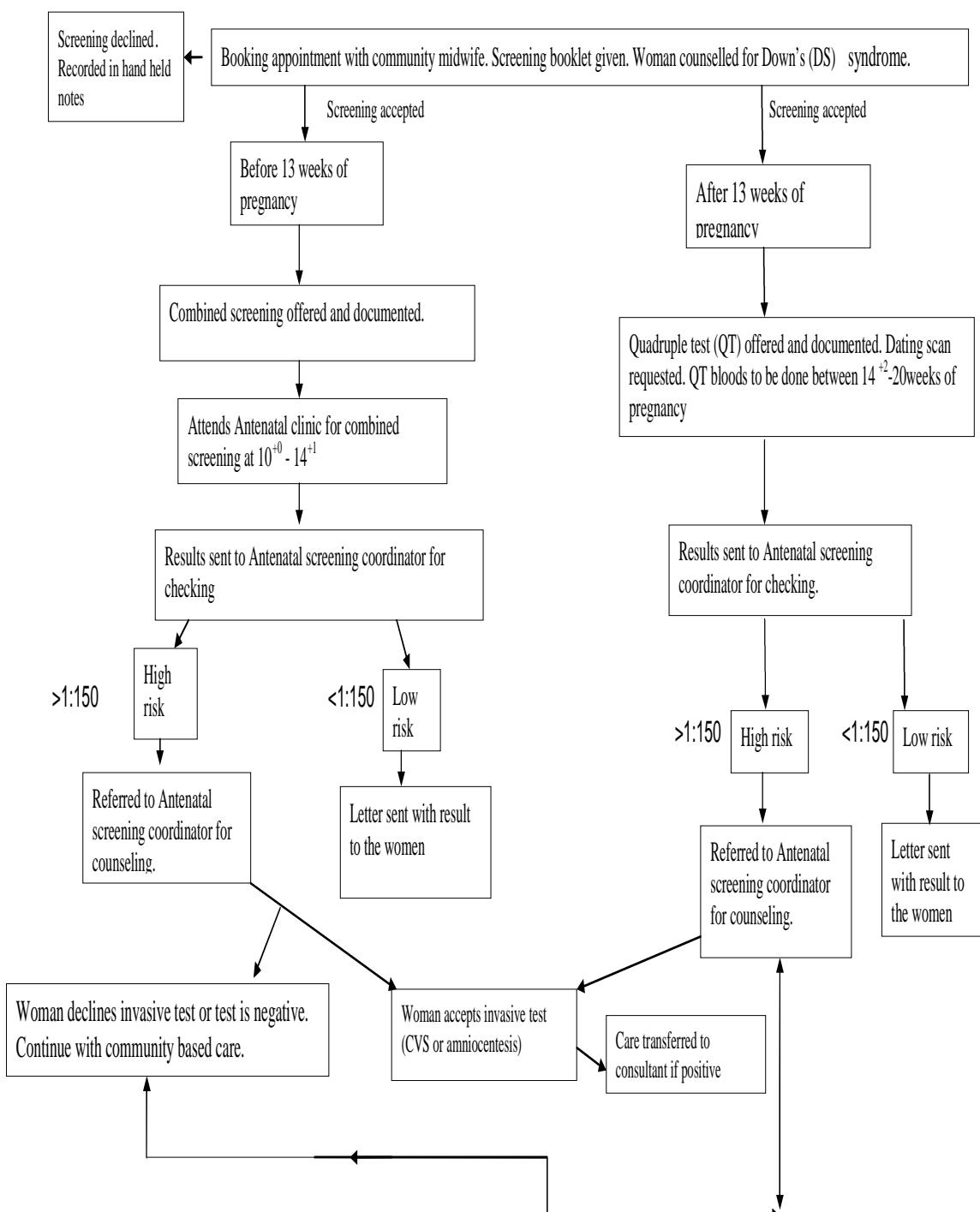
Trust 2  
HOMN  
↓  
2 matrons  
↓  
Specialist midwives  
↓  
bnd 7 (low fee leaders)  
↓  
bnd 6 (midwives & low midwives)  
↓  
bnd 5 (nursing qualified)  
↓  
↓  
+ Homs 6  
↓  
9  
↓  
18  
None NW Decline 1st trimester Combined Screening  
and see again by 2nd MN at 15 weeks where  
they offer 2nd trimester Screening (AT)

## Trust I

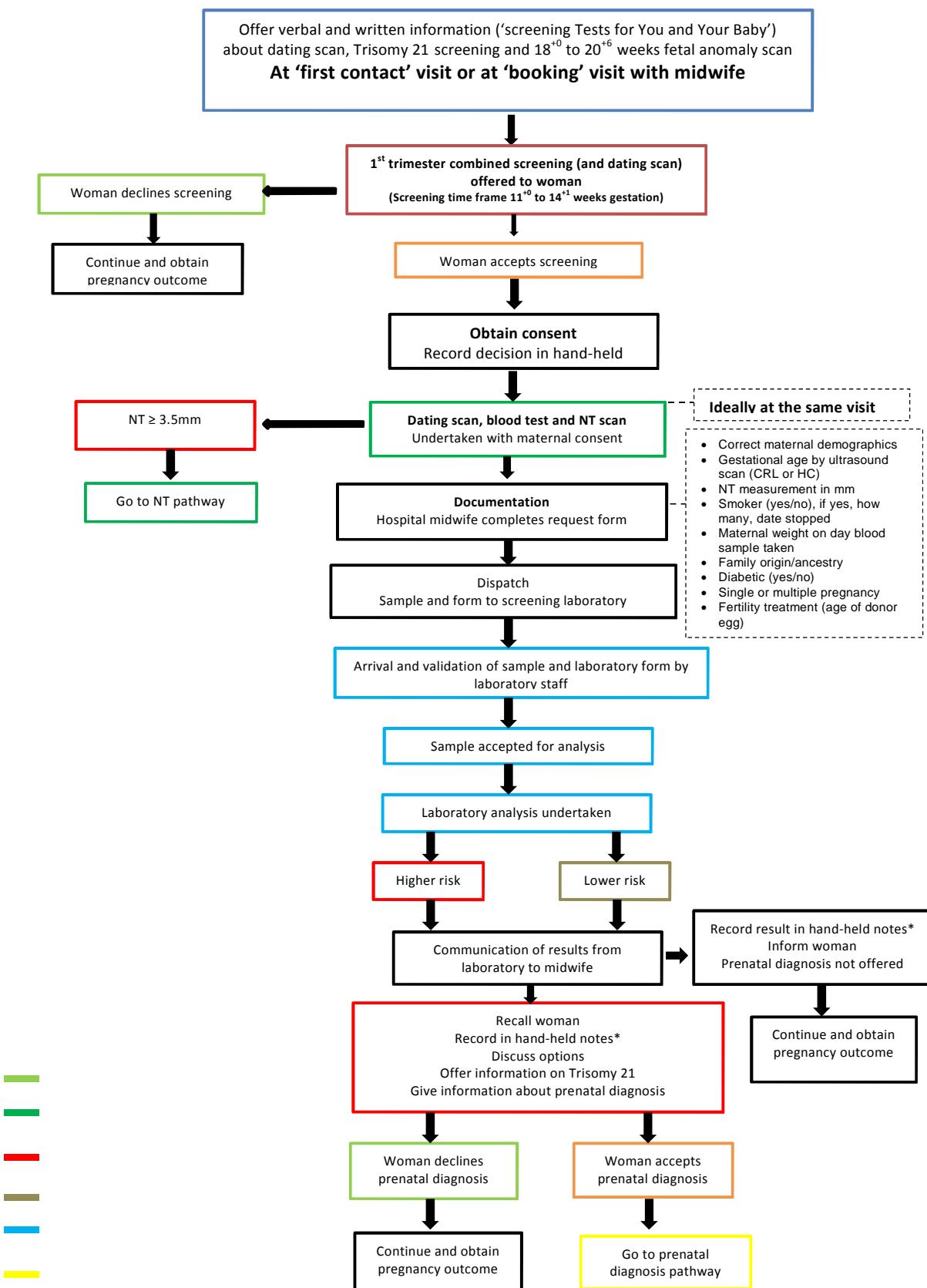
HOMN  
↓  
Matron (3)  
↓  
Specialist NW → low fee leaders  
every month the Ds report from the audits  
to find out if they were completed properly  
→ to achieving the national target of NSC whereby  
of 100% or acceptable of 97%. The MN will  
donate if the targets are not met.

## Appendix 10: Care pathways for Terrace and Landscape maternity services

### Care pathway for Down's syndrome screening test (Terrace maternity service)



### Care pathway for Down's syndrome screening (Landscape maternity service)



**Appendix 11: Abstract for oral presentation (8<sup>th</sup> Mixed Methods International Conference, University of Leeds, 19th June 2012)**

10:00 - 10:30 Baines Wing: 1.06

Philosophical and Methodological Issues

Hyacinth Ukuhor

**Methodological dilemmas in web-based mixed methods research**

Methodological concerns were highlighted in a study investigating the experiences of services users and frontline service providers involved in the prenatal Down's syndrome screening programme in the UK. This presentation draws on a multiple case study that seeks to describe the influence of service organization and delivery on women and partners' decisions to participate using the exploratory sequential design involving web-based interviews, discussion fora and survey questionnaire. The Office of National Statistics revealed that 6 million people accessed the internet using their mobile phones during 2010-2011 and 77% of UK households have access to the internet. This has opened up opportunities for data collection using the internet and World Wide Web in health services research, but within this exciting possibility are complex methodological issues. In order to sample participants in the qualitative phase of the study, a review of the literature on methodological issues in internet-based qualitative and mixed methods research was conducted. It became clear from the review that sample adequacy using a purposive sampling approach for on-line qualitative research was confusing and conflicting. Complexity was exacerbated by the fact that data could be collected asynchronously or synchronously in the qualitative phase of on-line studies. Whilst some contemporary on-line studies advocate the use of sample size as in traditional qualitative methods, others propose sample sizes that are larger due to anticipated high attrition. More research is needed, particularly to avoid sample inadequacy when collecting on-line qualitative data for mixed methods research. This presentation will elicit discussion on issues of recruitment and sampling in the qualitative phase of web-based mixed methods research based on a review of the literature and the experience from this study.

## **Appendix 12: Abstract for oral presentation (School of Healthcare Conference, October 2012)**

### **Oral Presentation 9**

**Hyacinth Ukuhor**

Year of study: 3 (Full Time)

Supervisors: Dr Janet Hirst<sup>1</sup>, Professor William Montelpare<sup>2</sup>, Professor Jose Closs<sup>1</sup>

#### **Using vignettes in an online qualitative study to explore participation in prenatal Down's syndrome screening.**

Participation rates in prenatal Down's syndrome screening in the UK are reported as 61% and in the North of England between 18%-80%. Studies have attributed such variation to psychosocial differences among women. Other studies have not found any association between these factors and uptake rates. This has led to an assumption that the characteristics of healthcare settings and service delivery may influence women and their partners' decisions to agree to screening.

The research questions are; 1) What is the experience of participating in prenatal Down's syndrome screening for pregnant women, their partners and community midwives? 2) Do the structures, processes and people in two healthcare settings influence women and partners' decisions to participate in prenatal screening for Down's syndrome? The study aims to explore the experiences of community midwives, pregnant women and partners and the influence of service organization and delivery on women and partners' decisions to participate in prenatal screening for Down's syndrome. An exploratory sequential case study design is being used, involving document review, web-based interviews and discussion forum in the pilot study which will be replaced by vignettes in the main online study. Purposive sampling will be used to invite 18-24 community midwives, 18-24 pregnant women and partners in each setting to respond online to vignettes with structured open-ended questions. Prompts will also be used with the main open-ended questions to elicit qualitative data. The data will be analysed using thematic analysis.

The presentation will focus on using vignettes constructed from the literature and pilot data as a methodological tool in the main online study. The vignettes will be adopted in the main study to address some of the issues raised in the pilot study. The criteria for the construction of the vignettes, types of vignette and their strengths and limitations will also be discussed.

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Prince Edward Island, Canada