Decision Making in Prenatal Testing.

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

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Abstract.

Introduction: One of the main objectives of prenatal diagnosis programmes is to ensure women make informed decisions about testing. As few studies have operationalised informed decision making, it is unclear whether or not this objective has been attained. However, evidence suggests that (a) insufficient information is provided to enable informed and autonomous decision making, and (b) women's knowledge of testing is often incomplete. It is unlikely, therefore, that all women make fully informed decisions.

Aims: To describe the conditions enabling informed decision making; to operationalise and assess the informed decision making process; to evaluate the efficacy of decision analysis to facilitate informed decision making; to describe the factors associated with prenatal diagnosis decisions.

Sample: Data from 128 prenatal diagnosis information giving consultations between a health professional and women receiving a screen positive triple test result were included for analysis.

Methods: Theoretical and integrative reviews summarising prior empirical research; observational designs assessing the decision making process; a randomised control trial design evaluating the decision analysis intervention. The following materials were piloted: a checklist of information provided; a coding frame of information utilised; a consent form; a questionnaire completed at two time points.

Results: Insufficient information about Down's syndrome and termination was provided by the health professional to enable informed decision making. As many women employed a 'screening out' strategy to limit the information for assimilation, not all women made fully informed decisions. In the RCT (n = 106), 17 women chose no further testing. Compared with women receiving routine information, those allocated to the decision analysis consultation made more informed decisions, experienced less decisional conflict, were less falsely reassured and had longer consultations. Perceived social norm, expected-utility values and anxiety predicted women's test decisions.

Discussion: Decision analysis consultations were associated with more informed decisions than routine care. However, additional empirical research is required to ascertain what aspects of the decision analytic technique were associated with the facilitation of informed decision making. Recommended changes to routine clinical practice and women's role in the consultation will be dependent on these subsequent findings.

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Abbreviations.

AFP	- Alpha Fetoprotein
CVS	- Chorionic Villus Sampling
DA	- Decision Analysis
DS	- Down's Syndrome
EDD	- Estimated Date of Delivery
ES	- Edward's Syndrome
EU	- Expected-Utility
EUT	- Expected-Utility Theory
EUV	- Expected-Utility Value
hCG	- Human Chorion Gonadtrophin
IDM	- Informed Decision Making
IDM:a	- Informed Decision Making: reasons against an alternative (measure)
IDM:i	- Informed Decision Making: information seeking (measure)
IDM:r	- Informed Decision Making: reasons for an alternative (measure)
IPDM	- Informed Patient Decision Making
LASS	- Leeds Antenatal Screening Service
LOT	- Life Orientation Test
LGI	- Leeds General Infirmary
MSAFP	- Maternal Serum alpha-fetoprotein
NDM	- Naturalistic Decision Making
NFC	- Need For Cognition
RCT	- Randomised Control Trial
SB	- Spina Bifida
TPB	- Theory of Planned Behaviour
TT	- Triple test
uE_3	- Unconjugated Estriols

1. Prenatal diagnosis and women's decision making: a theoretical review.

This chapter provides an overview of the application of psychological theory to understanding informed decision making in the context of prenatal diagnosis for Down's syndrome. The first section of the chapter describes the procedures offered during pregnancy to identify fetal abnormalities, summarises the findings from research exploring women's experiences of prenatal testing for Down's syndrome and outlines the objectives of prenatal diagnosis. The second part of the chapter focuses on some of the psychological models that have been used to explain individual's health decisions. In particular, this section assesses the usefulness of these models in understanding, operationalising and facilitating informed decision making. The final section includes a *theoretical review* (Cooper, 1989) that integrates the empirical evidence from research carried out to explain women's prenatal diagnostic test behaviour for Down's syndrome and evaluates the extent to which this testing decision can be said to be informed.

1.1 Prenatal testing.

A number of procedures offered during pregnancy are described by the umbrella term 'prenatal testing'. These procedures fall into three areas of testing: *prenatal screening (section 1.1.1)*; *genetic testing (section 1.1.2); prenatal diagnosis (section 1.1.3).* Prenatal diagnosis is only offered to a woman[1] if the risks of having a baby with an abnormality are sufficient to warrant the use of such an invasive and risky procedure (Harper, 1992; Royal College of Obstetricians and Gynaecologists, 1993; Hewison, 1996). Women with a sufficient or increased risk of having a baby with an abnormality are identified and distinguished from those with 'insufficient risk', either following a screening test or as a result of carrier testing for a genetic disease. Although the offer of a procedure may depend on the results of a previous test, each choice a woman makes about a prenatal testing procedure is a discrete decision event (Shiloh, 1995). The following section briefly describes the types of procedures used to identify fetal abnormalities within each of these three prenatal testing contexts.

1.1.1 Screening tests.

Screening tests provide women with a risk figure that estimates their likelihood of having a baby with an abnormality. Arbitrary cut-off points are used to dichotomise the

[1] *Woman* refers to the individual making the prenatal testing decision. For consistency, the term woman is employed when the literature refers to couples making the prenatal diagnosis decision.

range of risk figures provided by the screening test into either a screen negative or screen positive test result. A screen negative result is sometimes referred to as a low risk, a screen positive as an increased or high risk. Prenatal screening tests provide a non-invasive method of segregating the population so that those with an increased or sufficient risk may be offered prenatal diagnosis. Although screening tests are referred to as non-invasive, there is an extensive literature describing an increase in psychological morbidity amongst pregnant women following their experience of undergoing prenatal screening (Berne-Fromell, Uddenberg and Kessler, 1983; Burton, Dillard and Clark, 1985; Tymstra, 1989; Green, 1990; Green, Statham and Snowden, 1992; Marteau, 1992; Gregg, 1993; Marteau, Kidd, Michie, Cook, Johnston and Shaw, 1993; Green, 1994; Thornton, Hewison, Lilford and Vail, 1995; Green and Statham, 1996; Hewison, 1996; Michie, Smith, McClennan and Marteau, 1997). In addition, a large body of research has focused on factors associated with the uptake of prenatal screening procedures, including both women's and health professionals' understanding of and attitudes towards prenatal screening (Lippman-Hand and Cohen, 1980; Sikkink, 1990; Green, Satham and Snowden, 1992; Marteau, 1992; Marteau, Johnston, Kidd, Michie and Cook, 1992; Marteau, Slack, Kidd and Shaw, 1992; Roelofsen, Kamerbeek, Tymstra, Beekhuis and Mantingh, 1993; Marteau, 1994a; Khalid, Price and Barrow, 1994; Smith, Shaw and Marteau, 1994; Smith, Slack, Shaw and Marteau, 1994; Jorgensen, 1995; Thornton, Hewison, Lilford and Vail, 1995; Goel, Glazier, Holzapfel, Pugh and Summers, 1996; Hewison, 1996; Sullivan, 1996; Fairgrieve, 1997; Michie, Smith, McClennan and Marteau, 1997; Proud and Murphy-Black, 1997; Ryder, 1998). The issues of impact and uptake pertaining to screening procedures are not discussed further as they are outside the remit of a thesis focusing on prenatal diagnostic test decisions. However, as prenatal diagnosis is only offered to women with a sufficient risk of having a baby with an abnormality, three prenatal screening methods used to estimate the risk of fetal abnormality are described in more detail below.

Taking a medical history is the primary screening method used to identify some women at increased risk for fetal abnormalities (Royal College of Physicians, 1989; Gosden, Nicolaides and Whitting, 1994). A medical, reproductive and family history is routinely taken from all women attending their 'booking-in visit' at an antenatal clinic early in the pregnancy. For example, the following characteristics of women are associated with an increased risk of the baby having an abnormality: increased maternal age (35 years and over) is associated with an increase in the incidence of chromosomal abnormalities, in particular Down's syndrome; certain ethnic groups have a greater likelihood of inheriting specific genetic diseases such as Tay-Sachs in the

Askenazi-Jewish population and cystic fibrosis amongst northern Europeans (Mueller and Young, 1995); having already had a baby with an abnormality may be associated with a recurrence risk in subsequent pregnancies; maternal diseases such as rubella and diabetes.

Biochemical or maternal serum screening is routinely offered in most antenatal departments (Green, 1994)[2]. Risk figures for neural tube defects and chromosomal abnormalities are calculated from a number of biochemical markers found in the woman's blood or maternal serum (Wald and Cuckle, 1992; Aitken and Crossley, 1996). Whereas risk figures from the medical-history taking are associated with the enduring characteristics of a woman, risk figures calculated from serum screening are specific to the current pregnancy. The screening test for neural tube defects is sometimes referred to as maternal serum alpha-fetoprotein (MSAFP) and the tests for chromosomal abnormalities as maternal serum screening, the triple test or the Bart's test. The biochemical tests provide more accurate risk estimates for the chromosomal abnormalities than figures based on maternal age (Wald and Cuckle, 1991). Although blood for the MSAFP is usually taken after 15 weeks gestation, the triple test can be performed after 13 weeks gestation (Aitken and Crossley, 1995). A screen positive triple test result falls between a 1:5-1:250 risk of having a baby with Down's syndrome, a screen negative result between 1:251-1:50 000 (Wald and Cuckle, 1992; Royal College of Obstetricians and Gynaecologists, 1993)[3]. Although the test is technically non-invasive and is often presented as a routine procedure (Madlon-Kay, Reif, Mersy and Luxenberg, 1992; Marteau, Slack, Kidd and Shaw, 1992; Gosden, Nicolaides and Whitting, 1994), guidelines suggest that women should make an informed choice to accept or decline maternal serum screening (Royal College of Obstetricians and Gynaecologists, 1993).

The ultrasound scan is routinely used within the ante-natal clinic to monitor the growth or development of the fetus (Campbell and Smith, 1984; Royal College of Physicians, 1989; Wald and Cuckle, 1991; Whittle, 1995). A picture of the fetus, placenta and amniotic fluid surrounding the baby is built up by passing high-frequency sound waves through the woman's abdomen (Gosden, Nicolaides and Whiting, 1994). Initially the

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[2] In theory, maternal serum screening is available to any pregnant woman. However, some health regions offer maternal serum screening to women over a certain age to reduce laboratory costs (Royal College of Obstetricians and Gynaecologists, 1993).

[3] The risk level of 1: 250 was estimated to generate a 5% amniocentesis rate. This rate is similar to that generated from screening by maternal age and was considered a reasonable cut-off point (Royal College of Obstetricians and Gynaecologists, 1993). However, the cut-off point risk figure is based on medical and economic considerations and may vary between health regions.

ultrasound scan is used to ensure that a pregnancy is viable, to ascertain the number of fetuses and to estimate the gestational age of the fetus (es). The ultrasound scan is also used to screen for fetal abnormalities at two time points: fetal nuchal translucency after thirteen weeks gestation (Nicolaides, Brizot and Snijders, 1994); structural abnormalities after sixteen weeks gestation (Campbell and Smith, 1984; Royal College of Physicians, 1989; Wald and Cuckle, 1991; Green, 1994a; Chitty, 1995; Smith and Marteau, 1995; Whittle, 1995). In addition, the ultrasound scan is used as a diagnostic technique for some fetal abnormalities such as a cleft palate or heart defect (Royal College of Physicians, 1989; Wald and Cuckle, 1992; Chitty, 1995). However, the results of ultrasound screening provide only a risk figure for the likelihood of having a baby with Down's syndrome. The evidence suggests that, although the ultrasound scan is employed as both a screening and diagnostic technique, few women make an informed choice about whether or not to be scanned (Lippman, 1994; Chitty, 1995; Smith and Marteau, 1995; Proud and Murphy-Black, 1997).

1.1.2 Genetic tests.

Over the last thirty years, genes and genetic inheritance patterns have been implicated in the cause of a number of diseases to a greater or lesser extent (Mueller and Young, 1995). Of particular relevance to this discussion are the disorders known as single gene diseases such as Huntington's chorea, cystic fibrosis, Duchenne muscular dystrophy, Tay-Sachs, thalassaemia and sickle cell. For these diseases, a single gene is known to be the cause and manifestation of the disease in an individual. The disease information is coded within an individual's genes and can only be passed on to others at conception of a fetus. Single gene diseases are either recessive or dominant. A recessive gene disease requires both genes within a gene pair to have the mutation associated with the disorder before the individual manifests the disease. An individual with one 'disease gene' is usually asymptomatic and is known as a carrier. A recessive gene disease can only be passed on to the fetus if both parents are carriers of the 'disease gene'. If both parents are carriers of a recessive gene disorder then there is a 25% or 1:4 risk that the baby will inherit the disease. A dominant gene disease requires only one gene within a gene pair to have the mutation associated with the disorder before the individual manifests the disease, i.e. an individual with only one 'disease gene' will usually develop the disease. A dominant gene disease requires only one parent to have the gene for the disease to be passed on to the fetus. If one parent has the disease gene, then there is a 50% or 1:2 risk that the baby will inherit the disease. For a number of single gene disorders, ascertaining whether or not individuals carry the 'disease gene' requires an analysis of their blood or saliva. Although carrier testing is technically non-

invasive, it is recommended that carrier testing should be undertaken before pregnancy (Royal College of Physicians, 1989) because testing may be associated with increased psychological morbidity (Royal College of Physicians, 1993; Bekker, Denniss, Modell, Bobrow and Marteau, 1994; Marteau and Anionwu, 1996). However, a number of single gene carrier testing programmes have been offered both before and during pregnancy (Watson, Mayall, Chappie, Harrington, Williams and Williamson, 1991; Marteau, 1992; Mennie, Gilfillan, Compton, Curtis, Liston, Pullen and Brock, 1992; Bekker, Modell, Dennis, Silver, Mathew, Bobrow and Marteau, 1993; Bekker, Dennis, Modell, Bobrow and Marteau, 1994; Meidzybrodzka, Hall, Mollison, Templeton, Russel, Dean, Kelly, Marteau and Haites, 1995; Hewison, 1996; Marteau and Anionwu, 1996). In essence, women at risk of having a baby with a genetic disease are considered to be of 'sufficient risk' to be offered a prenatal diagnostic test.

1.1.3 Diagnostic tests.

Three test procedures are offered during pregnancy to *diagnose* whether or not a baby has a fetal abnormality: amniocentesis; chorionic villus sampling; fetal blood sampling. All three procedures are invasive and use a 'sampling needle' to remove some tissue or liquor from the fetus *in utero* (Gosden, Nicolaides and Whiting, 1994). The procedures are usually performed transabdominally in conjunction with an ultrasound scan to guide the position of a needle (Royal College of Physicians, 1989; Gosden, Nicolaides and Whiting, 1994). An additional rate of miscarriage (spontaneous abortion) over and above the miscarriage rate quoted for routine pregnancies is associated with the diagnostic test procedures. In addition, women may experience tenderness or bruising as a consequence of the 'sampling needle' insertion. The abnormalities identified by the tests, the techniques, the timing of test results and the risk of miscarriage associated with the procedure are described below.

Amniocentesis is usually performed after 14 weeks of pregnancy. A sampling needle is passed through the abdomen into the amniotic sac within the uterus (Gosden, Nicolaides and Whitting, 1994; Queenan, 1996). Between 10-20 ml of amniotic fluid are aspirated for analysis and test results obtained about 2-3 weeks later. The miscarriage risk figure associated with amniocentesis is between 0.5-1% (Royal College of Physicians, 1989). There is a small likelihood that the cells contained within the fluid may not culture or grow, requiring a repeat test to be carried out (Harper, 1992; Gosden, Nicolaides and Whitting, 1994). The following fetal abnormalities are diagnosed by amniocentesis: chromosomal disorders such as Down's syndrome, Edward's syndrome

and Turner's syndrome; single gene disorders such as cystic fibrosis, Duchenne muscular dystrophy and Huntington's chorea; neural tube defects such as spina bifida.

Chorionic villus sampling is carried out after 10 weeks of pregnancy. Two needles are commonly required for this procedure: the first is passed through the abdomen into the uterus to penetrate the surface of the placenta; the second is passed through the first needle and is used to scrape some tissue from the placenta for analysis. The reported risk of miscarriage from chorionic villus sampling ranges between 1-4% (Royal College of Physicians, 1989). In addition, carrying out chorionic villus sampling before 10 weeks gestation is associated with an increased risk of fetal limb defects. A chorionic villus sampling culture is twice as likely as amniocentesis to need repeating because of the likelihood of mosaicism (Tolmie, 1995). Mosaicism refers to the presence of different numbers of chromosomes or complements of chromosomes in two or more cell lines from the same sample (Mueller and Young, 1995; Tolmie, 1995). Mosaicism may be a result of either a true *in vivo* difference in the arrangement of chromosomes in the fetus or a phenomenon of the prenatal diagnostic test procedure; chorionic villus sampling takes cells from outside the fetus that may result in the extraction of both maternal and fetal cells and mosaicism being identified (Harper, 1992; Gosden, Nicolaides and Whitting, 1994; Roberts and Rodeck, 1995; Mueller and Young, 1995; Tolmie, 1995). Chorionic villus sampling provides a diagnosis for the chromosomal and single gene disorders described under the explanation of amniocentesis. Although preliminary test results are obtained after 24 hours, information concerning the presence of mosaicism is only available three weeks after the test has been undertaken.

Fetal blood sampling involves inserting a sampling needle through the abdomen into the umbilical cord within the uterus to extract the baby's blood. The procedure related risk of miscarriage is about 1-2% after nineteen weeks of pregnancy (Royal College of Physicians, 1989; Gosden, Nicolaides and Whitting, 1994; Roberts and Rodeck, 1995). Fetal blood sampling is commonly used to diagnose haemoglobin disorders and infections such as toxoplasmosis and rubella; it can also be used for the identification of chromosomal or single gene disorders. The test results are obtained between 1-2 days later. As fetal blood sampling is offered later in pregnancy, the majority of women having prenatal diagnosis will be offered the choice of amniocentesis or chorionic villus sampling (Ward, 1991; Williamson, Harris, Church, Fiddler and Rhind, 1996). For the purpose of this thesis, prenatal diagnosis refers to the offer of amniocentesis and chorionic villus sampling only.

1.1.4 Women's experience of prenatal diagnosis for Down's syndrome.

The focus of this thesis is to understand women's informed decision making about whether to have or not have prenatal diagnosis following receipt of a screen positive triple test for Down's syndrome. Down's syndrome (trisomy 21) is a chromosomal abnormality occurring spontaneously in about 1 in 700 births (Wald and Cuckle, 1992; Mueller and Young, 1995). Down's syndrome is the most common cause of mental retardation. The severity of Down's syndrome varies by individual as illustrated by the IQ scores which have a range of values from 25 to 75 (Mueller and Young, 1995). Although life expectancy is generally good, there are a number of associated physical abnormalities, such as malformations of the heart, digestive system, eyes and ears, which may lead to premature death (Wald and Cuckle, 1992; Leeds Antenatal Screening Service, 1993; Barnes and Bryan, 1997). There is a strong association between advancing maternal age and the risk of having a baby with Down's syndrome; between 35 and 45 years of age women's risk of having a baby with Down's syndrome increases from about 1:400 to 1:30 (Wald and Cuckle, 1992). The recurrence rate of having a second baby with Down's syndrome is related to maternal age and falls somewhere between a 0.5-1% risk (Wald and Cuckle, 1992; Mueller and Young, 1995). Until the early 1990s, maternal age was used as the screening test for Down's syndrome with women over 35 years of age being offered prenatal diagnosis for Down's syndrome (Royal College of Obstetricians and Gynaecologists, 1993). However, the development of a biochemical screening test for Down's syndrome, the triple test, has meant that all pregnant women can be offered a screening test for Down's syndrome regardless of maternal age (section 1.1.1). All women receiving a screen positive biochemical test result are now offered prenatal diagnosis.

The decision to have or not have a diagnostic test following receipt of a screen positive triple test result can be described as 'risky'. The decision is 'risky' because women have to choose an alternative from two options, both with uncertain and negative consequences: to have no diagnostic test and continue the pregnancy, knowing the risk of having a baby with Down's syndrome; to have a diagnostic test and risk miscarrying a baby without Down's syndrome. Furthermore, a woman's decision to have or not have a diagnostic test involves what are referred to as 'hot cognitions' (Abelson, 1963) i.e. cognitive processes about personal, affect-laden and important issues (Janis and Mann, 1977). The consequences of the prenatal diagnostic test options are serious and some are irreversible: management of the 'increased risk' information; birth of a healthy

baby[4]; miscarriage of a healthy pregnancy; birth of a baby with Down's syndrome; consideration of a termination of a pregnancy for Down's syndrome. It is likely that the prenatal diagnostic test decision would result in women experiencing intense conflict during decision making (Janis and Mann, 1977). Decisional conflict or ambiguity (Eagly and Chaiken, 1993) is present in a decision which generates the simultaneous desire to accept and reject an option (Janis and Mann, 1977). Where both alternatives involve negative consequences or losses, such as the prenatal diagnosis decision, decisional conflict intensifies. Decisional conflict is expressed by hesitation, vacillation, feelings of uncertainty, and acute emotional affect such as anxiety (Janis and Mann, 1977; Mann, 1992; Baron, 1994). The evidence to date suggests that women do find the prenatal diagnosis decision extremely distressing (Nielsen, 1981; Pauker and Pauker, 1987; Gregg, 1993). Women expressed concerns about having a baby with an abnormality, injuring the fetus following testing, interfering with nature, miscarrying the fetus, the pain of having a diagnostic procedure and waiting for test results (Nielsen, 1981; McGovern, Goldberg and Desnick, 1986; Sjorgen and Uddenberg, 1988; Green, Statham and Snowden, 1992; Burke and Kolker, 1993). In addition, anxiety levels measured before women have a diagnostic procedure tended to be extremely high, equivalent to those considered to be clinically abnormal (Beeson and Golbus, 1979; Burton, Dillard and Clark, 1985; Green, 1990; Marteau, Kidd, Cook, Michie, Johnston, Slack and Shaw, 1992). Although the majority of studies report that this high level of anxiety returned to levels considered normal for pregnant women upon receipt of a negative diagnostic test result (Burton, Dillard and Clark, 1985; Green, 1990; Marteau, Kidd, Cook, Michie, Johnson, Slack and Shaw, 1992), it is clear that women find the decision to have or not have prenatal diagnosis difficult and stressful.

1.1.5 Objectives of prenatal diagnosis.

The offer of testing during pregnancy is an aspect of preventive medicine. More specifically, secondary prevention - which aims to detect and cure disease at a stage before symptoms present in an attempt to reduce the morbidity and mortality associated with the disease (Royal College of Physicians, 1991; Marteau, 1994a) - Wilson (1965) developed a number of criteria to justify which diseases may be suitable targets for preventive medicine (from Ogden, 1996):

1. the disease must be sufficiently prevalent and/or sufficiently serious to make early detection appropriate;

[4] The term 'healthy' is used for convenience to refer to a fetus, baby or child unaffected by the abnormality for which it was tested. Although it is likely most babies are born healthy, there are instances where the baby may have or develop a condition despite undergoing prenatal testing.

2. the disease must be sufficiently well defined to permit accurate diagnosis;
3. there must be a possibility that the disease remains undiagnosed in many cases so that rapid diagnosis is not inevitable without intervention;
4. there must be a beneficial outcome from early diagnosis in terms of disease treatment;
5. there must be a screening test which has good sensitivity and specificity and a reasonable positive predictive value in the population to be screened.

There are some concerns about the use of these criteria to justify targeting some diseases and omitting others from the realm of preventive medicine. For example, a screening programme for a disease may be introduced because the technology is available to test for it rather than because it is 'sufficiently serious'. Equally, 'sufficiently serious' is a subjective evaluation of a disease state which may be susceptible to the attitudes of the professionals implementing the technology rather than populations' attitudes towards the disease (see Rose, 1992). It is beyond the remit of this thesis to explore these concerns in relation to the current diseases or abnormalities targeted within the context of prenatal testing. However, an issue particularly pertinent to the prenatal testing domain is that of ensuring that testing is associated with more benefits than costs. Undergoing testing in pregnancy is a voluntary behaviour of women who are essentially well. As previously described, the experience of undergoing prenatal diagnosis for Down's syndrome is associated with increased psychological morbidity and a consequence of testing may be to miscarry a healthy pregnancy. Of the number of women that accept the offer of prenatal screening, about 5% (Haddow, 1996) are offered a diagnostic test. About 2-4% of those offered prenatal diagnosis receive a diagnosis of Down's syndrome (Royal College of Obstetricians and Gynaecologists, 1993; Piggott, Wilkinson and Bennett, 1994). Once Down's syndrome has been identified there is no treatment to cure it. The 'treatment' offered to women at this stage is to terminate the pregnancy (therapeutic abortion). If women accept treatment, the current pregnancy is terminated and the birth of a baby with Down's syndrome is prevented; if treatment is declined, women have gained information about the health status of their baby during the second trimester of pregnancy, so enabling them to prepare for the birth of a baby with Down's syndrome. Taking into account the affect associated with making the prenatal diagnosis decision, the increased risk of miscarrying a healthy pregnancy and the likelihood of having to terminate a current pregnancy, it appears that, at least in the short term, prenatal testing may engender more harm than good. In an attempt to minimise these iatrogenic consequences, the Royal College of Physicians (1989) developed the following guidelines for health

professionals to consider when offering prenatal diagnosis. The objectives of prenatal diagnosis are:

- to allow the widest possible range of informed choice to women and couples at risk of having children with an abnormality;
- to provide reassurance and reduce the level of anxiety associated with reproduction;
- to allow couples at risk to embark on having a family knowing that they may avoid the birth of seriously affected children through selective abortion;
- ensure optimal treatment of affected infants through early diagnosis.

The same report makes it clear that at least from an ethical perspective, ensuring women make an informed choice to have or not have prenatal diagnosis is perhaps the most important objective for professionals to attain when offering prenatal diagnosis:

> Prenatal diagnosis should be undertaken within the general principles of informed consent, including the possibility that after testing, the question of terminating the pregnancy may have to be faced. Women must therefore have the right to refuse testing, even at a fairly preliminary stage, and must understand the implications of their decision (Royal College of Physicians, 1989: section 8.5).

1.2 Informed decision making.

Informed decision making, or informed consent, is a well established concept within the applied medical decision making literature (Mazur, 1986; Royal College of Physicians, 1989; Royal College of Obstetricians and Gynaecologists, 1993; Buchanen, 1995; Llewellyn-Thomas, 1995; Robinson, 1995; ASHG, 1996; Ubel and Loewenstein, 1997; Coulter, Entwhistle and Gilbert, 1999; Sugarman, McCroy, Powell, Krasny, Adams, Ball and Cassell, 1999). The following two components are usually referred to as necessary for informed decision making to occur (Emery, 1984; Mazur, 1986; RCP, 1989; Abramsky, 1994; Clark, 1994; Buchanen, 1995; Llewellyn-Thomas, 1995; Mueller and Young, 1995; O'Connor, 1995; ASHG, 1996; Ubel and Loewenstein, 1997). Firstly, it is the responsibility of the health professional to provide women with sufficient information to be able to make the decision. Sufficient information refers to all the facts a reasonable person would need to know to be able to make the decision. These facts should include information about the abnormalities being tested for, explanations of all the alternatives available and the risks and consequences associated with those alternatives. Secondly, informed decision making is concerned with the woman's autonomy in making the decision. The information should be presented neutrally so that

women may assimilate the information in accordance with their own values. The information consultation should be non-directive and the decision made without undue pressure from the health professional. However, it is the role of the health professional to facilitate the decision making process; to communicate the information in sympathy with the woman's values.

Despite consensus on the above factors required for informed decision making, there are few agreed criteria on what constitutes an informed decision (Marteau, 1995). Consequently, assessing whether or not women make informed decisions about prenatal diagnosis is complicated by the apparent difficulty in operationalising the concept. The following section of the chapter explores a few of the psychological theories and areas of research that have aimed to explain individual's decision making. One of the primary concerns of researchers within this area of psychology has been the issue of explaining and defining 'good' decision making. Discussed in more detail later within this section are some of the ways to operationalise and assess good decision making.

1.2.1 Decision making theories.

1.2.1.1 Classical decision theory.

The theoretical basis for informed decisions is to be found within the social sciences. For over forty years, models of decision making under uncertainty have been developed by economists and psychologists. The most dominant model is that of (subjective) expected-utility (EU) (Edwards, 1954; Savage, 1954), sometimes referred to as classical decision theory (Beach and Lipshitz, 1993) or rational choice theory (Zey, 1992, 1998). It is essentially a normative model of decision making, defining the characteristics that are necessary to make the most appropriate choice under uncertainty in an 'ideal world' (Bell, Raiffa, and Tversky, 1988; van der Pligt, 1998). Many texts describe the axioms and underlying principles of EU models in adequate detail (March, 1982; Eiser and van der Pligt, 1988; Yates, 1990; Baron, 1994; Garnham and Oakhill, 1994). For the purposes of this chapter, it is necessary to be aware of the assumptions of the models and how decision making is conceptualised. Some assumptions of EU theory are that the decision maker has volition over the decision making process, has access to complete information about the alternatives, has a stable ranking of preferences and will choose the 'best' alternative (Zey, 1992; Baron, 1994; Crozier and Ranyard, 1997). The best alternative is calculated after the decision has been broken down into its components. The components include the perceived risk

or probability of each consequence occurring and the decision maker's utility or evaluation of how good or bad that consequence is perceived to be should it occur. An expected-utility figure is calculated by multiplying these two figures together in accord with a rational decision rule for each consequence of the decision (van der Pligt, 1998). The expected-utility figures of all the consequences pertaining to each action or alternative are added together. The correct choice is the alternative with the maximum expected-utility value. Essentially the EU model provides a framework for a decision to be made based on the relationship between the decision maker's individual attitudes and evaluations of risk of the decision' consequences.

1.2.1.2 Describing decision making.

EUT was developed from mathematical theorems and axioms that aimed to explain risky choice within decision contexts specific to game theories (Lopes, 1994; Crozier and Ranyard, 1997). The decision making described is that of a hypothetical decision maker, Economic Man (Beach and Lipshitz, 1993), who is able to assemble and evaluate all the available information about each alternative systematically and according to the laws of logic and probability. However, empirical evidence from researchers aiming to explain individuals' actual decision making and problem solving established that EUT is not a descriptive theory of decision making (Simon, 1992). The research by Allais (1953), Simon (1955,1956), Eilsberg (1961) and Tversky (1969) illustrated that when making decisions involving risk, individuals violate both the axioms and assumptions of rational decision making as defined by EUT. Essentially, individuals do not have the mental capacity to assess the utilities and probabilities associated with the consequences of each alternative of a decision, to assimilate this information systematically and then to select the alternative with the greatest expected-utility. These cognitive limitations or 'bounded rationality' (Simon, 1955) have implications for the information attended to, the judgements made about the information and the processing strategies employed to make the decision. Two of the observed techniques by which individuals reduce the cognitive load during risky decision making, heuristic methods (see Tversky and Kahneman, 1974, 1988) and satisficing strategies (Simon, 1955, 1956), have implications for ensuring that individuals make informed decisions.

Heuristics are rules of thumb or principles (Tversky and Kahneman, 1974) that provide individuals with simpler judgements to assimilate when making a decision than the systematic evaluation of the probabilities and utilities of each attribute. Heuristics require less cognitive effort than the systematic evaluation of all the available

information (Eagly and Chaiken, 1993). The individual tends to focus on aspects of the decision context rather than the decision content or information when using an heuristic method (Chaiken, 1980; Eagly and Chaiken, 1993). These heuristic rules are learnt from past experience or observation (Eagly and Chaiken, 1993) and can mediate an individual's judgement during decision making. An example of an heuristic rule may be that 'health professionals can be trusted with regard to health care issues' (Eagly and Chaiken, 1993; Stroebe and Stroebe, 1995). A consequence of this rule may be the heavy weighting of information provided by a health professional during the decision making process, such as a personal aside like 'I had chorionic villus sampling' or 'most of my patients have amniocentesis'. This information may discourage a woman from surveying all the options available or may alter her perception of the risks involved. For some decisions the application of an heuristic to reach a decision is appropriate (Fischhoff, Slovic and Lichtenstein, 1980; Vlek, 1986; Eiser and van der Pligt, 1988; Baron, 1994; Ubel and Loewenstein, 1997). However, more 'unsatisfactory' decisions are likely to be derived from applying an heuristic rule than those made following systematic evaluations of the decision information (Tversky and Kahneman, 1974; Plous, 1993; Baron, 1994). Judgements influenced by the decision context rather than based upon the decision information are open to counter-arguments and are more likely to change (Eagly and Chaiken, 1993). For decisions that have serious negative consequences, an individual may experience greater post-decision regret or dissatisfaction when decisions are based on these labile judgements.

Decision making under uncertainty is one area where individuals are likely to employ a number of heuristics when making judgements about risks or probabilities (Tversky and Kahneman, 1974; Baron, 1994; Schwarzer, 1994; van der Pligt, 1994; Croyle, Sun and Hart, 1997). The concern is that the use of these heuristics may lead to biased judgements which, in turn, affect the quality of the decision made (Eiser and van der Pligt, 1988). There are a number of texts that adequately describe the heuristics and biases consistently employed by individuals when confronted with certain decision problems that lead to systematic errors in judgements and decision making (Tversky and Kahnemen, 1974; Tversky, 1978; Pious, 1993; Baron, 1994; van der Pligt, 1994; Croyle, Sun and Hart, 1997). Two examples are used to illustrate how aspects of the decision context may be associated with biases in women's judgement of the risk information referred to during decision making about diagnostic testing.

The anchor and adjustment heuristic refers to the phenomenon that individuals make judgements about risk with reference to information from outside the decision domain

(Tversky and Kahneman, 1974; Eiser and van der Pligt, 1988). To explain further, a woman's perception of her triple test risk figure is likely to be influenced by her experience of fetal abnormality risks. For example, there are a number of risks a woman may refer to when evaluating her triple test result, such as her age-related risk of Down's syndrome, the triple test screen positive cut-off point, a friend's triple test result or a previous triple test result. If a woman's age-related risk is greater than the triple test risk, she may perceive her triple test result to be a 'low' risk. However, if her triple test risk is greater than that of a friend's, she may perceive the result to be 'high'. Consequently, prior experience or information about risks of abnormality may be important in women's judgement of their triple test risk figures and subsequent decisions to have or not have prenatal diagnosis.

Framing concerns the presentation of the decision information (see Baron, 1994; Kuhberger, 1997). Individuals make different judgements about the decision information depending on whether it is 'framed' negatively or positively (see Eiser and van der Pligt, 1988; Pious, 1993; Kuhberger, 1997). For example, both of the following are explanations of the same triple test risk figure: there is a 1 in 100 chance that the baby has Down's syndrome; there are 99 chances out of 100 that the baby is healthy. It is likely that a woman's judgement of her risk figure would differ in response to the way the risk information was presented or framed.

The second finding of significance for the issue of informed decision making concerns the strategies employed by individuals to combine the decision information. One of the assumptions of EUT is that when individuals are making a decision they will be motivated to choose the 'optimal' choice, the alternative with the maximised expected-utility. Performing such a task is complex, requiring time and large cognitive effort (Janis and Mann, 1977). Considering individuals' cognitive limitations, it is unlikely that the decision maker has the resources available to weigh up systematically the advantages and disadvantages of each alternative. Simon (1955) suggested that individuals employ a less cognitively demanding strategy when evaluating alternatives, that of *satisficing* (see Simon, 1992; Janis and Mann, 1977; Garnham and Oakhill, 1994). When *satisificing*, individuals are believed to have a criterion with which they compare the information about the decision alternatives. When the criterion is met, the alternative is selected. As this strategy compares alternatives on only one dimension and screens out the other attributes it is cognitively less demanding than the proposed combining of components in EUT (Eiser and van der Pligt, 1988; Garnham and Oakhill, 1994). For example, if a woman's goal is to know for certain whether or not her baby

has Down's syndrome then a satisfactory alternative would be to choose to have a diagnostic test. As the 'no further testing' alternative does not meet the 'certainty' criterion, information about this alternative is screened out of the decision making process. The empirical evidence suggests that individuals do employ non-optimising strategies when assimilating or 'trading-off' information to make decisions (see Baron ,1994; Pious, 1993; Garnham and Oakhill, 1994). If women are screening out information about one of the alternatives in the diagnostic test decision, it is unlikely that they are making an informed decision to have or not have prenatal diagnosis.

In response to the descriptive evidence indicating that individuals do not make decisions in accord with EUT, research has progressed in three directions to explain decision making under uncertainty (Beach and Lipshitz, 1993; Neumann and Politser, 1994). Researchers in one area both accept the explanation for normative or ideal decision making provided by EUT and acknowledge that individuals' behaviour deviates from the optimum choice. However, these researchers or 'decision analysts' aim to bridge the gap between the actual and optimum decision by changing the individuals' behaviour following the application of a decision aid during decision making. A second area of research has developed following the rejection of EUT as an appropriate model from which to understand 'Psychological Woman's' decision making. The naturalistic decision making (NDM) researchers have developed a number of alternative models to understand and explain decision making by describing the processes individuals use when making 'real-world' decisions. A third area of research has focused on modifying the attributes and components of EUT, in light of the descriptive findings on individuals' decision making. Although these researchers want to retain the logic underlying the components of EUT they also want the theory to be more predictive of individuals' behaviour. Each of these responses is discussed in more detail below.

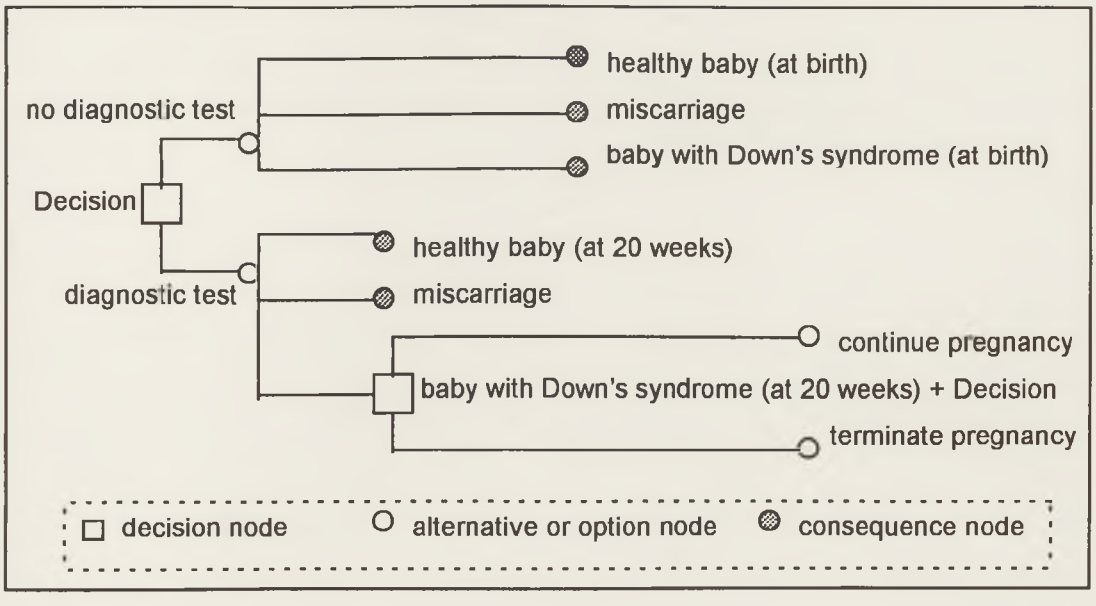
1.2.1.3 Using decision aids to modify behaviour.

Researchers supporting EUT as the standard with which to evaluate the quality of decisions made under uncertainty acknowledge that individuals make biased judgements that lead to poor choices, i.e. decisions that deviate from alternatives with the maximum expected-utility (Neumann and Politser, 1994; Golub, 1997). However, as behaviour is modifiable, it is reasonable to suggest that individuals can be encouraged to make decisions in accord with EUT. Within the medical context, both decision and information aids have been used to provide clear information that enables the individual to understand the decision alternatives and problems (O'Connor, Llewellyn-Thomas and Drake, 1995). Decision aids differ from information aids by: emphasising the individual's

responsibility to choose between alternatives, by explicitly representing all the alternatives and their consequences, by alerting the individual to the risks or probabilities of the consequences, by eliciting the individual's values associated with the consequences and by encouraging the individual to make a decision founded on this information (Homes-Rovner, 1995; O'Connor, Llewellyn-Thomas and Drake, 1995). In essence, the purpose of a decision aid, technique or support is to help the individual make a difficult or hard decision (von Winterfeldt and Edwards, 1986; Pitz, 1987; Eiser and van der Pligt, 1988; Homes-Rovner, 1995; O'Connor, Llewellyn-Thomas and Drake, 1995) rather than increase knowledge.

Decision analysis is the most established of the decision aids (Holmes-Rovner, 1995). It was developed to encourage individuals to make 'better' or more rational decisions (von Winterfeldt and Edwards, 1986; Clemen, 1990), i.e. decisions in accord with EUT (von Winterfeldt and Edwards, 1986; Jungermann and Schutz, 1992; Ubel and Loewenstein, 1997). It is beyond the remit of this chapter to explain the steps involved in applying decision analysis to real world decisions in detail (see von Winterfeldt and Edwards, 1986; Clemen, 1990; Baron, 1994; Golub, 1997; chapter 6). However, discussed briefly below are a number of aspects of the decision analytic technique that may enable individuals to employ more systematic processes and rely less on heuristic methods when making a decision, i.e. to facilitate good decision making.

Figure 1: Decision tree representation for the prenatal diagnosis decision.



First, as few 'real world' decisions present themselves as well defined choices between a given set of alternatives (von Winterfeldt and Edwards, 1986; Baron, 1994; Golub, 1997), some effort is spent on structuring the decision problem. Usually a third party or 'decision analyst' identifies a number of alternatives that may help the individual or 'decision maker' to make the decision (von Winterfeldt and Edwards, 1986; Pauker and Pauker, 1987; Golub, 1997). The decision problem is broken down into components: decisions or the choice between alternatives; consequences or uncertain events; objectives or goals of making the decision (von Winterfeldt and Edwards, 1986; Baron, 1994; Golub, 1997). The association between these attributes is usually summarised visually by the construction of a decision tree. For decisions made under uncertainty, the likelihood or probability for each consequence occurring is recorded (Pauker and Pauker, 1987). An example of how the decision to have a prenatal diagnostic test may be structured and represented is illustrated in figure 1.

This structuring of the decision problem ought to encourage the individual to evaluate all the alternatives that may help solve the problem rather than to focus on just one alternative that initially appears to satisfy some pre-defined criteria. In addition, the visual representation of the decision problem is continuously present during the decision making process, so enabling the individual to rehearse an exhaustive set of scenarios for each possible action (von Winterfeldt and Edwards, 1986). It is likely that the cognitive effort required to evaluate the decision information is reduced by having this visual prompt.

Second, the decision analyst asks the individual to make judgements about the decision information, i.e. elicits utilities about the consequences of each alternative (Pauker and Pauker, 1987; Baron, 1994). The utilities are influenced in part by the individual's judgement of the attractiveness or *aversiveness* of the consequence should it occur (Eiser and van der Pligt, 1988) and partly by their attitude towards the likelihood of the consequence occurring (von Winterfeldt and Edwards, 1986; Baron, 1994; Bromiley and Curley, 1994). The utility figures elicited are necessary for inclusion in the next stage of the decision analytic process: calculating the alternative with the greatest expected-utility.

The third stage is to calculate the expected-utility for each consequence node by multiplying the probability and utility values recorded on each arm of the decision tree. The expected-utility values associated with each alternative are added together. Selecting the alternative with the greatest expected-utility value is the correct or rational

choice. It is likely that less cognitive effort is required to make a decision when the probabilities and elicited utilities of the decision problem are integrated explicitly, i.e. external to an individual's information processing strategies (Pitz, 1986; Ubel and Loewenstein, 1997). Consequently, individuals should be able to evaluate the decision information more systematically. If individuals have not selected the option with the greatest expected-utility, they are encouraged to examine why their decision departed from the ideal decision. Such a process may lead to a re-evaluation of some of the decision attributes (Politser, 1981; Baron, 1994; Nease, 1996) and result in a decision more in accord with one theoretically derived from EUT.

Although there are several ways to evaluate utilities - including direct scaling, difference measurement, conjoint measurement and standard gambles (*see* Llewellyn-Thomas, Sutherlans, Tibshirani, Ciampi, Till and Boyd, 1982; Farquhar, 1984; von Winterfeldt and Edwards, 1986; Baron, 1994) - the most frequently used technique within the prenatal diagnosis context is that of the standard gamble (Pauker and Pauker, 1977; Pauker and Pauker, 1979; Pauker and Pauker, 1987; McNutt, 1989; Thornton, 1990a; Thornton, Lilford and Johnson, 1992; Baron, 1994). Individuals are asked to rank order the consequences of the decision from best to worse outcome (McNutt, 1989). Usually, the rank order of consequences for the prenatal diagnostic decision is: healthy child; spontaneous abortion; therapeutic abortion of affected fetus; therapeutic abortion of unaffected fetus; affected child (Pauker and Pauker, 1977; Pauker and Pauker, 1979; Pauker and Pauker, 1987). A continuous scale from 0 to 100 is used to provide a numerical value for the cost of the outcomes occurring. The gamble, reference lottery or hypothetical lottery is presented as follows: "If the probability of having a baby with Down's syndrome or without Down's syndrome is given as 50:50, would you (the woman) choose to carry on or terminate the pregnancy?" If the woman is able to make a choice between termination or continuing with the pregnancy, the gamble is varied until the woman is unable to answer the hypothetical question. For example, a woman may be unable to answer the question at a 25% chance of Down's syndrome and a 75% chance of the baby not having Down's syndrome; her level of indifference between the gamble and having prenatal diagnosis is 75%. The utility value associated with the prenatal diagnosis outcome elicited from this woman, then, is 0.75 on a scale from 0 to 1.0 (Pauker and Pauker, 1977; Pauker and Pauker, 1979; Pauker and Pauker, 1987; McNutt, 1989; Thornton, 1990b). Although a utility can be elicited for each consequence, Pauker and Pauker (1977) regard the cost or burden of a therapeutic abortion as the most important factor in women's decisions to have or not have prenatal

diagnoses. It is this 'therapeutic abortion utility' that is elicited during the prenatal diagnosis consultation.

The elicited utility value and the woman's probability of having a child with Down's syndrome are integrated with reference to a *threshold probability* (Pauker and Pauker, 1977, 1987; Thornton, 1990b). Pauker and Pauker (1977) used the equation for the threshold probability to create a *threshold graph* (see chapter 6). This graph provides the health professional with a visual prompt that is able to integrate the woman's utility of the therapeutic abortion (y axis) with her triple test risk figure (x axis). In brief, the point on the graph where the utility score and the probability figure meet either falls underneath or above the *threshold line*. The areas above or below the threshold line correspond to either the diagnostic test or to the no diagnostic test alternatives. In other words, a decision to have or not have prenatal diagnosis will be based on the woman's attitude towards prenatal diagnosis and her triple test risk figure.

Decision analysis has been applied to individuals' decision making within a number of clinical settings, including the domain of prenatal diagnosis (Pauker and Pauker, 1977, 1978, 1987; McNutt, 1989; Thornton and Lilford, 1990; Heckerling, Verp and Hadro, 1994; Morabia, Steinig-Stamm, Unger, Slosman, Schneider, Perrier and Junod, 1994; Verp and Heckerling, 1995). However, the findings of two recent systematic reviews on patient decision making (O'Connor, Llewellyn-Thomas and Drake, 1995; Bekker, Thornton, Airey M, Connelly, Hewison, Lilleyman, Macintosh, Maule, Michie, Pearman and Robinson, 1999) suggest that no studies have actually evaluated whether or not a consultation guided by the decision analytic technique is more likely to result in a 'better' or more rational decision than a decision made without assistance from a decision aid. It is equally feasible that, as individuals do not naturally make decisions in accord with EUT, encouraging them to do so may actually hinder the decision making process. Decision analysis, then, may be an inappropriate aid to facilitate decision making as it is based on EUT and aims to encourage accurate or rational decision making (Jungermann and Schutz, 1992; Beach and Lipshitz, 1993)[5]. The debate between proponents and critics of decision analysis needs to be addressed by the empirical investigation of decision quality following patient decision making, with or without the application of the decision analytic technique.

[5] The theoretical criticisms and clinical concerns pertaining to the decision analytic technique are described in more detail in chapter 6.

1.2.1.4 Naturalistic decision making (NDM) models.

As mentioned, classical decision theory was an untenable model for researchers interested in understanding how individuals make decisions and in describing the strategies and processes employed during decision making. Classical decision theory is concerned with only one aspect of the decision process, the decision event (Orasnu and Connolly, 1993). Its main focus is on ensuring that the predefined alternatives and consequences have been weighed up correctly, so achieving the optimum choice (Beach and Lipshitz, 1993). In 'real-world' settings, it is likely that information about the decision alternatives is unavailable, incomplete, irrelevant or too complex for rational decision making to occur. (Zey, 1992; 1998). Further, there are many different types of 'real-world' decisions and few require such a detailed trade-off between the benefits and costs of the decision attributes (Beach and Lipshitz, 1993).

Researchers interested in describing individuals' decision making perceive classical decision theory to be inappropriate, not because individuals are irrational but because it fails to capture the complex and adaptive nature of human decision making (Cohen, 1993). It is argued that, as classical decision theory does not describe individuals' decision making, it is an inappropriate standard with which to evaluate the quality of the decision made (Zey, 1992; Beach and Lipshitz, 1993; Cohen, 1993; Frisch and Clemen, 1994). A response of researchers to the limitations of classical decision theory has been to develop alternative models that describe the cognitive processes employed during decision making, i.e. naturalistic decision making (NDM) models. Although it is beyond the scope of this thesis to explore any one NDM theory in detail (see Klein, Orasanu, Calderwood and Zsambok, 1993; Crozier and Ranyard, 1997), two of the important contributions to understanding individuals' decision making as a result of the NDM research are discussed.

First, new methodologies have been developed to access the data with which to inform the NDM models. The models are concerned with understanding the process of decision making, i.e. tracing the train of thought or reasoning that led to the final decision (Payne, 1980; Harte, Westenberg and van Someren, 1994; Harte and Koele, 1997). As cognitions cannot be measured directly, 'process tracing' techniques for observable phenomena have been developed. Data are extracted from either verbal protocols or information search methods (Payne, 1980, Carroll and Johnson, 1990; Baron, 1994; Harte and Koele, 1997). The verbal protocol technique requires the individual to 'think aloud' while making a decision. Individuals are encouraged to verbalise whatever thoughts enter their heads and are discouraged from evaluating or introspecting about

the thoughts (Payne, 1980; Carroll and Johnson, 1990; Crozier and Ranyard, 1997). The information search methods record the information searched or utilised by the individual when making the decision. There are a number of techniques to document the information searched by the individual: recording of eye movements; information or decision boards; computer programs; audio and video tape-recordings of inter-personal interactions (Payne, 1980; Carroll and Johnson, 1990; Crozier and Ranyard, 1997; Harte and Koele, 1997). Information search techniques require less input from the decision maker than verbal protocols. Although time-consuming, these methods are able to record the decision making process over time, concurrently with the decision being made.

Second, by describing the decision making process, researchers have a more comprehensive understanding of how individuals make decisions and the factors that impact on the decision making process and outcome. One of the most significant observations from the wealth of research in this area is the interaction between the decision context and the decision making process. The strategies[6] and processes individuals employ when making decisions are a function of the demands of the decision and factors associated with the decision context, such as: how much expertise or decision-specific knowledge individuals have; their emotional state while making the decision; whether or not the decision is time-pressured; the seriousness and permanence of the consequences; the personal relevance of the decision consequences; the way the decision information is presented (see Klein, Orasanu, Calderwood and Zsambok, 1993; Lipshitz, 1993; Raynard, Crozier and Svenson, 1997). In addition, most NDM models suggest that individuals create mental images or schema of the decision which include information about the alternatives and consequences as well as their personal goals and preferences (Klein, Orasanu, Calderwood and Zsambok, 1993). The use of schema implies that decision making is a more dynamic process than selecting the best choice from given alternatives, as strategies and alternatives are appraised with reference to the schema and subsequently either rejected from or integrated into the decision image.

Although it is feasible that the strategy of maximising utility is employed to make some decisions (Ubel and Loewenstein, 1997), no single theory is able to understand and facilitate all the different types of real-world decisions (Lipshitz, 1993). NDM researchers argue that, by explaining how individuals actually make decisions and understanding why poor decisions occur, more appropriate aids and alternative standards can be

[6] The cognitive strategies employed during decision making are discussed further in chapter 5.

developed to facilitate and evaluate the quality of the decision made (Beach and Lipshitz, 1993). However, operationalising a NDM model for a decision aid may be difficult as the decision context and process are interrelated (Klein and Woods, 1993); it is possible that each decision could require a different decision aid. As yet, there is little empirical evidence to suggest that a decision aid grounded in NDM theory facilitates the decision making process or leads to more informed decisions than either one structured using decision analysis or one that is unaided (O'Connor, Llewellyn-Thomas and Drake, 1995; Bekker, Thornton, Airey M, Connelly, Hewison, Lilleyman, MacIntosh, Maule, Michie, Pearman and Robinson, 1999).

1.2.1.5 'Synthesis' models of decision making.

There are a number of psychological models that have modified EUT to account for the systematic variations in decision making as documented by empirical findings. Some examples are: prospect theory (Kahneman and Tversky, 1979); regret theory (Bell, 1982; Loomes and Sugden, 1982); the health belief model (HBM) (Becker, 1974); the theory of planned behaviour (TPB) (Ajzen, 1985). These models retain the assumption that an individual's behaviour is consistent with the goals, expectations and values of the decision maker (van der Pligt, 1998). The models are also descriptive because they attempt to identify some of the psychological processes associated with individual's decision making, such as perceptions of risks, attitudes, intentions, self efficacy, and perceptions of normative behaviours (see Conner and Norman, 1996). As these factors are implicit, measurement requires 'elicitation' using questionnaire scale methods. Several of the models have been applied within the area of health to understand and explain health decisions or behaviour. In addition, these models have been used to inform interventions with the purpose of changing behaviour (see Marteau, 1989; Stroebe and Stroebe, 1995; Conner and Norman, 1996). Although these 'synthesis' models are better able to describe the decision making process, they do not aim to explain or operationalise informed decision making. In essence, these models are concerned with predicting health behaviours and not providing a standard with which to evaluate the quality of the decision made.

1.2.2 Defining informed decision making (IDM).

One goal common to each of the research areas discussed is to facilitate 'good' decision making. Unfortunately there are no established, objective criteria with which to evaluate the quality of a decision (Vleck, 1987; Pitz, 1987; Llewellyn-Thomas, 1995; Broadstock and Michie, in press). The quality of a decision may be evaluated along the following dimensions: behavioural measures such as whether or not an alternative was

selected or length of decision making; cognitive measures such as knowledge, perceived risks, perceived benefits, accuracy of decision making and informed decision making; affective measures such as anxiety, satisfaction or decisional conflict (O'Connor, 1994; Llewellyn-Thomas, 1995). Selecting a measure to evaluate the decision quality depends largely on the theoretical perspective of the researcher rather than on an objective standard of good decision making (Llewellyn-Thomas, 1995).

The standard with which to evaluate the quality of the decision within the prenatal and genetic testing context is to ensure that individuals make informed choices (Royal College of Physicians, 1989; American Society of Human Genetics, 1996). As mentioned earlier, informed decision making has not been adequately operationalised within the applied setting (Marteau, 1995; Broadstock and Michie, in press). However, a standard of good or 'ideal' decision making has evolved from the research into theories of decision making, the components of which correspond with those criteria considered necessary for informed decision making to occur. The development of this standard is discussed in more detail below.

As mentioned, the criticisms of classical decision theory encouraged decision research to focus more on understanding the decision process than on the decision outcome (Baron, 1994). A good quality decision should be associated with a process of reasoned choice rather than with the selection of the rational alternative (Zey, 1992; 1998). Consequently, proponents of 'reasoned choice' models (Zey, 1992; 1998) suggest that an appropriate standard to evaluate decision quality ought to be based on the process of decision making (Janis and Mann, 1977; Beach and Lipshitz, 1993; Frisch and Clemen, 1994). After reviewing the literature on determinants of high quality decision making, Janis and Mann (1977) selected seven criteria for the 'ideal' decision making procedure:

1. to thoroughly canvass a wide range of alternative courses of action;
2. to survey the full range of objectives to be fulfilled and of the values implicated by the choice;
3. to carefully weigh whatever he [*sic*] knows about the costs and risks of negative as well as the positive consequences that could flow from each alternative;
4. to intensively search for new information relevant to further evaluations of the alternatives;
5. to correctly assimilate and take account of any new information or expert judgement to which he [*sic*] is exposed, even when the information or judgement does not support the course of action he [*sic*] initially prefers;

6. to re-examine the positive and negative consequences of all known alternatives, including those originally regarded as unacceptable, before making a final choice;
7. to make detailed provisions for implementing or executing the chosen course of action, with special attention to contingency plans that might be required if various known risks were to materialise.

More recently, criteria to evaluate the decision making process (Frisch and Clemen, 1994) suggest that a good decision ought to be based on (a) the relevant consequences of the different options *(consequentialism)*, (b) an accurate assessment of the world and consideration of all relevant consequences *(thorough structuring)* and (c) trade-offs of some kind *(compensatory decision rule)*. This standard of good or reasoned decision making provides an adequate operationalisation of informed decision making. To explain further, evaluating whether or not an individual has made an informed decision requires evidence that (a) the final decision was founded on the consideration of the desirability and likelihood of the consequences of all alternatives, (b) the structure of the decision information encouraged a more accurate consideration of the relevant consequences and reduced the impact of biasing processes that may alter evaluations of likelihood and desirability and (c) the decision was made as a result of trade-offs between these factors. In other words, an assessment of informed decision making requires a description of the process employed by the decision maker when reaching the decision.

1.3 Prenatal diagnosis and informed decision making.

The final section of this chapter summarises the empirical findings from research into women's prenatal diagnosis test decisions. The results are discussed with reference to the two factors considered necessary for informed decision making to occur: the provision of sufficient information by health professionals; the utilisation of consultation information by women to make a decision in accordance with their values.

1.3.1 The provision of sufficient information: empirical evidence.

Over the last twenty years, most of the research into prenatal diagnosis information giving has been within the context of genetic testing, counselling and decision making (Pauker and Pauker, 1978; Kessler, 1981; Lippman, 1991; Verjaal, Leschot and Treffers, 1982; Royal College of Physicians, 1989; Donnai, 1992; Harper, 1992; Clarke, 1994; British Medical Association, 1998). The genetic counselling information provision literature falls into three areas: the principles of genetic counselling and the type of information to be included within the ideal consultation (Emmery, 1984; Royal College

of Physicians, 1989; Harper, 1992; Clarke, 1994; British Medical Association, 1998); self-reports by genetic counsellors of the information they offer (Czeizel, Metneki and Osztovics, 1981; Verjaal, Leschot and Treffers, 1982; Wertz, Sorenson and Heeren, 1988; Rapp, 1989; Burke and Kolker, 1994); the impact of genetic counselling on women's reproductive decision making, perceptions of risk, attitudes and knowledge (Evers-Kiebooms and van den Berghe, 1979; Lippman-Hand and Fraser, 1979; Lubs, 1979; Swerts, 1987; Somer, Mustonen and Norio, 1988; Lippman, 1991; D'Amico, Jacopini, Vivona and Frontali, 1992; Frets, Duivenvoorden, Verhage and Niermeijer, 1992). Despite this breadth of research, few, if any, studies have described the information provided during the genetic counselling consultations (Kessler, 1992; Marteau, Plenicar and Kidd, 1993). In consequence, whether or not the information provided within the genetic counselling context was sufficient to enable individuals to make an informed decision remains an empirical question.

Although the majority of research into prenatal diagnosis information giving has taken place in the domain of genetic counselling, women deciding to have or not have a diagnostic test following a screen positive triple test result do so in the context of 'routine' prenatal care. There is a paucity of research describing and evaluating the provision of information within this health care setting. However, it is likely that the provision of prenatal diagnosis information will vary between this health care setting and that of the genetic counselling clinic, for at least two reasons. First, non-directive information giving about genetic diseases and risk figures is a necessary skill acquired by genetic counsellors during their professional training (Emmery, 1984; Rapp, 1988; Wertz and Fletcher, 1988; Clarke, 1994; Marteau, Drake and Bobrow, 1994; British Medical Association, 1998). Obstetricians and midwives do not receive the same education as genetic counsellors with regard to the communication of risk, explanation of genetic and chromosomal diseases, evaluation of reproductive alternatives and ethical principles of non-directive consultations. Empirical evidence does suggest that knowledge of genetic disorders, attitudes towards testing, termination and prenatal diagnosis, confidence in discussing these new technologies and directiveness of information giving does differ by medical discipline (Rapp, 1988; Firth and Lindenbaum, 1992; Green, 1994; Khalid, Price and Barrow, 1994; Marteau, Drake and Bobrow, 1994; Smith, Slack, Shaw and Marteau, 1994; Bernhardt, Geller, Doksum, Larson, Roter and Holtzman, 1998). Second, a woman receiving a screen positive triple test result for Down's syndrome requires different result-related information than a woman who has received a positive carrier test result. To explain further, a screen positive triple test result usually occurs within the context of a 'normal', low risk pregnancy. The test result

is a 'one-off' with few implications for subsequent pregnancies and relatives. In contrast, the result of a screen positive carrier test has life-long implications for an individual, each pregnancy and the carrier's relatives. For the purposes of this thesis, the following discussion focuses on describing the information considered necessary to ensure informed prenatal diagnosis decision making upon receipt of a screen positive triple test result and summarises the empirical research carried out to evaluate the quality of this information.

There is no one definitive set of guidelines describing the information considered sufficient for women to make an informed decision about prenatal diagnosis following receipt of a screen positive triple test result. In consequence, the literature on information provision for both serum screening (Royal College of Obstetricians and Gynaecologists, 1993; Kennard, Goodburn, Golightly and Piggott, 1995; Sheridan, Williams, Caine, Morgan, Mason and Mueller, 1997) and prenatal diagnosis and genetic testing (Emmery, 1984; Harper, 1992; Royal College of Physicians, 1989; Donnai, 1992; Kessler, 1992; Abramsky, 1994; Clarke, 1994; Marteau, Shaw and Slack, 1995; British Medical Association, 1998) was referred to when compiling the list of information to be discussed with women before they decide, to accept or decline the offer of prenatal diagnosis. The common themes extracted from this literature suggest that the consultation content should include the following information:

- a description of the cause, prognosis and prevalence of Down's syndrome;
- an explanation of the triple test result with reference to the meaning of a screen positive, screen negative, 1 in 250 cut-off point, other abnormalities tested for, woman's actual risk figure for having a baby with Down's syndrome and the chances of receiving a screen positive in subsequent pregnancies;
- a discussion of the options to have or not have further diagnostic testing. Information about the subsequent procedures associated with each option (fetal anomaly scan, amniocentesis and chorionic villus sampling) should include details about the accuracy of a test, the likelihood of a test identifying an abnormality, the abnormalities identified, the associated risks, a description of the test procedure, the meaning of a positive and negative test result, confirmation about the method of receiving the results and timing of the test results;
- a discussion of the options available should women receive a positive diagnostic test result, i.e. the choice to terminate the pregnancy or continue with the pregnancy;

This information should be presented neutrally or non-directively to ensure the autonomy of the individual when making an informed decision about prenatal diagnosis

(Royal College of Physicians, 1989). That is to say, the health professional should be required to refrain from persuading a women to pursue a particular test alternative (Kessler, 1992). Directive information can be expressed both explicitly and indirectly. An example of an explicit directive statement is, 'I think it would be best if you were to have chorionic villus sampling'. There are several more subtle ways for health professionals to be directive, such as spending more time talking about one alternative, making value judgements about the woman's choice, expressing a preference for an alternative and implying that one alternative is the normative or more common choice (Kessler, 1992).

There is a paucity of research evaluating the provision of information by health professionals when offering prenatal diagnosis. Few, if any, studies have described the information given during a prenatal diagnosis consultation following receipt of a screen positive triple test result. However, the content of the information given during consultations with women at increased risk of having a baby with Down's syndrome because of raised maternal age has been described (Kessler, 1981; Marteau, Plenicar and Kidd, 1993; Bernhardt, Geller, Doksum, Larson, Roter and Holtzman, 1998). The findings from these studies suggest that although the risk of Down's syndrome and procedure related miscarriage of amniocentesis were consistently mentioned, issues pertaining to the condition of Down's syndrome and termination were discussed infrequently (Marteau, Plenicar and Kidd, 1993; Bernhardt, Geller, Doksum, Larson, Roter and Holtzman, 1998). In addition, consultations included misrepresentations of information or misinformation and an implicit assumption that 'at risk' women should undergo prenatal testing (Marteau, Plenicar and Kidd, 1993; Bernhardt, Geller, Doksum, Larson, Roter and Holtzman, 1998). Further empirical evidence suggests that not all obstetricians and midwives have complete knowledge of prenatal testing (Smith, Slack, Shaw and Marteau, 1994) From these findings, it seems unlikely that all women have received sufficient information to make informed choices about the decision to have or not have prenatal diagnosis and it is likely that women's decision making autonomy was, in part, compromised by the directiveness of some test information.

1.3.2 Women's utilisation of consultation information: empirical evidence.

As mentioned, there is limited empirical research describing women's prenatal diagnosis decision making following receipt of a screen positive triple test result. In consequence, the findings from studies assessing the decision to have prenatal diagnosis upon receipt of either a screen positive triple test result or because of raised maternal age were integrated. These two decision contexts were sufficiently similar as the women's choice

was between a risk of miscarrying following prenatal diagnosis and the 'increased' risk of having a baby with Down's syndrome as a consequence of either age or biochemical screening. However, differences between the two groups of women have been observed for some variables. Unsurprisingly, the mean age of women was lower in the biochemical screening group and the reproductive history more varied in the raised maternal age group (Evans, Pryde, Evans and Johnson, 1993; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994). In addition, women had lower anxiety scores when offered prenatal diagnosis as a consequence of raised maternal age (Beeson and Golbus, 1979). The relationship between maternal age and accepting or refusing prenatal diagnosis was unclear (Evans, Pryde, Evans and Johnson, 1993; Beekhuis, de Wolf, Mantingh and Heringa, 1994; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994); one study suggested that the tendency to refuse a diagnostic test increased with age (Beekhuis, de Wolf, Mantingh and Heringa, 1994) whereas another implied that younger women were more likely to refuse testing (Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994). There was no empirical evidence to suggest that women's decision making processes differed when choosing to have or not have prenatal diagnosis as a consequence of Down's syndrome screening test. In consequence, the following discussion includes empirical evidence from studies describing women's diagnostic choices as a consequence of raised maternal age and upon receipt of a screen positive triple test result.

The prenatal diagnosis studies have investigated factors associated with women's decisions to have: a diagnostic test or not (Lippman-Hand and Cohen, 1980; Bernhard and Bannerman, 1982; Verjaal, Leschot and Treffers, 1982; Swerts, 1987; Pauker and Pauker, 1987; French, Kurczynski, Weaver and Pituch, 1992; Beekhuis, de Wolf, Mantingh and Heringa, 1994; Julian-Reynier, MacQuart-Moulin, Aurran, Chabal and Ayme, 1994; Piggot, Wilkinson and Bennet, 1994; Halliday, Lumley and Watson, 1995; Jorgensen, 1995; Williamson, Harris, Church, Fiddler and Rhind, 1996; Kenen, Smith, Watkins and Zuber-Pittore, 1997); chorionic villus sampling or amniocentesis (Lippman, Perry, Mandel and Cartier, 1985; McGovern, Goldberg and Desnick, 1986; Sjogren and Uddenberg, 1988; Evans, Bottoms, Critchfield, Greb and Laferla, 1990; Evans, Pryde, Evans and Johnson, 1993; Burke and Kolker, 1993; Kolker and Burke, 1993; Heckerling, Verp and Hadro, 1994; Halliday, Lumley and Watson, 1995); amniocentesis only (Farrant, 1985; Nielsen, 1991); no diagnostic test (Volodkevich and Huether, 1981). As the main findings from these studies have been outlined in numerous texts (Green, 1990, 1994; Marteau and Slack, 1992; Marteau, 1995; Green and Statham, 1996;

Hewison, 1996; Shilou, 1996), the subsequent discussion focuses on how these empirical findings can inform the following: a description of how women make the decision to have prenatal diagnosis; the adequacy of the methodologies employed to assess informed decision making; the conclusions that may be drawn about women's informed decision making.

1.3.2.1 Describing women's decision making processes: empirical evidence.

Although none of the published studies have described the decision making process concurrently with women making the decision (Marteau, 1995), the findings do provide an idea of the factors considered by women when making prenatal diagnosis decisions. As the decision making models suggest (see Marteau, 1989; Baron, 1994; Shafir, Simonson and Tversky, 1994; Conner and Norman, 1996), women do appear to make decisions by weighing up the advantages and disadvantages of the alternatives available. Reasons given by women for choosing to have no diagnostic test were: not being offered the tests; negative attitudes towards termination of pregnancy; low perception of risk of having a child with an abnormality; high perception of the procedure-related risk of miscarriage; low perceived benefit of diagnosis (Volodkevich and Huether, 1981; Verjaal, Leschot and Treffers, 1982; French, Kurczynski, Weaver and Pituch, 1992; Julian-Reynier, MacQuart-Moulin, Aurran, Chabal and Ayme, 1994). Reasons given by women for deciding to have a diagnostic test were: high perceived risk of having a child with an abnormality; positive attitudes to diagnosis and termination of pregnancy; relief of anxiety or uncertainty; experience of an acquaintance with an abnormality; reassurance of a negative result (Nielson, 1982; Verjaal, Leschot and Treffers, 1982; Farrant, 1985; McGovern, Goldberg and Desnick, 1986; Swerts, 1987; Sjogren and Uddenberg, 1988; French, Kurczynski, Weaver and Pituch, 1992; Julian-Reynier, MacQuart-Moulin, Aurran, Chabal and Ayme, 1994; Jorgensen, 1995). Reasons for choosing amniocentesis over chorionic villus sampling were: lower risk of test-related miscarriage; less risk of fetal damage; confidence in physicians test-related skills (Lippman, Perry, Mandel and Cartier, 1985; McGovern, Goldberg and Desnick, 1986; Heckerling, Verp and Hadrow, 1994). Reasons for the choice of chorionic villus sampling over amniocentesis were: quicker notification of results so reducing period of uncertainty; earlier diagnosis resulting in first trimester terminations (Lippman, Perry, Mandel and Cartier, 1985; Burke and Kolker, 1993; Evans, Pryde, Evans and Johnson, 1993; McGovern, Goldberg and Desnick, 1986; Heckerling, Verp and Hadrow, 1994).

The empirical evidence from these studies suggested that other factors external to women's cognitive processes were also associated with women's prenatal diagnosis

decisions. First, some studies observed demographic variations in women choosing whether to have or not have a diagnostic test (Nielsen, 1982; Beekhuis, de Wolf, Mantingh and Heringa, 1994; Halliday, Lumley and Watson, 1995) and those choosing between chorionic villus sampling or amniocentesis (Evans, Bottoms, Critchfield, Greb and Laferla, 1990; Kolker and Burke, 1993; Heckerling, Verp and Hadrow, 1994; Halliday, Lumley and Watson, 1995). These variations in decision making by demographic characteristics may be better understood by explaining the psychological pathways that mediate the relationship (Rutter and Quine, 1994, 1996). Although most of these studies described the demographic variations by test decision, they did not adequately operationalise models of decision making to further knowledge of the psychological processes that may mediate the observed association. The findings suggest that women choosing to have no further diagnostic testing tended to have had more children and were older than women having the diagnostic tests; women choosing to have chorionic villus sampling rather than amniocentesis tended to be better educated, older, have a lower parity and a fewer number of miscarriages. This pattern of demographic differences by test decision was not observed across all studies (Sjogren and Uddenberg, 1988; Heckerling, Verp and Hadrow, 1995). It is unclear from these studies how the demographic variables and reproductive experiences of women were associated with women's decision making strategies and subsequent decisions.

Second, in some studies describing women's decisions to have prenatal diagnosis, women felt obliged to have a test (Farrant, 1985; Sjogren and Uddenberg, 1988; Santalahti, Hemminki, Latikka and Ryynanen, 1998). In other words, women who had prenatal diagnosis perceived the social or health professional norm to be in favour of testing. From the research to date it is unclear whether this finding implies that health professionals encourage women towards the prenatal diagnosis option or that women having prenatal diagnosis perceive the test information differently from those who choose to have no diagnostic test. It is likely that women's uptake of prenatal diagnosis is partly influenced by the health professional, as associations have been found between characteristics of obstetricians and referral patterns for amniocentesis (Lippman-Hand and Cohen, 1980; Bernhardt and Bannerman, 1982; Heckerling, Verp and Hadrow, 1994; Verp and Heckerling, 1995). However, the strength of this association has not been established for women's uptake of prenatal diagnosis following triple test screening within the UK. In addition, women's perceived directiveness of the prenatal diagnosis information provided by health professionals has not been adequately addressed in studies to date. It remains an empirical question as to whether

or not women perceive information provided about prenatal diagnosis as directive and what factors may be associated with this perception.

An associated research question concerns the relationship between women's perceived directiveness of prenatal diagnosis information and the observed directiveness of the information provided. Few, if any, studies have compared measures of the information's directiveness with women's perception of directiveness. As mentioned (section 1.1), guidelines suggest that health professionals provide non-directive information to encourage women's autonomy to make the decision (Royal College of Physicians, 1989; Harper, 1992; Clarke, 1994; Kessler, 1992). However, there is a line of argument to suggest that women may prefer the health professional to be directive (Czeizel, Metneki and Osztovics, 1981; Savage and Armstrong, 1990). Although few studies within the prenatal diagnosis context have adequately addressed this empirical question, the issue of preference for information and decision making has been investigated in other health contexts (Ende, Kazis, Ash and Moskowitz, 1989; Llewellyn-Thomas, McGreal, Thiel, Fine and Erlichman, 1991; Borgers, Mullen, Rijken, Eussen, Plagge, Visser and Blijham, 1993; Verheggen, Jonkers and Kok, 1996; Thompson, Pitts and Schwankovsky, 1993; Avis, 1994; Fallowfield, Hall, Maguire, Baum and A'Hern, 1994; Silverman and Altman, 1996). In brief, the findings suggest that most patients want to be informed about subsequent treatment but vary in their preferences for taking the responsibility for the treatment choice (Ende, Kazis, Ash and Moskowitz, 1989; Thompson, Pitts and Schwankovsky, 1993; Fallowfield, Hall, Maguire, Baum and A'Hern, 1994). This preference for decision making autonomy was lower in more severe illness and higher in decisions requiring less medical knowledge. In other words, patients have preferences about making treatment decisions but not regarding the receipt of information. It seems reasonable to assume, therefore, that women would want complete information about prenatal diagnosis but may differ in their preferences for making the testing choice.

1.3.2.2 Assessing informed decision making: empirical evidence.

This section describes the most frequently employed measures reported in these studies of women's prenatal diagnosis decision making. A number of methodological issues are discussed that suggest that informed decision making has not been adequately assessed within the published empirical research to date.

- All studies employed questionnaire or interview techniques to assess women's cognitions and characteristics either before or after the prenatal diagnosis decision. Although some studies evaluated these cognitions more than once (Verjaal, Leschot

and Treffers, 1982; Swerts, 1987; Sjogren and Uddenberg, 1988), no studies described women's utilisation of information during the prenatal diagnosis consultation. As no studies have described the decision making process it is unlikely that informed decision making has been evaluated.

- The majority of designs evaluated women's cognitions retrospectively, i.e. after the prenatal diagnosis decision had been made (Volodkevich and Huether, 1981; Nielsen, 1982; Farrant, 1985; Burke and Kolker, 1993; Julian-Reynier, MacQuart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994; Jorgensen, 1995). Studies assessing memory for medical information (see Ley, 1988) suggest that it is unlikely that women were able to remember all the factors important to the decision in retrospect (Carroll and Johnson, 1990). Further, it is uncertain whether the reasons provided and recorded to support a decision made a week, a month, or a year earlier were the same as the reasons alluded to during the decision making period (Nisbett and Wilson, 1977; Yates, 1990). There is a body of evidence documenting the association between the performance of an action and subsequent changes to attitudes, such that the individual's attitudes become congruent with the behaviour (Eagly and Chaiken, 1993). This process of reason re-alignment is founded on the concept of cognitive dissonance (Festinger, 1957) and suggests that post-hoc evaluations of the decision are unlikely to be comprehensive representations of the decision making process and, therefore, of informed decision making.
- Only a small proportion of the studies evaluated the psychological processes associated with the decision to have or not have prenatal diagnosis (Verjaal, Leschot and Treffors, 1982; Swerts, 1986; French, Kurczynski, Weaver and Pituch, 1992; Beekhuis, de Wolf, Mantingh and Heringa, 1994; Julian-Reynier, MacQuart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994; Jorgensen, 1995). As mentioned, most studies investigated factors associated with only one prenatal diagnosis alternative, either the decision to have a diagnostic test or the choice of having no further testing. A prerequisite of informed decision making is the evaluation of the consequences associated with both testing alternatives. As a significant proportion of the studies did not assess the decision to have or not have prenatal diagnosis, it is unlikely that researchers described all the attributes associated with the informed decision making processes.
- The measures referred to within studies were not designed to assess informed decision making. Most studies reported the behaviour or test decision women made, i.e. the decision outcome. Some studies employed measures to assess the decision making process, such as perceptions of risk (Volodkevich and Huether, 1981; McGovern et al, 1986; Swerts, 1986; Sjogren and Uddenberg, 1988; French,

Kurczynski, Weaver and Pituch, 1992; Burke and Kolker, 1993; Kolker and Burke, 1993; Heckerling, Verp and Hadrow, 1994; Julian-Reynier, MacQuart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994), perceived benefits, attitudes, utilities and reasons (Volodkevich and Huether, 1981; Swerts, 1986; French, Kurczynski, Weaver and Pituch, 1992; Burke and Kolker, 1993; Heckerling, Verp and Hadrow, 1994). Others reported women's knowledge of the testing procedure, perceived autonomy in decision making and need for further information (Volodkevich and Huether, 1981; Nielson, 1982; Verjaal, Leschot and Treffors, 1982; Farrant, 1985; Swerts, 1986; French, Kurczynski, Weaver and Pituch, 1992). No studies within the prenatal diagnosis context have yet reported a process tracing technique or measure that assesses informed decision making (Marteau, 1995). In consequence, the exact relationship between the current measures of the decision making process and that outcome with assessments of informed decision making remains unclear.

1.3.2.3 Women's informed decision making: empirical evidence.

From the previous discussion of research to date, it is clear that no studies have assessed women's informed decision making in the context of prenatal diagnosis. That is to say, there is no published empirical evidence that women utilised the consultation information and integrated this with their values about prenatal diagnosis before making a decision. However, some studies aimed to describe the quality of women's prenatal diagnosis decision making by assessing the accuracy of the decision, women's knowledge of testing and the degree of autonomy in decision making. The evidence suggests that more women made prenatal diagnosis decisions in accord with EUT or 'accurately' than did not (Pauker and Pauker, 1986; Heckerling, Verp and Hadrow, 1994), that women's knowledge of prenatal diagnostic tests was found to be varied and often incomplete (Volodkevich and Huether, 1981; Farrant, 1985; Swerts, 1986; French, Kurczynski, Weaver and Pituch, 1992) and that the final decision to have or not have testing was influenced or made by the health professional (Verjaal, Leschot and Treffers, 1982; Julian-Reynier, MacQuart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994). Whether or not these findings imply that women's prenatal diagnosis decisions were informed or not is unclear. Certainly, women's final decisions appear to be based on evaluations of test specific information but their knowledge of testing was incomplete and the decision was partly influenced by the health professional's opinion. It was unlikely, therefore, that all women made fully informed decisions to have or not have prenatal diagnosis.

1.4 Chapter summary.

One objective for health professionals offering prenatal diagnosis is to ensure that women make an informed decision about testing (Royal College of Physicians, 1989). This chapter has discussed the issues pertaining to informed decision making in the context of women choosing to have or not have prenatal diagnosis upon receipt of a screen positive triple test result. Both psychological theory and medical guidelines on informed decision making were referred to in order to provide a working definition of informed decision making. Briefly, an informed decision is one where the final decision was based on an accurate evaluation of the advantages and disadvantages of the consequences of all the alternatives and that the decision was associated with a trade-off between these factors (section 1.2). Two conditions are considered necessary to enable women to make an informed decision: the provision of sufficient information by health professionals for women to assimilate; the provision of non-directive information to encourage women's autonomy in making the decision. The theoretical review summarising the empirical evidence of women's prenatal diagnostic decision making indicated that few, if any, studies had defined and operationalised informed decision making, had described the information provided by health professionals, had assessed the information utilised by women when making the decision to have or not have testing or had evaluated factors associated with the facilitation of informed decision making. It remains an empirical question, therefore, as to whether or not women make an informed decision about prenatal diagnosis.

2. Facilitating informed patient decision making: an integrative review.

The purpose of the preceding theoretical review (see chapter 1) was to provide an overview of the main issues involved in describing, assessing and facilitating informed decision making within the context of prenatal diagnosis. A theoretical review is most frequently used to assimilate empirical evidence with theoretical assertions to inform subsequent research questions and provide the rationale for further investigation (Cooper, 1989). Theoretical reviews do not aim to be inclusive summaries of all the available findings; the empirical evidence is utilised selectively to support, counter and develop theories about the phenomenon under investigation. As a consequence, theoretical reviews focus only on prior research that is of direct relevance to the new study's research agenda (Cooper, 1989).

This chapter contains a description of the methods, results and conclusions derived from an *integrative review* (Cooper, 1989) of studies carried out to assess the facilitation of informed patient decision making (Bekker, Thornton, Airey, Connelly, Hewison, Lilleyman, MacIntosh, Maule, Michie, Pearman and Robinson, 1999). The purpose of an integrative, research or systematic review is to efficiently summarise a large amount of empirical evidence from many separate studies that are believed to address a similar research question (Cooper, 1989). An integrative review aims to include the findings from all empirically based studies fulfilling a given set of criteria to provide a comprehensive knowledge base of the quantity and quality of research addressing a particular research question (Cooper, 1989; Mulrow, 1995). The information contained within each study in an integrative review is approached and integrated more systematically than findings are utilised in a theoretical review. In essence, each study within an integrative review is the equivalent of the participant in primary, empirical research. To explain further, the information documented in an article[1] provides the data by case for a review; the cognitions of the participant are the data by case for a primary empirical study. Both cognitions and article information are extracted from the data source following the application of a questionnaire or *review coding sheet*. The questionnaire or review coding sheet is informed by the operationalisation of a research agenda. As each participant answers all the items within a questionnaire, so all items within the coding sheet are completed by the researcher utilising the information

[1] An article may contain more than one empirical study. However, for congruity in this text it is assumed that an article describes only one empirical study.

contained within each article. The extracted data from both sources can be synthesised following the application of either qualitative or quantitative statistical methods (Cooper, 1989; Clarke and Stewart, 1995; Eysenck, 1995).

This chapter describes in more detail the rationale for, and findings from, one of the systematic reviews referred to in the preceding chapter, *Informed decision making: an annotated bibliography and systematic review* (Bekker, Thornton, Airey, Connelly, Hewison, Lilleyman, MacIntosh, Maule, Michie, Pearman and Robinson, 1999). This review provided evidence to support the following assertions: no published studies have empirically evaluated interventions aimed at facilitating informed decision making within the prenatal diagnosis context (section 1.3.2); despite the rhetoric of informed consent and decision making, there is a dearth of research assessing techniques to facilitate informed decision making by patients within most health care areas (section 1.2.1.3). The aim of the informed patient decision making (IPDM) review, carried out by the author of this thesis and colleagues, was to provide a comprehensive bibliography of empirical research of interventions that might have an effect on informed health care decision making by patients. The IPDM review was commissioned and, after peer review, published by the UK Health Technology Assessment (HTA) National Health Service Research and Development (NHS R&D) programme. The *executive summary* of the review's report appears in Appendix I.

The paucity of studies explicitly evaluating interventions aimed at the facilitation of informed patient decision making resulted in the IPDM review being qualitative or 'illustrative' rather than quantitative (Cooper, 1989). The information systematically extracted from each article provided a summary of the types of interventions, measures, theories, decisions, study designs and health areas researched rather than concise conclusions based on the results of meta-analyses. Primarily, this review is a resource, a bibliography of articles to refer to when embarking on empirical research aimed at the explanation and/or facilitation of informed patient decision making. It is unlikely that any one research question or agenda would demand reference to all the categories of all the articles classified in the final IPDM review. For example, the categories in the IPDM review pertinent to this thesis are those that classify article information according to the use of decision aids within the intervention and prenatal testing as the context of health care. The main focus of this chapter is to synthesise the findings from those IPDM review articles of pertinence to this thesis. The findings will be integrated and reported in a manner consistent with the presentation of results in the published IPDM report (Bekker et al, 1999).

The structure of the remainder of this chapter begins with a description of the theoretical issues that informed the IPDM review's research questions and coding sheet (section 2.1). Section 2.2 describes the methodology of the IPDM review: the criteria for inclusion of articles; the development of search strategies to identify articles that may be associated with the facilitation of informed patient decision making; the information categories developed to extract data systematically from articles. Section 2.3 outlines the IPDM review procedure for extracting information from those articles that fulfilled the IPDM review's criteria. In addition, section 2.3 describes the procedure for the integration of findings from those IPDM review articles that were of pertinence to this thesis, i.e. interventions designed to facilitate informed decision making and/or interventions carried out in the context of prenatal testing. This chapter's results section (section 2.4) focuses on describing and synthesising the findings of those articles selected from the IPDM review that were of pertinence to this thesis. The final section (section 2.5) discusses the findings from the further analysis of those articles selected from the IPDM review that were of pertinence to the thesis. Some general observations are made concerning the effectiveness of published empirical research to facilitate informed decision making.

2.1 IPDM systematic review: background.

The purpose of this IPDM review was to provide an integrated summary of information from intervention studies that may plausibly influence informed patient decision making (Bekker et al, 1999), so highlighting those areas that have been well- or under-researched (Mulrow, 1995). Following numerous meetings, reference to the decision making literature (see chapter one) and utilisation of the IPDM review members' expertise[2], several categories of information were identified as useful in understanding and facilitating informed patient decision making: factors associated with a patient's ability to make informed decisions; ways of measuring or assessing informed decision making; the theoretical framework or model of the primary research; the study quality; a summary of the empirical findings. These pre-defined categories were used to guide the identification of appropriate articles, inform the structure of the review coding sheet and classify information extracted from published empirical research. The operationalisation of these categories should enable the systematic extraction and integration of

[2] The IPDM steering group included a multi-disciplinary team of senior researchers with expertise in decision making and health services research. I was employed on the review to integrate these different perspectives and operationalise the ideas formulated in meetings by defining the inclusion criteria of articles, developing the coding frame, extracting article information, inputting the data, analysing results and co-writing the final report (see acknowledgements).

information from published empirical research. The pre-defined categories were not designed to identify an exhaustive list of factors associated with informed decision making. However, the data provided by this process should be sufficient for a review of interventions associated with the facilitation of informed patient decision making. A more detailed explanation of the type of information contained within these categories is described below.

2.1.1 Factors associated with informed decision making: IPDM review.

Prior evidence suggested that a number of factors might be associated with a patient's ability to make an informed decision, those pertaining to the *decision context*, the *decision maker*, and *other influences* (see Yates, 1990; Baron, 1994; Ranyard, Crozier and Svenson, 1997; chapter 1). Systematically extracting information about these factors from each study might be useful in subsequent explainations of the effectiveness of interventions to facilitate informed patient decision making.

The following features of the *health decision context* might be associated with the likelihood of patients making more or less informed decisions:

- **type of health decision.** For example, it is likely that the following decisions differ in the degree of motivation and responsibility required to carry out the action: smoking or drinking alcohol; attending a health-care appointment; adhering to medication; having a diagnostic test; participating in choice of treatment and so on;
- **seriousness of the outcome.** For example, choosing whether or not to take a headache pill has less serious consequences than deciding to donate an organ;
- **familiarity with the decision.** For example, deciding to exercise or not is more commonly experienced by most individuals than choosing to have or not have a genetic test;
- **level of certainty.** For example, the consequences of declining insulin always makes diabetics ill whereas declining prenatal screening only carries a risk of having a Down's syndrome child;
- **health domain.** For example, differences in the decision context, such as the type of medical speciality - medicine, surgery or primary care;
- **recipient.** For example, an individual making a decision that affects the individual's health may require different decision making strategies compared with an individual making a decision for a third party such as a child.

Factors pertaining to the individual that might be associated with the likelihood of making an informed decision are: preferences for information; preferences for involvement in the decision making process; beliefs about and attitudes towards health professionals, health and illness (see chapter 1; Bekker et al, 1999). In other words, whether or not a person utilises the information considered necessary to be able to make an informed decision depends in part on their motivation to engage in the decision making process and their beliefs about health, illness and the doctor-patient role.

Other influences that might be associated with the likelihood of a decision being informed are those that impact on strategies employed to process information and those that limit the availability of options from which to make the decision. Such factors as the presentation and quantity of information, an individual's mood state and time pressure might influence whether information was processed systematically or whether an heuristic was employed (see Ranyard, Crozier and Svenson, 1997; chapter one). On the other hand, legislation, provision of services and payment for services are factors that might limit the alternatives available for inclusion in some patient's decision making (Bekker et al, 1999).

2.1.2 Measures of informed decision making: IPDM review.

Prior reviews suggested that the most frequently employed measures used to assess informed decision making were: behaviour; knowledge; utilities and attitudes; affect; satisfaction or regret (Llewellyn-Thomas, 1995; Marteau, 1995; Bekker et al, 1999; see chapter 1). In general, most of these measures evaluate the outcome of the decision made, whereas informed decision making requires some evaluation of the process of reaching the decision (see chapter 1). It is unclear how many studies have employed a process tracing technique to evaluate the information utilised during decision making. It therefore remains an empirical question as to how many studies have actually assessed informed decision making. Without adequate operationalisation or measurement, it is unlikely that studies have comprehensively investigated the facilitation of informed decision making. The limitations specific to each measure in providing an assessment of informed decision making are described in more detail below:

- recording a health behaviour indicates that a decision was made but no more than this;
- knowledge assesses the individual's ability to recall information provided by the health professional, not the information utilised when making a decision. The

relationship, if any, between measures of knowledge and informed decision making has yet to be established;

- utilities assess an individual's values or attitudes towards the choices available. These measures evaluate an attribute that may be used during the decision making process. As an informed decision is made following the integration of an individual's attitudes with the knowledge of the consequences of the available alternatives (see chapter 1), it is likely that measures of utility assess some aspect of the decision making process. However, a measure of utility cannot evaluate whether or not that attitude or value was utilised in the decision making process or allude to other information referred to when making the decision;
- affect such as anxiety, satisfaction, regret and decisional conflict are measures of affect taken before and after decisions have been made. As discussed, there may be associations between increased affect and the likelihood of employing systematic information processing strategies. However, these are measures of outcome which may have some clinical significance but do not assess either the utilisation of information or integration of attitudes during the decision making process.

2.1.3 Models of decision making: IPDM review.

A number of decision making models have been developed from both theory and empirical evidence to explain an individual's behaviour (see chapter 1). Facilitating informed patient decision making depends, in part, on explaining how patients currently make decisions and how they can be assisted to make better decisions (Bekker et al, 1999). These models have described several psychological processes associated with an individual's health care decision making and factors that may hinder or facilitate informed decision making. It is argued that interventions aimed at changing or facilitating decision making are more likely to obtain their objectives if they have been informed by such theories of behaviour (Marteau, 1995). In addition, it is likely that studies operationalising a decision making model within the applied clinical setting provide a more comprehensive explanation of the intervention and associated (non-) significant effects with which to inform subsequent research and/or modify existing models. As yet, the extent to which decision making theories have been operationalised or used to inform studies within the context of patient health behaviour is unclear.

2.1.4 Application of the IPDM review report.

As stated, the aim of the IPDM review was to provide a systematic integration of information from interventions that may plausibly inform patient health care decision

making (Bekker et al, 1999). The authors of the IPDM review anticipated that the review would be utilised in one of two ways. First, researchers might refer to the review's synthesis of findings to inform or address research questions raised with regard to informed decision making. The discussion of the IPDM review findings were structured with reference to five 'naive' questions. These questions were considered by the IPDM review's authors to be representative of the type of issues with which informed decision making researchers may be concerned:

- Can decision making be facilitated?
- Do people who have more information make better decisions?
- Does the way in which information is presented change the effectiveness of the decision?
- Does the context affect decision-making?
- Does the effectiveness of information on decision making vary by medical setting?

Second, that researchers might select articles from the bibliography for secondary or additional analysis to inform a more specific research agenda.

This chapter provides an example of how a selection of articles from the IPDM review has been analysed in more detail to inform a research agenda concerned with the facilitation of informed decision making in the context of prenatal testing. In particular, this secondary analysis focuses on the facilitation of informed decision making following the application of a decision aid to a health care decision. In consequence, articles were selected from the IPDM review's bibliography either because they included information about decision making in the *context of prenatal testing* or they *evaluated the effectiveness of a decision aid* to facilitate patient decision making (see section 2.4).

2.2 IPDM systematic review: methodology.

2.2.1 Study inclusion criteria

The study criteria were developed over four months, during which over 300 abstracts downloaded from the pilot Medline and Psychlit search strategies were circulated to all project group members. At monthly meetings, each IPDM review member fed back the inclusion or exclusion status of each abstract. When members disagreed, a discussion took place until a set of working rules was developed. At the end of this development phase, agreement between members on the inclusion and exclusion criteria was good. Nevertheless, in this area there are no pre-defined or obvious boundaries to be used for selection criteria and some of the 'cut off' categories may appear arbitrary. For inclusion, each study published within an article had to fulfil the following criteria:

- Study participants must be *patients*. A patient was defined as any individual making a health decision. Studies using university student participants were excluded. Studies assessing health professional decision making about another individual's care were excluded.
- A study must report a *behavioural measure of the health decision made*. This measure of reported or observed behaviour assesses decision making, a more inclusive category than informed decision making. The health behaviour outcome may include actual, intended or hypothetical decisions. This broad health behaviour outcome measure incorporates many decisions, including: smoking or not; adherence to medication; attendance for screening; choices between treatments like chemotherapy or surgery. Studies were excluded if the health behaviour measure only assessed the effectiveness of a pharmacological treatment such as one comparing the effects of placebo gum with nicotine gum on smoking cessation. In addition, studies that reported outcomes such as bio-physiological levels, knowledge, satisfaction, preferences, utilities or affect were excluded. Although it is likely that some of these measures are associated with decision making and are frequently used to infer informed decision making (see chapter 1, section 2.1), they are not direct measures of decision behaviour.
- A study assessing an *experimental intervention*. The definition of an experimental study was interpreted broadly to include any design that evaluated an 'experimental' group with a comparison group. Studies with one of the following six designs were included: randomised controlled trial (RCT) with a low risk of bias (RCTa, the method of randomisation was delivered by a third party, a computer or on opening a previously numbered, sealed, opaque envelope); RCT with an unknown risk of bias (RCTb, the method of randomisation was not clearly described); RCT with a high risk of bias (RCTc, approaching patients on alternate days or following the toss of a coin); non-randomised concurrent; historical studies or 'before and after intervention' with different samples; 'before and after intervention' studies with the same sample. Studies assessing predictors of a behaviour such as factors associated with breast screening attendance or preferences for treatment were excluded. The definition of an intervention was broad, to incorporate the many factors that may alter decision making: offering additional information; framing figures in different ways; patient use of a decision aid; comparison of information mediums; changes in the delivery of the health care service such as the provision of mobile units for mammography screening; legislation such as compulsory wearing of seat-belts and so on.
- The study was published in *English*.

2.2.2 Development of the search strategy.

Studies considered for inclusion within the IPDM review were first identified from abstracts generated from one of two sources: electronic databases and hand-searches of complete sets of journals. Articles were retrieved if the IPDM review criteria were met or the abstract contained insufficient information to assess the criteria.

Over six months a search strategy was developed for the *electronic databases* Medline, Psychlit and BIDS (social science). A pilot strategy had been developed for inclusion within the initial protocol from a set of keywords chosen by the project group. Additional keywords were included within the strategy, derived from 17 articles considered relevant to the field of informed patient decision making, 100 articles considered for inclusion within the review following application of the pilot strategy to Medline and hand-searching of the journal *Medical Decision Making*. The keywords of the final strategy were modified and expanded within each of the electronic databases. The final strategy included terms from three categories: decision making; health care users; comparative study designs (Bekker et al, 1999). The sensitivity of the search strategy was evaluated by comparing the number of articles identified by hand-searching a second journal, *Patient Education and Counselling*. The pilot strategy identified 7% of articles considered for inclusion while the final strategy identified 62%. This sensitivity figure was considered satisfactory, as electronic searches identify between 17%-82% of relevant articles (Cooper, 1995). However, these efforts to increase sensitivity resulted in significantly reducing the specificity of the final strategy as 17,860 abstracts were generated for the years 1991-1996 alone. Consequently, the application of the strategy was limited to articles published between these years to ensure completion of the IPDM review given the resources available.

Three journals were selected for *hand-searching* because of their relevance to the health care users decision making area: *Medical Decision Making*, *Patient Education and Counselling*, and *Preventive Medicine*. In the hand-search, every abstract within every volume was read and considered for inclusion. All volumes were hand-searched for the years 1986 to 1996 inclusive.

2.2.3 Development of the coding form.

To ensure that consistent information was extracted from each study of each article, a data extraction sheet or coding form was developed over a six month period (see Appendix II). The categories within the coding form were informed with reference to the

decision making and integration of research literature, and to the expertise of members of the project group. Each draft coding frame was piloted on articles derived from the search strategies and modifications agreed within the group monthly meetings. Following piloting, several categories were simplified or omitted because it proved impossible to achieve consistency between articles as the information published was neither described nor presented in a format compatible with the review's research questions.

2.3 IPDM systematic review: procedure.

2.3.1 Article identification.

All abstracts generated from the search strategies were assessed for inclusion by the first author. Articles were retrieved if the IPDM review criteria were met or the abstract contained insufficient information to assess the IPDM review criteria. The original research questions investigated by the authors of the primary articles were not evaluated, i.e. articles were included in this review independent of the primary authors' research aims. In other words, articles included in the IPDM review may not have been designed to evaluate interventions aimed at the facilitation of informed patient decision making. Batches of ten articles were sent in rotation to each member of the project group. The inclusion criteria were again assessed by that member of the project group and, if included, s/he completed a coding form for each study of each article. Completed forms were returned to the first author and checked. Classification disagreements were resolved by discussion. All IPDM review details and extracted information were entered into the project electronic database.

2.3.2 Data synthesis.

The results of the IPDM review were presented in four ways:

- descriptive summaries and listings of articles by study number, grouped by health domain, decision and theoretical context;
- identification of 'good' studies. Good studies reported an RCT design with a low risk of bias (RCTa), referred to a theory to inform the intervention and assessed at least one decision making measure. As an insufficient number of studies (n=5) fulfilled these criteria, no meaningful quantitative meta-analysis were applied;
- non-parametric analyses were used in an attempt to answer the five questions raised at the end of section 2.1;

- a trajectory of knowledge tracing the number of studies using different types of methodology, decision making theory and recording an informed decision outcome were analysed by publication year.

Most results were generated directly from variables that classified information extracted by the IPDM review coding form. However, two additional variables were created by combining data from these information category variables: theory operationalisation and reported effects. These variables were created to facilitate the integration of the IPDM review findings with the IPDM review research questions (section 2.1). The variables are described in more detail below:

- the *theory operationalisation* variable combined information with reference as to whether or not the authors mentioned a theory to inform their study or described measures to assess the decision making process. Four classifications were created: theory and decision making process measure; theory only; decision making process measure only; neither theory nor decision making process measure.
- the summary of *reported effects* variable was a crude record of whether or not the authors reported a significant effect between the intervention and health behaviour outcome measure.

The extracted information included within this chapter was derived from fewer articles than those for the IPDM review. The application of statistical analysis to these findings was not appropriate because of the small sample size. As a consequence, the content and structure of this chapter's results section differs from that of the IPDM review and includes:

- descriptive summaries of the following extracted information by article: authors, titles and journal; health care area and type of decision; study design and intervention description; theory reference and description of measures; summary of findings.
- frequency summaries of the information extracted from articles grouped by: study design; health care context; type of decision; content of intervention; reference to a theory; other measures assessed; theory operationalisation; theory operationalisation by reported effect size. As this chapter is concerned with the facilitation of informed decision making, an additional *reported effect* variable was created to assess whether or not the primary research reported an association between the intervention and measures of the decision making process;
- a qualitative synthesis of findings based on a more detailed description of the extracted information from those selected articles that operationalised a theory.

2.4 Integrative review: results.

The criteria for inclusion in the final report was met by 547 articles, published between 1991-1996[3]. The main findings from the IPDM systematic review are described briefly in its executive summary (Appendix I). The results reported in this chapter are specifically concerned with the findings of articles that either included a decision aid in the intervention or investigated decision making in the context of prenatal diagnosis.

2.4.1 Inclusion of articles: decision aids and prenatal testing.

As mentioned, articles of pertinence to this thesis are those that assess the facilitation of informed patient decision making and /or evaluate interventions carried out in the prenatal testing health context. No studies within the IPDM review were designed specifically by the primary-source authors to facilitate informed decision making in the prenatal testing context. However, a number of interventions were designed by the primary-source authors to evaluate changes either in the patient's utilisation of consultation information or in prenatal testing decisions.

Obstetrics, gynaecology and midwifery (n=31) and *genetics* (n=7) were the two IPDM review classification categories of the health context that may have included decisions about prenatal diagnosis (Bekker et al, 1999). The IPDM review tables summarising the information extracted from articles were referred to for further details about the health decision being made. The types of health decisions described in the *obstetrics, gynaecology and midwifery* category were associated with: breast feeding (n=9); substance use and/ or risk factor reduction (n=9); prenatal care (n=4); other post-partum care (n=3); menopause (n=2); issues about service provision changes (n=4). The types of health decisions described in the *genetics* category were associated with: uptake of carrier testing (n=4); awareness and coping with genetic diseases (n=2); genetic counselling and reproduction (n=1). Articles were selected if the health care context was associated with women's prenatal testing behaviour. Five articles described decisions carried out in the context of either prenatal care (n=4) or genetic counselling and reproduction (n=1) (table 1; Appendix III). These articles were selected for inclusion in this chapter's more in-depth analysis of the IPDM review bibliography.

Patient prompt (n=55) was the IPDM review classification category of interventions that aimed to change the patient's decision making processes or utilisation of consultation

[3] Articles extracted from the electronic database searches were published between 1991 and 1996. Articles identified after handsearching journals dated from 1986.

information. The tables summarising the information extracted from all 55 patient prompt articles in the IPDM review report were referred to for a more detailed description of the interventions. On further investigation, the patient prompt interventions were classified as: prompts to facilitate decision making (n=5); prompts to encourage active or shared participation in the doctor-patient consultation (n=5); reminder or memory prompts to increase adherence with medical regimens (n=45). Articles were selected if the intervention either facilitated patient decision making or increased active participation in the consultation. Of these studies, less than a third evaluated interventions aimed at encouraging patient participation in the consultation. In total, thirteen articles were selected for further analysis as their interventions included either a decision aid or specific information on being pro-active during a consultation (table 1, Appendix III). As none of these thirteen articles were carried out in the context of prenatal testing, eighteen articles were included in this chapter's further analysis of the IPDM review bibliography.

2.4.2 Summary of information: extracted information by study.

A description of the information extracted systematically from each article is summarised in Appendix III: table 2 includes information about the health context and the type of decision made; table 3, the study design, description of the intervention, number of experimental groups and sample size; table 4, the theory referred to and measures assessed; table 5 summarises the articles' main findings, i.e. the association, if any, between the intervention and the decision behaviour. In essence, this information provides the raw data by case for use in the following analyses.

2.4.3 Summary of information: frequencies.

Frequency analyses provide a simple integration of findings i.e. what proportion of studies assessed which variables. Frequencies of following classification categories are described: the health care context; the type of intervention; the study design; the type of decision; the theory used to inform study; the measures employed; the reported effects of the intervention on the decision behaviour.

Health care context (table 2.1)*:* five articles were selected for inclusion in this chapter on the basis of their health care context (either from *obstetrics, gynaecology and midwifery* or *genetics*). Of those studies selected for inclusion on the basis of their intervention, primary health care was the most frequent health care context, followed by oncology and then mental health.

Table 2:1 frequency of studies by health care context.

	n	(%)
obstetrics, midwifery & gynaecology and genetics	5	(28)
primary care	5	(28)
oncology	3	(17)
mental health	2	(10)
other (arthritis, geriatrics, diabetes)	3	(17)

Content of intervention (table 2.2)*:* most studies included in this results section were selected on the basis of their intervention (patient prompt). Despite this selection criteria, only a fifth of studies (4/18) evaluated the effectiveness of a decision aid in the facilitation of patient decision making. The majority of interventions included more than one manipulation, such as a change in the delivery of information and the use of a memory prompt. The most frequently employed intervention was to alter the amount of information the patient received.

Table 2:2 frequency of studies by content of intervention.

	n	(%)
change in number of visits	3	(17)
additional information (face-to-face, telephone, written, video)	16	(89)
group delivery information	1	(6)
decision aid	4	(22)
patient participation / information prompt	7	(39)
memory prompt	1	(6)

Study design (table 2.3)*:* one of the IPDM review criteria was to include only articles with an experimental study design (see section 2.2). The majority of articles subsequently selected for this chapter's analysis employed a randomised control trial design (16/18). However, only four adequately described the randomisation procedure and/ or used a procedure with a low risk of bias.

Type of decision (table 2.4): question asking during the consultation and utilisation of services were the most frequently assessed decision behaviours. Few studies systematically investigated patient choices between treatment options.

Table 2:3 frequency of studies by design.

RCTa low bias		RCTb unknown bias		RCTc high bias		Concurrent		Historical		Before/after same sample	
n	(%)	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)
4	(22)	3	(16)	9	(50)	1	(6)	0	(0)	1	(6)

Table 2:4 frequency of studies by decision investigated.

		n	%
utilisation services	(attendance appointments, call-out services)	6	(33)
question asking	(information seeking)	5	(28)
adherence regimens	(medication, diet, recommendations)	3	(17)
decisions about treatment		2	(10)
making living wills		1	(6)
reproductive decisions	(to have more children or not)	1	(6)

Theory driven research (table 2.5)*:* half of the articles did not refer to any theory when describing the study aims or methods. Only one study was informed by a theory of decision making.

Table 2:5 frequency of studies by type of theory.

	n	%
No theory	9	(50)
Doctor-patient communication	8	(44)
EUT	1	(6)

Type of measures evaluated (table 2.6)*:* all studies assessed a number of other measures besides the decision behaviour (see table 2.4). The most frequently reported measures concerned the demographic details of the patients. Less than half the studies employed measures to assess psychological strategies that might be associated with the process of decision making (decision making).

Theory operationalisation (table 2.7)*:* information from tables 2.5 and 2.6 were combined to create a variable assessing the operationalisation of theoretical driven research (see section 2.3.2). Less than half of the selected articles had used a theory to

inform the study *and* assessed either some aspect of the information utilised during the consultation or a decision making process measure.

Table 2:6 frequency of studies assessing other measures.

		n	(%)
demographics	age, sex, level education, employment	17	(94)
personal history	medical and family history	13	(72)
decision making	attitudes, self-efficacy, reasons, perception of risk, decision autonomy, decision confidence,	8	(44)
knowledge	knowledge, recall of information	5	(28)
satisfaction	perceived usefulness, satisfaction	11	(61)
affect	anxiety, depression, worry	5	(28)
individual differences	need for cognition, optimism, locus of control	2	(11)
other	social support, coping, clinician measures	4	(22)

Table 2:7 frequency of studies by operationalisation of theory.

	n	(%)
Theory + decision making process measure/ question asking outcome	8	(44)
Theory only	2	(11)
Decision making process measure/ question asking outcome only	3	(17)
No theory or decision making process measure/ question asking outcome	5	(28)

Reported effect (table 2.8)*:* two additional variables were created from the summary of the studies' findings to assess whether or not the intervention was associated with differences in the decision behaviour or decision making process measures (see section 2.3.2). Eleven studies (61%) reported a change in the decision behaviour by intervention group; seven studies (39%) reported a change in the measures of information utilisation or decision process by intervention group. Most of the studies (7/11) reporting an association between the intervention and decision behaviour did not operationalise a theory and/or assess the decision making process. All studies (7/7) reporting an association between the intervention effect and decision making process measures had operationalised a theory.

Table 2:8 frequency of studies reporting an association between the intervention groups and the decision behaviour or decision process measures.

	Behaviour Effect		*Process* Effect	
	n	(%)	n	(%)
Total number of studies.	11	(61)	7	(39)
Operationalised theory and measures (n=8)	*4*	*(17)*	*7*	*(39)*
Not operationalised theory and measures (n=10)	*7*	*(44)*	*0*	*(0)*

2.4.4 Summary of information: synthesis of findings.

The main purpose of this final results section is to integrate the findings of those intervention studies that may plausibly influence informed patient decision making. As stated, most of the articles included in this chapter were selected on the basis of their intervention (table 2.2). However, as the focus of this thesis is on understanding informed patient decision making within the context of prenatal diagnosis, five additional studies associated with women's prenatal testing behaviour were also selected for inclusion in this chapter's analysis (table 2.1). None of these five articles evaluated interventions aimed at changing women's involvement in the decision making consultation. In consequence, the subsequent synthesis of findings first briefly summarises the results of the five articles selected on the basis of their health care context and then focuses on those studies that may address issues pertaining to informed patient decision making.

2.4.4.1 Interventions in the prenatal testing context: synthesis of findings.

The two health care categories of the IPDM review most likely to involve a decision about prenatal testing were (a) *obstetrics, gynaecology and midwifery* and (b) *genetics*. Following reference to the IPDM review report, five articles were selected for inclusion in this chapter's analysis of articles (Binstock and Wolde-Tsadik, 1995; Rowley, Hensley, Brinsmead and Wlodarczyk, 1995; Shiloh, Reznik, Bat-Miriam-Katznelson and Goldman, 1995; Sikorski, Wilson, Clement, Das and Smeaton, 1996; Thornton, Hewison, Lilford and Vail, 1995). No studies in this health care context assessed an intervention aimed at increasing patient involvement in a decision making consultation or referred to a decision making theory to inform the research. The interventions assessed can be broadly divided into those that evaluated either the delivery of care (Binstock and Wolde-Tsadik, 1995; Rowley et al, 1995; Sikorski et al, 1996) or

information provided (Shiloh et al, 1995; Thornton et al, 1995). Although no studies operationalised or evaluated informed decision making, two studies did assess measures associated with the decision making process (Shiloh et al, 1995; Sikorski et al, 1996). Most of the studies (4/5) reported an association between the intervention and decision behaviour. However, as these studies did not evaluate an intervention aimed at facilitating the patient's participation in the decision making consultation, it is beyond the scope of this thesis to explore these findings in more detail.

2.4.4.2 Interventions facilitating informed decision making: synthesis of findings.

As previously discussed (see sections 1.2; 2.1), informed patient decision making requires some assessment of the decision making *process*. In addition, it is likely that a better understanding of the decision process and outcome may be achieved if empirical research is informed with reference to a theory. In consequence, only the eight studies that operationalised a theory (table 2.7) are referred to in this synthesis of findings (Brennan et al, 1995; Butow et al, 1994; McCann and Weinman, 1996; Rost et al, 1991; Sander et al, 1996; Shepperd et al, 1995; Whelan et al, 1995; Tabak, 1988). A quantitative integration of results would be appropriate were the data within these studies homogenous, i.e. if the interventions, patients, decision, decision contexts, research questions and results were similar or comparable (Cooper, 1989; Eysenck, 1995). However, the studies were too few and disparate to carry out meaningful quantitative analyses. In consequence, the information extracted from each study is discussed in more detail with reference to: the design, theory, and measures employed; the decision behaviour and context; the type of intervention; the article's findings.

Design, theory, and measures: most of the studies (Brennan et al, 1995; Butow et al, 1994; McCann and Weinman, 1996; Rost et al, 1991; Sander et al, 1996; Whelan et al, 1995; and, Tabak, 1988) employed a randomised control design to evaluate the intervention. Only one study was designed to encourage 'better' decision making (Brennan et al, 1995); the remainder were designed to facilitate shared doctor-patient decision making and consultations. In addition, Brennan et al (1995) was the only study to refer to a specific theory of decision making; no studies operationalised informed decision making. Most studies (Butow et al, 1994; McCann and Weinman, 1996; Rost et al, 1991; Sander et al, 1996; Tabak, 1988) assessed question asking or information seeking during the consultation. Three additional variables were employed to evaluate the quality of decision making: confidence with the decision made (Brennan et al, 1995; Shepperd et al, 1995); perceived participation or autonomy in the decision making

(Butow at al, 1994; Whelan et al, 1995; Sander et al, 1996); individual preferences for information (Rost et al, 1991; Butow et al, 1994).

Decision behaviour and context: although it was argued in some articles that increased patient participation was associated with changes in lifestyle and medical regimen adherence, the primary decision behaviour for most studies (6/8) was the facilitation of information seeking or question asking (Butow et al, 1994; McCann and Weinman, 1996; Rost et al, 1991; Sander et al, 1996; Tabak, 1988). Only Whelan et al (1995) and Shepperd et al (1995) assessed differences in the choice of treatment by intervention group. Although Brennan et al (1995) assessed decision making on behalf of a third party, most studies focused on the decision made by the patient for the patient. Half of the studies (McCann and Weinman, 1996; Sander et al, 1996; Shepperd et al, 1995; Tabak, 1988) were carried out in the primary health care context. The remaining studies investigated decision making in the context of cancer (Butow et al, 1994; Whelan et al, 1995), diabetes (Rost et al, 1991) and Alzheimer's (Brennan et al, 1995). No studies operationalising a theory were carried out in the context of prenatal or genetic testing.

Type of intervention: all the interventions included changes in the provision of information, five using leaflets, one using information cards attached to a decision board, one using a computer link and one using a video-tape recording. Four studies included information prompts to specifically encourage patients to ask questions about their health issue during the consultation (Butow et al, 1994; McCann and Weinman, 1996; Sander et al, 1996; Tabak, 1988). The four studies classified as employing decision aids provided additional information about the consequences of the alternatives available structured, either by a decision tree or by a visual representation of the choices available (Brennan et al, 1995; Rost et al, 1991; Shepperd et al, 1995; Whelan et al, 1995). Although the focus of the decision aid interventions was the facilitation of patient decision making, only the computer link decision aid (Brennan et al, 1995) was concerned with helping the patient reach a decision. The computer link decision aid provided a technique to associate factual information about the consequences of alternatives with a patient's individual evaluations of these outcomes to obtain a 'best' or 'correct' decision as defined by expected utility theory.

Integration of findings: the results associating the interventions and facilitation of decision behaviour were mixed: two of the information prompt studies (McCann and Weinman, 1996; Sander et al, 1996) and two of the decision aid studies (Rost et al,

1991; Shepperd et al, 1995) reported an increased effect on patient participation within the consultation. Butow et al (1994) found that although the total number of questions asked was not associated with the information prompt intervention, the type of information sought did differ by group. Of those studies that assessed perceived autonomy and confidence in decision making, all of the decision aid studies reported a positive association (Brennan et al, 1995; Shepperd et al, 1995; Whelan et al, 1995) but the one information prompt study found no such relationship (Butow et al, 1994). Brennan et al (1995) found no association between the skill or 'correctness' of the decision made with the decision aid intervention. It seems unlikely that patient preference for information was associated with differences in the intervention groups (Rost et al, 1991; Butow et al, 1994).

2.5 Discussion.

The aim of the IPDM review was to provide a comprehensive bibliography of empirical research of interventions that might have an effect on informed health care decision making by patients. Between the years 1991-1996, 17, 860 abstracts were generated by the IPDM review's electronic search strategy. Following evaluation of each abstract, 547 articles were identified by members of the IPDM review working group as fulfilling the review's criteria. This chapter has been concerned with focusing on those articles that might facilitate informed patient decision making following the application of a decision aid to a health care decision. In total, 18 articles classified in the IPDM review's bibliography were pertinent to this chapter. The results reported in this chapter were based on a detailed analysis of the information extracted from these 18 selected articles.

As mentioned, the main advantage of carrying out an integrative, systematic or research review is to efficiently integrate findings from a large number of studies. Articles are selected for inclusion in the review according to the research agenda of the review. The review's research agenda also informs the coding sheet applied to each article used when systematically extracting study information. The integration of this information provides a comprehensive knowledge base for researchers to address the questions posed by the review (Cooper, 1989; Mulrow, 1995). The conclusions based on the findings of this type of review may be seen to be less biased than those derived from theoretical reviews. Integrative reviews include all articles that fulfil their inclusion criteria, whereas theoretical reviews select articles to support or counter their argument. In addition, it is likely that the article inclusion technique of integrative reviews encourages a wider range of articles to be considered for the final review than that of a

theoretical review, as integrative review articles are judged on their information content rather than on the intended aims of the primary-source authors. In other words, articles were included in the IPDM review because the information contained within an article fulfilled the review's criteria and not because the article's authors intended to evaluate the facilitation of informed decision making.

The main disadvantage of integrative reviews is the systematic exclusion of articles that do not fulfil the review's criteria. There are several areas of empirically-based studies that may have informed some of the research questions raised in the IPDM review, including: studies of patient's preferences for health care; comparative studies assessing predictors of behaviour; studies evaluating changes in an alternative outcome measure such as knowledge; experimental or student-based studies operationalising decision making theory (Bekker et al, 1999). It is likely that reviews in all of these areas would increase researchers' understanding of informed decision making and the strategies employed by individuals to make decisions. However, specific issues concerning the facilitation of informed patient decision making are most likely to be answered by a review carried out in the area of research identified by the IPDM review.

The remainder of the discussion focuses on general conclusions drawn from this integrative review's results pertaining to the facilitation of informed patient decision making in the context of prenatal diagnosis. The discussion is structured to address the following questions adapted from those described in the IPDM review report (see section 2.1.4):

- Can informed decision making be facilitated?
- Do decision aids facilitate informed decision making?
- What other factors are associated with informed decision making?

2.5.1 Can informed patient decision making be facilitated?

Although the interventions of studies included within the IPDM review may be associated with issues of informed decision making, none were explicitly designed to evaluate the facilitation of informed patient decision making. The purpose of those 13 articles selected for further analysis in this chapter on the basis of their intervention content was to either facilitate active patient participation in the consultation or to encourage better decision making. The aims of the five studies selected for further analysis in this chapter on the basis of their health care area were more varied and included assessing changes in attendance, reproduction and other pregnancy outcomes. None of the 18 studies defined informed decision making or attempted to

measure the process of informed decision making. Nine articles referred to a theory when designing the study but only one was a theory of decision making. In essence, there is little evidence that informed decision making was operationalised. It remains an empirical question as to whether or not informed patient decision making has or can be facilitated. However, findings from the 13 studies aiming to facilitate patient participation in the consultation can be used to inform some general observations concerning the association between the experimental interventions and changes in both decision behaviour and decision process measures.

The findings suggested a mixed association between the patient prompt interventions and changes in the decision process and outcome measures. About half of the studies (7/13) reported an association between the intervention and decision behaviour, 4/8 of those that operationalised a theory and 3/5 of those that did not. Of those studies aiming to facilitate active patient participation in the consultation, 3/5 reported an associated increase. Less than half of these studies (6/13) included other measures to evaluate the process of decision making. Most of these interventions (4/6) were associated with an increase in the quality of the decision making process. These results suggest that interventions aimed at encouraging active patient participation in the consultation were likely to be associated with reliable and positive changes in the quality of the decision making process rather than with increasing or decreasing the decision behaviour.

2.5.2 Do decision aids facilitate informed patient decision making?

There is insufficient empirical evidence to address this question. As stated, none of the 547 IPDM review articles systematically selected from 17,680 abstracts had operationalised informed patient decision making and designed an intervention to facilitate informed patient decision making. Of the eight articles that had described a patient prompt to facilitate active patient participation in the consultation and used a theory to inform the study, only four were classified as decision aids. All of the articles classified as decision aids did make explicit the issue of choice, the presentation of options, information about the risks and benefits of the options and the individual tailoring of information to the patients needs. Only one of these decision aids (Brennan et al, 1995) was used to explicitly gauge the patient's values associated with the proposed outcomes and assist the patient in reaching a decision. As all of these components are considered necessary for a patient prompt to be classified as a decision rather than information aid (see chapter 1; O'Connor et al, 1994), it is likely that the IPDM review's classification of interventions as decision aids was generous. In

other words, few studies have adequately operationalised a decision aid in an intervention-based study. Furthermore, no studies included in the IPDM review had adequately assessed the relationship, if any, between the employment of a decision aid and changes in patient's decision making processes.

Some general observations about the efficacy of decision aid interventions can be made with reference to the findings from the eight studies that operationalised a theory and assessed patient prompt interventions. In brief, four interventions were essentially information aids or prompts to encourage active consultation participation and four were decision aids to facilitate treatment decisions. Only half of these studies reported an association between the patient prompt and decision outcome measure, two studies included information prompts and two studies incorporated decision aid interventions. The results suggest that the information prompts and decision aids were equally likely to be associated with changes in the decision behaviour outcome measures and that factors other than the patient prompt interventions were associated with changes in the decision behaviour. Only four studies assessed some aspect of the decision making process, such as decision autonomy and confidence with the decision made. All three decision aids reported a positive association with these measures; the information prompt did not. These findings suggest that decision aid interventions are likely to facilitate some aspect of the decision making process but there is little empirical evidence to support such an association with information prompt interventions. Only one study (Brennan et al, 1995) assessed the 'theoretical correctness' of the decision made; no differences in measures of decision correctness as a consequence of using a decision aid were found. Until other studies have adequately operationalised and evaluated decision aids, this finding suggests that theory driven decision aids are not associated with the facilitation of theoretically correct decisions.

2.5.3 What factors are associated with informed patient decision making?

During the development of criteria for article inclusion and coding form content by IPDM review members, a number of pre-defined categories were suggested as factors that might be associated with a patient's ability to make an informed decision (see section 2.1; Bekker et al, 1999): aspects of the decision context, such as the type of health decision or riskiness of the decision; individual differences, such as preferences for information or attitudes towards health care; other factors, such as legislation which may limit the number of options available to a patient. Information pertaining to these factors was systematically extracted from each study and summarised in the IPDM review. As previously mentioned, no studies within the IPDM review adequately

operationalised informed patient decision making. In addition, few studies included measures of the decision making process. In other words, there is insufficient empirical evidence to adequately address this question. However, it is likely that this particular research question may be better addressed by a review with a different set of inclusion criteria. For example, a subsequent review might include studies designed to assess factors associated with the decision behaviour, i.e. a study measuring predominantly cognitive mechanisms.

2.6 Chapter summary.

The IPDM review provided a comprehensive bibliography of empirical research carried out between 1991-1996 that might be associated with the facilitation of informed health care decision making by patients. Of the 547 articles included in the IPDM review, 18 articles were considered of relevance to this thesis and analysed in more detail. There were no studies that directly evaluated the facilitation of informed decision making in the context of prenatal diagnosis. Further, no studies operationalised or measured informed decision making in any health care domain. Nine studies referred to a theory to inform their research questions and interventions but only one was a theory of decision making. Four studies evaluated decision aid interventions but none of them were carried out in the context of prenatal diagnosis. Only four studies included other measures of the decision quality such as decision autonomy and confidence. Despite the dearth of theoretically operationalised studies assessing informed patient decision making, the findings were integrated and suggest that patient prompt interventions were more likely to be associated with changes in measures of the process of decision making than the decision behaviour outcome; decision aid interventions were more consistently associated with changes in decision autonomy and confidence than information prompt interventions; the application of decision aids to health decisions was not associated with changes in the correctness of the decision made.

3. Research questions and thesis overview.

The application of psychological theory to understanding risky decision making in a 'real-world' health-care setting provides the main theoretical context for this thesis. The real world decision is one confronted by a significant minority of pregnant women, the choice to have or not have prenatal diagnosis following a screen positive triple test result for Down's syndrome. The decision is termed risky because both alternatives offered to women have a probability of a negative consequence occurring (see section 1.1.4). Prenatal diagnosis carries between a 0.5%-2% risk of miscarrying the pregnancy, while continuing the pregnancy upon receipt of a screen positive triple test result carries a risk of between 0.4%-20% of the baby having Down's syndrome. As prenatal and genetic testing are associated with iatrogenic consequences (see section 1.1.5), one of the primary objectives of offering testing in this real world context is to ensure that women make informed decisions to have or not have tests (Royal College of Physicians, 1989). The empirical evidence summarised in both the theoretical and integrative reviews (chapters one and two) suggested that few studies have adequately addressed the issues pertaining to informed decision making in women choosing to have or not have prenatal diagnosis. The preceding overviews have been used to inform the research questions for this thesis. Section 3.1. summarises the main research questions addressed in this thesis. Section 3.2 outlines the four subsequent empirical chapters and contains descriptions of the research aims, objectives and study design for each.

3.1 Research questions.

As stated, this thesis is concerned with the application of psychological theory to women's decision making about prenatal diagnosis. The theoretical review (chapter 1) integrated findings from empirically based studies of women's prenatal diagnosis decisions (section 1.3) with models of decision making (section 1.2.1). A definition of informed decision making was operationalised (section 1.2.2) with reference to reasoned-based theories of decision making and criteria identified as significant by prenatal and genetic testing committees. The facilitation of individuals' decision making was also discussed as decision aids, particularly the decision analytic technique, have been used in the prenatal diagnosis context (section 1.2.1.3). The findings summarised in the theoretical review (chapter 1) suggested that few, if any, studies had operationalised informed decision making within the prenatal diagnosis context. From the integration of empirical research in chapter 2, it was evident that informed decision

making has not been adequately defined or assessed in any health care context. Further illustrated by the integrative review (chapter 2) was the dearth of studies evaluating decision aids as a means of facilitating decision making. In addition, there is a paucity of research to indicate whether or not women have been provided with sufficient information to enable the process of informed decision making (section 1.3.1). Most of the applied studies have been carried out to understand the factors associated with women's prenatal diagnosis decisions (section 1.3.2). However, a number of methodological concerns were discussed (section 1.3.2) which may limit the explanations provided of women's (informed) decision making about prenatal diagnosis.

The following research questions have been broadly separated into those that address the objective of encouraging women's informed decision making about prenatal diagnosis and those that may inform psychological explanations of decision making under risk.

Ensuring informed decision making in the context of prenatal diagnosis:

- Do women receive sufficient information to be able to make an informed decision about prenatal diagnosis?
- Are women making informed decisions about prenatal diagnosis?
- Can informed decision making be facilitated?

Understanding decision making under risk:

- What information processing strategies are employed by women when deciding to have or not have prenatal diagnosis?
- What psychological factors are associated with the decision to have or not have prenatal diagnosis?
- Does the application of the decision analytic technique to the prenatal diagnosis decision modify decision behaviour and lead to more accurate or better decisions?

3.2 Overview of empirical studies included in the thesis.

The following four chapters describe the empirically based studies designed to provide evidence to address the proposed research questions. Brief summaries of each chapter are provided below.

Chapter 4: this chapter describes the prenatal diagnosis information provided to women upon receipt of a screen positive triple test result. The study employed a non-experimental, cross-sectional, structured observational design. A checklist was

designed to categorise the information provided by the health professional during the consultation. Over a three month period, all the information giving consultations provided by health professionals at the Leeds General Infirmary (LGI) for women who had received a screen positive triple test result were audio tape-recorded. The purpose of this study was to assess whether or not women had received sufficient information to enable them to make an informed, autonomous choice about the decision to have or not have prenatal diagnosis.

Chapter 5: this chapter describes the processes employed by women when making the decision to have or not have prenatal diagnosis. The study employed an observational, cross-sectional design. The 22 transcripts from chapter 4 and an additional 22 transcripts from a further three months of audio tape-recording the information giving consultations were used in this chapter. The first aim of this study was to describe the information utilised by women when making the decision to have or not have prenatal diagnosis. A coding frame was developed using content analysis to classify women's utterances during the information-giving consultation. The findings from this process tracing technique provided evidence of the way women processed or at least utilised information when making this risky decision. To ascertain whether or not there was an association between the utilisation of consultation information and test decision, the pattern of information utilisation was presented by prenatal diagnosis test decision. The second aim of this study was to operationalise the concept of informed decision making (see chapter one) and develop a measure with which to assess the informed decision making process.

Chapter 6: this chapter evaluated the application of the decision analytic technique to the prenatal diagnosis consultation. The purpose of the consultation was to compare the efficacy of a consultation structured by decision analysis with that of a routine information-giving consultation to facilitate informed decision making. The study employed a randomised control trial (RCT) design to evaluate the efficacy of the decision analytic technique. Both process tracing techniques and questionnaire-based measures were used to evaluate differences in the decision making process and outcome by intervention group. In total, complete data sets for 106 women randomised to either the decision analysis or routine consultation were analysed. The study findings were used to inform research questions about the facilitation of informed decision making and the ability of decision analysis to modify the decision quality and outcome.

Chapter 7: this chapter describes the psychological factors associated with women's decision to have or not have prenatal diagnosis. The data obtained from the RCT in chapter 6 were used in this chapter to provide a more comprehensive understanding of women's prenatal diagnostic test behaviour. The findings from the following process tracing and questionnaire measures were used to assess factors associated with the decision to have or not have prenatal diagnosis: informed decision making; cognitive mechanisms such as utilities and perceived social norms; knowledge; demographic characteristics.

Chapter eight discusses the findings summarised in these four empirical chapters with reference to the research questions of this thesis. The chapter focuses on describing the extent to which the research questions were addressed by these empirical studies, the strengths and limitations of the methodologies employed and outlines areas for further decision making research in the context of prenatal diagnosis decision making.

4. Prenatal diagnosis: describing the provision of information.

This chapter is concerned with the provision of information by health professionals for women choosing to have or not have prenatal diagnosis upon receipt of a screen positive triple test result. To enable informed decision making about prenatal diagnosis, it is the responsibility of health professionals to provide sufficient information about the test decision and to communicate this information in a neutral or 'non-directive' manner (section 1.2). These conditions should provide women with the opportunity to integrate the test information with their beliefs and values, so encouraging informed and autonomous decision making. As mentioned in chapter one, there is a paucity of research evaluating the provision of information by health professionals for women choosing to have or not have prenatal diagnosis upon receipt of a screen positive triple test result (section 1.3). A few studies described the content of the prenatal diagnosis consultation between health professionals and women with an increased risk of Down's syndrome as a consequence of raised maternal age (Kessler, 1981; Marteau et al, 1993; Bernhardt et al, 1998). The findings suggest that women did not receive sufficient information about the test alternatives and that health professionals did not impart all information in a neutral manner (section 1.3).

The purpose of the study reported in this chapter is to describe the information provided by the health professional for women making the decision to have or not have prenatal diagnosis upon receipt of a screen positive triple test result. The main aim is to assess whether or not the information-giving consultation enables women to make an informed and autonomous decision about prenatal diagnostic testing.

4.1 Methodology.

4.1.1 Sample.

The triple test is routinely offered to all pregnant women aged 29 years or over at estimated date of delivery at the Leeds General Infirmary (LGI). For the year preceding the study, 63% (1044/1661) of pregnant women offered the triple test at the LGI were tested. Thirteen percent (133/1044) of screened pregnant women received a positive triple test result. Prenatal diagnosis was offered to the 133 women who screened positive and 90% (120/133) chose to have a diagnostic test. All women who received a screen positive triple test result from the Leeds General Infirmary (LGI) from April to

June 1995 were invited to take part in this study. Over this three month period, none of the 22 women who received a screen positive triple test result refused to take part in the study.

4.1.2 Design.

A non-experimental, cross-sectional, structured observational study design (Robson, 1993) was employed to describe the information provided by health professionals for women deciding to have or not have prenatal diagnosis following receipt of a screen positive triple test result. An audio tape-recorder was used to record the content of the prenatal diagnosis information-giving consultations. The consultation audio tape-recordings were transcribed fully by a third party (KM). The transcribed health professional's utterances provided the data source for this study. Any statement proffered by the health professional during the consultation was classified, including: triple test result information; details concerning the decision to have or not have prenatal diagnosis options; responses to women's questions. This observational technique enabled the actual information given during the consultation to be coded for analysis rather than relying on the health professional's or woman's recall of the consultation information (Robson, 1993). It is argued that an observer or audio tape-recorder may subtly alter the woman-health professional interaction (Robson, 1993). Alternatively, such a direct technique may increase the validity and reliability of the findings by reducing the likelihood of memory, reporting and social desirability biases (Robson, 1993; Smith, 1995; Bowling, 1997). In an effort to reduce any 'observer' effects, the author (HB) was introduced as a member of the ante-natal care team, initially as a trainee and later as the main information-giver for this prenatal diagnosis consultation. On balance, a structured observational design was considered the most appropriate to achieve the aims of this study.

4.1.3 Materials.

4.1.3.1 Triple test result sheet.

During each consultation, the health professional structured the consultation with reference to the woman's triple test result sheet (Appendix IV). The following details were recorded on the triple test result sheet: the woman's name, address and LGI maternity reference number; two gestation dates informed by the first day of her last period and the LGI's dating scan; maternal serum levels for the hormones unconjugated oestriol (uE_3), human chorionic gonadotrophin (hCG) and alpha-fetoprotein (AFP); maternal age; maternal weight at time of screening; estimated date of delivery; risk

figure for Down's syndrome based on the triple test calculation; risk figure for Down's syndrome based on maternal age; risk figure for Edward's syndrome; risk figure for spina bifida.

4.1.3.2 Information giving checklist: categories and inter-rater reliability.

A structured observational study design requires the development of a checklist or coding scheme to classify the information observed in the 'real world' setting (Robson, 1993). The categories developed for this type of checklist were informed more by the guidelines outlining criteria to enable informed decision making in the context of prenatal diagnosis (Royal College of Physicians, 1989; Royal College of Obstetricians and Gynaecologists, 1993; section 1.3) than by the development of themes from the health professional's utterances (Robson, 1993). In other words, most of the categories included within the checklist were previously identified within the literature as necessary to ensure informed decision making (Royal College of Physicians, 1989; Royal College of Obstetricians and Gynaecologists, 1993; Kennard, Goodburn, Golightly and Piggott, 1995; Marteau, Shaw and Slack, 1995; section 1.2). In brief, the consultation information should contain *sufficient information* to enable an informed decision to be made and this information should be presented *neutrally* to encourage women's autonomy in decision making (section 1.3). However, as the checklist was used to classify all the health professional's utterances, three further categories were incorporated within the checklist. These additional categories classified information provided by the health professional that were specific to this health care context but not directly associated with women's informed prenatal diagnosis decision making.

Coding the information giving consultation requires the comparison of information contained within the checklist to be compared with that documented in the transcript. The transcript either contains the information outlined in the checklist or not. In total, seventeen categories were identified to classify the content and delivery of the information contained within the transcripts (Appendix V). Fourteen categories classified information pertaining to the provision of sufficient information and the neutral communication of information, while three categories classified additional information specific to this consultation context. The content of these categories are discussed in more detail below.

Sufficient information.

As mentioned, one of the conditions considered necessary to enable women to make an informed decision about the decision to have or not have prenatal diagnosis following

receipt of a screen positive triple test result for Down's syndrome is the provision of sufficient information (Royal College of Physicians, 1989; Royal College of Obstetricians and Gynaecologists, 1993; Kennard, Goodburn, Golightly and Piggott, 1995; Marteau, Shaw and Slack, 1995; section 1.2). This information should include a discussion of the consequences of all the decision alternatives and the benefits and risks associated with each alternative. The information included within the following categories has been identified within the literature as sufficient to enable informed decision making (section 1.3).

- *A description of the abnormality being tested for*, in this instance Down's syndrome. The description should include the following information: the population incidence (1 in 650 live births); the life expectancy (between 55 and 60 years); the prognosis (a range of severity which may lead to premature ageing and signs of Alzheimer's disease); the impairment (many who have Down's syndrome lead independent lives, are usually very loving and caring, have a slower mental and physical development, have 'typical' facial features and require possible medical interventions for physical abnormalities); cause and recurrence (spontaneous chromosomal abnormality located on chromosome 21 with a recurrence in subsequent pregnancies of approximately 1%) (Leeds Antenatal Screening Service, 1993; Royal College Obstetricians and Gynaecologists, 1993; Kennard et al, 1995; Marteau, Shaw and Slack, 1995; Barnes and Bryan, 1996; section 1.1). Any information provided by the health professional concerning Down's syndrome was classified in category *hp7* of the checklist (Appendix V).
- *An explanation of the maternal serum screening test result.* Women should be aware of the following information explaining the triple test maternal serum screening result: the meaning of a screen positive and negative result; an explanation of the cut-off risk figure between screen positive and negative result; the likelihood of detecting a fetus with Down's syndrome; the abnormalities identified by the triple test; an explanation of the details used to calculate the risk figure included in the triple test equation (Leeds Antenatal Screening Service, 1993; Royal College of Obstetricians and Gynaecologists, 1993; Kennard et al, 1995; Marteau, Shaw and Slack, 1995, section 1.1). Triple test screening information was classified in category *hp1* of the checklist (Appendix V). In addition, information given regarding two other first trimester screening tests, maternal age and nuchal pad risk figures for Down's syndrome, was classified in category *hp1* of the checklist (Leeds Antenatal Screening Service, 1993; section 1.1). Any actual risk figures mentioned by the health professional during the consultation were coded separately in category *hp9* of the checklist (Appendix V).

- *An explanation of the 'no test' alternative.* Women are only able to make an informed decision about prenatal diagnostic testing if they are aware that there is a choice between having or not having prenatal diagnosis (Marteau, Shaw and Slack, 1995). In consequence, health professionals ought to make this choice explicit during the information-giving consultation. Category *hp2* of the checklist classifies any utterance from the health professional that explicitly refers to the consequences associated with not having a diagnostic test (Appendix V). The information classified in this *no test alternative* category may include reference to: not having an invasive test; the 19 week fetal anomaly scan; delaying the prenatal diagnosis decision until after the results of the 19 week fetal anomaly scan. This 19 week fetal anomaly scan is seen as a likely course of action or alternative following a screen positive triple test result for spina bifida (Royal College of Physicians, 1989; Royal College of Obstetricians and Gynaecologists, 1993; Marteau, Shaw and Slack, 1995). In addition, this scan may be used as another screening test for Down's syndrome, identifying physical abnormalities or 'markers' associated with an increased risk of Down's syndrome (Leeds Antenatal Screening Service, 1993; Marteau, Shaw and Slack, 1995; Barnes and Bryan, 1996; section 1.1).
- *A description of the diagnostic test alternative.* Women need to have information about the risks and benefits associated with each diagnostic test if they are to make an informed choice about prenatal diagnosis. For each diagnostic test offered, health professionals should include the following information: the condition(s) testing for; the test procedure; the timing of the test; the meaning of a diagnostic test result; the risk of procedure related miscarriage; the options available should the test result be positive (Marteau, Shaw and Slack, 1995). At the LGI, both amniocentesis and chorionic villus sampling were offered to women upon receipt of a screen positive triple test result. Details specific to these two diagnostic tests have been described in section 1.1. Category *hp3* of the checklist classified utterances pertaining to the provision of amniocentesis information, category *hp4* chorionic villus sampling information (Appendix V). Information given about the miscarriage consequence of testing was classified in category *hp5* of the checklist (Appendix V). Specific information provided about termination as an option following receipt of a positive diagnostic test result was coded in category *hp6* of the checklist (Appendix V).
- *The provision of misinformation about prenatal diagnosis.* Prior empirical research has documented the provision of inaccurate information about prenatal diagnosis (Marteau, Plenicar and Kidd, 1993; Bernhardt, Geller, Doksum, Larson, Roter and Holtzman, 1998). As the provision of inaccurate information may be associated with women's ability to make an informed decision about prenatal diagnosis, category

hp8 of the checklist coded any reference to misinformation by the health professional (Appendix V).

Neutrally communicated information.

The second component enabling the making of informed decisions is to ensure women's autonomy in the decision making process. In other words, allowing women the opportunity to assimilate the 'sufficient information' with their own attitudes or values (section 1.2). Communicating *directive information* is regarded as exerting undue pressure on the individual; whereas communicating *neutral* or *non-directive* information is believed to encourage the individual's ability to make the decision autonomously (Kessler, 1993, section 1.3). In other words, directive information alludes to or explicitly states a desirable course of action, so reducing the opportunity for an informed decision to be made, while non-directive information requires the individual's assessment of the information, so encouraging the process of informed decision making. The directiveness of information can be communicated subtly or overtly (Kessler, 1993; Marteau, Plenicar and Kidd, 1993; Bernhardt, Geller, Doksum, Larson, Roter and Holtzman, 1998). An example of an overt, non-directive communication is the health professional explicitly stating that, 'the decision is yours [the woman's]'; an overt, directive communication may be 'chorionic villus sampling is not the right choice for you'. Some examples of subtle, non-directive communications are: focusing equally on the reasons for and against each option; encouraging women to take time to evaluate the information; rephrasing risk figures. Examples of subtle, directive communications are: the health professional's value judgements about the 'sufficient' information; omitting to discuss the advantages or disadvantages of one alternative; alluding to a 'norm' for decisions about prenatal screening, diagnosis and termination (Donnai, 1992; Kessler, 1993; Marteau, Plenicar and Kidd, 1993; Bernhardt, Geller, Doksum, Larson, Roter and Holtzman, 1998). Within the checklist, categories *hp10* - *hp13* (Appendix V) classify the way in which information was communicated during the consultation. An additional category, *hp14* (Appendix V), was included in the checklist to classify the direct questions posed by the health professional. These direct questions provided women with the only opportunity to express their attitudes or opinions about the consequences of the prenatal diagnosis decision during the consultation. The elicitation of these attitudes may encourage women to integrate their opinions about prenatal diagnosis with the consultation information and facilitate the informed decision making process.

Additional categories.

Although the aforementioned fourteen categories of the checklist were developed with reference to the literature on informed decision making, three additional categories were also included to accommodate information provided by the health professional specific to the setting of this study. In brief, category *hp15* of the checklist (Appendix V) classifies information provided by the health professional that pertains to the prenatal diagnosis decision, but the relationship of these factors to issues of informed decision making is unclear. For example, the health professional may compare the advantages and disadvantages of the two diagnostic tests offered rather than provide the details of each test sequentially. Category *hp16* of the checklist (Appendix V) classifies information provided by the health professional during the consultation that was not directly associated with the prenatal diagnosis decision, such as the results of routine antenatal tests. Finally, category *hp17* (Appendix V) classifies information pertaining to the elicitation of utilities following application of the decision analytic technique to the prenatal diagnosis consultation (section 1.2, chapter 6). The decision analytic technique was not applied routinely by the health professionals at LGI. However, the author was permitted to practise the elicitation of utilities with consenting women after they had made their decision to have or not have prenatal diagnosis. The piloting of this elicitation technique will be discussed in more detail in chapter 6.

Inter-rater reliability.

As stated, most of the checklist' categories were developed with reference to the literature on informed decision making. This literature broadly defined the content and style of information considered necessary to enable women to make an informed prenatal diagnosis decision. The checklist modified these criteria for application to the prenatal diagnosis decision upon receipt of a screen positive triple test result. In consequence, additional categories were included to accommodate health professional's utterances specific to this health context. As such, the checklist was a non-standardised instrument with which to classify qualitative data. To assess the reliability of the checklist to accurately classify health professional's information giving, a test for inter-rater reliability was carried out (Robson, 1993). Ten per cent of the 22 transcripts (n=3) were selected for use in the inter-rater reliability test. The three selected transcripts represented each of the following prenatal diagnosis test decision outcomes: no further diagnostic testing; amniocentesis; chorionic villus sampling. The transcripts were coded separately by the author (HB) and a third party (SA). In total, 137 statements were coded by both the author and SA. Cohen's Kappa coefficient was calculated to assess the degree of concordance between the two raters' applications of the checklist to the three transcripts. The value of the Kappa coefficient was 0.73. As

this figure falls between 0.60 and 0.75, it suggests a good degree of agreement between the two raters (Robson, 1993). This finding provides some evidence that the checklist may be a robust classification system for the categorisation of health professional's information-giving within the context of prenatal diagnosis.

4.1.4 Procedure.

The routine practice of the ante-natal staff at the LGI for informing women of their triple test results and subsequent prenatal diagnostic test decisions was maintained throughout the study period. Women with a screen negative triple test result were not routinely informed of their risk figure, although a few individual consultants sent letters to women under their care informing them of their triple test status. All women with a screen positive triple test result were contacted by telephone or letter if a telephone number was not available. Contacting women about a screen positive triple test result and informing them of their reproductive options during a consultation was the responsibility of one midwife within the ante-natal team. Minimal information was provided to women over the telephone: the purpose of the phone call; the triple test figure, both as an odds number and percentage; the appointment time for a further information consultation; addressing the immediate concerns raised by women. On attending the antenatal clinic for the prenatal diagnosis information giving consultation, women were informed of the study. Participation in the study involved having the author sitting in the room and audio tape-recording the consultation. No women refused to take part in the study.

4.1.5 Analysis.

The results are summarised using descriptive statistics. Frequencies are recorded for the number of consultations that referred to a particular category of information. Although some categories of information were mentioned more than once during a consultation, this results section focuses only on whether or not the necessary information to enable informed decision making was provided. Text considered to be representative of the 22 transcripts is used to illustrate the information categories of the checklist when describing the study's findings. The main purpose of the study was to describe the information provided by the health professional during the consultation. As a consequence, no formal statistical analysis was applied to the data. However, to illustrate the pattern of information given by test decision[1], the category frequencies

[1] The test decision refers to those who chose either to have or not have prenatal diagnosis. It is outside the remit of the thesis to assess differences between those who chose chorionic villus sampling rather than amniocentesis.

were reported for both the complete sample and separately by whether or not women decided to have a subsequent prenatal diagnostic test.

4.2 Results.

Of the 22 women participating in the study, five chose not to have a diagnostic test and seventeen chose a diagnostic test (ten amniocentesis and seven chorionic villus sampling). The categorised information provided by the health professional during the prenatal diagnosis consultations is summarised in table 4:1 and table 4:2. The findings from this study are discussed in more detail below.

4.2.1 Provision of sufficient information.

4.2.1.1 Description of the condition.

Information about Down's syndrome, the abnormality being tested for, was mentioned in 27% (6/22) of consultations (hp7, table 4:1). The following are examples of the information given about Down's syndrome:

description: "Well, it's 20 or 30 years down the line when you've got what is essentially a baby still."

description: " But yes, there are varying degrees of Down's syndrome so you're not going to know until the baby gets here."

cause: "It's a one off. Nobody knows exactly why Down's syndrome occurs. But what it is, is an extra number 21 chromosome. When the sperm and the egg meet instead of dropping the chromosome for some reason whenever number 21 occurs, it's kept hold of three not two and that's what causes the features of Down's syndrome. 'Cos it's the sperm and the egg, it's decided at conception. So don't think it's anything you've done, could or should have done."

Table 4:1 frequency summary of 'sufficient information' provided by the health professional during the prenatal diagnosis consultation.

Information category	Total (n=22)		No test (n=5)		Test (n=17)	
	n	%	n	%	n	%
hp1 Triple Test	**22**	***100%***				
hp1.1 meaning / describing tt	22	100%	5	100%	17	100%
hp1.2 Edward's syndrome	16	73%	0	0%	16	94%
hp1.3 Spina Bifida	16	73%	0	0%	16	94%
hp1.4 age screening	15	88%	0	0%	15	88%
hp1.5 nuchal pad screening	3	18%	0	0%	3	18%

Table 4:1 continued...

Information category	Total (n=22)		No test (n=5)		Test (n=17)	
hp2 No test option	***22***	***100%***				
hp2.1 explicit "do not have to have test"	21	96%	4	80%	17	100%
hp2.2 19 week scan / screening DS	10	45%	5	100%	5	29%
hp3 Amniocentesis	***22***	***100%***				
hp3.1 procedure	22	100%	5	100%	17	100%
hp3.2 timing results	22	100%	5	100%	17	100%
hp3.3 abnormalities testing for	17	77%	2	40%	15	88%
hp3.4 certainty diagnostic tests	18	82%	4	80%	14	82%
hp4 Chorionic villus sampling	***22***	***100%***				
hp4:1 procedure	22	100%	5	100%	17	100%
hp4:2 timing results	19	86%	4	80%	15	88%
hp4.3 Mosaicism	19	86%	3	60%	16	94%
hp5 Miscarriage	***15***	***68%***				
hp5.1 reasons for miscarriage	15	68%	2	40%	13	76%
hp5.2 other miscarriage	1	4%	0	0%	1	6%
hp6 Termination	***10***	***45%***				
hp6.1 procedure termination	9	41%	2	40%	7	41%
hp6.2 other termination	5	23%	2	40%	3	18%
hp7 Down's syndrome	***6***	***27%***				
hp7.1 description / severity DS	5	23%	2	40%	3	18%
hp7.2 cause / recurrence DS	1	4%	0	0%	1	6%
hp7.3 more information DS/ other service	0	0%	0	0%	0	0%
hp8 Misinformation	***16***	***73***				
hp8.1 tt cut off point - calculation	7	32%	1	20%	6	35%
hp8.2 mosaicism risk figure - 3/300000	7	32%	1	20%	6	35%
hp8.3 tt unreliable	2	9%	0	0%	2	12%
hp8.4 other screening tests more reliable	4	18%	3	60%	1	6%
hp8.5 other	2	9%	1	20%	1	20%
hp9 Risk figures						
hp9.1 tt risk Down's syndrome	22	100%	5	100%	17	100%
hp9.2 age risk Down's syndrome	14	64%	2	40%	12	71%
hp9.3 amniocentesis miscarriage	22	100%	5	100%	17	100%
hp9.4 chorionic villus sampling	21	96%	4	80%	17	100%
hp9.5 1: 250 cut off triple test	7	32%	2	40%	2	12%
hp9.6 1:100 background risk miscarriage	3	14%	1	20%	2	12%
hp9.7 other	3	14%	1	20%	2	12%

Table 4:2 frequency summary of the 'directiveness dimension' communicated by the health professional during the prenatal diagnosis consultation.

Information category	Total (n=22)		No test (n=5)		Test (n=17)	
	n	%	n	%	n	%
hp10 Comparison risks						
hp10.1 comparison tt and age risk	6	27%	1	20%	5	29%
hp10.2 comparison tt and amniocentesis	14	64%	3	60%	11	65%
hp10.3 comparison tt and CVS	11	50%	3	60%	8	47%
hp10.4 other comparisons	4	18%	0	0%	2	12%
hp11 Perception risk						
hp11.1 minimise Down's syndrome risk	15	68%	5	100%	10	59%
hp11.2 augment Down's syndrome risk	8	36%	2	40%	6	35%
hp11.3 minimise miscarriage risk	2	9%	0	0%	2	12%
hp11.4 augment miscarriage risk	1	5%	0	0%	1	6%
hp12 Facilitate IPDM	***22***	***100%***				
hp12.1 diagnostic patient's choice	17	77%	2	40%	15	88%
hp12.2 termination patient's choice	2	9%	1	40%	1	6%
hp12.3 think about it / reduce time pressure	12	54%	3	60%	9	53%
hp12.4 some have test, others not	10	45%	1	20%	9	53%
hp12.5 invert miscarriage risk / %	21	96%	5	100%	16	94%
hp12.6 invert Down's syndrome risk / %	21	96%	4	80%	17	100%
hp13 Impair IPDM	***17***	***77%***				
hp13.1 health professional suggests option	6	27%	1	20%	5	29%
hp13.2 clinician's own test decision	8	36%	1	20%	7	41%
hp13.3 increase time pressure / don't wait	3	14%	0	0%	3	18%
hp13.4 why have test if not terminate	14	64%	3	60%	11	65%
hp13.5 woman 'x' had test, baby fine	6	27%	1	20%	5	29%
hp13.6 most have test - not balanced	2	9%	0	0%	2	12%
hp13.7 had triple test so have diagnostic	1	4%	0	0%	1	6%
hp14 Direct questions						
hp14.1 ask about termination	13	59%	1	20%	12	71%
hp14.2 ask about having baby abnormality	0	0%	0	0%	0	0%
hp14.3 how Down's syndrome affect family	2	9%	0	0%	2	12%
hp14.4 how miscarriage affect family	1	4%	0	0%	1	6%
hp14.5 do you need to know for certain	7	32%	3	60%	4	24%
hp14.6 can you live with not knowing	2	9%	1	20%	1	20%
hp14.7 other	0	0%	0	0%	0	0%
hp15 Miscellaneous						
hp15.1 compared CVS and amniocentesis	12	54%	2	40%	10	59%
hp15.2 discuss scan/ tt date discrepancy	2	9%	1	20%	1	6%
hp15.3 worst - no test / DS baby	6	27%	2	60%	4	24%
hp15.4 worst - test / miscarry healthy baby	8	36%	2	60%	6	35%

4.2.1.2 Explanation of a screen positive triple test result.

All women received an explanation of their screen positive triple test result (hp1, table 4:1). Most women (16/22) were informed that the triple test also screened for Edward's syndrome and spina bifida (hp1, table 4:1). The cut-off risk figure between a screen positive and negative was mentioned in just seven consultations (hp9, table 4:1). Typical explanations provided for the triple test result are illustrated below:

screen positive calculation: "I'll go through your report with you as it's come back to me... The reason your weight goes into the calculation is you're gonna have less circulating hormone if you're six stone wet through than if you're 20 stone wet through. So it's to work out the hormones per body weight. That's why that's gone in. The triple test is so called because it's worked out on three hormones: alhpa-fetoprotein coming from the baby; unconjugated oestriol that is coming from the placenta; and human chorion-gonadtrophin that is coming from you."

screen positive meaning: "What they do in it's simplest terms, it's a diagram, is if we plot a graph which is what they do in the labs, we know that there is a normal curve for a normal baby. And all the values are gonna cluster within that curve somewhere. And what they're going to do is read off how many weeks pregnant you are and what your risk is and that's how they work it out. In retrospect if we were to test all the babies that we know have Down's syndrome they would show a completely different curve and the two overlap but you see the curve pushed this way for the babies that have got Down's syndrome. If it's screened you at risk, it's not saying that baby has got Down's syndrome, we're a long, long way off that, but they've seen it in this bit of the curve. So they've seen it in this half of the curve if you like. So that's in its simplest terms how it's worked out."

4.2.1.3 Explanation of the choice to have or not have prenatal diagnosis.

The health professional stated that the choice to have or not have a prenatal diagnostic test was the woman's in 77% (17/22) of consultations (hp12, table 4:2). In almost all of the consultations (21/22) women were explicitly told they did not have to have a diagnostic test (hp2, table 4:1). Typical examples from the health professional's transcript are:

choice: "I'm not here to say 'do this, this and this' 'cos those days are long gone. I give you all the information and support you in the decision you make but it's up to you what you want to do."

no further testing: "So what I'll do is I'll go through the options with you as to what you can do about it... now the first thing you could do is nothing. You could say 'Fine I know that I've got a 1 in 140 risk of having a baby with Down's and I've a 139 out of 140 risk of the baby being normal, so I'm not going to do anything'."

fetal anomaly scan: "Taking you back to scanning when you said could you see things on scan. It would be wonderful if we could diagnose conclusively from scan. We know that babies have got 17 markers for Down's syndrome that you can pick out on the scan. And they're things like: they tend to have heart defects but you get a normal baby with a heart defect; they tend to get a fatty pad at the back of the neck; they tend to get ... they hold their thumb well away from the fingers; they hold their big toe well away from the other toes. One of them in isolation means absolutely nothing but if you saw two or three of those things alarm bells would start to ring and we would say at that stage, 'Perhaps it might be worth having testing done'. Now we wouldn't bring your scan forward because it's not big enough to see. While at this stage you scan see its heart and you can see it flicking, you can't see if all the chambers and the vessels leading from it which is what you're going to need to do to look for a defect. But we also know that 50% of babies with Down's syndrome don't show anything on the scan at all. So 50% of babies will be missed on scan. So you could say that we pick up the worst ones on the scan. If we don't see any, it's thought to reduce your odds of you having a baby with Down's, but nobody knows to what."

4.2.1.4 Describing the available diagnostic test procedures.

All women received information about the two diagnostic tests, amniocentesis and chorionic villus sampling (hp3 and hp4, table 4:1). The extract below shows the information about the test procedure and timing of results that was consistently given. In addition the procedure-related miscarriage risk figures were mentioned in most consultations (hp9, table 4:1). About two thirds (15/22) of the women received an explanation about the reasons for a miscarriage (hp5, table 4:1). Just under half (10/22) were given information on termination (hp6, table 4:1). However, the health professional also asked women a direct question about termination of pregnancy (hp14, table 4:2). When references to both the direct question about termination of pregnancy and providing termination information were combined, the issue of termination was raised in 86% (19/22) of consultations (hp6, table 4:1; hp14, table 4:2).

description tests: "The second would be to have an amniocentesis, which involves putting a needle through the wall of your tummy and taking off 15 mils of fluid from around the baby. Within that fluid are cells from the baby, both living and dead. By taking off 15 mils we're not

going to harm the baby but within that fluid we can hopefully get some cells to grow. So we would put that fluid in a flask in an incubator and shut the door for three weeks. We're simulating conditions inside you and getting those cells to multiply and divide. Hopefully at the end of three weeks there will be enough cells grown to look at all the cells and to analyse them. Now we can tell you with 99 point whatever percent certainty that baby is normal or baby has got Down's syndrome. Nothing in medicine is 100% certain, but as near as we can get to it. The risk of you losing a baby from having that done is 1 in 200 or half a percent. So you're weighing up 1 in 130 with 1 in 200. So you could have it done and you could miscarry and we could tell you it was normal. The other option that you've got is something called a CVS or a CVB and it means exactly the same thing, chorionic villus sampling or biopsy. Instead of going into the fluid around the baby we can go into the placenta. Within the placenta are rapidly dividing cells that you can set up and analyse straight away. We could look at those within 24 hours and we could get you a provisional result 24 hours later but they'd be 98% accurate. The reason it's only 98% accurate is because we can miss something called mosaic downs and those cells don't rapidly multiply and divide. So, to capture that, we put the other half of the culture to long term culture the same as the amnio, and we leave it for three weeks the same. The backup result that you get at 3 weeks is the same as the amnio, 99 point whatever. But the advantage is that you get a faster result in 24 hours. The disadvantage is it's got a higher risk of miscarriage, it's 1% or 1 in 100."

miscarriage: "It is - the biggest time is up to ten days after having it done. Once you get beyond ten days it's unlikely that you're going to miscarry. I mean it could happen but it's unlikely that it's going to happen and if it does you've got to question whether it's the amnio that's caused it or whether you were gonna miscarry any way. There are two reason that you might, are more likely to miscarry. One is thought to be just the trauma of having the test done and that you'd disturbed things within the uterus really, which is unlikely. The biggest thing is infection and that's not because we've not cleaned everything and not cleaned your tummy but if you're putting a needle through somebody's skin you've got about seven layers to get down into the baby, so if there's bacteria sat between the layers then the needle is gonna push it through and it's gonna be bacteria that you're never gonna see so it's no good saying, 'I think I've got a chest infection or I think I've got a urine infection' because it's bacteria that are unharmful in the right place but harmful in the wrong place. So, if you're pushing a needle through various layers if bacteria gets in your uterus that should be there, bacteria in your uterus are gonna cause an infection and an infection is gonna cause you to miscarry. And we know it would take 10 days for the infection to set up. Now, after having either done you don't need to take to your bed or rest completely 'cos if it's gonna happen it's

gonna happen, whatever. And if infection has gone through, you're not going to stop it getting through by resting and carrying on as normal."

termination: "We'll bring you in as soon as possible but I've got to say to you, it's labour. There is no other way to terminate beyond 12/13 weeks. So, I mean, I'll tell you that now. I need to put the cards on the table, really, if the worst should come to the worst. And the reason is the baby is too big to do it any other way. We can't do like a D&C, suction termination, because we'd have to stretch the neck of your womb far too much, so it's putting you at risk and it's potentially putting any future babies at risk. I mean, you might say, 'I don't want any more after this' but we never know what's around the corner for us. So it would stretch the neck of your womb too far. The only other way would be to operate, which is virtually a caesarean section; so we're putting you at risk for a non-viable baby. So the only other way, I mean it's not a full blown labour like it would be if you were at term, but nevertheless that's the only way to do it, if the worst should come to the worst."

4.2.1.5 Misinformation about prenatal diagnosis.

Misinformation about some aspect of the prenatal diagnosis decision was given in 73% (16/22) of the consultations (hp8, table 4:1). The inaccuracies most consistently referred to by the health professional were explaining the reason for the triple test screen positive cut-off point and the likelihood of mosaicism occurring.

explanation cut-off risk: "Every risk comes back individual and it depends on your hormones and the baby's hormones. Anything less than 250 we consider high risk. The reason, where we drum up the magic 250, is the risk of miscarriage at amnio which is half a per cent or 1 in 200 and we put the 50 on for people that might have a discrepancy between dates and scans, and then you're capturing those three or four days. So that's where we've drummed up this magic 250."

mosaicism: "In all the reported literature, 3 mosaic Down's have been missed in 300,000. So it's remote but nevertheless it's there and I've got to tell you about it."

In addition, the health professional consistently (22/22) referred to the lowest procedure-related miscarriage rates associated with the prenatal diagnostic tests. These figures are half the risk of the rates suggested in the appropriate guidelines (Royal College of Obstetricians and Gynaecologists, 1993; Marteau, Shaw and Slack, 1995).

4.2.2 Neutrally communicated information.

To enable informed decision making, it has been suggested that the health professional communicate the decision information in a neutral or non-directive style (see section 1.3). The following results suggest that the health professional communicated information in a manner that might encourage woman's autonomy in decision making. As mentioned, in 77% (17/22) of consultations the health professional explicitly stated that the prenatal diagnosis decision was the woman's choice (hp12.1, table 4:2). In addition, the health professional rephrased the risk figures as percentages or inverse risks in most consultations (21/22) (see exerpts in 4:2.1.3 and 4:2.1.5). This rephrasing of risk is likely to reduce biasing in the appraisal of risk information. Finally, in just over half the consultations (13/22), the health professional directly asked women to think about the consequences of a positive diagnostic test result, i.e. encouraging discussion of the consequences of testing (hp14, table 4:2). These techniques might facilitate a more informed decision making process. However, the results also suggest that some statements were communicated in a more directive, less neutral style. In each consultation, the health professional expressed at least one personal judgement, belief or attitude that may be seen as directive (hp10, hp11 and hp13 in table 4:2). The directive statements included: direct advice about the options available; reference to personal experience of undergoing chorionic villus sampling; judgements about the risk figures; inferences as to a 'correct' decision or normative behaviour. These directive statements may impair women's decision making autonomy.

direct advice: "To be honest, with it being Bank Holiday, if you have the CVS today you're not going to get it while Wednesday. So you've almost waited a week. So with the CVS you've taken a high risk and it's not doing what you wanted to do [early result]."

personal experience: "I had chorionic villus sampling... Mine was 1 in 20 when I had mine done. And he's fine, normal, healthy."

risk judgements: "So, I mean, we're talking about a fairly low risk if you turn the figures round... the odds are in your favour at 230 to 1."

risk judgements: "You can get a risk of 1 in 10,000 and somebody has got to be the one."

correct choice: "Now, I mean, I always said if my risk is less than a hundred then I would probably go for the CVS because I've got more risk of a Down's than losing it at CVS and I get a faster result. But if it was over a hundred then I would sit tight and have the amnio because I'd have the one with the least risk."

correct choice: "What I would say to you is, if there is no way you would have a termination, there is little point in putting a pregnancy at risk if you're not going to do anything about it, ultimately, at the end of the day."

correct choice: "I would say to you, 'why have you had the triple test done if you're not going to do anything about it?'"

4.2.3 Additional classification categories.

This chapter is concerned with the provision of sufficient information by the health professional to ensure women's informed and autonomous decision making. Of the additional information categories incorporated in the checklist, only the utterances classified by category *hp15* (Appendix V) were associated specifically with the prenatal diagnosis decision information (see section 4:1.3.2). This section focuses on describing the type of information classified by category *hp15*. The health professional referred to one of the following additional, decision-related statements in most consultations (14/22): comparing the risks and benefits of the two diagnostic tests; discussing discrepancies in the gestation dates based on either the first day of the last period or the dating scan; rephrasing the negative consequences of both testing alternatives.

comparison diagnostic tests: "So, with your amnio, you get over 10 days and you think, 'Thank goodness for that, I'm over it' and then you think, 'Oh my God, the results are looming in another ten days time'. With the CVS you get the results in 24 hours, probably find out it's normal and then think, 'Oh my God I could miscarry up to ten days and I know it's normal'."

gestation date: "Now that's based on your scan. So that's presuming that you were 15 weeks and 6 days. You think that's [scan date] more accurate? Right. If you said to me that I know I'm 17 weeks then it would be a completely different risk. It would probably give you a slightly higher risk ... because you're further on in pregnancy. But if you think 15 and 6 is more accurate then we'll stick to the 15 and 6. So that's what that's saying, it's based on scan."

rephrasing negative consequences: "The worst things that can happen to you is you can do nothing about this and you can be the one at 40 weeks that gets a baby with Down's syndrome. And it's how would that affect you two and how would it affect your marriage. Is it your first baby? How it would affect you two, and you three as a family. At the other end of the scale, you could do everything possible and you could have all the tests going and you could miscarry and you could lose this baby and we could ring you up and tell you it was

normal. And it's how would that affect you two, you know, if you'd gone through all the testing then they tell you that the baby is perfectly normal."

4.2.4 Information giving differences by test decision

The frequency pattern of information classified by categories was presented both as a complete sample and by test decision (tables 4:1 and 4:2). No formal statistical analyses were carried out on these data for two reasons: the main purpose of the study was to describe the information provided, not to assess differences or associations; the results of an analysis would be meaningless because of the small sample size and large number of coding categories. However, broad observations made from the frequency tables suggest that women deciding to have or not have a diagnostic test generally received similar consultation information. Specifically, women choosing to have no further testing were: provided with more information about the fetal anomaly scan (hp2, table 4:1); more likely to be informed that the triple test was less reliable than other screening tests (hp8, table 4:1); more likely to have the risk of Down's syndrome minimised (hp11, table 4:2); less likely to be explicitly informed that the decision was their choice; more likely to be asked if they needed to know for certain whether or not the baby had Down's syndrome (hp14, table 4:2). Whereas women choosing to have a diagnostic test were: provided with more detail about diagnostic testing and miscarriage (hp3-hp5, table 4:1); more likely to be asked whether or not they would terminate for Down's syndrome (hp14, table 4:2).

4.3 Discussion.

The purpose of this study was to describe the information provided by the health professional when women were making the decision to have or not have prenatal diagnosis following receipt of a screen positive triple test result. In particular, the aim of the study was to assess whether or not sufficient information was being communicated in a neutral manner to enable informed decision making. From the few empirical studies that have described the prenatal diagnosis information-giving consultation (Kessler, 1981; Marteau et al, 1993; Bernhardt et al, 1998), findings suggest that there was incomplete information about the testing alternatives to enable informed decision making by women. The study reported in this chapter was one of the first to systematically document the provision of prenatal diagnosis information by the health professional following a screen positive triple test result. There are at least three consistencies between the findings of this study and those of prior empirical research.

First, there was little variation in the consultation content between women; and the information content of the consultations was similar to that of prior research (Marteau et

al, 1993; Bernhardt et al, 1998). Most women received the following information: an explanation of the triple test result and risk figure; details about amniocentesis and chorionic villus sampling, including information about the procedure, timing of the results, procedure-related miscarriage rate and identification of chromosomal disorders other than Down's syndrome; explicit reference to the woman taking responsibility for the final decision; explicit reference to the possibility of termination following a positive diagnostic test result. Further, the type of information routinely omitted was similar to that of previous research (Marteau et al, 1993; Bernhardt et al, 1998): a description of Down's syndrome that Included details of its prognosis, occurrence and recurrence rate; descriptions of the type of abnormalities not tested for, such as genetic disorders; explicit statements about the option to have a termination or not following a positive diagnostic test result. In addition, Marteau et al (1993) noted some misinformation given during the prenatal diagnosis consultations. This study also described some misinformation routinely mentioned during the consultation, the explanation provided for the screen positive cut-off point and the likelihood of mosaic Down's syndrome being identified; procedure-related risks of miscarriage.

Second, the communication of some information in all the consultations was directive (Marteau et al, 1993; Bernhardt et al, 1998). It was clear from the consultation transcripts that the health professional had a belief about a correct course of action and this was implicitly communicated during the consultation. To explain further, the health professional asserted in the majority of consultations that having a prenatal diagnostic test and terminating a pregnancy were associated decisions, i.e. that if a woman had a diagnostic test she *should* be considering termination of pregnancy for Down's syndrome. Although less frequently stated, the health professional also made a connection between having the triple test and choosing to have a diagnostic test, i.e. that if a woman has a screening test she *should* have a diagnostic test. There were a number of more subtle communications that may be seen as directive and suggestive of a 'correct' course of action (Kessler, 1992; Marteau et al, 1993; Bernhardt et al, 1998): more information was provided about the diagnostic test options and consequences than the no further testing alternative; the test alternative was framed as an active choice whereas the no test alternative was framed as 'doing nothing'; the procedure-related risks of miscarriage were half those documented in guidelines and were consistently compared with the triple test risk, so implying that the tests were a less risky option than no testing; concrete examples were provided of women having a screen positive test result, choosing a diagnostic test and the baby being 'healthy', but

no such comprehensive scenarios were given about women choosing to have no further invasive testing.

Third, despite the directiveness of some of the communications, the health professional attempted to promote autonomy in women's decision making by explicitly stating that the decision to have prenatal diagnostic testing was their choice (Bernhardt et al, 1998). Further, two additional communications within the consultation suggest that the health professional was providing conditions amenable with informed decision making: 54% of women were encouraged to take their time making a decision; risks were rephrased as both percentages and inverse risks in the majority of consultations. To explain further, reducing time pressure to make a decision is associated with the employment of more systematic decision making strategies (Donnai, 1992; Maule and Edland, 1997). In addition, the rephrasing of the risk figures is likely to reduce the effect of 'framing' and subsequent employment of heuristic processing strategies (section 1.2). Both of these factors are akin to a more informed process of decision making (section 1.2).

The structured observational study design was employed to assess the content and style of the information communicated by the health professional during the prenatal diagnosis decision making consultation. The checklist was developed to classify only the health professional's utterances during the prenatal diagnosis consultation. As a consequence, the findings reported in this chapter have limited application to understanding the relationship, if any, between the provision of information and women's prenatal diagnosis decision outcomes. For example, the frequency summaries suggested that women received broadly similar information by test decision. However, frequency patterns from a few sub-categories appeared to differ by test decision. From this analysis of the data it is difficult to ascertain whether the health professional modified the information given in response to comments from the women stating a preferred course of action or whether the women's prenatal diagnosis decision was made following changes to the information provided by the health professional. There is little empirical research systematically evaluating the relationship between the style and content of information with subsequent decision making outcomes in the context of prenatal diagnosis for Down's syndrome (Marteau et al, 1993; Figueires and Marteau, 1999). It is likely that some directively communicated information is detrimental to the informed decision making process, whereas another communication provides appropriate reassurance (Kessler, 1992). An example might be the health professional's description of the triple test risk for Down's syndrome (section 4:2.2): stating a value judgement for a 1 in 230 risk as 'low' may influence the woman's evaluation of the risk

and, therefore, the subsequent decision about testing; stating that most women will have a normal baby or "the odds are in your favour at 230 to 1" is a more accurate evaluation of the risk and may be appropriately reassuring. As this thesis is concerned with informed rather than shared doctor-patient decision making (Coulter, Entwhistle and Gilbert, 1999), it is beyond its scope to explore the woman-health professional interaction further.

4.4 Chapter Summary.

The main aim of the study was to assess whether or not the information-giving consultation would enable women to make an informed and autonomous decision about prenatal diagnostic testing. The findings reported in this chapter suggest, then, that women had sufficient information to have a diagnostic test but not to make a decision to have or not have a diagnostic test for Down's syndrome. In addition, some of the information communicated implied a correct course of action, that of having a diagnostic test. However, the health professional did explicitly specify the women's role in and responsibility for making this prenatal diagnosis decision. Although these findings indicate a comprehensive provision of test information for women, it is unlikely that the conditions have been met to enable women to make a fully informed decision about prenatal diagnosis following receipt of a screen positive triple test result. For the consultation to contain 'sufficient' information, a description of Down's syndrome and a more detailed discussion of the decision to terminate or not should be included. Reducing the directiveness of the information communicated requires a number of changes: rephrasing the no invasive testing option as an active choice; the provision of accurate information or reduction of misinformation; explicit emphasis on any course of action being acceptable, such as whether to have no invasive testing or testing for information only.

5. Prenatal diagnosis: describing the process of women's decision making.

This chapter focuses on describing the decision making process of women choosing to have or not have prenatal diagnosis upon receipt of a screen positive triple test result. In particular, this chapter is concerned with describing whether or not women's prenatal diagnosis decision making was informed. One of the main objectives for those offering prenatal diagnosis is to ensure that women make an informed decision about testing (Royal College of Physicians, 1989). As there are few agreed criteria on what constitutes an informed decision within the medical literature (Marteau, 1995), there is little empirical research operationalising and assessing patients' informed decision making (see chapter two). However, proponents of reasoned-choice theories of decision making (Janis and Mann, 1977; Zey, 1992, 1998; Baron, 1994; Frisch and Clemen, 1994) have developed a standard with which to evaluate the quality of individuals' decision making. This standard of good decision making provides an adequate operationalisation of informed decision making (see section 1.2). In brief, assessing the quality of a decision requires an appraisal of the decision making process, i.e. a description of the strategies employed to reach a decision. A good decision ought to be based on an evaluation of the consequences of the different options, an accurate assessment of the consequences and a trade-off between the consequences of the decision alternatives (Frisch and Clemen, 1994; section 1.2).

Few, if any, empirical studies evaluating the quality of women's prenatal diagnosis decision making have employed a methodology to assess the decision making *process* (see section 1.3.2). The majority of studies used a prospective or retrospective study design. In other words, women completed questionnaires or were interviewed before or after they had made their decision to have or not have prenatal diagnosis. No published studies assessed the decision making process concurrently with women making the prenatal diagnosis decision. In addition, the dominant decision making theory or paradigm underpinning the development of these questionnaires and semi-structured interviews is that of expected-utility theory (EUT). That is to say, there is an assumption amongst researchers that women's prenatal diagnosis decisions are logically associated with evaluations of the likelihood of the consequences occurring and the value attached to their occurrence. In consequence, most empirical studies within the prenatal diagnosis context have assessed women's attitudes, perceptions of risk and measures of decision outcome, such as knowledge, affect and satisfaction. The findings from

these studies suggest that women do make decisions about prenatal diagnosis by weighing up the advantages and disadvantages of the two testing alternatives (see section 1.3.2). In addition, factors such as attitudes towards termination of pregnancy, perception of the risk of abnormality and perceptions of the miscarriage risks are associated with the decision to have or not have testing. However, there is little empirical evidence assessing whether or not women made informed decisions, as no published studies to date have operationalised informed decision making.

The naturalistic decision making (NDM) approach to understanding individuals' decision making has been to describe the strategies employed by individuals when making real-world decisions (Lipshitz, 1993). In part, the development of NDM research was a response to findings of earlier studies which ascertained that individuals have a 'bounded rationality' (Simon, 1955). To explain further, it is unlikely that individuals have the cognitive capacity to assimilate evaluations of the probabilities and values associated with the consequences of all decision alternatives as proposed by EUT (Allais, 1953; Ellsberg, 1961; Tversky, 1969; see section 1.2). Indeed, evidence suggests that individuals employ information processing strategies such as *heuristics* and *satisficing* to limit the amount of information assimilated when making a decision (see section 1.2.1.2). If individuals selectively attend to aspects of the decision information, it is unlikely they are making fully informed decisions. As no published studies to date have described the process of women's prenatal diagnosis decision making, it is unclear what strategies women employ when assimilating the test information and whether or not these strategies facilitate or impair informed decision making.

The purpose of the study reported in this chapter is to describe the process of women's decision making about prenatal diagnosis following receipt of a screen positive triple test result. As cognitions cannot be directly measured, a *process tracing* technique was employed to assess the information utilised by women during the prenatal diagnosis information giving consultation. Two common techniques employed to trace the decision making process are *verbal protocols* and *information tracing techniques* (Payne, 1980; Harte, Westenberg and van Someren, 1994; Harte and Koele, 1997). As discussed in chapter 1, the most direct process tracing technique is that of the verbal or 'think-aloud' protocols. The researcher encourages the individual to verbalise the thoughts raised while making the decision. Verbal protocols do not require the individual to reflect on the decision making process but to talk through normally silent cognitions.

Although it is unlikely that the generation of verbal protocols alters the decision made, there is evidence that verbal protocols slow down the decision making process (Payne, 1980). In additional, there is evidence that introspecting about the decision making process may be associated with greater post-decision regret or dissatisfaction (Wilson, Lisle, Schooler, Hodges, Klaaren and Lafleur, 1993; Wilson, Hodges and Lafleur, 1995). As women find the prenatal diagnosis decision distressing (section 1.1.4), such an active tracing technique was not considered appropriate for this health context and decision.

The information search methods are less interactive and require the researcher to document the information utilised or referred to by individuals when making their decision. The use of an audio or video tape-recorder would record the information referred to by women concurrently with their decision making. The information giving consultation women receive prior to making their prenatal diagnosis decision provides a good opportunity to employ an information search technique. In each consultation, the information provided by the health professional is consistent (see chapter four) and the purpose of the consultation well defined. That is to say, the conditions of the decision context are comparable for each woman making the decision to have or not have prenatal diagnosis upon receipt of a screen positive triple test result.

The aims of this study are (a) to describe the information referred to by women when making the prenatal diagnosis decision, (b) to assess differences in the pattern of information referred to by women choosing to have or not have prenatal diagnosis and (c) to evaluate whether or not the decisions made by women were informed.

5.1 Methodology.

5.1.1 Sample.

Forty-six women who received a screen positive triple test result from the Leeds General Infirmary (LGI) were invited to take part in this study. Forty-four women agreed to participate (section 4.1.1 provides a more detailed explanation of the triple test screening protocol for the LGI).

5.1.2 Design.

A non-experimental, cross-sectional observational study design was used to describe the process of women's decision making during this information giving consultation. An audio tape-recorder was used to record the content of the prenatal diagnosis

information-giving consultations (see section 4.1.2). The consultation audio tape-recordings were transcribed fully by a third party (KM). The transcribed women's utterances provided the data source for this study.

The demands of this study's research questions were important in selecting an appropriate sample size for subsequent analysis. Studies aiming to generate data-driven theories about women's decision making are inclined to focus on between five and twelve transcripts (Payne, 1980: Smith, 1995). The transcripts may require detailed analysis on a number of different levels such as: noting the occurrence of certain words or phrases; carrying out a phenomenological interpretation of utterances to suggest associations between phrases with underlying meanings of pertinence to the individual; coding of paralinguistic features such as interruptions, intonation, pauses and repetition; applying a theoretical analysis to make associations between the different patterns coded from within the data (Robson, 1993; Charmaz, 1995; Smith, 1995). However, the purpose of this study was to describe the process of women's decision making concurrently with the decision to have or not have prenatal diagnosis.

A quantitative content analysis (Millward, 1995) was applied to the consultation transcripts to identify themes or categories classifying women's utilisation of information when making the prenatal diagnosis decision. This type of analysis provides a numerical summary of the presence or absence of theoretically driven categories within the real-world setting rather than the generation of a new theory. In this decision context, theories of decision making and empirical evidence from prenatal diagnosis studies were used to inform the identification of themes and subsequent categories. Each consultation transcript was assessed for the presence or absence of these themes. In other words, the transcript data was systematically coded and interpreted with reference to an appropriate literature (Millward, 1995; Bowling, 1997). Although no optimum sample sizes were provided for this type of analysis, data from 20 individuals is referred to as small and from more than 60 as unlikely to contribute more to the interpretation of the data (Millward, 1995). In essence, the sample size for this study was based on a compromise between adequately answering the research questions within the permitted study time and consideration of the aforementioned range of participants.

5.1.3 Procedure.

The procedure employed for this study is the same as that described in chapter four. In brief, all women with a screen positive triple test result were informed of their result by telephone or letter if a telephone number was not available. On attending the antenatal clinic for the prenatal diagnosis information giving consultation, women were informed of the study. The consultations of women agreeing to participate were audio tape-recorded. The utterances of 22 women included in this study were transcribed from the same consultations described in chapter four.

As mentioned, this sample size was an insufficient number to carry out a quantitative content analysis. To increase the number of participants in this analysis, transcripts from another study audio tape-recording this information giving consultation were added to the analysis. To explain further, the author (HB) carried out a randomised control trial to assess the effectiveness of decision analysis in facilitating women's decision making about prenatal diagnosis. Although this study is described in greater detail elsewhere (see chapter six), it is necessary to mention its control and experimental groups. The information giving consultation for women randomised to the control group was the same as that described in chapter four, i.e. routine care offered by the LGI. The information giving consultation for women randomised to the experimental group received information structured by the decision analytic technique (section 1.2.1.3). As mentioned, the author (HB) was introduced to women as a member of the ante-natal team throughout the data collection period of this thesis. Initially, the author observed the content and structure of the information giving consultation in order to be acquainted with the routine delivery of information in this health context. Once the ante-natal team was confident of the author's ability, the author became the primary information-giver for women receiving a screen positive triple test result. The author aimed to follow the same pattern of information giving and to provide the same type of information during the control arm of the trial as was offered routinely by the ante-natal staff at LGI. In essence, the consultation described in chapter four and the control group of the randomised control trial were comparable consultations. To increase the sample size for this chapter's analysis, the consultation transcripts of the first 22 women allocated to the randomised control trial's control group were also included for assessment. During this six-month data collection period of the randomised control trial, two women invited to take part declined participation.

5.1.4 Coding frame development.

Transcripts of consultations generate a large amount of qualitative data. The purpose of a coding frame is to develop themes to summarise this plethora of information in a meaningful manner. By applying the same coding frame to each transcript, information can be extracted and classified systematically. The most important aspect of content analysis, then, is the generation of themes (Robson, 1993; Bowling, 1997). The themes or categories group items together, thus enabling the data to be managed more efficiently. The theme content may be defined according to an aspect of theory or following assessment of similarities between items within the transcript data (Millward, 1995). Themes are designed to be both exhaustive and mutually exclusive (Robson, 1993; Smith, 1995). That is to say, all items within a transcript can be assigned to one theme only. In practice, the items derived from conversational transcripts are often inter-related and not independent utterances. Therefore, in some situations, the researcher is either required to make judgements about an item to ensure exclusivity or to occasionally multiply categorise an item (Robson, 1993). The generation of themes is laborious and time-consuming as it requires continual reference to the transcript, items extracted, working themes and theoretical basis of categories (Robson, 1993; Smith, 1995).

The themes for this study were developed in five stages. First, the unit of analysis was defined, i.e. the aspect of the text to be classified. Second, the items identified from preliminary analysis of the text were clustered by similarity of meaning. Third, preliminary themes and theme titles were generated from these item clusters and piloted on a half the transcripts. Fourth, the themes and theme titles were revised to incorporate changes from the first piloting and applied to remaining transcripts. Finally, the themes and theme titles were modified again to form the final coding frame. The final coding frame was applied to all forty-four transcripts. These five stages are discussed in more detail below.

5.1.4.1 Unit of analysis: coding frame development.

So far, the woman has been referred to as the decision maker because (a) the woman experiences the consequences of any prenatal diagnosis decision and (b) it is the woman's consent that is required before any procedures can be carried out. However, it is unlikely that the decisions made about tests, terminations or continuations of a pregnancy are made solely by the woman. Indeed, most women (40/44) attended this information giving consultation with a companion to whom the purpose of the

consultation had been disclosed. During the consultation, it was frequently observed that the companion and woman decision-maker talked with each other about the decision and finished off the other's sentences. In addition, the companions raised questions and concerns of their own as well as referring to issues expressed previously by the woman decision-maker. The woman decision-maker was exposed to both the reasoning, questions and additional information following a question of the companion during the consultation. The purpose of the study was to describe the information referred to during the consultation and not the nature of the interactions between the health professional, woman and companion. As the woman decision-maker and companion were both focused on issues associated with making the decision to have or not have a test, utterances made by either party were coded as information utilised during the consultation. For consistency, these collective utterances were referred to as those of the woman decision-maker.

The unit of analysis, coding unit (Millward, 1995) or recording unit (Robson, 1993) is the verbal utterance that is used to form the content of the categories in the coding frame. This unit of analysis may be anything from counting the occurrence of words to the number of stories on a page within a newspaper. Again, the researcher defines the unit of analysis in accord with the aims of the research questions. Complete phrases were used as the unit of analysis within this study. As the transcripts were of conversational interactions, a phrase could be part of a sentence or include more than one sentence. The phrase ought to provide a unit of analysis that unambiguously referred to a piece of information used by a woman while making the prenatal diagnosis decision. All complete phrases or 'items' from each transcript were recorded on a separate document referred to as the 'item-list' (see Appendix VI). This document was updated following analysis of each transcript. Once all the items had been generated, an item-list was completed for each transcript so documenting the number of times an item was referred to by a woman. Some examples of the types of items included on the item-list are: "Down's syndrome is not in our family"; "I had the triple test to find out so I should have a diagnostic test"; "my husband would not cope with a Down's syndrome child"; "Does the test hurt?".

5.1.4.2 Item clustering: coding form development.

Broad titles were created to aid the first author in locating items coded when updating the item-list (see Appendix VI). Items were clustered under the following titles: questions asked; comments about the triple test; use of risk figures; perception of risk; causes of Down's syndrome; reasons for not having a diagnostic test; reasons for having a

diagnostic test; reasons for choosing one diagnostic test over another; planning to have a test or not; perceptions of having a child with an abnormality; comments about termination; perceptions of norms; experience of Down's syndrome; experience of prenatal and genetic testing; comments about pregnancy and health; expressed affect and dissonance with decision making; previous advice or comments by health professionals.

5.1.4.3 Preliminary themes and titles: coding frame development.

The preliminary themes were informed with reference to both decision making theory and empirical evidence from studies of women's prenatal diagnosis decision making previously summarised in chapter one. The preliminary themes focused on issues of women's informed decision making within the context of prenatal diagnosis and are described in more detail below.

Classical decision making theory: The expected-utility explanation of decision making has influenced much of the prior research assessing women's prenatal diagnosis decision making. In consequence, attributes of EUT, such as evaluations of attitudes and risks associated with the decision consequences of alternatives, provided themes for subsequent categorisation of items.

Decision making strategies: The NDM models and evidence from studies describing the decision making process suggest that a number of strategies are used by individuals when making decisions. The coding frame included themes to classify examples of either an heuristic or systematic evaluation of the decision information. In addition, the evidence from real-world decision making research suggests that people use reasons rather than attitudes to rationalise their decisions (Shafir, Simonson and Tversky, 1993). In consequence, reasons for and against attributes of the prenatal diagnosis decision were included as themes.

Informed decision making: The seven stages of vigilant information processing (Janis and Mann, 1977) provided additional themes for classification of women's utterances. In brief, these seven stages include: canvassing a wide range of alternatives; identifying values associated with the consequence of the choice; weighing the costs and benefits of each consequence; search for new information; correctly assimilating new information; re-examining the costs and benefits of the consequences; making provision for the implementation or execution of the chosen alternative.

Autonomy in decision making: Empirical evidence that suggests women may receive directive information from others that compromises their ability to make an informed decision. A theme was included to classify utterances of women which indicated that the woman had received advice from a third party about a particular course of action.

Decisional conflict: Empirical evidence suggests that women find making the decision to have or not have prenatal diagnosis difficult. The decision making literature refers to this difficulty in choosing between two risky alternatives, both with negative consequences, as decisional conflict. One theme classified items as pertaining to explicit utterances about the experience of making a decision.

Affect: Aside from decisional conflict, there is empirical evidence to suggest that women are distressed upon receiving a screen positive triple test result. In addition, some women expressed reassurance upon receipt of tests during pregnancy. One theme classified utterances that expressed some dimension of affect.

The items from the first set of 22 transcripts were grouped with reference to these themes. During this process, it was clear that the Janis and Mann stages of systematic decision making and the expression of attitudes towards the consequences of the decision were inappropriate themes to categorise women's utterances. First, women's information seeking about alternatives could be classified as both information seeking and/or planning to implement a chosen course of action. Second, women seldom made explicit reference to the utility or attitude of a consequence; consequences were more likely to be referred to within either a positive, neutral or negative frame. For example, women were more likely to say "I don't want to miscarry" rather than "miscarrying is bad" or "I couldn't cope with a Down's syndrome child" rather than "I perceive the severity of Down's syndrome to be great". These themes were revised during the next stage of the coding frame development.

5.1.4.4 Revised themes and theme titles: coding frame development.

The revised themes simplified the categories describing informed decision making. Items about the 'no test' and 'test' alternatives were grouped into one of four categories: information seeking about the alternative; consequences of the alternative; systematic reasons for selecting or rejecting the alternative; heuristic reasons for selecting or rejecting the alternatives. A systematic reason was one that referred to the consequences of the alternative, such as "I want a diagnostic test because I would terminate for Down's syndrome"; a heuristic reason was one that did not refer to the

consequence, such as "I had the triple test, so I should have the diagnostic test". The two themes that grouped items by actual and perception of risk were maintained. In addition, a number of themes were developed to account for the information referred to during the consultation outside the parameters of informed decision making: further information about the triple test; affect about the triple test; affect about decision making; reflections on the decision making process; questions about Down's syndrome; reference to their personal prenatal testing experience; reference to others' prenatal testing experience; reference to other sources of information. These themes provided a more comprehensive classification system for coding all women's utterances.

Each phrase on the item-list was allocated to a theme. The appropriateness of the item included within a theme was discussed with a colleague familiar with the applied literature on prenatal testing (JH). The revised coding frame was applied to the second set of 22 transcripts. In addition, a colleague familiar with the area of genetic counselling (AM) along with the first author applied the coding frame to three transcripts selected at random from the first set of twenty two transcripts. The discussions from these activities and the difficulties in allocating items to themes initiated further changes within the coding frame:

- The informed decision making themes were simplified to reflect the three stage standard proposed by Frisch and Clemen (1994): an evaluation of the consequences of the different options; an accurate assessment of the consequences; a trade-off between the consequences of the decision alternatives.
- The distinction between 'heuristic' and 'systematic' reasons for selecting an alternative was unreliable and, in consequence, removed from the coding frame. The simplified categories classified reasons for an alternative as either reasons for or against a course of action (Shafir, Simonson and Tversky, 1993). An additional category was required to classify the seeking of information rather than the generation of reasons for or against an alternative.
- Previously, the consequences associated with an alternative were attributed to either the 'test' or 'no test' alternative and classified accordingly. However, the consequences may be associated with both decision alternatives. For example, a woman may have a Down's syndrome baby whether or not she chooses to have a diagnostic test. Three themes were subsequently included to classify items pertaining to the consequences of the prenatal diagnosis decision independent of the two decision alternatives. The consequence themes contained three sub-categories to classify utterances as either information seeking, positively framed or negatively framed references.

5.1.4.5 Final themes and titles: coding frame development

The revised coding frame was applied to all transcripts (see Appendix VII). Twenty themes were developed to include issues of informed decision making and other factors referred to when making the prenatal diagnosis decision. Five transcripts were selected at random to assess the inter-rater reliability of coding between the first author and a colleague (AM). In total, ninety statements were coded by both the author and AM. Cohen's Kappa coefficient was calculated to assess the degree of concordance between the two raters' application of the checklist to the five transcripts. The value of the Kappa coefficient was 0.83. As this figure falls above 0.75, it suggests an excellent degree of agreement between the two raters (Robson, 1993). The themes and sub-categories that classify women's utterances during the information giving consultation about prenatal diagnostic testing are described in more detail below. Quotes from the transcripts of women's utterances are used to illustrate further the theme's content.

1. No test option: The theme included items referring to alternatives other than the diagnostic tests offered, such as having no invasive testing, and to queries about the non-invasive alternatives such as the nineteen-week fetal anomaly scan. The theme had three sub-groupings: information seeking or planning for this alternative; reasons in favour of this alternative; reasons against this alternative. The following are examples of the information seeking items coded within this theme: "If we went on, can they test the baby when it is born?"; "Would it be possible to wait and see from the scan?"; "What sort of markers do they look for in the scan for Down's syndrome?"; "But there might be markers if it's an ordinary person?". The following are examples of reasons generated for an alternative; "So we have the reassurance of the scan anyway"; "I would like to have the scan and know how much information you can get from the scan"; "The advantage is that there is no risk to the child". The following are examples of reasons generated against an alternative: "The scan might not tell us for definite"; "I definitely couldn't go on for six months without knowing"; "There's also that risk that if you don't, for the full term baby [being Down's syndrome]".

2. Test option: The theme included items about diagnostic tests in general. The same three sub-groupings as the 'no test' option were retained. The following are examples of the information seeking items coded within this theme: "So if you decided to have the test, how soon can it be done?"; "Are the tests a hundred per cent reliable?"; "I read the more experienced the person the less risk there is so obviously we wouldn't want anyone inexperienced doing the test"; "Do they scan while they are doing it?". The following examples as reasons generated for this alternative: "I think for our own peace

of mind we have got to know"; "But I would like the choice, I think I would like to know, I think I would like to make an informed choice"; "I think it's better to know. It's better to prepare ourselves"; "I know that there is that risk you can miscarry but I think it outweighs the problem having, at nine months, a baby that is going to be [Down's syndrome]". The following are examples of reasons generated against this alternative: "I'm pretty sure in my mind I don't want amniocentesis or chorionic villus sampling, I suppose my reasoning is that it is likely to be a normal baby and there is quite a high risk of miscarriage"; "the thought of having needles stuck in my stomach is completely horrible"; "there is a bit of me that doesn't want to know"; "I think I would prefer not to have an invasive test".

3. Choice between tests: This theme included items that made neutral comments about choosing between tests, for example: "It gives us time to think about it and discuss which test we would want to have then"; "So both tests is what, a three week period waiting for them [the results]"; "Why would you want to do a chorionic villus sampling rather then an amniocentesis?" .

4. Chorionic villus sampling: Items referring explicitly to chorionic villus sampling were classified within this theme. The sub-groupings were the same as the previously defined. So, examples of information seeking utterance about this testing option are: "I'm just going through it in my head . . . the next day you would be able to say yes it definitely has got it but you wouldn't be able to say no it definitely hasn't until three weeks afterwards?"; "Is the risk of miscarriage with chorionic villus sampling less or greater if you're further, you know, for your pregnancy"; "Are the chances of miscarriage the same as with the other [amniocentesis] test?". The following are examples of reasons for the test alternative: "I can see the attraction of the chorionic villus sampling with it being so quick"; "The only advantage of that is you get a quicker result?". The following are examples of reasons against the alternative: "You are taking an additional one per cent risk just for that [quicker result] and there is no need for that at this stage anyway"; "You would have to wait three weeks anyway for the chorionic villus sampling to be absolutely positive"; "But the miscarriage risk is a lot higher for that isn't it".

5. Amniocentesis: Items referring explicitly to amniocentesis were classified within this theme maintaining the same sub-groupings as previously used. Examples of the information seeking utterances are: "Do you have a per centage on how many people have the amniocentesis?"; "You know the amniocentesis, how long do you wait for that one [the results]?"; "[Does] the risk of amniocentesis vary between clinics?". The

following are examples of reasons generated for the alternative: "You've still got to go through the same outcome [termination] whether it was tomorrow or three weeks, so I would rather cut down the risk"; "It's worth it, just for the peace of mind, it's worth waiting those three weeks"; "I suppose my initial reaction would be to sit with the amniocentesis"; " the reasons that we had the amniocentesis last time are even stronger this time". The following are examples of reasons generated against the test alternative: were "Three weeks is a long time to wait"; "The problem we have got is that with the amniocentesis there is an elevated risk of miscarriage"; "I hate the three or four weeks waiting".

6. Down's syndrome (consequence): Items referring explicitly to the consequence of having a baby with Down's syndrome were classified within this theme. The three sub-classifications were maintained but were referred to as information seeking, positive or neutral reference and negative reference rather than information seeking, reasons for and reasons against. The following are examples of information seeking items about the consequence: "Does it make any difference that we are half cousins, I mean there is no history of disabilities on either side of the family?"; "The thing is that if we are told at some point that this is likely to be a Down's syndrome baby then obviously I would want to know a lot more about life expectancy and the quality of life"; "Have you any idea what actually causes this extra [chromosome]?"; "Can they tell straight away when it's born?". The following are examples of neutral or positive comments about Down's syndrome: "A kid with Down's syndrome is fair enough . . . it's still a child"; "I have heard them on the radio, they can read"; "We know what a Down's syndrome baby is . . . It's a very wide ranging disability"; The following are examples of negative references or images of the consequence; "I'd be hysterical if I had a baby like that, I just couldn't cope with one"; "We wouldn't be happy with the responsibility of a Down's and we'd always be worried"; "I know this sounds awful but it [baby with Down's syndrome] is imperfect and that is how we would see it".

7. Miscarriage (consequence): All items referring to miscarriage were included within this theme with the sub-classifications of information seeking, positive or neutral comments and negative comments. The following are examples of information seeking itmes about the consequence: "What creates the risk of miscarriage?"; "How long would it be if I were to miscarry, within days?"; "Can I ask an ironic question? If I were to miscarry as a result of having the amniocentesis, do they tell you whether the baby was OK or not?"; The following are examples of the neutral or positive comments about the consequence: "I'd rather take the chance [miscarry as a result of test] than worry for the

next few months"; "If we have a miscarriage, we have a miscarriage and that's it. We'll try again". The following are examples of negatively classified items: "It's the risk of miscarriage that bothers me"; "I'm not scared of amniocentesis, I'm scared of miscarrying as a result".

8. Termination (consequence): All items referring to termination were included within this theme, with the same sub-groupings as the other consequence themes. Examples of information seeking items are: "If it's got Down's, now am I right in saying that in three weeks time if it's got Down's and we have to terminate we have to go through labour, now is that the case?"; "Let us assume that it is Down's syndrome, how soon after that would I be booked in for termination"; "What's the last date you can terminate?". Positive or neutral items about termination were: "Well, we said we would have a termination"; "Because as I say, I don't think we would have gone through with the tests if we weren't going to do something about it"; "I'd consider termination I think". Examples of negative references to termination were: "I wouldn't terminate, I don't think I could"; "So that's [labour termination] going to be pretty horrendous"; "I don't think we would want an abortion for the sake of Down's syndrome".

9. Risk figure reference: Simple categories of reference to risk were developed for this study in an attempt to describe general patterns of information used by women when making the decision to have a diagnostic test. This theme's sub-groupings were reference to a single risk figure for Down's syndrome, a single risk figure for miscarriage and a comparison of risks. Examples of the single risk reference to Down's syndrome are: "Did you say the borderline was 1 in 250?"; "So basically we've got less than a 1% chance having a Down's syndrome child?"; "I just wanted to know exactly how sort of you knew it was 1 in 250?". Examples of the single risk figure for miscarriage were: "So that 1 in 100 is a national figure?"; "The current risks, do amniocentesis run into 1 in 100 or 1 in 200?". Examples of the comparison between risks are: "It's the balance of the risk of not knowing against the risk of [miscarriage], it's not brilliant"; "I think the risk of Downs of 1 in 10 is slightly higher than that of the procedures we are talking about".

10. Perception of risk reference: Four sub-groupings were used to classify perception of risk items: minimising Down's syndrome risk; augmenting Down's syndrome risk; minimising risk miscarriage; augmenting risk miscarriage. Examples of minimising risks are: "I wouldn't bet on that"; "It's still pretty good odds though"; "It's only 1%"; "So it's a very small chance". Examples of augmenting risks are: "But you never know if you're

going to be that 1%"; "I know but it's still a lot"; "There is quite a high risk of miscarriage".

11. Decision making process (decisional conflict): Women's utterances pertaining to the decision making process were classified by this theme. The sub-categories reflected decision making about the following: the triple test decision; making a diagnostic test decision before the information giving consultation; expressed delay in making the diagnostic test decision. Examples of each of these categories are: "We wanted the triple test for our own personal knowledge"; "We had made it [decision to have diagnostic test] before we had the triple test"; "I think we might need to think about it for a bit".

12. Expressed affect: This theme incorporates three sub-groupings pertaining to items of affect mentioned by women during the consultation: negative comments about receipt of the triple test result; phrases reflecting difficulty or 'decisional conflict' in making the diagnostic test and termination decisions; references to reassurance or positive phrases about testing. Examples of items within each category are: (receipt triple test), "It's a bit of a shock really", "It's hard just to take it in isn't it?" and "I mean, it's just another knock is this"; (decisional conflict), "It's a hard decision", "I mean all the way through you're thinking we'll face that when we come to it", "I don't want to do it but that is the decision, it sounds ambiguous doesn't it?" and "morally it's the hardest decision because technology puts that decision upon you doesn't it?"; (reassurance about tests), "It [triple test screen negative] might be reassuring for some women".

13. Triple test: This is a broad theme incorporating all the items expressed by women about the triple test. No sub-classifications of this theme have been created as the decision to have or not have a diagnostic test should, theoretically, be independent of the previous decision to have the triple test. Some examples of the items within this category are: "What do they actually test for, it's Down's and what else?"; "I never got screened with the first baby and if I had of been would that have been high as well?"; "Would they have told us if it was over 250?"; "I'm just clutching at straws here but can the bleeding I had, can that have an abnormal reading on the blood results?".

14. Confidence with screening results: This theme includes items mentioned by women when discussing the reliability of screening tests and their results. Some examples are: "But the hospital she had the triple test done in is not doing it anymore because they say there were such incoherent inaccuracies in the triple test"; "So that

[triple test risk result] is more factual than that [age related risk result]?"; "Yes it's strange that just a week could make such a difference . . . you know it's just curious".

15. Comparison with norms: The items within this theme were mentioned by women trying to establish a consensus with the normative prenatal diagnosis response. For example: "On the whole what, if people are at this stage, what do they do?"; "Which is the best one to have then? What would you recommend?"; "Out of those people could you actually give a percentage who have actually had the test that have said 'yes, it's a Down's' or 'not it's not'?"; "Am I common or not [triple test result risk figure]?".

16. Personal experiences: Within this theme the four sub-groupings were items referring to the woman's experience: physical changes with pregnancy; complications with pregnancy; previous pregnancies and prenatal testing; familiarity with abnormalities or genetics. Examples for each category are: "It's different once you start feeling something" and "The worse thing was I saw it last week on the scan"; "It is difficult because we have lost three pregnancies" and "I've been to the doctor's this morning because I had a bleed yesterday and a slight bleed this morning"; "We had a child about 18 months ago and we had the triple test done with her" and "We hit a Bank Holiday last time . . . nearly four weeks before we got the results"; "I've had quite a few scans and the sort of genetic risk was 50-50".

17. Others experience: Items were coded under two sub-groups within this theme, prenatal testing and abnormalities or genetics. Examples of each sub-category are: "A friend recently who was pregnant with twins and she had amniocentesis and she lost them both and they were normal girls" and "My sister had a baby . . . and she had a high result as well"; "I mean 'cause my friend had a Down's" and "My cousin is Down's syndrome, I know how hard it is".

18. Preparation pregnancy and birth: Examples of items are: "The other thing I would be scared of is an epidural"; "It's a shock finding out I'm pregnant, it's going to be hard with a healthy baby to look after and work . . . "; "I just don't think there's any point in telling the parents because, you know, if the worst comes to the worst . . . I think we'll hang, wait three weeks".

19. Health professionals' advice: The items within this theme were classified under the following sub-groupings: neutral-positive comments about consultations; negative-dissatisfied comments about consultations; contradictory information to current

consultation. Neutral-positive items included the following examples: "I was very lucky when I was first pregnant and went to see my GP, he was very good and talked me through"; "It's really helped to talk it through"; "'Cause the doctor said they can't do a lot of tests 'til you're so far on [in pregnancy]". Negatives or dissatisfied items included: "I've just heard a lot of midwives say they wouldn't have it [the triple test]"; "I just don't think there was enough feedback about the test, I mean how vital it [dating scan] would have been". Examples of items pertaining to contradictory information are: "That's interesting 'cause I was given different information when I asked before about the triple test. I said 'does it screen for anything else other than Down's syndrome' [and she said] 'no'".

20. Other sources of information: Women mentioned referring to other sources of information about pregnancy and prenatal tests, such as the media and hospital literature. The phrases that follow are examples of these items, "I've read it all before I came" and " I mean I've looked it up a lot and there was that programme recently".

5.1.5 Analysis.

The preliminary aim of this analysis was to establish what information was referred to by women when making the decision to have or not have a diagnostic test. In other words, the subsequent findings will illustrate a pattern of information searched by women concurrently with their decision making. In consequence, the subsequent analyses are based on data that represent whether or not a woman referred to an item or theme. That is to say, a woman could score only '0' or '1' for each theme or sub-categorisation, i.e. a woman either mentioned an item within the theme during the consultation or not. In this instance, the data do not report how many times a woman referred to the themes. The analyses and results are presented in three sections:

- Descriptive statistics are used to summarise the frequency with which themes were referred to by women when making the decision to have or not have testing (table 5:1).
- Descriptive and univariate analyses are used to illustrate differences in the pattern of information utilised by women choosing to have either a diagnostic test or no further invasive testing (table 5:2).
- Four informed decision making variables were created from the data contained within the following themes: no test alternative; test alternative; amniocentesis; chorionic villus sampling; choice between diagnostic tests; Down's syndrome (consequence); miscarriage (consequence); termination (consequence). To explain further, an informed decision is based on an evaluation of the information about the

consequences of each alternative and a trade-off between the advantages and disadvantages of the consequences. These theme titles of the coding frame classify the information required to assess whether or not women made informed prenatal diagnosis decisions. In other words, summarising the information referred to under these themes provides an operationalisation of informed decision making. Four variables were calculated from these themes: a total informed decision making score (IDM-T); a score summarising the information seeking utterances of women about the test alternatives (IDM-I); a score summarising the generation of reasons for selecting an alternative (IDM-F); a score summarising the generation of reasons against the selection of an alternative (IDM-A). The IDM-T score was a global measure assessing whether or not women made reference to any items pertaining to the no test alternative, the test alternative, Down's syndrome, miscarriage and termination. For the purposes of the IDM-T score, items referring to all utterances about the test alternatives were aggregated (aggregated themes were the test alternative, amniocentesis, chorionic villus sampling and comparison between tests), If, for example, a woman mentioned any item classified under the 'no test' theme she scored '1', if she did not say anything about the 'no test' alternative she scored '0'. The IDM-T score was calculated by addition of the five recoded alternative and consequence themes (range 0-5). The remaining three informed decision making variables provided summaries more representative of the decision making process: information seeking; reasons for an alternative; reasons against an alternative. Scores for these variables were generated as follows: if women referred to an item classified as information seeking for the above themes, the score was '1' for each theme; if the woman did not refer to an information seeking item, the score was '0'. These recoded information seeking sub-category scores were added together to form the IDM-I score (range 0-8). The same process was carried out twice more for women referring to items classified either as reasons for or reasons against alternatives under each of the aforementioned themes. However, omitted from the IDM-F and IDM-A calculations was the theme that classified utterances comparing the two diagnostic tests. As these comparisons simultaneously provided both a reason for selecting one test and for rejecting another, they were incorporated in the information seeking informed decision making variable only. In consequence, the range of the IDM-F and IDM-A scores was 0-7. Univariate and multivariate analyses were carried out to (a) describe women's informed decision making (table 5:3 and table 5:4) and (b) assess differences in informed decision making between those choosing to have a test with those choosing to have no further invasive testing (table 5:5).

5.2 Results.

5.2.1 Describing the pattern of information utilised by women: results.

The average consultation length was 22.7 minutes (s.d. 7.0; range 8.6 - 40.7). Eight women chose to have no further diagnostic testing; thirty-six women had a diagnostic test (twenty-nine had amniocentesis, seven chorionic villus sampling). Table 5:1 illustrates the pattern of information utilised by women when making the decision to have or not have prenatal diagnosis. The frequency of reference to theme titles suggests that over two-thirds of women utilised the following information during decision making: screening and the calculation of the triple test result; the test alternative; termination consequences; risk figures; perception of risk; reflection on the making of prenatal testing decisions; personal reproductive experiences. Fewer women referred to themes incorporating items about: the 'normative' prenatal testing behaviours, i.e. what other people do in this situation; the no test alternatives; Down's syndrome; miscarriage; expression of affect; the validity of screening test results; others' reproductive experience; preparation for the birth of a child; the advice of other health professionals; additional sources of prenatal diagnosis information.

Table 5:1 number of women referring to one or more phrases within a theme

Theme titles	women (n=44)	%
1. No Test Option (non invasive/ scan)	**26**	**59 %**
1.1 information seeking or planning about non-invasive option	17	39 %
1.2 reasons for not having a diagnostic test	7	16 %
1.3 reasons against not having a diagnostic test	18	41 %
2. Test Option	**44**	**100 %**
2.1 information seeking or planning about tests in general	39	89 %
2.2 reasons for having a test	38	86 %
2.3 reasons against having a test	12	27 %
3. Choice between tests - neutral comment	**13**	**30 %**
4. Chorionic villus sampling choice	**31**	**71 %**
4.1 information seeking about chorionic villus sampling	22	50 %
4.2 reasons for chorionic villus sampling	13	30 %
4.3 reasons against chorionic villus sampling	27	61 %
5. Amniocentesis choice	**40**	**91 %**
5.1 information seeking about amniocentesis	35	80 %
5.2 reasons for having amniocentesis	17	39 %
5.3 reasons against amniocentesis	21	48 %
6. Down's syndrome consequence	**23**	**52 %**
6.1 information seeking/ planning	12	27 %
6.2 neutral-positive reference	6	14 %
6.3 negative reference	13	30 %

Table 5:1 continued . . .

Theme titles	women (n=44)	%
7. Miscarriage consequence	**25**	**57 %**
7.1 information seeking/ planning	22	50 %
7.2 neutral - positive reference	4	9 %
7.3 negative reference	2	4 %
8. Termination consequence	**31**	**71 %**
8.1 information seeking/ planning	20	45 %
8.2 neutral - positive comment	17	39 %
8.3 negative reference	13	30 %
9. Risk figure reference	**31**	**71 %**
9.1 single risk Down's syndrome	19	43 %
9.2 single risk miscarriage	16	36 %
9.3 comparison risks	17	39%
10. Perception of risk reference	**33**	**75 %**
10.1 minimised Down's syndrome risk	12	27 %
10.2 augmented Down's syndrome risk	19	43 %
10.3 minimised miscarriage risk	11	25 %
10.4 augmented miscarriage risk	4	9 %
11. Decision Making	**31**	**71 %**
11.1 triple test	12	27 %
11.2 pre-consultation discussion of decision outcome	17	39 %
11.3 delay in making decision	14	32 %
12. Expressed Affect	**25**	**57 %**
12.1 triple test (shock)	19	43 %
12.2 diagnostic / termination (hard decision / stage at a time)	12	27 %
12.3 reassurance testing	1	2 %
13. Triple test - further explanation	**32**	**73 %**
14. Confidence with screening results	**17**	**39 %**
15. Comparison norms (others / you do)	**27**	**61 %**
16. Personal experiences	**29**	**66 %**
16.1 physical aspects of pregnancy (e.g. scan, baby moving)	8	18 %
16.2 complications with pregnancy (e.g. miscarriage, infertility)	15	34 %
16.3 prior pregnancies and testing	11	25 %
16.4 familiarity abnormalities or genetics	3	7 %
17. Others' experience	**17**	**39 %**
17.1 prenatal testing and pregnancies	15	34 %
17.2 familiarity abnormalities or genetics	4	9 %
18. Preparation for pregnancy (birth, telling others)	**12**	**27 %**
19. Health professionals' advice	**18**	**41 %**
19.1 neutral or positive comments about advice given	14	32 %
19.2 dissatisfaction or directive advice given	4	9 %
19.3 inaccurate or conflicting information	5	11 %
20. Other sources of information (e.g. media, books)	**18**	**41 %**

5.2.2 Differences in information utilised by decision outcome: results.

The purpose of this analysis was to look for differences in the pattern of information referred to by those choosing to have or not have a diagnostic test. A Chi-square analysis is the most appropriate test of significance for differences in frequency data (Howell, 1985; Coolican, 1990). However, there are two limitations with applying Chi-square statistics to this data. First, only eight women chose not to have a test, meaning that any analysis will result in at least one cell having fewer than the recommended five cases. The Chi-square statistic may not be a reliable or meaningful statistic in such situations (Howell, 1985). Second, the content analysis was detailed and generated a large number of themes with sub-categorisations. Although these classifications were independent categories, occasionally items within phrases were related to each other. Carrying out multiple analyses on related data may lead to the reporting of statistical significance when no such association exists (Tabachnick and Fidell, 1989).

There are a number of methods that, when applied, may increase the reliability of statistics generated. The first method combines categories or levels of variables to reduce the number of cells within the model to increase the number of cases per cell (Coolican, 1990). As all the variables presented are dichotomous, categories cannot be further combined. The second method employs a multivariate statistic such as a logit analysis (Tabachnick and Fidell, 1989) to reduce the degree of error generated by performing multiple uni-variate analyses. A logit analysis is still sensitive to the minimum five cases per cell but, provided there are an equal number of cases within each category of the dependent variable, such an analysis may be acceptable. Unfortunately, the split of cases 'no test / test' was 18% to 82%, suggesting that such an analysis was not appropriate. Thirdly, if cell frequencies are less than five cases, then it is acceptable to employ a Fischer's exact test of significance (Coolican, 1990). Finally, to reduce the margin of error generated from performing multiple analyses, variables can be deleted or collapsed on theoretical grounds to reduce the number of analyses carried out (Tabachnick and Fidell, 1989). The data and analyses of this section of results incorporated these final two points.

It is unlikely that any analysis performed on the data from this study could be regarded as particularly robust. However, in an attempt to illustrate differences in the pattern of responses by those who chose to have or not to have a test, statistical analyses have been applied to the data. The data was modified to reduce the number of statistical tests. Themes were aggregated in accord with the demands of the research aims, to assess patterns of informed decision making (see Appendix VII). The simplification of

the coding form is described in brief. First, only sub-categories of themes associated with informed decision making were used in subsequent analyses: no test alternative; test alternative; Down's syndrome consequence; miscarriage consequence; termination consequence; risk reference; perception of risk; comments about the decision making process. In addition, the test alternative was aggregated to one theme and three sub-categories to include all references to the invasive testing alternatives. All other sub-categories of themes were aggregated so that if a woman mentioned any one of the items within the sub-classification, this was recorded as '1', whereas if the woman had not referred to any of these items this was recorded as '0'. As only one woman commented that testing may be reassuring, this item was removed from further analyses.

Table 5:2 differences in utilisation of information by test decision.

Theme Title	no test n = 8	%	test n=36	%	Chi^2 / Sig.
1. No Test Option	7	**88 %**	19	**53 %**	
1.1 information seeking non-invasive	7	**88 %**	10	**28 %**	9.8 <0.01
1.2 reasons for non-invasive	7	**88 %**	0	**0 %**	37.5 <0.01
1.3 reasons against non-invasive	5	**63 %**	13	**36 %**	NS
(2.) Test Option (combined 2+3+4+5)	8	**100 %**	36	**100 %**	
information seeking about test	8	**100 %**	35	**97 %**	NS
reasons for test	6	**75 %**	35	**97 %**	NS
reasons against test	7	**88 %**	30	**83 %**	NS
6. Down's syndrome consequence	6	**75 %**	17	**47 %**	
6.1 information seeking about Down's	5	**63 %**	7	**19 %**	6.12 0.02
6.2 neutral-positive reference Down's	3	**38 %**	3	**8 %**	NS
6.3 negative reference Down's	1	**13 %**	12	**33 %**	NS
7. Miscarriage consequence	4	**50 %**	21	**58 %**	
7.1 information seeking miscarriage	2	**25 %**	20	**56 %**	NS
7.2 neutral/ positive reference miscarriage	2	**25 %**	2	**6 %**	NS
7.3 negative reference miscarriage	0	**0 %**	2	**6 %**	NS
8. Termination consequence	6	**75 %**	25	**69 %**	
8.1 information seeking termination	4	**50 %**	16	**44 %**	NS
8.2 neutral/ positive comment termination	3	**38 %**	14	**39 %**	NS
8.3 negative reference termination	3	**38 %**	10	**28 %**	NS
9. Risk figure reference	5	**63 %**	26	**72 %**	
9.1 single risk Down's syndrome	3	**38 %**	16	**44 %**	NS
9.2 single risk miscarriage	0	**0 %**	16	**44 %**	5.59 0.02
9.3 comparison risks	5	**63 %**	12	**33 %**	NS
10. Perception of risk reference	8	**100 %**	25	**69%**	
10.1 minimised Down's syndrome risk	7	**88 %**	5	**14 %**	17.8 <0.01
10.2 augmented Down's syndrome risk	1	**13 %**	18	**39 %**	NS
10.3 minimised miscarriage risk	1	**13 %**	10	**28 %**	NS
10.4 augmented miscarriage risk	3	**38 %**	1	**3 %**	4.0 0.05

Table 5:2 continued . . .

Theme Title	no test n = 8	%	test n=36	%	Chi² / Sig.
(11.2) pre-consultation discussion decision	3	38 %	14	39 %	NS
(11.3) delay in making decision	5	63 %	9	25 %	4.24 0.05
(12.) Negative affect	6	75 %	19	52 %	NS
13. Triple test - further explanation	6	75 %	26	72 %	NS
14. Confidence with screening results	6	75 %	11	31 %	5.45 0.03
15. Comparison norms	4	50 %	23	64 %	NS
16. Personal experiences	6	75 %	23	64 %	NS
17. Others experience	4	50 %	13	36 %	NS
18. Preparation pregnancy	3	38 %	9	25 %	NS
19. Health professionals' advice	4	50 %	14	39 %	NS
20. Other sources information	4	50 %	14	39 %	NS

Chi-square statistics were applied to all the informed decision making and aggregated themes (table 5:2). Fischer's exact significance tests were reported for differences between themes if the probability was ≤ 0.05. Any significance value should be interpreted with caution as the sample is small and multiple analyses have been carried out. Despite these limitations, there do appear to be differences in patterns of information utilised by those choosing to have or not have a diagnostic test. Those women who chose not to have an invasive test were more likely to enquire about the no test alternatives ($p < 0.01$) and the consequences of having a baby with Down's syndrome ($p = 0.02$) than those who chose to have a diagnostic test. However, both sets of women were as likely to enquire about the test alternatives, miscarriage and termination consequences. Women choosing the no test alternative were less likely to refer to the risk of miscarriage figure alone ($p = 0.02$), more likely to interpret this risk as large ($p = 0.05$) and more likely to minimise the risk of Down's syndrome ($p < 0.001$). Additionally, women choosing not to have a further invasive test were more likely to question the validity of the triple test screening result and method ($p = 0.03$). Finally, women choosing not to have an invasive test were more likely to comment on needing more time to make the decision ($p = 0.05$). However, there was no significant difference in consultation length between the no test and the test decision (respectively: mean = 25.6 minutes (s.d. = 8.1), mean = 21.3 minutes (s.d. = 6.6); $t = 1.6$; d.f. = 42; $p = 0.12$).

5.2.3 Describing informed decision making: results.

The purpose of this analysis was to describe whether or not women were utilising sufficient and appropriate information during the consultation to ensure informed decision making. Four informed decision making variables were created from eight

themes included within the coding frame (see section 5.1.5): total informed decision making (IDM-T) (Table 5:3); information seeking (IDM-I); generation of reasons for alternatives (IDM-F); generation of reasons against selecting an alternative (IDM-A) (table 5:4).

Table 5:3 frequency of women referring to the informed decision making themes.

	1 theme		2 themes		3 themes		4 themes		5 themes	
	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)
Total	8	(18%)	12	(27%)	16	(36%)	6	(14%)	2	(5%)
No test	0	(0%)	2	(25%)	3	(38%)	2	(25%)	1	(12%)
Test	8	(22%)	10	(28%)	13	(36%)	4	(11%)	1	(3%)

Only five per cent of women (2/44) utilised information about both test alternatives and all three consequences of testing when making their decision to have or not have prenatal diagnosis (table 5:3). Most women referred to information classified by two or three of the informed decision making themes (table 5:3). Women tended to seek for more information than they used when reasoning about decision consequences and alternatives (table 5:4).

Table 5:4 description of the informed decision making scores.

Informed decision making (IDM) measures	range	min-max	x (s.d.)
IDM-I: seek information about alternatives (from 1.1, 2.1, 3.0, 4.1, 5.1, 6.1, 7.1 & 8.1)	0 - 8	1 - 5	2.6 (1.1)
IDM-F: reasons for selecting alternatives (from 1.2, 2.2, 4.2, 5.2, 6.2, 7.2 & 8.2)	0 - 7	1 - 4	1.7 (0.8)
IDM-A: reasons against selecting alternatives (from 1.3, 2.3, 4.3, 5.3, 6.3, 7.3 & 8.3)	0 - 7	0 - 4	1.9 (1.1)

A multiple analysis of variance (MANOVA) was carried out to assess differences in informed decision making by women choosing to have or not have prenatal diagnostic testing. The three dependent variables included in the analysis were the 'process measures' of informed decision making, IDM-I, IDM-F and IDM-A. The global measure of informed decision making (IDM-T) was not included in this analysis as it summarised the same information referred to by women as that classified separately in the three 'process' measures. As the sample size of the test and no test group was unevenly split,

a Bartlett-box M test of significance was included within the analyses to assess the homogeneity of variance of the group distributions.

Table 5:5 differences in informed decision making by test decision.

	No test (n=8)		Test (n=36)		MANOVA (1,42)	
	mean	(s.d)	mean	(s.d)	F	Sig.
IDM-I: information seeking	3.3	(1.0)	2.4	(1.1)	3.9	0.06
IDM-F: reasons for	2.6	(0.7)	1.9	(1.2)	20.6	<0.001
IDM-A: reasons against	2.0	(0.8)	1.5	(0.6)	0.10	NS

The tests for homogeneity of variance within the MANOVA were not significantly different suggesting that subsequent findings were robust. No differences by test decision were observed for the reasons generated against the decision alternatives (table 5:5). However, women choosing not to have any further testing were likely to generate reasons for more of the alternatives and consequences than those deciding to have a diagnostic test ($p<0.001$, table 5:5). In addition, there was a trend towards significance suggesting that women who decided to have no further testing were likely to ask for information about more of the alternatives and consequences than women who chose to have a diagnostic test ($p=0.06$, table 5:5).

5.3 Discussion

The purpose of this study was to describe the process of women making the decision to have or not have prenatal diagnosis upon receipt of a screen positive triple test result. Most, if not all, of the previously published empirical research in this health context had used questionnaires or semi-structured interviews to assess factors associated with women's decision making. This study employed a non-experimental, cross-sectional observational design to describe women's decision making processes when deciding to have or not have prenatal diagnosis. The use of an information search strategy should enable the following: an evaluation of the factors involved in women's decision making concurrently with the making of the decision; an opportunity to operationalise the criteria of informed decision making. In consequence, the results should provide a more accurate representation of the decision making processes employed by women than previously described and an evaluation of whether or not women's decision making was informed. The following discussion focuses on the extent to which this study design has adequately addressed these research aims. This section is divided into four parts: a discussion of the reliability and validity of the findings; a description of the processes

employed by women to reach the prenatal diagnosis decision; an assessment of women's informed decision making; implications of these findings for changes in the provision of prenatal diagnosis information giving.

5.3.1 Information search strategy: reliability and validity.

The advantage of an observational study design is that it enables the direct observation of an individual's behaviour to be made without relying on the interpretation of that behaviour from the individual (Robson, 1993). However, this type of design has an associated number of problems. First, it is suggested that an individual's behaviour may subtly alter or change as a consequence of being observed (Robson, 1993; Bowling, 1997). Second, as the researcher is required to interpret the individual's behaviour, it is possible that the subsequent interpretation will reflect the researcher's perception of the behaviour alone and the findings are not replicable or reproducible by a third party. Third, cognitions can only be inferred from the verbal behaviour of an individual (see introduction). Any difficulties in interpreting another's actions are likely to be augmented in a study aiming to assess the psychological process involved in making decisions. In consequence, issues pertaining to the reliability and validity of measures assessing observational data are paramount. In other words, are researchers able to replicate the coding frame developed for this study and does the coding frame assess factors associated with the process of women's decision making?

In practice, it is not possible to assess the effects, if any, of observation on individuals' behaviour (Robson, 1993). However, an effort was made to reduce the profile of the study and minimise the impact of an observational technique on women's consultation behaviour. First, the author (HB) was introduced as a member of the ante-natal team with an interest in the prenatal diagnosis consultation rather than a 'psychologist' or 'researcher' observing the woman's behaviour. Second, the information giving consultation was an established part of the routine care provided by the LGI for women receiving a screen positive triple test result and not an interview conducted specifically for the study. Finally, the consultation was audio tape-recorded and transcribed at a later date. The effects of this observational technique on participants' behaviour are believed to be minimal (Bowling, 1997). Although women were aware that the consultation was being audio tape-recorded, few comments were made about its use before, during or after the consultation. In consequence, it is fair to assume that the observational design of this study had a minimal impact on women's behaviour and decision making about prenatal diagnosis.

The method carried out to develop the coding frame was rigorous. The coding frame was developed in stages, requiring continual reference to the consultation transcripts, the literature on decision making and prior empirical research of women's prenatal decision making. During its development, the themes and rationale used to classify women's utterances were discussed with two colleagues (JH and AM) familiar with the issues of prenatal diagnosis. The coding frame was piloted twice and, where appropriate, revised. In addition, a Cohen's Kappa coefficient was carried out to assess the inter-rater reliability of the final coding frame. The resulting coefficient (0.83) indicated an excellent rate of agreement between the two raters (HB and AM). In consequence, there is evidence to suggest that the coding frame developed to classify women's utterances during the information giving consultation was a highly reliable instrument.

As discussed, the process tracing technique employed in this study described the information utilised by women during the information giving consultation. It is argued that process tracing techniques do provide a more comprehensive understanding of individuals' decision making than questionnaires informed by EUT (Carroll and Johnson, 1990). However, there are at least two concerns with using an information search technique to assess psychological processes. First, it has been suggested that individuals may be unable to verbalise the cognitions that underpin a final decision (Harte, Westenberg and van Someren, 1994). Second, although individuals acquire information when making decisions, this information is not processed or utilised to inform the decision (Payne, 1980). Although it is difficult to address these concerns, the context of the real-world decision described in this chapter may minimise their impact on subsequent findings. It is worth noting that these concerns have arisen from research investigating individuals' acquisition of information (Payne, 1980; Carroll and Johnson, 1990; Baron, 1994; Harte, Westenberg and van Someren, 1994; Harte and Koele, 1997). In this chapter's decision context, the health professional provided women with most of the information considered necessary to make the prenatal diagnosis decision (see chapter four), i.e. woman did not have to actively search for information. In addition, the audio tape-recording of the consultation recorded all women's utterances, not just those pertaining to information seeking. In other words, questions asked to obtain information, statements referring to peripheral decision information and the verbalisation of reasons were all recorded. It is feasible to suggest that within this context, women verbalised information they were assimilating when making a decision. It is possible that women may have utilised information when making the decision without verbally referring to it. In consequence, the data summarised by the coding

frame may under-represent the cognitions employed by women when making their prenatal diagnosis decisions. Despite this under-representation, a number of findings from this study replicate results from prior empirical research that have used alternative methodologies to assess factors associated with women's prenatal diagnosis decision making. The following discussion briefly summarises the similarities between the findings from this study and prior empirical evidence.

Reasons: The questionnaire and interview based studies summarised in section 1.3.2.1 identified a number of reasons provided by women supporting their prenatal diagnosis decision, such as: needing to know for certain whether or not the baby has Down's syndrome; not believing in termination of pregnancy; the timing of the diagnostic test results (Nielsen 1981; Volodkevich and Huether, 1981; Verjaal, Leschot and Treffers, 1982; Farrant, 1985; Lippman, Perry, Mandel and Cartier, 1985; McGovern, Goldberg and Desnick, 1986; French, Kurczynski, Weaver and Pituch, 1992; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994). The information utilised by women during this study included these reasons for selecting a decision alternative (themes 1-8; table 5:1). In addition, women generated a number of other reasons for rejecting or selecting an alternative, such as: not being able to cope with a Down's syndrome baby; waiting for the results of the fetal anomaly scan; a child with Down's syndrome being 'fair enough'.

Perception risk: Prior research also identified women's perception of risk as important in women's prenatal diagnosis decision making, both the risk of miscarriage and the likelihood of having a baby with Down's syndrome (see section 1.3.2.1). The information search technique coded 71% of women referring to an actual risk figure and 75% evaluating the risks figures (themes 9 and 10; table 5:1).

Timing of decision: Raised in a few of the interview based studies were issues reflecting women's perception of the decision making process: making a decision about testing prior to the offer of prenatal diagnosis (Sjogren and Uddenberg, 1988); feeling obliged to have the test (Farrant, 1985; Sjogren and Uddenberg, 1988). Although these statements were raised by women during the information giving consultation of this study, fewer women referred to them (theme 11; table 5:1). This difference may support the assertion that retrospective study designs are subject to biases in the recall of information. That is to say, women interviewed after having a test perceived themselves to be more certain of their choice in retrospect than woman observed concurrently with their decision making.

Comparison norms: The observational study design illustrated that 61% of women asked for information about the normative behaviour, i.e. what other people tend to choose in this situation (theme 15; table 5:1). This search for normative behaviour may reflect findings from choice of treatment studies within cancer suggesting that patients want to be informed about the medical options but find the responsibility for making the final treatment choice difficult (Fallowfield, 1997). Ascertaining what others do in the same situation may serve to diffuse the responsibility for any negative consequences of their final choice.

Affect: As discussed in section 1.1.4, women are anxious upon receipt of a screen positive triple test result and concerned about the prenatal diagnosis decision (Beeson and Golbus, 1979; Nielsen, 1982; Verjaal, Leschot and Treffers, 1982; Berne-Fromell, Uddenberg and Kjessler, 1983; Fava, Trombini, Michelacci, Linder, Pathak and Bovicelli, 1983; Burton, Dillard and Clark, 1985; Farrant, 1985; Pauker and Pauker, 1987; Sjogren and Uddenberg, 1988; Marteau, Kidd, Cook, Michie, Johnston, Slack and Shaw, 1992; Evans, Pryde, Evans and Johnson, 1993; Burn, Fairgrieve, Franks, White and Magnay, 1996). Indeed, Burn et al (1996) reported that, in retrospect, 78% of women receiving a screen positive triple test result were worried or devastated by their test result. Forty-three per cent of women in this study expressed worry at their triple test result (theme 12; table 5:1). In addition, 27% made a comment about the difficulty of making the prenatal diagnosis decision.

Triple test: Although all women chose to have the triple test and were provided with information about screening, 73% requested further information about the triple test and calculation of screening results (theme 13; table 5:1). This finding may support evidence from empirical work on serum screening suggesting that women do not have sufficient information to enable them to choose whether or not to have a screening test (Marteau, Johnston, Plenicar, Shaw, Slack, 1988; Smith, Shaw and Marteau, 1994; Michie, Smith, McClennan and Marteau, 1997).

5.3.2 Describing women's decision making.

If, as inferred in section 5.3.1, the coding frame is a reliable and valid instrument with which to classify women's utterances, the following suggestions can be made about the processes employed by women when making the decision to have or not have prenatal diagnosis.

First, although the majority of women in this study referred to most of the information provided by the health professional when making their decision (themes 1-10; table 5:1), few utilised information pertaining to all the consequences of both alternatives. That is to say, most women mentioned the testing option, details about amniocentesis and chorionic villus sampling, termination and risk figures for either Down's syndrome or miscarriage. This finding suggests that most women employed a systematic strategy to process the decision information. As the prenatal diagnosis decision is a risky decision, has two well-defined alternatives and has serious consequence for the individual, it is likely that an individual would employ such a cognitively demanding strategy to process the information (Chaiken, 1980; Eagly and Chaiken, 1993; Ubel and Loewnstein, 1997).

Second, most women also referred to information outside that provided by the health professional when making the decision. In particular, most women referred to information about the triple test and their own experiences of testing, pregnancy and abnormality (themes 13 and 16; table 5:1). From the perspective of a classical decision approach to decision making, the consequences of a previous decision ought not to be associated with a current decision. This finding suggests that women also employ some heuristic strategies to process the decision information. For example, they may refer to a previous experience or scenario when assimilating information. Such a scenario may act as a 'cognitive short-cut' for reaching a decision. It is likely that both processes need to be employed to enable an individual to reach a decision (Vlek, 1987; Eagly and Chaiken, 1993).

Third, most women referred to both a risk figure and an evaluation of the risk when making the prenatal diagnosis decision (themes 9 and 10; table 5:1). The empirical evidence from the majority of interview or questionnaire based studies finds that the perception of risk and not the actual risk predict behaviour (section 1.3.2.1). One explanation for the discrepancies in findings is that women utilise the risk information during the information giving consultation, evaluate the risk when assimilating it with existing beliefs and recall only their perception of risk when asked in retrospect.

Fourth, although most women are likely to have employed a systematic strategy to process the information attended to, the pattern of results suggests that most women do not apply an optimising strategy when appraising the decision information (themes 1-10; table 5:1). To explain further, the pattern of information utilised suggests that half of the women *screened out* the no test alternative, focusing mainly on the test alternatives. Further, the sub-categories of these themes suggest that trade-offs between the

advantages and disadvantages of consequences were minimal. As suggested by prior research, describing individuals' decision making processes, it is likely that women used a satisficing strategy to limit the amount of information required for assimilation (see section 1.2)

Fifth, although the sample size was small and the use of multiple analyses limited the interpretation of findings, some differences were observed in the decision making processes by test decision (table 5:2). Eight significant differences were reported by decision outcome, suggesting a coherent difference in the pattern of information utilised by women choosing to have or not have a diagnostic test. Women choosing not to have a diagnostic test were: more likely to search for information about the non-invasive test option and Down's syndrome; less likely to refer to the actual risks of miscarriage but more likely to describe this particular risk as high; more likely to refer to the risk of Down's syndrome as low and further reduce the threat of Down's syndrome by expressing dissatisfaction with screening tests and results; more likely to mention the need for more time to think about the information before making a decision. These findings suggest that most women choosing to have no further testing were more likely to survey the consequences of both prenatal testing alternatives. One explanation for these differences in information utilisation by prenatal testing decision is that women choosing to have no further testing have to actively 'opt out' of the testing process whereas women choosing to have a test merely agree with the offer of a test (Marteau, Johnston, Kidd, Michie and Cook, 1992).

This quantitative content analysis described the information utilised by women during the information giving consultation. It was beyond the remit of this chapter's aims to explore the interaction between the health professional's provision of information and its subsequent utilisation by women. As mentioned in chapter four, a more in-depth analysis of the transcripts would be required in order to assess whether it was the health professional's information that encouraged women to utilise that information in their decision making or whether the women's questions prompted the health professional to impart an appropriate response (table 4:1 and table 5:1). In addition, the data extracted from the transcripts might be used to explore more detailed relationships between the decision making themes, for example: relationship between affect and type of information processing strategy employed (themes 12.1 and themes 1-10; table 5:1); association between information utilised and whether or not the decision was made before the consultation (theme 11.2 and themes 1-10, table 5:1); the association between Down's syndrome and reduced confidence in the triple test screening test

result (Croyle, Sun and Hart, 1997) (themes 14 and 10.1; table 5:1). However, as stated, the main purpose of this chapter was to describe the processes employed by women when making the prenatal diagnosis decision upon receipt of a screen positive triple test result.

5.3.3 Assessing informed decision making.

The final aim of the study was to ascertain whether or not women were making informed decisions to have or not have a diagnostic test. This study is one of the first within the prenatal diagnosis context to operationalise criteria associated with the making of an informed decision (Frisch and Clemen, 1994; section 1.2); a good decision ought to be based on an evaluation of the consequences of the different options, an accurate assessment of the consequences and a trade-off between the consequences of the decision alternative. The final coding frame included themes to classify information pertaining to the two decision alternatives and three consequences of the prenatal diagnosis decision. In addition, the themes' sub-categories were informed by the *reason-based choice* (Shafir, Simonson and Tversky, 1994) explanation for how individuals 'trade-off' information when making a decision. Four informed decision making scores were created from the data contained within these themes: a global measure (IDM-T); one assessing the information searched for an alternative or consequence (IDM-I); one assessing the reasons provided to support an alternative or consequence (IDM-F); one assessing the reasons provided to reject an alternative or consequence (IDM-A).

The findings reported that only five per cent of women (2/44) referred to information from all five themes included in the IDM-T measure (table 5:3). Most women utilised information pertaining to two or three of the themes when reaching their prenatal diagnosis decision (table 5:3). In addition, women were more likely to search for information than provide reasons for or against the alternatives or consequences (table 5:4). One explanation for these findings may be that women do not verbalise the process of information assimilation when making the decision. However, the pattern of information referred to by women as described in table 5:1 and the findings from previous empirical research in this health context suggest an alternative explanation, that most women were not making fully informed decisions about the decision to have or not have prenatal diagnosis. This explanation is further supported by this chapter's findings evaluating the difference in informed decision making by whether or not women had had diagnostic tests. Women choosing to have no further testing generated more reasons and tended to search for more information about test alternatives and

consequences than women having diagnostic tests (table 5:5). In other words, 'no test' women were utilising and assimilating more information to reach a decision than 'test' women.

5.3.4 Implications for the provision of prenatal diagnosis information.

These findings describing the information utilised by women when making their prenatal diagnosis decisions have specific implications for the provision of information about prenatal diagnosis and screening tests within this health context. Few women were utilising information during the consultations to indicate that they were making fully informed decisions. In particular, most women were not referring to information about the no test alternative, the consequence of miscarriage and the condition of Down's syndrome. It was clear that women arrived at this consultation with different experiences of, information about and opinions towards prenatal testing: some had discussed the diagnostic test decision although most had not; others had 'stories' about their own and acquaintance's prenatal testing experiences; some referred to advice proffered by other health professionals; some women had searched for additional information via the media and information leaflets. However, it is the responsibility of the health professional to provide information about all the consequences of both alternatives and to address women's *a priori* beliefs, thus enabling women to make an informed decision to have or not have a diagnostic test (Royal College of Physicians, 1989; American Society of Human Genetics, 1996). As this prenatal diagnosis information giving consultation is the last opportunity for the health professional to establish whether women have received and evaluated all the appropriate information, these consultations ought to provide a more detailed discussion of the 'no test' alternative, the consequence of miscarriage and the consequence of having a Down's syndrome child.

In addition, the findings from this study and previous empirical research evaluating knowledge of women undergoing serum screening tests, suggest that a significant proportion of women did not have sufficient information to be making informed choices to have or not have screening tests (Marteau, Johnston, Plenicar, Shaw and Slack, 1988; Smith, Shaw, Marteau, 1994; Michie, Smith, McClennan and Marteau, 1997). All of the women in this study knew they had undertaken the triple test and that the test was 'looking for' Down's syndrome. However, over seventy per cent of these women asked for further information about screening and the triple test result. The types of questions raised involved information about: clarification on the meaning of a screening test result; how the triple test risk figure was derived; the association between fertility

treatment, familial and personal pre-dispositions to hormone generation on receipt of a screen positive result; the likelihood of recurrence of a screen positive result in subsequent pregnancies; the justification of the screen positive cut-off point; the validity of the triple test result compared with age and nuchal-pad screening figures. The nature of these questions and the development of more screening technologies suggests that the content of prenatal testing information needs to be modified. Those receiving screening test results appear to require a more sophisticated explanation of the risk figure generation than is currently provided. Additionally, a more comprehensive explanation of the screening tests available during pregnancy, with the advantages and disadvantages of each test, may help women to make more informed choices about which screening test is best for them. This information may reduce the confusion of comprehending which risk figure for Down's syndrome should be used during the decision to have a diagnostic test or not.

As yet, there is little evidence to suggest that altering the information giving consultation would be associated with changes in women's informed decision making. For example, Pauker and Pauker (1977; 1978; 1987) have used decision analysis as a technique to facilitate women's prenatal diagnosis decision making (see chapter one). During a consultation, women are encouraged to systematically utilise and evaluate information pertaining to both the test and no test alternatives and assimilate this information with their *a priori* attitudes. To date, there is no published evidence suggesting that this technique is associated with the making of a more informed or better decision (Bekker et al, 1999). It is possible that if health professionals provide women with a more structured discussion of both decision alternatives, women may utilise information pertaining to both these alternatives when making their prenatal diagnosis decision. However, it is as likely that changing the structure of an information giving consultation will not lead to a more balanced appraisal of the decision information. Further, there is some evidence to suggest that poorer decision outcomes, such as post-decision satisfaction, are associated with a more systematic evaluation of the decision information (Wilson, Lisle, Schooler, Hodges, Klaaren and Lafleur, 1993). Empirical research is required to evaluate the consequences of altering the prenatal diagnosis information on measures of women's decision making processes and outcomes.

5.3.5 Chapter summary.

In brief, few, if any, published empirical studies have assessed the processes employed by women concurrently with their decision to have or not have prenatal diagnosis. In addition, little empirical research has operationalised informed decision making in the

context of prenatal diagnosis. This study employed an observational design to classify the information utilised by women when making the prenatal diagnosis decision. The coding frame developed to classify women's utterances satisfactorily summarised the information referred to by women when making the decision to have or not have a diagnostic test. Most women did not utilise all the necessary information within a consultation to ensure fully informed decisions. Although 70% of women referred to the test alternative, termination, actual risk figures and evaluations of risk, over 40% of women did not refer to the no test alternatives, miscarriage or condition of Down's syndrome. Women used both systematic and heuristic information processing strategies to assimilate information about testing. Consistent differences in the pattern of information referred to were observed by decision outcome, women choosing to have a test appeared to employ a satisficing strategy to screen out the 'no test' information. The findings suggest that those choosing not to have a test were making a more informed decision than those deciding to have prenatal diagnosis. It was suggested that information about all the consequences of both diagnostic test alternatives should be presented equally during the consultation and that the *a priori* beliefs of women discussed to ensure that women make an informed choice whether or not to have a test.

6. Facilitating informed decision making in the prenatal diagnosis context: a randomised control trial.

This chapter describes the findings from a randomised control trial carried out to evaluate the effectiveness of decision analysis in the facilitation of women's informed decision making within the prenatal diagnosis context. The theoretical review, described in chapter one, found there to be little empirical research that had operationalised informed decision making in the context of prenatal diagnosis. In addition, few empirical studies have developed interventions designed to facilitate patient decision making in the health care domain (see chapter two). The findings from over 500 intervention studies were summarised in an integrative review described in chapter two (Bekker et al, 1999). In total, just 18 studies evaluated interventions aimed at encouraging patient participation in the consultation. None of these studies had assessed whether or not their interventions were associated with changes in patient's informed decision making. In other words, there is a paucity of research assessing the facilitation of individuals' informed decision making in the health care domain.

There is empirical evidence from studies describing women's experiences of and reasons for undergoing prenatal diagnosis which suggests that women are not making informed decisions about testing (see chapter one). Within the literature, at least two explanations have been proposed as to why women are not making informed decisions about prenatal diagnosis (Marteau, 1995). First, health professionals are not providing women with sufficient information to enable informed decision making. Second, the health professionals have provided women with sufficient information but women have not utilised all the necessary information to ensure that the decision was informed. The empirical findings described in chapter four provide some support for the assertion that health professionals are not providing sufficient conditions to enable women to make an informed decision about prenatal diagnosis. However, there is also evidence to suggest that individuals employ cognitive strategies to reduce the information to be assimilated when making risky or difficult decisions (see chapter one). The findings discussed in chapter five provide support for this explanation of decision making, i.e. that women do not utilise all the available information when making the prenatal diagnosis decision. In essence, facilitating informed decision making requires a technique that both improves the quality of the information provided by the health professional and encourages a more systematic evaluation of this information by women.

A technique that enhances the decision making process is known as a decision aid (Pitz, 1987). Most decision aids have developed from decision analysis (Holmes-Rovner, 1995). As mentioned in chapter one, decision analysis is a formal technique based on expected-utility theory (EUT) that both restructures the decision information and encourages women to explicitly explore the risks and consequences of each alternative. In addition, the decision analytic technique enables individuals to assimilate their utility values of the decision consequences with their perceptions of the likelihood of these consequences occurring. This process culminates in the identification of the optimum, accurate or correct choice for the individual (Thornton and Lilford, 1990; Lilford and Thornton, 1994; Pauker and Pauker, 1987; Pitz, 1987; O'Connor, 1995; Holmes-Rovner, 1995; Nease, 1996; Ubel and Loewenstein, 1997; see chapter one).

In brief, the purpose of applying decision analysis to real-world decision making is to encourage individuals to make 'better' decisions. The limitations of human information processing abilities suggest that intuitive decision making leads to poorer decisions (Politser, 1981; Ubel and Loewenstein, 1997). Decision analysis is supposed to lead to better decision making for a number of reasons: it provides the structure with which to identify the decision-specific information (Dowie, 1995); the provision of a decision tree reduces the cognitive load for the decision maker as the information is constantly visually available in the form of a visual prompt (Arkes, 1981; Pitz, 1987); the technique allows the elicitation of utilities and the opportunity to integrate these values during decision making, a process not incorporated by more intuitive approaches to decision making (Politser, 1981; Baron, 1994; Ubel and Loewenstein, 1997); it is argued that these conditions encourage more informed, systematic or rational decisions as all the information can be attended to without bias and the technique encourages trade-offs between attributes (Arkes, 1981; Travis, Phillipi, Tonn, 1989; Ubel and Loewenstein, 1997); the technique is able to represent the conflicting information that makes many risky decisions hard or difficult (Pitz, 1987; Ubel and Loewenstein, 1997); finally, by explicitly discussing all the information associated with the decision, the individual is less likely to make a choice based upon an expert's opinion (Ubel and Loewenstein, 1997).

A number of concerns have been raised regarding the application of decision analysis to the clinical or 'real-world' setting. The first set of concerns to be discussed focus on the more 'theoretically-based' issues. First, the real-world setting is such that it is unusual for individuals to be faced with a well-defined, single decision. Most real-world decisions are dynamic and involve a number of related decisions over a period of time

(Politser, 1981). In consequence, it may be unrealistic to define one discrete decision event appropriate for decision analysis in the real-world setting. Second, many decisions are made with incomplete or inaccurate information. Applying a decision analytic technique to incomplete information may lead to an erroneous decision being made. Third, some attributes within the decision structure are difficult to quantify (Vlek, 1987; Ubel and Loewenstein, 1997) which may result in an over-representation of the attributes that are easier to quantify. Fourth, decision analysis is concerned with emphasising the systematic processing of information. As most decisions require the application of both systematic and intuitive processes (Vlek; 1987), it is unclear how effective decision analysis would be in facilitating the whole decision process. Equally the efficacy of the decision analytic technique relies on the notions that all individuals would prefer to make decisions systematically and that all decisions are best made systematically. It is likely that the preference for style of information processing differs between individuals, with some favouring a more predominantly heuristic approach to decision making while others a more systematic style (Cacioppo, Petty and Kao, 1984; Scheier and Carver, 1985; Burnett, 1991; Scheier, Carver and Bridges, 1994; Webster and Kruglanski, 1994; Sorrentino, Holmes, Hanna and Sharp, 1995). Finally, there is evidence that different decisions are associated with different types of processing. In other words, the uniform application of decision analysis and the encouragement of systematic processing may not always result in decision facilitation or better outcomes (Lipshitz, 1993; Ubel and Loewenstein, 1997).

The application of decision analysis to the health-care context raises a number of 'clinically-based' concerns in addition to the theoretical issues previously discussed. Some argue that, as all decisions are approached uniformly, the technique is insensitive to the clinical situation and that this consultation style may de-humanise the patient (Schwartz, 1979). Others suggest that the technique devalues the expert's clinical judgement by regulating all consultations as well as needlessly increasing consultation time (Schwartz, 1979). Decision analysis may also be perceived as directive since the expert calculates the optimum decision for the individual and the expression of this decision may be seen to compromise the individual's autonomy in decision making (Falek, 1984; Kessler, 1992). Equally, the application of decision analysis may be stressful for some individuals as they may not understand the justification for being asked such explicit and difficult questions during the consultation (Pauker and Pauker, 1987; Ubel and Loewenstein, 1997). In addition, such a consultation may raise issues that the expert is unable or unwilling to address (Schwartz, 1979). The decision analytic technique is not regarded as being able to fully incorporate the emotions encountered

within many clinical situations, such as anxiety, regret and uncertainty (Ubel and Loewenstein, 1997). Finally, there is some evidence to suggest that encouraging individuals to think systematically about their reasons for a decision may be associated with an increase in post-decisional regret or dissatisfaction (Wilson, Lisle and Schooler, 1993), i.e. decision analysis may be associated with worse rather than better decision outcomes.

Although decision analysis has been applied to patient decision making for a number of decades (Pauker and Pauker, 1979, 1987; Thornton, 1990; Morabia, Steinig-Stamm, Unger, Slosman, Schneider, Perrier and Junod, 1994), there is little published evidence to suggest whether or not decision analysis leads to better patient decision making than decisions made during routine clinical care (Pitz, 1987; O'Connor, Llewellyn-Thomas and Drake, 1995; Bekker et al, 1999). Few, if any studies, have employed a randomised control trial (RCT) design to evaluate the effectiveness of decision analysis to facilitate patient decision making. Most studies have described either the application of the decision analytic technique within the health care setting (Pauker and Pauker, 1977; 1979; 1987) or assessed the degree to which an individual's decision deviates from that predicted by EUT (Heckerling, Verp and Hadro, 1994; Verp and Heckerling, 1995). Few if any studies have addressed the issue of whether decisions based on decision analysis based decisions are 'better' than unaided decisions.

This dearth within the literature may in part be attributable to the difficulty of evaluating the quality of the decision and of defining what is meant by a good decision (Pitz 1987; Frisch and Jones, 1993). From a theoretical or decision analytic perspective, a 'good' decision is the choice with the maximum expected utility. Several studies have applied decision analysis within the prenatal diagnostic testing context to examine whether or not women were making optimal decisions (Pauker and Pauker, 1987; Thornton and Lilford, 1990; Heckerling, Verp and Hadro, 1994; Verp and Heckerling, 1995). The studies suggest that not all women make decisions consistent with the theoretically accurate choice. However, an accurate decision is not necessarily the best or good decision (Frisch and Jones, 1993). Other studies within the clinical setting assess the quality of the decision by the decision outcome, such as the prognosis of the disease (Whitbeck and Brooks, 1983; Morabia, Steinig-Stamm, Unger, Slosman, Schneider, Perrier and Junod, 1994). The main limitation of this evaluation for most individual-based or value-laden decisions is that there is no objective assessment of an individual's judgement, i.e. no external reference with which to ascribe a 'correct' or 'incorrect' choice (Frisch and Jones, 1993; O'Connor, 1995). As there is no objective

standard with which to assess the quality of a decision, the choice of measure to evaluate good decision making is usually determined by the researchers' aims and theoretical perspective (Llewellyn-Thomas, 1995). In the context of prenatal diagnosis testing, one of the main objectives of prenatal diagnosis is to encourage women to make *informed decisions*.

The numerous measures employed by researchers to assess the quality of decisions fall into three broad categories: behavioural, cognitive and affective (Llewellyn-Thomas, 1995). Behavioural assessments include, recording the number of people choosing each of the alternatives and recommending the decision to others (Pitz, 1987). Cognitive measures may include evaluations of both the process and outcome, such as: utilisation of information during decision making (see chapter five); reasons for and against alternatives (Whelan, Levine, Gafni, Lukka, Mohide, Patel and Streiner, 1995); recording of perceptions of risk (Pitz, 1987) and utilities or attitudes (Pauker and Pauker, 1987; d'Ydewalle and Evers-Kiebooms, 1987; Heckerling, Verp and Hadro, 1994: Heckerling and Verp, 1995); ranking of the order of utilities (McNutt, 1989; Thornton and Lilford, 1990; Nease, 1996); knowledge or understanding of the information (Pitz, 1987; Holmes-Rovner, 1995; O'Connor, Llewellyn-Thomas and Drake, 1995). Affective measures may include: decisional satisfaction (Holmes-Rovner, Kroll, Schmitt, Rovner, Breer, Rothert, Padonu and Talarczyk, 1996); confusion or conflict in making the decision (O'Connor, 1995); confidence with the final decision (Pitz, 1987); anxiety (O'Connor, 1995); worry; regret with the decision. Arguably, a fourth area of evaluation is that of the experience of the clinical situation such as satisfaction with the care provided. As discussed in chapters one and five, few studies have assessed the process of informed decision making.

The purpose of the current study is to evaluate the effectiveness of the decision analytic technique to facilitate women's decision making in the context of prenatal diagnosis. The aim is to compare the quality of decisions made during the routine information giving consultations with those made during a consultation structured by the decision analytic technique. Behavioural, affective, cognitive and clinical measures were assessed to evaluate any differences in the quality of the decision process and outcome.

6.1 Methodology.

Over a six month period, the study was integrated into the routine care provided by the antenatal department at Leeds General Infirmary (LGI) following: the application for

ethical approval from the Leeds and St James' ethical and scientific committee; training and advice from those obstetricians and midwives actively involved in the provision of information about prenatal diagnosis (see chapters four and five); piloting of the information giving techniques (see chapters four and five); piloting of study materials (see section 5.1). During the data collection period of this thesis, the author (HB) was responsible for the follow-up care of all women receiving a screen positive triple test result. These duties included contacting women about their triple test result (chapter four) and providing women with the prenatal diagnosis information during the consultation described in chapter four. For consistency, the author (HB) is subsequently referred to as the study health professional.

6.1.1 Design.

The most appropriate method for evaluating the effectiveness of any intervention is to employ a true experimental design or randomised control trial (Bowling, 1997). Ideally, both the health professional and the woman should be 'blind' to the allocated experimental condition in order to reduce experimenter and sampling biases. However, as this intervention requires the health professional to deliver information in one of two styles, it was necessary for the health professional to be aware of the experimental condition. Equally, the nature of human experimental research requires participants to be informed about the study. The women were aware that the information they received was delivered in one of two ways but they were not informed of the experimental group they had been randomised to. As this particular decision was a 'one-off' for most of the women, it was unlikely that they were aware of the differences between a routine and an experimental consultation.

This study was conducted within a real-world situation and concerned a particularly distressing test result and decision. It was not appropriate, therefore, to include a 'no treatment' group within the design. In consequence, a two-condition randomised control trial was employed. As mentioned, over a three-month piloting period, the study health professional (HB) was integrated into the LGI antenatal team. Further, the study health professional had been trained to provide women with information that was considered routine practice by the LGI antenatal team. In addition, the study health professional was also trained to elicit women's utilities firstly, by observing an obstetrician proficient in the application of the decision analytic technique apply the *lottery question* to three women making the decision to have or not have prenatal diagnosis (Thornton, 1990a; 1990b; Thornton, Lilford and Johnson, 1992) and, secondly, by practising the elicitation method on five women who had already made their prenatal diagnosis decision during

the information giving consultation (see chapters four and five). The two conditions or study groups are described briefly below.

Routine information giving consultation: the consultation between the health professional and the woman was conducted in accord with the routine practice of the antenatal staff at the LGI (see chapter four). The consultation was structured with reference to the triple test result sheet. The following information was mentioned in each consultation: the triple test result and risk; the choice between diagnostic testing and no diagnostic testing; the risks and procedures of the fetal anomaly scan, amniocentesis and chorionic villus sampling; a direct question enquiring about termination for Down's syndrome.

Decision analysis consultation: in essence, the consultation content was the same as that provided in the routine consultation. However, the consultation was structured with reference to: the triple test result sheet; the decision tree visual aid; the lottery decision visual aid; the threshold graph visual aid (Appendix VIII). Although, the elicitation of utilities in the context of prenatal diagnosis was mentioned in section 1.2.1.3, the structure of the decision analysis consultation is described in more detail. First, women in the decision analysis group were provided with the same triple test result information and visual prompt as women in the routine group, i.e. the triple test result sheet. Second, the decision tree representing the alternatives and consequences of the prenatal diagnosis decision was shown to women when the study health professional provided information about the choice to have or not have prenatal diagnosis. Third, women were informed that they were going to be asked a hypothetical question about terminating for Down's syndrome. The study health professional stated that the purpose of this exercise was to gauge the value women attached to the offer of prenatal diagnosis. Women were shown the lottery decision visual aid and asked the following hypothetical question, "If the probability of having a baby with Down's syndrome or without Down's syndrome is given as 50:50, would you (the woman) choose to carry on or terminate the pregnancy?". As mentioned in section 1.2.1.3, the gamble is varied until the woman is unable to answer the hypothetical question. This figure for the 'level of indifference' between terminating and continuing with the pregnancy is the utility figure (range 0 - 100). Fourth, women were shown the threshold graph visual aid. The point of intersection between their utility value and triple test risk was identified. This point usually fell above or below the threshold line, so indicating whether or not the woman should be thinking about having a prenatal diagnostic test. Finally, the resulting decision analysis decision was discussed with women. It was emphasised that the final

decision to have or not have prenatal diagnosis was entirely the woman's, regardless of the decision analysis decision.

The information for each condition was delivered by the same health professional. The same health professional was responsible for all consultations within which the women discussed the receipt of a screen positive triple test result and subsequent testing decisions. The disadvantage of using the same health professional for each condition was that there was a potential increase in experimenter bias. However, had two health professionals had been used, one for each condition, there would have been potential for differences in information-giving to occur as a function of individual style. As the experimental condition requires the manipulation of information, it was considered more important to control for the information given during the consultation than introduce techniques that may or may not have reduced the likelihood of experimenter bias.

6.1.2 Measures.

The following section is concerned with discussing all the measures that were used to evaluate the effectiveness of decision analysis in facilitating women's decision making to have or not have prenatal diagnosis.

6.1.2.1 Consent form.

Ethically, all research involving humans requires the individual to provide informed consent of participation (Bowling, 1997). Written information ought to be provided to the participant about the study purpose, the demands of participation, the associated risks, the voluntary nature of participation, the confidentiality of participation as well as indiciating that it is appropriate to withdraw from the study at any point during the research (Coolican, 1990; Bowling, 1997). Written informed consent should be obtained from the individual after receipt of this study information.

A consent form was developed for this study which included the following information: a measure of anxiety (Short-form Spielberger STAI: Marteau and Bekker, 1992); four likert-scale items assessing perception of risk for having a healthy child, a child with Down's syndrome, miscarrying a fetus with Down's syndrome and miscarrying a healthy fetus; three open-ended questions for the women to note any other concerns they had about their health, pregnancies and babies (Appendix IX). The following factors prompted the inclusion of these measures on the consent form. First, there is some evidence that those who decline to participate differ from those who do not and documentation of these differences is necessary for accurate interpretation and

generalisability of results (Marteau, Johnston, Kidd, Michie and Cook, 1992). Second, there is evidence that anxiety and perception of risk are two factors associated with the decision making process (Janis and Mann, 1977; Marteau, Johnston, Kidd, Michie and Cook, 1992; Van der Pligt, 1988; Van der Pligt and de Vries, 1998). Third, an indication of the pre-consultation levels of these factors was considered useful by the LGI antenatal staff.

6.1.2.2 Consultation.

Within this clinical setting, the most efficient and least obtrusive method of observing the decision making *process* was to audio tape-record the consultation (see chapter five). The resulting transcripts provided a rich source of data. There are numerous techniques that can be employed to analyse transcript data, the choice of which depends on the study aims and research questions (see chapters four and five). The main purpose of this study was to evaluate the effect of decision analysis on women's informed decision making. However, one of the functions of employing a decision aid is to decrease the confusion or conflict associated with making a difficult decision (O'Connor, 1995). In consequence, two techniques were applied to the consultation transcripts to assess both the reasoning and affective content of women's utterances: a theme-based coding frame assessing informed decision making and a text analysis programme assessing the emotional and cognitive content.

Theme-based coding frame: Chapter five described the development of the theme-based coding frame to assess informed decision making. The application of the coding frame to each consultation transcript systematically classified all the information the women referred to during the consultation when making the decision to have a diagnostic test or not. By assessing references to these themes, it was possible to ascertain whether the women were utilising sufficient information to make an informed decision to have or not have a diagnostic test.

The Linguistic Inquiry and Word Count (SLIWC): This text analysis package was developed specifically to look at the emotional and cognitive processes within written texts, such as essays, but has since been applied to transcripts of speech (Pennebaker and Francis, 1997). The package has a powerful dictionary that classifies words in four ways: a linguistic analysis such as counting words and identifying pronouns or articles; psychological constructs, such as affect and cognition; concepts of 'relativity', such as time, space and motion; issues of a personal nature, such as work, home and leisure. The external validity of the package has been reported to be good in a number of

populations and countries (Pennebaker and Francis, 1997). As this programme was not developed specifically for this study, not all of the categories coded by SLIWC were appropriate for this analysis. The categories that were consistent with the research aims included the number of words in the transcript, the percentage of positive emotion-laden words, the percentage of negative emotion-laden words, the percentage of cognitive associated words and the percentage of words referring to social or communication issues (Appendix X).

6.1.2.3 Post-decision Questionnaire - time 1 (T1).

As discussed, the choice of measure to assess the quality of value-laden decisions depends upon the research aims of the study (Llewellyn-Thomas, 1995; O'Connor, 1995). The purpose of this study was to evaluate the effectiveness of decision analysis to facilitate women's decision making within the prenatal diagnosis consultation. As this is one of the first studies to address this issue, there are few validated or reliable measures to assess the decision quality within this decisional context. However, empirical research on decision making and the application of psychological theory to understanding health behaviours have generated a number of measures that could be modified to address this study's aims. Behavioural, affective, cognitive and clinical measures of decision quality were developed with reference to the literature on: the application of decision analysis to women's prenatal diagnostic decision making (Pauker and Pauker, 1978; Pauker and Pauker, 1987; Thornton, 1990; Heckerling, Verp and Hadro, 1994; Verp and Heckerling, 1995); explaining women's prenatal screening and diagnostic behaviour (Marteau, Johnston, Plenicar, Shaw and Slack, 1988; Marteau, Cook, Kidd, Michie, Johnston, Slack and Shaw, 1992; Bekker, Modell, Denniss, Silver, Mathew, Bobrow and Marteau, 1993; Evans, Pryde, Evans and Johnson, 1993; Smith and Marteau, 1995; Michie, Smith, McClennan and Marteau, 1997); models of decision making and behaviour (Eagly and Chaiken, 1993; Shafir, Simonson and Tversky, 1994; Conner and Sparks, 1996).

As the final questionnaire (Appendix XI) was developed with reference to a number of diverse areas and sources, the items included within it were selected following three stages of piloting: the development of a semi-structured interview; the development of a draft questionnaire for peer review: the piloting of the draft questionnaire on a sample of women similar to the population of the main study. The semi-structured interview was completed by five women in the delivery ward. All women included in the pilot of the semi-structured interview were under 30 years of age. Triple test screening is only offered to women at the LGI who will be 30 years of age at estimated date of delivery.

Consequently, no women in this pilot had been offered prenatal screening, which meant that the resulting interview focused mainly on women's attitudes and beliefs towards prenatal screening, testing, abnormalities and termination rather than on decision making during pregnancy. In response to this pilot, a questionnaire was drafted to focus more on items evaluating the quality of women's prenatal diagnosis decision making and less on questions pertaining to their attitudes and beliefs towards triple test screening, abnormalities and termination. As the structure or template of many of these items was informed by previous research, the resulting measures have likert scales with different lengths, with either five-, six- or seven- point scales. This version of the questionnaire was circulated for comments to colleagues with a range of expertise: AP and JM in theoretical and applied decision making research; RP and CA in the application of social cognition models to food health behaviours; JGT and JH in prenatal care and application of psychological theory to health services research. In addition, the questionnaire was completed by fourteen women waiting to undergo diagnostic testing in the LGI antenatal clinic. That is to say, the pilot sample were sufficiently similar to the target sample of the main study.

The women completed the questionnaire (T1) after they had made their decision to have or not have further diagnostic testing but before they had had the diagnostic test or the nineteen week fetal anomaly scan. Most of the women made this decision after the information giving consultations described in chapters four and five. The final items included within the study questionnaire are discussed in detail below, with reference to the appropriate literature and comments from the piloting exercises. The items are grouped by type of measure: behavioural; affect; cognitive processes; clinical quality; socio-demographic characteristics and individual differences.

6.1.2.3.1 Behavioural measures.

The actions of undergoing diagnostic testing or of continuing the pregnancy without diagnosis provided the main behavioural measures. In addition, the questionnaire included three 'intention to behave' items: intention to have a test (7 point likert scale); intention to recommend diagnostic testing (5 point likert scale); intention to recommend the triple test (5 point likert scale) (Appendix XI: Q17, Q31, Q39). In the first instance, the intention to behave item was informed by the literature on reasoned and rational choice models of behaviour which propose that intention and actual behaviour are causally related (Conner and Sparks, 1996). Intended behaviour has frequently been used in studies as the main measure of behaviour when it has not been possible to assess actual behaviour. These items were informed by the literature aiming to evaluate

the quality of decision making (Pitz, 1987). Assessing whether or not a course of action would be recommended by the individual is an indirect measure of satisfaction (Pitz, 1987). There are at least two problems with attempting to assess satisfaction directly in this context. First, evaluations were made by the women after a decision was reached. It is likely that some cognitive readjustments could have taken place such as those described by dissonance theory and thus biasing the response (Festinger, 1957; Olson and Zanna, 1993; see chapter one). Second, the women were being asked to comment on their satisfaction with their health care half-way through their prenatal care. It is likely that women would be encouraged to provide a socially desirable answer (Bowling, 1997), one that did not jeopardise subsequent care.

6.1.2.3.2 Affect measures.

The emotional response of the women to the prenatal diagnosis decision making process was assessed by a number of measures of affect. It has been well documented that prenatal screening and diagnosis are associated with short-term variations in anxiety and worry about the procedure and subsequent results (Farrant, 1980; Nielson, 1981; Verjaal, Leschot and Treffers, 1982; Burton, Dillard and Clark, 1985; McGovern, Goldberg and Desnick, 1986; Evans, Pryde, Evans and Johnson, 1993; Green, 1990, 1994). In addition, other studies report the distress of making a difficult decision about such value-laden choices (Pauker and Pauker, 1977; Farrant, 1980; Sjorgen and Uddenberg, 1988). Measures of affect were included within the questionnaire to evaluate whether or not decision analysis was associated with a decrease in distress.

There are a number of established, validated and reliable measures of mood states including: the Zung Self-Rating Depression Scale (SDS: Zung, 1965); the Leeds Hospital Anxiety and Depression Scale (HADS: Zigmond and Snaith, 1983); the Beck Depression Inventory (BDI: Beck, Ward, Mendelson, Mock and Erbaugh, 1961); General Health Questionnaire (GHQ: Goldberg, 1987); the Spielberger State-Trait Anxiety Inventory (STAI: Spielberger, 1976). The majority of these measures were developed to assess the mood states of clinical populations to evaluate the prognosis of patients. As the study sample were not patients, the most appropriate measure was one validated within a well population. Also, the discrete nature of the test context suggests that anxiety rather than depression was the more appropriate emotional response to assess. The STAI has been validated within a number of populations (Spielberger, Gorsuch, Lushene, Vagg and Jacobs, 1976; Knight, Waal-Manning and Spears, 1983) and applied within the prenatal testing context for over ten years (Beeson and Golbus, 1979; Marteau, Kidd, Cook, Michie, Johnston, Slack and Shaw, 1992; Marteau, Cook,

Kidd, Michie, Johnston, Slack and Shaw, 1992; Michie, Marteau and Kidd, 1992; Evans, Pride, Evans and Johnson, 1993; Thornton, Hewison, Lilford and Vail, 1997). In addition, the original twenty items of the STAI (state scale) have been shortened to a six-item version and validated within a pregnant population (Marteau and Bekker, 1992). In consequence, the questionnaire included the short-form STAI as a general measure of state anxiety.

It has been suggested that general measures of affect may not be sensitive enough to the emotional responses of specific events such as pregnancy and prenatal testing (Green, 1990; Marteau, Kidd, Michie, Cook, Johnston and Shaw, 1993). Consequently, some researchers have developed context specific measures of affect (Green, 1990;). Items from the Worries Questionnaire (Thornton, Hewison, Lilford and Vail, 1997) were modified for inclusion in the pilot questionnaire. The content of the original measure included items about testing and attachment with a multiple choice response layout. Measures of attachment were not considered appropriate for inclusion within the questionnaire. The main reason for this omission was the ever-increasing length of the questionnaire. In essence, the purpose of the study was to evaluate the effectiveness of decision analysis on the decision making process. Measures of the impact, coping and adjustment following the decision were beyond the primary concern of this study. The items included were: worry that the scan, medication, emotion, amniocentesis and chorionic villus sampling may harm the baby; worry that the baby would be physically ill, mentally ill or have a serious health complaint. An alternative seven-point likert scale response layout for the selected items was piloted (Appendix XI: Q43 and Q44). This likert scale format was shared with several other items within the questionnaire and no adverse comments were made about their completion. The main advantage of converting from a multiple choice to likert scale format was the generation of continuous rather than categorical data for subsequent analyses.

There are fewer measures of affect associated with the process of decision making. At the time of questionnaire development, two measures were being referred to within the decision making literature: the Flinders Decision-Making Questionnaire, now published as the Melbourne Decision-Making Questionnaire (MDMQ: Mann, Burnett, Radford and Ford, 1997); the Decisional Conflict Scale (DCS: O'Connor, 1995). The theoretical foundations of both the MDMQ and DCS derive from conflict theory (Janis and Mann, 1977). However, the MDMQ is concerned with individual coping styles and the DCS with the affective state associated with the decision making process. Although the MDMQ has been applied to health care decisions, it was developed within the context of

career and business decision making amongst student populations. The DCS was developed within the context of health care treatment choices by patients. As the DCS met the research aims more fully than the MDMQ, the measure was included within the questionnaire. Since development of the questionnaire, a Satisfaction with Decision Scale (SDC: Holmes-Rovner, Kroll, Schmitt, Faan, Padonu and Talarczyk, 1996) has been published. Subsequent studies may wish to consider the inclusion of this measure to assess the decision quality.

6.1.2.3.3 Cognitive measures.

Measures aimed at inferring the cognitive processes employed by women when making the decision to have or not to have a diagnostic test were particularly important to the evaluation of decision analysis as a technique to facilitate decision making. The purpose of decision analysis is to change the way an individual understands the decision (Pitz, 1987) by aiming to encourage a more systematic, analytic or rational processing of information. As discussed, there are a number of cognitive changes that may be expected following the application of the decision analytic technique: increased reasoning; greater understanding of the decision attributes; a more realistic interpretation of the risks; a greater awareness of the utilities and attitudes. The measures of cognitive process considered for inclusion in the T1 questionnaire are discussed in more detail below. The T1 questionnaire is reproduced in Appendix XI.

Assessing reasoning of the information used to make a decision is an item common to the different literatures. Several studies have included a simple, open-ended item to ascertain what the reasons were for the final choice (Verjaal, Leschot and Treffers, 1982; Roelofsen, Kamerbeck, Tymstra, Beekhuis and Mantingh, 1993; Julian-Reyner, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994). An open-ended question was included within the questionnaire to document the respondent's main reason for having the triple test. However, it is likely that individuals hold both reasons for and against the choice they accept or reject (Shafir et al, 1994). That is to say that they have an ambivalent attitude towards the behaviour (Eagly and Chaiken, 1993; Olson and Zanna, 1993; Conner and Sparks, 1996). Certainly the decision making and social cognition literature suggest that decisions are made following a trade-off between the reasons for and against alternatives (Eagly and Chaiken, 1993; Frisch and Clemen, 1994; Shafir et al, 1994; Conner and Sparks, 1996). Consequently, an open-ended item was developed from a study assessing attitudes to breast cancer screening (Bekker, Morrison and Marteau, 1999) to assess the individual's perception of reasons for and against prenatal testing. An open-ended question design was selected to ensure that the

reasons generated were salient to the respondent rather than to an evaluation of the researcher's agenda. The reasons for having tests in pregnancy were referred to as 'advantages' and reasons against as 'disadvantages'. On the same line as each space for the advantages and disadvantages responses was a five-point likert scale (either small to great advantage or small to great disadvantage) informed by a study assessing ambivalent attitudes to food (Povey, 1997). These evaluations of the respondent's reasons may go some way towards describing the trade-off between reasons.

Most of the empirical research within decision analysis and social cognition applications is concerned with assessing utilities or attitudes and perceptions of risk or probability, that is to say, with cognitive processes that are less easily expressed than reasons. Chapter one highlighted the breadth of issues researched under the umbrella term 'prenatal testing', such as attitudes and perception of risks towards screening, prenatal diagnosis, abnormality, termination and new technologies. As demonstrated by piloting of the semi-structured interview, the attempt to assess all the different prenatal testing decisions within one research project proved to be an unrealistic aim. The purpose of the study was to evaluate the effectiveness of decision analysis to facilitate prenatal diagnosis decision making. Consequently, the final questionnaire contained only ***attitude*** and ***perception of risk*** items associated with the four outcomes of the prenatal testing decision: the birth of a healthy baby; a miscarriage (spontaneous abortion); a termination (elective abortion); the birth of a child with an abnormality (Pauker and Pauker, 1978).

The subjective norm is a concept within the social cognition literature which represents an individual's evaluation of a third person's opinion about the individual performing the action (see Conner and Sparks, 1996; Trafimow, 1998). That is to say, it provides an evaluation of the perceived normative action. Some argue that the perceived social norm is a concept distinct from measures of attitudes; others argue that attitudes and subjective norms are measures of the same underlying construct (see Trafimow, 1998). For the purposes of this study, the subjective norm was operationalised as an independent predictor of behaviour, distinct from other evaluations of the behaviour. A single item (on a seven-point likert scale) was included within the questionnaire to assess the women's perceived social norm towards prenatal diagnosis.

As mentioned in chapter one, there are many methods of ***eliciting utilities*** or ***measuring attitudes***. During the consultation, those within the decision analysis group provided a utility following the lottery or standard gamble technique (Pauker and

Pauker, 1978; Thornton and Lilford, 1990; Baron, 1994). However, such a technique could not be used within the questionnaire without potentially confounding the decision making process of the routine consultation group. A direct scaling technique would be less confrontational (Pauker and Pauker, 1978; Baron, 1994). One direct scaling method requires the respondent to place on a line, labelled from zero to one hundred, the desirability of all the possible consequences of the alternatives (Pauker and Pauker, 1977, 1978, 1987). The 'best' consequence is placed at one hundred, the 'worst' at zero and the other consequences somewhere between the two points. The best consequence usually refers to the birth of a healthy child, the worst consequence the birth of a child with Down's syndrome. This measure provides both a utility figure and the relative relationship of the consequences to each other. During piloting, this question had to be modified. First, the demands of this question are unusual for standard self-completion questionnaires. The written explanation was longer than other items and required additional verbal prompting to encourage the placing of consequences at '0' and '100'. Second, respondents frequently mentioned the difficulty in ranking the consequences as some were perceived to be equally good or as bad as others. The final utility measure simplified the demands of the task for respondents by placing a line labelled zero to one hundred alongside each consequence of the prenatal diagnosis decision. The respondent placed a mark along each line to represent how bad or good that consequence was perceived to be. It was the responsibility of the researcher to note both the utility and ranked position of the consequence in relation to the others. This revised item allowed the respondent to answer the question in a more 'naturalistic' manner without enforcing an EUT structure on the subsequent response.

In addition to the above measure of utility, a number of more routine ***belief items*** were included within the questionnaire. In an attempt to assess respondents' beliefs about the cause of Down's syndrome, an open-ended question was provided for respondents to suggest what may increase or decrease the likelihood of having a child with Down's syndrome. A question was developed to assess respondents' beliefs about a child with Down's syndrome. From preliminary observations within consultations (chapter five), comments concerning Down's syndrome focused on either coping with a child with Down's syndrome or the quality of life a child with Down's syndrome might experience. The questionnaire contained two seven-point likert scales to assess the quality of life and amount of care of three abnormalities: a predominantly mental abnormality, Down's syndrome; a physical abnormality, Spina Bifida; a less severe physical abnormality, cleft palate. The aim of the last four items was to assess respondents' general beliefs about the purpose of testing during pregnancy. Following the semi-structured interviews,

four statements from the women's responses were selected: that tests are necessary for the woman's well-being; that tests improve the chance of having a healthy baby; that tests are necessary for the well being of the fetus; that tests decrease the chances of miscarrying. Five-point likert scales were used to assess the degree of agreement with each statement. Although these items were informed by the literature and piloted, they have not been validated or their reliability established. The items were included within the questionnaire as a preliminary stage in the assessment of their feasibility as more established or standardised measures.

Perception of risk is the other cognitive process of importance when assessing decision making. Numerous studies have reported a relationship between individuals' perception of risk and subsequent health behaviour (see Van der Pligt, 1998; chapter one). However, it is well documented that an individual's judgement of risk information is sensitive to the context of presentation (Vlek, 1987; Croyle, Sun and Hart, 1996; Van der Pligt, 1998). The subsequent utilisation of biased information may lead to poorer decision outcomes. As decision analysis visually represents all the risks associated with the decision, it is argued that an individual's subsequent perceptions should be more accurate than a method which may or may not focus equally on all the information (Yates and Stone, 1994). As perception of risk is a multi-attributed concept, there is no optimum measure to assess evaluations of risk (Vleck, 1987; van der Pligt, 1998). This questionnaire included three assessments for perception of risk: an evaluation of the personal risk for Down's syndrome as low, medium or high; estimates for the likelihood of occurrence of seven possible prenatal diagnosis consequences; five items to assess the comparative likelihood of the respondents developing health or pregnancy problems compared with other similarly aged women. For each item contained within the last two questions, respondents circled numbers on verbally labelled likert scales. This type of scale has been found to be a good predictor of behavioural intentions (Van der Pligt, 1998).

An assessment of ***knowledge*** of the test procedures is one of the most frequently used measures within the applied literature (French, Kurczynski, Weaver and Pituch, 1992; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994; Jorgensen, 1995). Knowledge is an indirect measure of informed decision making or of understanding of information, as such measures assess whether or not respondents remember information they may or may not have been provided with. These measures are unable to assess what information was used by the women when making their prenatal diagnosis decision. The format for most measures of knowledge is to include a

number of multiple choice items based on factual information considered necessary to enable informed decision making (French, Kurczynski, Weaver and Pituch, 1992; Bekker, Modell, Denniss, Silver, Mathew, Bobrow and Marteau, 1993; Marteau, Kidd, Michie, Cook, Johnston and Shaw, 1993; Michie, Smith, McClennan and Marteau, 1997). The study questionnaire included ten multiple choice knowledge items, which included questions about factual information associated with diagnostic testing, miscarriage and Down's syndrome.

6.1.2.3.4 Clinical quality measures.

The following measures were included in order to address some of the concerns expressed when decision analysis is applied to a real-world clinical situation: that it is time consuming; that it confuses the lay population; that it compromises the individual's autonomy when making decisions. As this was one of the first studies to evaluate decision analysis within the prenatal testing context, there were few examples of measures employed to provide evidence supporting or counteracting these concerns. In addition, it is unclear whether all these clinical concerns are adverse or undesirable. For example, an increased length of time to make a decision may be associated with a more systematic evaluation of the information whereas a short consultation length may suggest that an heuristic was applied when making the decision (Eagly and Chaiken, 1993; Michie, Smith, McClennan and Marteau, 1997). Equally, within a paradigm of shared doctor-patient decision making, directiveness is perceived negatively. However, there is some evidence that patients want to be informed but do not necessarily want to make decisions about their treatment (Fallowfield, 1997). Finally, it is likely that some patients may not be aware that information or advice provided by health professionals about a desired course of action might be considered directive or inappropriate.

The questionnaire included a seven-point likert scale to estimate the perceived length of time taken to make the decision and a categorical variable to assess whether the decision was made before or after the consultation. The consultation length had been noted during transcription of the audio-tapes of consultations. As perceived autonomy in decision making is seldom assessed (Sjorgen and Uddenberg, 1988), an item was developed and piloted specifically for this study. A five-point likert scale assessed whether or not a number of health professionals and significant others were forthcoming about their choice of an appropriate course of action. Usefulness of information was evaluated with a single seven-point likert scale. In an attempt to avoid the interviewer and social desirability bias raised earlier in this section, the wording to elicit reponses about the quality of the consultation was carefully selected. For example, the terms

'directive' and 'satisfied' imply a certain or socially desirable response. Consequently, the more positive and less ambiguous term 'encouraged' was used in the autonomy in decision making question. Equally, the term 'satisfied' requires the patient to comment directly on the care they have received. As most of the women will continue to be seen by the same health care team, this type of question may have resulted in a response bias. The term 'useful' replaced the 'satisfied' term as it required the women to focus on an aspect of the care provided and not on an evaluation of the service.

6.1.2.3.5 Profile characteristics of women.

In addition to measures evaluating the effectiveness of the intervention on women's decision making, the questionnaire included a set of variables to describe the following profile characteristics of participants: socio-demographic variations; reproductive history; individual differences in information processing. A number of studies have reported associations between these types of measures and subsequent prenatal diagnosis decisions (see chapter one). Consequently, descriptions of these differences may assist in the interpretation and generalisability of findings. In an attempt to restrict the length of the questionnaire, items most closely associated with the research aims were selected from the range of available measures.

Socio-demographic characteristics included four items: marital status; level of education; religious activity; ethnic origin. A four-category, single item assessed whether a woman was married, living as married, single or another category. A measure for level of education was included rather than socio-economic status (SES). As the intervention was concerned with information presentation and processing, an item describing respondents' educational training seemed a more useful measure than SES. In addition, accurately evaluating the SES of female 'reproductive' respondents is more difficult as their employment status is likely to be influenced by having had children. The item assessing religious activity had been developed for use in a multi-cultural, primary-care based population (Bekker, 1994). The first part of the item allowed respondents to indicate which religion, if any, they followed. The second part of the item rated how often they attended religious gatherings. The item aimed to differentiate those respondents with an active set of religious beliefs from those with an affiliation to a particular faith. The measure of ethnic origin was informed by those categories of the Officie of Population, Censuses and Surveys in the UK (1993).

Reproductive history included three items assessing prior experience of prenatal testing and Down's syndrome. These single items were developed for the study and

piloted following a review of the appropriate literature. There is evidence that prior behaviour is a predictor of future behaviour (Van der Pligt, 1994). It is likely, therefore, that choice of diagnostic test may be associated with past reproductive decisions, such as previous prenatal tests, experiences of abnormality and terminations. Information about the previous number of pregnancies, miscarriages and terminations was derived from the women's notes.

There are numerous measures assessing ***individual differences***, personality traits or dispositional constructs that have been developed within psychological research. Previous research suggested an association between preferences for a style of judgement and decision making (Schwarzer, 1994; Van der Pligt, 1994; Webster and Kruglanski, 1994; Sorrentino, Holmes, Hanna and Sharp, 1995). Considered for inclusion within this questionnaire, then, were standardised measures assessing differences in style of judgement only, in particular the Need for Cognition (NC: Cacioppo, Petty and Kao, 1984) and the Need for Cognitive Closure (NFC: Webster and Kruglanski, 1994); Life Orientation Test (LOT: Scheier and Carver, 1985). The NC assesses "an individual's tendency to engage in and enjoy effortful cognitive endeavours" (pg. 306; Cacioppo, Petty and Kao, 1984); the NFC a "desire for predictability, preference for order and structure, discomfort with ambiguity, decisiveness, and close-mindedness" (page. 1049, Webster and Kruglanski, 1994); the LOT "dispositional optimism, a habitual style of anticipating favourable outcomes" (page. 41, Johnston, Wright and Weinman, 1994). The NFC was not included within the final questionnaire because it was reported to be highly correlated with the NC (Webster and Kruglanski, 1994) and also included 42 items within the measure. An eight-item short-form measure of the NC scale modified from the eighteen-item short-form measure (Cacioppo, Petty and Kao, 1984) was included within the questionnaire. The development of the eight-item measure is reported in Appendix XII. The short-form LOT (Johnston, Wright and Weinman, 1994) was also included within the final questionnaire.

6.1.2.4 Follow-up questionnaire - time 2 (T2).

The literature suggests that the effectiveness of decision analysis to facilitate decision making should have post-decisional advantages. Essentially, the more accurate judgements of risk information and explicit evaluations at the time of decision making should result in more robust attitudes (Eagly and Chaiken, 1994) and greater satisfaction, or less regret, with the final choice (Pitz, 1987; O'Connor, 1994). The questionnaire T1 was modified and used as a follow-up questionnaire (Appendix XIII). The following items were omitted from the T2 questionnaire because they should have

remained constant over the one-month time period: socio-demographic characteristics; reproductive history; individual differences. As the purpose of T2 was to evaluate the long-term impact of decision analysis on decision quality, only items pertaining to the prenatal diagnosis decision and information consultation were evaluated at T2. All the cognitive, affective and clinical measures of decision quality were included within the final questionnaire.

6.1.3 Procedure.

The LGI's routine clinical practice for contacting women, informing them of their triple test result and providing the prenatal diagnosis information giving consultation was maintained throughout the fifteen-month study period (March 1996 - June 1997). Chapter four described this routine clinical practice in more detail. All the women attending the clinic for the prenatal diagnosis information giving consultation were informed of the study both verbally and with written information before the start of the consultation. All women completed a consent form stating whether or not they agreed to participate in the research. The women declining to participate in the study received the routine care consultation from the study health professional. No further contact was made with those women declining to participate in the study once they had reached a decision about prenatal diagnosis.

The women agreeing to participate in the study were allocated to one of the study consultations following the opening of a previously sealed, numbered, opaque envelope. Within the envelope was an instruction stating whether the consultation was to be conducted according to routine care or structured using decision analysis. The differences in the structure of the two consultations have been described in section 6.1.1. This simple random sampling technique ensured that each woman had an equal chance of allocation to one of the two experimental conditions. The audio tape-recorder was switched on as soon as the woman agreed to participate in the study. At the end of the consultation, most women made a decision to have or not have a diagnostic test. The women were given the questionnaire (T1) to complete at this point. To reduce the influence of the author on the women's responses, they completed the questionnaire (T1) alone. Ideally, all the women would have completed the questionnaire in the clinic after the consultation and before undergoing any procedures. However, this was a real decision being made within a clinical setting. Occasionally, the women either required more time to reach a decision or wanted to discuss the decision made with a partner not present during the consultation. There were also instances of some women undergoing the diagnostic test procedure before completion of the questionnaire because of the

availability of the other health care professionals. These women were provided with stamped addressed envelopes and encouraged to return the questionnaires (T1) as soon as was convenient.

Those women who had participated in the study were sent a follow-up questionnaire (T2) and stamped addressed envelope four to five weeks after the screen positive triple test consultation. Originally the follow-up questionnaire (T2) was to be sent at a time when all the consequences of the prenatal diagnosis alternatives would have occurred: after the birth of a baby with or without Down's syndrome; after termination; after miscarrying. However, following discussions with the antenatal team it was considered inappropriate and insensitive to send questionnaires to those women who had had an undesired consequence. In addition, the questionnaire (T2) would have had to have been sent six months after the women made the decision not to have or to have a diagnostic test to ensure that all the consequences had occurred, i.e. after the birth of the baby. It is likely that the evaluation of the information intervention at this stage would be particularly sensitive to the memory and cognitive readjustment biases of human processing raised earlier in this thesis (see chapter one). Most of the following consequences of the prenatal diagnosis decision apart from the birth of a child with or without Down's syndrome would have occurred four to five weeks after the screen positive triple test consultation: diagnostic test result; nineteen-week fetal anomaly scan; test-related miscarriage; termination. In consequence, those women who had not miscarried or received a positive diagnostic test result within this time period were sent a follow-up questionnaire (T2). If the women had not returned the follow-up questionnaires (T2) within two weeks, reminder questionnaires (T2) and stamped addressed envelopes were sent in an attempt to decrease attrition rates.

In theory, all the women who received screen positive triple test results throughout the study period were eligible for inclusion in the study. However, there were a number of situations which meant that some women were subsequently excluded from the study both pre- and post-randomisation. These situations meant that it was either difficult to control the amount and quality of information the women were exposed to or that the decision process and outcome could not be evaluated. The reasons for exclusion fell into two categories: those women that did not fulfil the study criteria; factors associated with women's decision making. The criteria issues were: those women with a complication that influenced the interpretation of the triple test result, such as diabetes and twins; those women informed about their test result by another member of the antenatal team on occasions when the author was not available; those women receiving

a screen positive triple test result for either spina bifida or Edward's syndrome; those women who participated in the study but during the course of the consultation found an error on their triple test result sheet and, following re-calculation, received a screen negative result; those women who could not read and write English. The factors associated with the women's decision making were: those women paying for prenatal care; those women who made a decision about prenatal diagnosis over the telephone; those women who participated in the study but did not return their post-decision questionnaire (T1). It was likely that the women paying for treatment, those deciding not to have a test or have a test over the telephone and those not returning their post-decision questionnaire (T1) differed in their decision making from those agreeing to take part in the study who had completed the post-decision questionnaire (T1). However, it was beyond the scope of this study to adequately explore this assertion.

Power calculations were used to estimate a sufficient sample size. As previously alluded to, there were few standardised measures at the time of questionnaire development to assess the quality of the decision. Only anxiety (STAI: Spielberger et al, 1976; Marteau and Bekker, 1992) and decisional conflict (DC: O'Connor, 1994) had been validated. Of these, the most widely applied measure within the prenatal testing and other contexts is anxiety (STAI). Consequently there are a number of examples of expected anxiety levels in various situations (Spielberger et al, 1976; Beeson and Golbus, 1979; Marteau and Bekker, 1992; Marteau, Kidd, Cook, Michie, Johnston, Slack and Shaw, 1992). In a non-pregnant sample of women aged between 19-39, the mean anxiety score was recorded as 36.2 and the standard deviation 11.0 (Spielberger et al, 1976). Within a pregnant population a similar value is recorded, mean 37.1 and standard deviation 11.0 (Marteau and Bekker, 1992). In women undergoing amniocentesis, higher mean anxiety scores are reported, between about 42 and 52 points, with no standard deviation scores reported (Beeson and Golbus, 1979; Marteau, Kidd, Cook, Michie, Johnston, Slack and Shaw, 1992). As decision analysis has not yet been evaluated, how much anxiety can be estimated to decrease from routine practice is unclear. For the purposes of the power calculation, a six-point difference in mean anxiety scores between groups was estimated to be a reasonable clinical decrease. The standard deviation reported within the shortened and full form STAI publications of eleven points was also used. The power was set at 80% and the degree of significance at 5% (0.05). An estimated sample size, then, would be approximately 52 women in each arm of the trial.

6.1.4 Analysis.

6.1.4.1 Missing values.

Frequencies were run for all the variables contained within the data files. The study numbers of the women were noted for any erroneous figures or missing data entry points. Questionnaires and transcripts were used to correct any mis-typed figures. Missing data can be dealt with in a number of ways: deletion of the whole case; using the mean to estimate a missing value; repeating the analysis with and without missing data; analysing all cases by missing data (Tabachnick and Fidell, 1989; Robson, 1997). The method used for this data was to replace the missing values with a mean value. As the subsequent analysis was carried out to evaluate differences between the routine and decision analysis groups, the estimated mean value of the appropriate group was used. If the missing value was pertained to a single item measure, the inserted mean was calculated from all the values of the remaining sample. If there was a missing value for an item within a standardised measure and more than 50% of the measure had been completed by the participant, then the mean value was calculated 'pro-rata' from the completed items. If less than 50% of the items of the measure had been completed by the participant, then the mean replacement was calculated from the remaining group data. However, if the same value was missing from more than 10% of the sample (n = 11), the measure was omitted from further analysis.

6.1.4.2 Outliers and normality of distribution

Outliers are cases with values on a variable or variables that can unduly influence statistics (Tabachnick and Fidell, 1989). Dichotomous variables with uneven splits between the two categories are regarded as outlier variables; 90% to 10% splits should not be used in subsequent analysis (Tabachnick and Fidell, 1989). How outliers are identified depends on the analysis to be carried out. The purpose of this analysis was to assess differences in measures by grouped data, routine consultations compared with decision analysis information giving. Consequently, the SPSS explore command was run by group to identify any outliers. The values of all outliers were checked with the raw data and altered where appropriate.

The SPSS explore command was used on the complete data set of both questionnaires to identify variables with distributions that varied significantly from the normal distribution. A z-value of 3.67 ($p < 0.001$) was used to determine which variables required transformation and/or elimination from further analysis (Tabachnick and Fidell, 1989).

6.1.4.3 Calculating Standardised Measures.

The post-decision questionnaire included items from four standardised measures: anxiety; decisional conflict; need for cognition; life orientation test (Appendix XI). The manipulation of the raw data to form these scores is discussed below.

Anxiety (Q9 - 14) - the positively framed items 'calm', 'relaxed' and 'content' were recoded in reverse order. All values from the six items were combined and the total pro-rated to be compatible with the complete form, i.e. multiplied by twenty and divided by six (Marteau and Bekker, 1993). The higher the score, the greater the expressed anxiety (range 20 - 80).

Decisional conflict (Q19 - 29) - the negatively framed items were recoded in reverse order. Three scores were calculated following addition of the items according to O'Connor, 1996: uncertainty with decision; informed decision making; efficacy in decision making. The higher the scores, the greater the expressed decisional conflict (ranges from 3 - 15, 4 - 20 and 4 - 20 respectively)

The need for cognition scale (eight item) was calculated in accord with Cacioppo, Petty, Kao (1984) (see Appendix XII). The values of items reflecting little desire to think in depth were recoded in reverse order. All values were combined and the total pro-rated to be compatible with the complete 34-item form. The lower the score, the greater the need for cognition (range 34 - 170).

The life orientation test (short-form) score was calculated in accord with Scheier and Carver (1985). The value of the negatively framed item was recoded in reverse order. The two items of the measure were combined and the final score pro-rated to be compatible with the complete 8-item scale. The lower the score, the greater the optimism outlook of the participant (range 8 - 40).

6.1.4.4 Calculating Study Specific Measures.

Measures from consultation transcripts: three informed decision making variables were calculated from the results of the application of the theme-based coding frame to the consultation transcripts: information seeking informed decision making score (IDM-I); reasons for pursuing an option or alternative informed decision making score (IDM-F); reasons against pursuing an option or alternative informed decision making score (IDM-A) (see chapter five).

Measures from post-decision questionnaire (T1): Fourteen measures were calibrated from items within the questionnaire: number of advantages and disadvantages of testing in pregnancy; strength of advantages and disadvantages; expected utility scores associated with the five consequences of testing; rank ordering of the values of the consequences of testing; directiveness of the diagnostic test information; knowledge about testing. The calibrated measures enabled some of the open-ended responses to be used within more formal and robust analyses.

Number of advantages and disadvantages of testing in pregnancy. Although the question permitted up to six responses for both advantages and disadvantages, the women reported no more than three advantages or disadvantages. Six new dichotomous variables for the advantages and disadvantages were calculated, representing whether or not an advantage or disadvantage had been referred to. From these new variables two scores were calculated: an addition of the dichotomous advantages variables to obtain the *number of advantages total* and an addition of the dichotomous disadvantages variables to obtain the *number of disadvantages total* (ranges 0 - 3).

Strength of perceived advantages and disadvantages of testing in pregnancy. Two continuous scores were calculated by combining all the evaluations of the listed advantages or disadvantages and dividing this total score by the frequency of advantages or disadvantages recorded (range 0 - 5).

Expected utility score for the five consequences of testing. Five variables were calculated from two questions to form the expected-utility of a consequence. The utility or value of the consequence was multiplied with the perceived likelihood of the consequence occurring. For example, the responses to "having a healthy baby is good - bad" and "the likelihood that I will have a healthy baby is 0 - 6" were multiplied together. Before multiplication of the values, the utility score was divided by ten resulting in an expected-utility score for each consequence of between 0 - 60. The higher the score, the greater the expected-utility of the consequence. Following exploration of the distribution of scores, a dichotomous variable was created for the expected-utility value associated with terminating a child with Down's syndrome. As the data suggested a bi-modal distribution, a median split was used as the cut-off for the for the dichotomous variable.

Rank ordering of utility scores. A dichotomous score was calculated to represent whether or not the women were able to rank in order the utilities of the prenatal testing consequences. Those women not completing this question or who gave the same value to two or more of the consequences scored zero, those marking a different utility for each consequence scored one (categorical variable 0/1).

Directiveness of information given about diagnostic tests. The information contained within this question was simplified to form three variables: perceived directiveness of researcher information; directiveness of other health professional information; directiveness of friends and family information. First, the values for each item were recoded to form four categories from the original six: not discussed (0); neutral information (1); encouragement towards an option (2); discouragement from an option (3). Second, responses to the general practitioner, obstetrician and midwife items were collapsed to form one health professional variable (categorical 0 - 3); partner, friends and others were collapsed to form one friends and family variable (categorical 0 - 3).

A total knowledge score was calculated from the multiple choice questions about diagnostic tests, miscarriage and Down's syndrome. Answers to question fifty-three were not included within the final score because the forced responses were not sufficiently different to generate a meaningful response. A correct response to all parts of a question scored 'one', an incorrect response 'zero'. Most questions were worth a maximum score of two, one correct response for amniocentesis and one correct response for chorionic villus sampling. Ideally there would have been only one correct response for each multiple choice item. However, as medicine is an imprecise science, there are occasions when more than one response may be correct. For example, within the prenatal testing context women are exposed to a range of risk figures for miscarriage and Down's syndrome. In addition, verbal expressions of probability are frequently used with some having subjectively similar evaluations. In consequence, a response was scored as correct for either of the following responses: it is unlikely the baby has Down's syndrome or the baby might have Down's syndrome; the risk of amniocentesis is 1 in 100, 1 in 150 or 1 in 200; the risk of chorionic villus sampling is 1 in 50 or 1 in 100; the average population risk of a baby being born with Down's syndrome is either 1 in 600 or 1 in 1000. A higher score was associated with a greater number of correct responses (range 0 - 20).

6.1.4.5 Excluding variables from further analysis.

As reported in the previous sections, the post-decision questionnaire (T1) was designed with reference to theories of decision making and prior empirical research within the context of prenatal testing. The T1 questionnaire was also piloted in an appropriately representative sample. However, despite the careful selection of items included within the T1 questionnaire, a number of measures were not included in this chapter's analyses. The two main reasons for excluding measures are discussed in more detail below.

First, a number of the non-standardised measures that appeared acceptable during the pilot study raised some concern during the main study. The data exploration showed the distribution of scores to be not normally distributed. A closer examination of the questions and replies suggested that either the wording of the question was ambiguous or the validity of the response was uncertain, resulting in the data being meaningless. Fortunately, a number of observational or standardised measures included in the evaluation assessed similar constructs to those of the study-specific measures to be excluded. The excluded variables were: the study-specific measure of intention to have a test; the perceived length of time to make the diagnostic test decision; the qualitative variables assessing helpfulness of information; the 'worry' questions; the prenatal testing belief items. The measures with similar constructs were respectively the actual behaviour to have or not to have testing, the actual length of the consultation, the likert scale assessing usefulness of information, the anxiety measures and the measure of decisional conflict.

Second, a number of questions were included in T1 that may be considered to be associated with the decision to have a diagnostic test or not but were not direct evaluations of the decision making process or outcome. It was thought that such items would encourage the women to focus on the context of the prenatal diagnosis decision rather than pregnancy *per se*. These items were not essential to the analyses evaluating an association between decision analysis and the facilitation of prenatal diagnosis decision making. The items excluded were: all questions exploring the triple test decision and experience; questions about coping with and the perceived quality of life of children with a cleft palate and Spina Bifida.

6.1.4.6 Structuring the Analyses.

The variables selected for analyses either provided information to evaluate the quality of the decision or helped to interpret the generalisability of subsequent findings. The

analyses included descriptive, univariate and multivariate tests. Two tailed tests of significance were used because there was no *a priori* evidence to indicate that the application of decision analysis would lead to 'better' decisions, 'worse' decisions or no difference in outcomes from routine consultations. A brief overview of the analyses are described below. More specific details about the tests employed are described in more detail under each of the subsequent results sections.

1. Over the fifteen-month study period, 178 women received a screen positive triple test result. Reasons for exclusion from the study have been described above. The following variables extracted from the women's notes were used to assess the representativeness of the final sample compared with women not included in the study: age, gestation, number of children, family history of abnormality, number of miscarriages, number of terminations and test decisions. In addition, an analysis was carried out to assess differences in pre-consultation anxiety by those that completed a consent form (n = 132): those agreeing to take part (n = 106); those agreeing to take part but later excluded post randomisation (n = 11); those declining participation (n = 15).
2. In total, 106 women were included within the final study sample: 56 women were randomised to the routine consultation; 50 women to the decision analysis consultation. The following variables were used to assess the comparability of the women within each of the study groups: age, gestation, number of children, family history of abnormality, number of miscarriages, number of terminations, previous diagnostic test experience, marital status, level of education, religious activity, ethnicity, pre-consultation measures of anxiety, optimism (LOT), need for cognition and return rate of post-decision questionnaire (T1).
3. Analyses were carried out to assess any differences in the process of decision making measures by study group (n = 106). The data used within this section were extracted from the consultation transcripts. The results of the theme-based coding frame assessing informed decision making and the computer based text analysis SLIWC (Pennebaker et al, 1997) were presented.
4. Analyses were carried out to assess any differences in the post-decision questionnaire (T1) measures by study group (n = 106). The variables included within these analyses were grouped by the type of measure: behaviour; affect; cognitive processes; consultation quality. The variables were: the test decision; post consultation anxiety and decisional conflict; expected-utilities of the consequences of prenatal diagnosis, rank ordering of the values of the testing consequences, perception of personal risk of Down's syndrome, perceived advantages and

disadvantages of prenatal screening, perceived social norm and knowledge; perceived directiveness of information, perceived usefulness of information and consultation length.

5. Six women miscarried within four weeks after the study consultation or received a true positive diagnostic test result. These six women were not sent a follow-up questionnaire (T2). Of the remaining 100 study sample, 68 women returned a follow-up questionnaire (T2). All these women had received either a true negative diagnostic test result and/or the results of a 'normal' 19 week fetal anomaly scan. The variables used to assess the representativeness of the women completing the T2 questionnaire with those not completing it were grouped by: socio-demographic characteristics, reproductive history and individual differences; behavioural measures; measures of affect; measures of cognitive processes; measures of consultation quality. All the variables mentioned in the previous three paragraphs were assessed by completion of T2 (n = 100).
6. The variables included to evaluate the long term effect of decision analysis on the quality of decision making were classified by the type of measure (n = 68): affect; cognitive processes; clinical quality. The variables assessed at T2 by study group were: anxiety and conflict in decision making; knowledge, expected utility of the consequences of prenatal diagnosis, ranking of utilities of the consequences of prenatal diagnosis, perception of personal risk for Down's syndrome, perceived advantages and disadvantages of testing in pregnancy, and perceived social norm; usefulness of information and directiveness of information.
7. The final analysis carried out looked at the pattern of responses for measures assessed more than once (n = 68). Repeated measures analyses were carried out to evaluate the group, time and group by time associations for the following interval measures: anxiety and decisional conflict; advantages and disadvantages of prenatal testing and knowledge; usefulness of information.

6.2 Results.

6.2.1 Assessing the representativeness of the study sample.

During the fifteen-month study period at the LGI teaching hospital, 178 women received a screen positive triple test result. Of these 178 women, 28 (16%) were not invited to participate because they did not fulfil the study criteria for one of the following reasons: they were private patient; they were unable to write in English; the information giving consultation was led by a health professional other than the author; the triple test result screened positive for either Edward's syndrome or Spina Bifida. A further 18 (10%)

women were not invited to participate in the study because they made a decision to have or not have testing during the phone call informing them of their test result. These women declined the invitation to attend the clinic for further information and either made an arrangement to have a prenatal diagnostic test immediately or stated that they did not desire a diagnostic test. Of those women attending the clinic for further information all were informed of the study. Fifteen women (8%) declined to participate in the study, 117 (66%) agreed to participate and were randomised accordingly. However, during the consultation seven women (4%) identified discrepancies in the triple test result sheet and following re-calibration of their result subsequently screened negative. A further four (2%) women were excluded because they failed to complete and return the questionnaire at the end of the information consultation (Appendix XIV: table 1). The following analyses examine the representativiness of the study sample. Systematic differences for the following variables, age, reproductive and test decision, are assessed for differences between those who declined or accepted the invitation to participate.

6.2.1.1 Age, reproductive characteristics and test decision by participation.

The data extracted from the women's notes describing their age, reproductive history and test decision appear in Appendix XIV. The findings describing this population and assessing differences by participation are summarised below. Complete data pertaining to these characteristics were not available for five per cent (9/178) of the women. These nine women were private patients and their notes were not immediately accessible. The following summary refers to those women for whom there was complete information.

Sixty-four per cent of women had one or more children, 20% had had a previous miscarriage, 21% a family history of abnormality and six per cent had had a termination. The mean age of women receiving a screen positive triple test result was 35.3 years, mean gestation 14.7 weeks (Appendix XIV: table 3). Eighty per cent (135/169) of women with a screen positive result chose to have a diagnostic test. Twnety per cent (34/169) chose no further testing (Appendix XIV: table 4).

To assess the representativeness of the study sample, tests for statistical significance between the above variables were carried out using Chi square analyses for the categorical variables and a single multi-variate analysis of variance (MANOVA) for the interval measures. A single MANOVA was used to reduce the likelihood of type I errors occurring. The five levels of invitation were: exclusion because the study criteria were not fulfilled; exclusion because women made a decision over the telephone; exclusion

after invitation because the test result was actually negative or the post-decision questionnaire (T1) was not returned; declining participation; participation (Appendix XIV: table 1). One variable was found to differ significantly by level of invitation: the decision to have a diagnostic test or not (Appendix XIV: table 5 - $Chi^2 = 9.4$, d.f. = 4, p = 0.05). The data suggested that the women declining to participate were more likely to have a diagnostic test than those completing the study whereas the women making a decision over the telephone were less likely to have a diagnostic test (Appendix XIV: table 4). It was beyond the remit of this study to satisfactorily explain or explore the differences in attendance for diagnostic testing by level of invitation.

6.2.1.2 Pre-consultation anxiety by study participation.

The women invited to participate in the study (n = 132) were asked to complete a consent form which contained a pre-consultation measure of anxiety (Appendix XIV: table 6). A single analysis of variance (ANOVA) was carried out to ascertain whether there were differences in anxiety between the women agreeing to participate with those declining participation and those who were subsequently excluded from the analysis. There was no significant difference in anxiety observed following this analysis (participated, mean = 62.1 (s.d. = 13.6); excluded because re-calculated negative result, mean = 53.3 (s.d. = 14.3); excluded because not returned T1, mean = 60.7 (s.d. = 23.4); declined participation, mean = 69.9 (s.d. = 10.2), (f = 2.12, d.f. = 3, p = 0.10).

6.2.2 Comparability of women in each study group.

One hundred and thirty-two women were invited to take part in the study. Of these, fifteen women declined to participate. The remaining 117 women were allocated to either the routine or decision analysis group following the opening of sealed, numbered, opaque envelopes. Six women (five in the decision analysis group, one in the routine group) were excluded post randomisation because their triple test result was re-calculated during the consultation and found to be screen negative. These six women were subsequently classified as not fulfilling the study criteria and excluded from further analysis. Five women were excluded post randomisation (four from the decision analysis group, one from the routine group) because they did not complete the post-decision questionnaire (T1). Those women not returning the T1 questionnaire were subsequently treated as declining to take part in the study. Taking these adjustments into consideration, the study response rate was 84% (106/126: six women were excluded from these figures because their triple test result changed from positive to negative during the information giving consultation).

The majority of the women were Caucasian (103/106) (Appendix XIV: table 7). Most were married or living as married (103/106) (Appendix XIV: table 7). Few were actively religious with only 8% (9/106) frequently attending a place of worship (Appendix XIV: table 7). A quarter of the women had degree-level qualifications (27/106); just under a third (33/106) had 'A' level equivalent qualifications; the remainder had GCSE equivalent qualifications or less (46/106) (Appendix XIV: table 7). Of those who had been pregnant before (81/106), 75% (61/81) reported having had a scan in the previous pregnancy, 38% (31/81) the triple test and only 5% (4/81) a diagnostic test (Appendix XIV: table 8). Seventy-four per cent (78/106) of women attended the information giving consultation with a companion (Appendix XIV: table 9).

Of those 106 women agreeing to participate and for whom full data were available, 56 received the routine consultation information, 50 the decision analysis consultation information. The independent samples design ought to have ensured a comparable set of women in each study group. The following variables were used to assess the similarity of the profiles of the women in each study group: socio-demographic characteristics; reproductive history; prenatal testing experience; measures of individual differences; pre-consultation anxiety. The prenatal testing experience variables and the two measures of individual differences, optimism (LOT) and the need for cognition (NFC), were obtained at the end of the consultation following completion of the post-decision questionnaire (T1). Chi square analyses were used to assess differences in the categorical variables by group allocation (Appendix XIV: tables 7 - 9); a single MANOVA was used for the interval measures (Appendix XIV: table 10). There were no differences by study group for age, pre-consultation anxiety, marital status, level of education, religious activity, number of children, number of miscarriages, family history of abnormality, previous prenatal testing experience, presence of a companion and need for cognition. Two significant differences by study group were observed for gestation (Appendix XIV: table 10; $f = 6.0$, d.f. = 1, 104, $p = 0.02$) and optimism (Appendix XIV; table 10; $f = 5.2$, d.f. = 1, 104, $p = 0.03$). Those in the routine group had a greater gestation figure and a lower level of optimism than those in the decision analysis group.

Any significant difference in the profiles of the women within the two study groups has implications for the interpretation of subsequent analyses evaluating the impact of the intervention on women's decision making. For example, should subsequent analyses reveal an association between the quality of decision making variables with the study group, then it could be argued that optimism or gestation were the underlying factors of the relationship rather than the attributes of the information manipulation intervention. In

consequence, in the two following sections describing the analyses evaluating the impact of the intervention on women's decision making, preliminary correlations were carried out between gestation and optimism with the study's process and outcomes variables. Where significant associations were observed, subsequent multi-variate analyses statistically controlled for the effect of either gestation or optimism by including them within the analyses as co-variates.

6.2.3 Analysis of the information giving consultation by study group.

This section summarises the information utilised by the women when making the prenatal diagnosis decision. The verbal utterances of the women were transcribed from audio tape-recordings of the information giving consultation. Both the theme-based coding frame describing women's decision making and the SLIWC text analysis programme were applied to the data. This section describes the findings from the subsequent analyses. First, summaries of the themes classifying the information utilised, the categories coding the emotional and cognitive content and the measures of informed decision making for the complete sample are described (n = 106). Second, findings from the analyses assessing differences in informed decision making by study group are described. Finally, the results from the analyses evaluating differences by study group in the cognitive and emotional content of the verbal utterances are reported.

6.2.3.1 Describing the decision making process, the emotional and cognitive content, and the informed decision making of the information giving consultations.

As previously discussed (see chapters one, four and five), the analysis of the information utilised by women during the consultation provides one of the most direct ways to access the process of decision making. The theme-based coding frame developed for this study categorised the information utilised by women during the consultation under twenty themes (see chapter five). The theme-based coding frame was applied to all the transcripts of the consultations within this randomised control study. Table 6:1 summarises the number of women referring to each of the themes of the coding frame, as a total sample and by study group. The frequencies presented in this table illustrate the pattern of information utilised by the women during the consultation. The purpose of this chapter's analyses was to evaluate differences in informed decision making by study group. In consequence, no formal analysis was carried out to evaluate differences in cognitive strategies employed by women to assimilate the decision information. However, there follows a brief description of the

findings summarising the frequency with which the women utilised the consultation information when making the decision to have or not have prenatal diagnosis.

Table 6:1 frequency summary for the theme-based analysis of transcripts.

Theme	Total n = 106	Routine n = 56		Decision Analysis n = 50	
	%	n	%	n	%
1. No Test Option (non-invasive/scan)	***69%***	*38*	***68%***	*35*	***70%***
1.1 information seeking/planning	**53%**	26	**46%**	30	**60%**
1.2 reasons for	**29%**	17	**30%**	14	**28%**
1.3 reasons against	**43%**	23	**41%**	23	**46%**
2. Test Option combined	***100%***	*56*	***100%***	*50*	***100%***
2.1 information seeking/planning	**99%**	55	**98%**	50	**100%**
2.2 reasons for	**94%**	52	**93%**	48	**96%**
2.3 reasons against	**88%**	50	**89%**	43	**86%**
6. Down's syndrome consequence	***69%***	*37*	***66%***	*73*	***69%***
6.1 information seeking/planning	**36%**	24	**43%**	14	**28%**
6.2 neutral-positive reference	**31%**	17	**30%**	16	**32%**
6.3 negative reference	**43%**	18	**32%**	28	**56%**
7. Miscarriage consequence	***61%***	*33*	***59%***	*32*	***64%***
7.1 information seeking/planning	**52%**	29	**52%**	26	**52%**
7.2 neutral-positive reference	**17%**	6	**11%**	12	**24%**
7.3 negative reference	**18%**	6	**11%**	13	**26%**
8. Termination consequence	***81%***	*42*	***75%***	*44*	***88%***
8.1 information seeking/planning	**43%**	23	**41%**	23	**46%**
8.2 neutral-positive comment	**57%**	22	**39%**	38	**76%**
8.3 negative reference	**21%**	9	**16%**	13	**26%**
9. Risk figure reference	***77%***	*43*	***77%***	*39*	***78%***
9.1 single risk Down's syndrome	**65%**	35	**63%**	34	**68%**
9.2 single risk miscarriage	**37%**	19	**34%**	20	**40%**
9.3 comparison risks	**44%**	24	**43%**	23	**46%**
10. Perception of risk reference	***75%***	*39*	***70%***	*41*	***82%***
10.1 minimised Down's syndrome risk	**38%**	20	**36%**	20	**40%**
10.2 maximised Down's syndrome risk	**42%**	23	**41%**	22	**44%**
10.3 minimised miscarriage risk	**19%**	9	**16%**	11	**22%**
10.4 maximised miscarriage risk	**11%**	6	**11%**	6	**12%**
11. Expressed Affect					
11.1 triple test: shock, worry	**44%**	22	**39%**	25	**50%**
11.2 diagnostic test: hard, conflict	**43%**	23	**41%**	23	**46%**
11.3 termination: hard, conflict	**33%**	13	**23%**	22	**44%**
11.4 testing reassurance	**6%**	2	**4%**	4	**8%**
11.5 anticipated regret	**19%**	11	**20%**	9	**18%**

Table 6:1 continued . . .

Theme	Total n = 106	Routine n = 56		Decision Analysis n = 50	
	%	n	%	n	%
12. Decision Making					
12.1 triple test	**26%**	13	**23%**	15	**30%**
12.2 test decision made before consultation	**38%**	22	**39%**	18	**36%**
12.3 each prenatal testing stage a different decision/termination decision not made	**29%**	16	**29%**	15	**30%**
12.4 delay in making decision	**32%**	18	**32%**	16	**32%**
12.5 confidence with decision	**23%**	10	**18%**	14	**28%**
12.6 discussed decision with other	**42%**	22	**39%**	23	**46%**
13. Triple Test: further explanation	***83%***	*44*	***79%***	*44*	***88%***
14. Confidence with Screening Results	***37%***	*23*	***41%***	*39*	***37%***
15. Comparison Norms	***36%***	*21*	***38%***	*17*	***34%***
16. Personal Experiences	***67%***	*37*	***67%***	*34*	***68%***
16.1 physical aspects pregnancy	**34%**	18	**32%**	18	**36%**
16.2 complications in pregnancy	**24%**	13	**23%**	13	**26%**
16.3 prior testing and pregnancies	**39%**	19	**34%**	22	**44%**
16.4 abnormalities or genetics	**18%**	10	**18%**	9	**18%**
17. Others Experience	***45%***	*25*	***45%***	*23*	***46%***
17.1 prenatal testing	**36%**	20	**36%**	18	**36%**
17.2 abnormalities or genetics	**20%**	9	**16%**	12	**24%**
18. Preparation Pregnancy	***15%***	*10*	***18%***	*6*	***12%***
19. Health Professionals' Advice					
19.1 neutral or positive comments	**34%**	20	**36%**	16	**32%**
19.2 dissatisfaction or directive	**27%**	13	**23%**	16	**32%**
19.3 inaccurate or conflicting consultation	**8%**	8	**14%**	1	**2%**
20. Other sources information	***35%***	*21*	***37%***	*16*	***32%***

The pattern of information utilised by the women during this randomised control trial showed some similarities to the pattern of responses in chapter five. All the women referred to information about the 'test option' during the consultation (theme 2, table 6:1) but not all the women (69%) referred to items classified under the 'no test' option (theme 1, table 6:1). In addition, fewer of the women utilised information about Down's syndrome (69%) and miscarriage (61%) than termination of pregnancy (81%) (themes 6, 7 and 8, table 6:1). About a third of the women commented on various aspects of the prenatal testing decision making process (themes 12.1 - 12.6, table 6:1). Most of the women (83%) wanted further information about the triple test result (theme 13, table

6:1). Forty-four percent (theme 11.1, table 6:1) mentioned the screen positive test result was a shock and about the same number referred to difficulty in making the diagnostic test decision (theme 11.2, table 6:1). About a third expressed some concern over the reliability of the triple test result (theme 14, table 6:1) and about the same proportion tried to compare their own situation with that of a 'norm' (theme 16, table 6:1). Sixty-seven percent of women reflected on their own prenatal experiences (theme 16, table 6:1) and 45% on others prenatal experiences (theme 17, table 6:1). About a third referred to other health professionals' advice (themes 19.1 - 19.3, table 6:1) and other sources of information (theme 20, table 6:1) during the consultation.

In summary, this pattern of utilised information suggests that most of the women referred to information about the alternatives and consequences necessary to make an informed decision during the consultation. In addition, a significant number of the women referred to information associated with more heuristic type processes, such as reference to prenatal testing experiences, others' advice and comparison with social norms. It was also evident that at least half of the women found the prenatal testing experience emotionally difficult. In essence, the pattern of findings described in table 6:1 are similar to those reported in table 5:1.

6.2.3.2 Differences in informed decision making by study group.

Three informed decision making scores were calculated from the application of the theme-based coding frame to the consultation transcripts: information seeking or planning for a consequence score (IMD-I: range 0 - 5); reasons for or favourable comments about a consequence score (IMD-F: range 0 - 5); reasons against or unfavourable comments about a consequence score (IMD-A: range 0 - 5). As these three variables provided an efficient summary of the informed decision making references by the women during the consultation, they were used in the analysis to ascertain whether or not decision analysis facilitated informed decision making. A preliminary correlation was carried out between the informed decision making variables with the two profile characteristics of the women found to differ significantly by study group allocation, gestation and optimism (see section 6.2.1). Only one of the profile characteristics, gestation, was found to be significantly associated with one of the informed decision making variables, IDM-F (table 6:2). In consequence, gestation was included in subsequent analysis as a co-variate to adjust for the effect between gestation and the informed decision making variables.

Table 6:2 correlation matrix for gestation and optimism with IDM measures.

	IDM-I		IDM-F		IDM-A	
	Pearson R	**Sig.**	Pearson R	**Sig.**	Pearson R	**Sig.**
gestation	0.005	**0.96**	-0.19	**0.05**	-0.06	**0.51**
optimism	-0.04	**-0.68**	-0.13	**0.20**	0.06	**0.56**

A multiple analysis of co-variance (MANCOVA) was carried out to assess the relationship between the informed decision making variables and the information intervention. A single equation was used in an attempt to decrease the likelihood of type I errors occurring. The Bartlett-box M criteria for homogeneity of variance for the dependent variables was not significant, suggesting no threat to the interpretation of the main effect results (Box's M = 5.8, Chi ^{2} = 5.6, p = 0.47). Main effects were observed for IDM-F and IDM-A but not for IDM-I (table 6:3). In essence, the women in the decision analysis group referred to more reasons for and against alternatives during the consultation than the women in the routine consultation. No difference in information seeking about alternatives was observed by the women in the two study groups. These findings suggest that reasoning rather than information seeking was greater in the decision analysis consultation.

Table 6:3 informed decision making variables by study group.

	Total n = 106		**Routine** n = 56		**Decision Analysis** n = 50		**MANCOVA** d.f. (1,103)	
	x	(s.d)	**x**	(s.d)	**x**	(s.d)	f	**Sig.**
informed decision making - **information seeking (IDM-I)**	**2.8**	(1.1)	**2.8**	(1.1)	**2.9**	(1.1)	0.08	**0.78**
informed decision making - **reasons for option (IDM-F)**	**2.3**	(1.0)	**2.1**	(1.0)	**2.6**	(1.0)	5.24	**0.02**
informed decision making - **reasons against option (IDM-A)**	**2.1**	(1.1)	**1.9**	(0.9)	**2.4**	(1.2)	5.33	**0.02**

6.2.3.3 Differences in emotional and cognitive content by study group.

The five SLIWC ways of classifying the content of consultations used in this analysis were: the number of words within the consultation; the percentage of positive emotion-laden words; the percentage of negative emotion-laden words; the percentage of cognitive mechanisms or 'thinking' words; the percentage of social processes or 'communication with others' words. As the purpose of the analysis was to ascertain

differences in the content of the consultation by study groups, preliminary correlations were carried out between the SLIWC variables and the two profile characteristics found to differ significantly by group allocation, gestation and optimism. In addition to these two profile characteristics, it was considered appropriate to include the consultation length in a preliminary correlation. The previous text analysis (theme-based informed decision making) counted the number of women referring to a theme, whereas the SLIWC analysis counts the number of utterances made by a woman during the consultation. It was feasible to suggest that the length of consultation would be associated with the number of utterances recorded.

Table 6:4 correlations between gestation, LOT and consultation length with the SLIWC variables.

	word count		positive emotion		negative emotion		cognitive mechanism		social processes	
	R	Sig.	R	Sig.	R	Sig.	R	Sig.	R	Sig.
gestation	0.12	**0.20**	-0.05	**0.60**	0.04	**0.70**	-0.15	**0.13**	-0.08	**0.41**
optimism	0.17	**0.08**	0.10	**0.31**	0.06	**0.54**	-0.01	**0.94**	0.19	**0.05**
consultation length	0.81	**<0.001**	-0.09	**0.93**	0.15	**0.13**	0.12	**0.21**	0.32	**0.001**

Eleven per cent of the words mentioned during the consultation were associated with cognitive mechanisms or 'thinking', 9.5% with social processes such as communicating with others and about 3% with affect-laden words. Only one of the profile characteristic, optimism (LOT), was associated with one of the SLIWC variables, social processes (table 6:4). The length of consultation was significantly correlated with both the number of words mentioned during the consultation and the percentage of words about social processes (table 6:4). In consequence, optimism and consultation length were included in subsequent analysis as co-variates to adjust for the effect between optimism and consultation length with the SLIWC variables.

A MANCOVA was used to evaluate the relationship between group allocation and the five SLIWC variables with length of consultation and the LOT score as co-variates. The Bartlett-box M criteria for homogeneity of variance for the dependent variables was not significant (Box's M = 23.3, Chi^2 = 22.1, p = 0.10). After controlling for the effects of consultation length and differences in optimism, two main effects were observed for differences in the percentage of positive emotion-laden words used and the percentage of cognitive mechanisms or thinking words used by study group (table 6:5). The women

in the decision analysis group were less likely to use language associated with positive emotions such as 'good' or 'happy' and more likely to use words associated with cognitive processes such as 'think' and 'cause'. There was also a trend towards significance, suggesting that the women in the decision analysis group were more likely to use negative emotion-laden words such as 'sad' or 'tense (p = 0.09, table 6:5). There was no difference in the number of words uttered or references to social processes by study group.

Table 6:5 SLIWC text analysis variables by study group.

	Total n = 106		Routine n = 56		Decision Analysis n = 50		MANCOVA d.f. (1,102)	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	Sig.
word count	**1117.8**	(917.6)	**966.2**	(785.8)	**1287.5**	(1027.4)	0.12	**0.73**
positive emotion	**1.9**	(0.7)	**2.0**	(0.8)	**1.8**	(0.6)	4.7	**0.03**
negative emotion	**0.9**	(0.5)	**0.8**	(0.4)	**1.0**	(0.5)	2.8	**0.09**
cognitive mechanism	**11.3**	(1.7)	**10.8**	(1.8)	**11.8**	(1.5)	6.5	**0.01**
social processes	**9.5**	(2.2)	**9.5**	(2.1)	**9.5**	(2.2)	1.0	**0.33**

6.2.4 Analysis of post-decision questionnaire (T1) by study group.

The first part of this results section provides a summary of the women's responses to the post-decision questionnaire (T1) as a complete sample. The results of the analysis assessing differences in post-decision questionnaire measures by study group are described in the second part of this section. The measures included within the T1 questionnaire fall into one of four categories: behaviour; affect; cognitive processes; clinical quality. The findings in each of the two sections are grouped and reported by category of measure. For example, all the cognitive process variables with their different levels of measurement and different types of analyses will be described together.

6.2.4.1 Descriptions of behaviour, affect, cognition and clinical quality measures.

The following descriptions are verbal summaries of all the women's responses to items asked within the questionnaire (T1). The raw data supporting these summaries are reported in the tables presented in the next section, i.e. the discussion of differences in measures by study group.

Behaviour: Deciding whether to have a diagnostic test or not was the behavioural measure for this study. 84% (89/106) of women decided to have a diagnostic test; 16% chose to have no diagnostic test. Of those women choosing to have a diagnostic test, 12 opted to have chorionic villus sampling and 77 amniocentesis.

Affect: Anxiety and decisional conflict were the measures of affect for this study. The sample mean for anxiety was high at 60.1 (s.d. = 14.8) points on the STAI scale (range 20 - 80). The mean values for the three decisional conflict sub-category scores were: uncertainty with the decision made, 8.4 (s.d. = 2.8); perception made an informed decision, 6.8 (s.d. = 1.7); perceived efficacy in decision making, 7.5 (s.d. = 2.2).

Cognitive process: Measures of the cognitive processes in this study number were: the perceived advantages and disadvantages of testing; the strength of advantages and disadvantages of testing; the perception of the triple test risk; the perceived social norm for testing behaviour; the expected-utility values associated with the consequences of prenatal testing; the ability to rank order the consequences of testing; knowledge of the tests.

Number of perceived advantages and disadvantages of prenatal testing: 12% of the women listed no advantages; 27% listed one advantage; 46% listed two advantages; 15% three advantages to prenatal testing. 18% of women listed no disadvantages; 30% one advantage; 35% two advantages; 17% three advantages to prenatal testing. The advantages and disadvantages listed by the women are described in table 6:6. The frequency scores do not total 100 as the women were able to identify more than one advantage or disadvantage.

Table 6:6 the number of women identifying advantages and disadvantages of prenatal testing.

Number of women stating an advantage			Number of women stating a disadvantage		
	n	%		n	%
check baby's health, OK	51	**48%**	worry, stress, waiting for results	49	**46%**
informed decision making	39	**37%**	miscarriage	48	**45%**
find abnormality	29	**27%**	making difficult decisions	29	**27%**
certainty	17	**16%**	testing unreliable	15	**14%**
discuss Down's syndrome	10	**9%**			
other (sex, scan, mother's health)	18	**17%**	other (pain, late gestation)	11	**10%**

Strength of advantages and disadvantages of prenatal testing: 87 of the women provided both at least one advantage and disadvantage of testing and evaluated the strength of that advantage or disadvantage on a five-point likert scale. The mean strength of advantages was 4.5 (s.d. = 0.9) and of disadvantages was 3.8 (s.d. = 1.2). The perceived advantages of prenatal testing were significantly stronger than the perceived disadvantages of testing (paired t-test: $t=4.6$, d.f. = 86, $p < 0.0001$). As not all women in the sample completed this question, this measure was not included in subsequent multi-variate analyses assessing differences in cognitive processes by study group.

Perception of triple test risk: 50% of women rated their triple test risk as high; 43% rated the risk as medium and 12% rated their risk as a low risk.

Perceived social norm for testing behaviour: The responses to this single item assessing women's perception of other's attitude to prenatal testing were skewed to the 'should have testing' end of the scale. The mean score was 1.5 (s.d. = 1.6) on a scale from 0 - 6.

Expected-utility values associated with the consequences of prenatal testing: The mean values for the expected-utility values (EUV) of the five consequences of testing listed in order were:

having a healthy baby, mean 39.5 (s.d = 10.9);
terminating a baby with Down's syndrome, mean 28.7 (s.d.= 21.7);
miscarrying a baby with Down's syndrome, mean 13.0 (s.d. = 11.1);
having a baby with Down's syndrome, mean = 3.5 (s.d. = 6.6);
miscarrying a healthy baby, mean 1.2 (s.d. = 2.1).

Ability to rank order the consequences of testing: Although the means of the whole sample's EUVs suggest a pattern to the women's evaluation of the consequences of testing, most of the women (55%) were unable to rank order the consequences of prenatal testing so that each consequence was perceived as better or worse than the other four consequences.

Knowledge of the tests: 10% of women scored less than half of the correct responses; 54% scored between half and three-quarters of the correct responses and 36% scored more than three-quarters of the correct responses on the multiple-choice knowledge questions.

Clinical quality: Consultation length, usefulness of information and perceived directiveness of study (HB) and other health professionals' information were the measures of clinical quality. The mean length of consultations was 29.0 (s.d. = 12.2) minutes; the information was perceived to be useful, the mean being 5.2 (s.d. = 1.1) on a scale from 0-6. 8% of the information provided by both the study and other health professionals was perceived to be 'encouraging'.

6.2.4.2 Assessing differences in behavioural, affect, cognitive processes and clinical quality by study group.

This section of the results describes the analyses that evaluated the impact of the decision analysis consultation on the quality of women's decision making. Preliminary correlations were carried out between the two profile characteristics found to differ significantly by study group allocation, gestation and optimism, with all the interval measures of decision quality. Optimism was significantly associated with two of the decision quality measures, EUV of having a healthy baby and knowledge (table 6:7). In consequence, optimism was included in subsequent analysis as a co-variate to adjust for the effect of optimism with the measures of decision quality.

Table 6:7 correlations between gestation and optimism with decision quality measures.

	Gestation n = 106		**Optimism (LOT)** n = 106	
	R	**Sig.**	R	**Sig.**
anxiety after consultation	0.12	**0.22**	0.23	**0.02**
conflict - uncertainty	-0.002	**0.98**	0.08	**0.44**
conflict - informed	0.09	**0.37**	0.13	**0.17**
conflict - efficacy	0.14	**0.14**	0.10	**0.33**
EUT1 - healthy baby	-0.16	**0.09**	-0.26	**0.007**
EUT2 - misc. Down's baby	0.06	**0.55**	0.09	**0.34**
EUT3 - term. Down's baby	-0.03	**0.80**	-0.12	**0.21**
EUT4 - have Down's baby	-0.11	**0.26**	0.08	**0.40**
EUT5 - misc. normal baby	-0.004	**0.97**	-0.04	**0.65**
advantages of testing	0.08	**0.40**	-0.16	**0.11**
disadvantages of testing	-0.005	**0.96**	-0.18	**0.06**
knowledge	-0.09	**0.37**	-0.20	**0.04**
length consultation	0.04	**0.67**	0.11	**0.24**
usefulness information	0.03	**0.73**	-0.03	**0.80**

The following analyses were employed to evaluate the information intervention: all the interval measures were entered into a single MANCOVA to reduce the likelihood of type I errors; Mann-Whitney tests of significance were used for interval measures not

normally distributed; Chi square analyses were employed for categorical variables. The variables included within the MANCOVA were: post-decision anxiety; decisional conflict; number of advantages and disadvantages of prenatal testing; knowledge; consultation length; usefulness of information; the co-variate optimism. Robustness of findings is not guaranteed if the Bartlett-box M test for homogeneity of variance is significant at $p < 0.001$ (Tabachnick and Fidell, 1989). For this analysis, the Bartlett-box M was not sufficiently significant to affect the interpretation of findings ($M = 72.1$, $Chi^2 = 65.5$, d.f. = 45, $p = 0.02$). The distributions of six interval variables were not normally distributed, all the EUVs for the consequences of prenatal testing and the perceived social norm item. Differences by study group for all but one of the EUV variables, termination for Down's syndrome, and the perceived social norm were assessed following the application of a Mann-Whitney analysis. The differences by study group for the EUV for termination for Down's syndrome, the perception of triple test risk, the ability to rank order the consequences of prenatal testing and the perceived encouragement of the study and other health professionals' advice were assessed using Chi square analyses. The findings are grouped by type of measure: behaviour; affect; cognitive processes; clinical quality.

Behaviour: There was no significant difference in the testing decision or behaviour by study group. Fourteen per cent of women in the decision analysis group and eighteen per cent of women in the routine group chose to have no further testing (table 6:8).

Table 6:8 to have or not to have testing by study group.

	Total (n = 106)		Routine (n = 56)		Decision Analysis (n = 50)		Chi ^{2} Sig.	
	n	%	n	%	n	%	d.f.	(1)
no test	17	**16%**	10	**18%**	7	**14%**		
test	89	**84%**	46	**82%**	43	**86%**	0.29	**0.59**

Affect: there was no significant main effect observed by group allocation with either anxiety or the three decisional conflict sub-categories (table 6:9).

Table 6:9 differences in measures of affect by study group.

	Total n = 106		Routine n = 56		Decision Analysis n = 50		MANCOVA (1, 103)	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	Sig.
T1 anxiety	**60.1**	(14.8)	**61.2**	(13.4)	**58.9**	(16.3)	0.09	**0.77**
conflict - uncertainty	**8.4**	(2.8)	**8.4**	(3.0)	**8.4**	(2.6)	0.01	**0.91**
conflict - informed	**6.8**	(1.7)	**6.8**	(1.7)	**6.9**	(1.7)	0.51	**0.48**
conflict - efficacy	**7.5**	(2.2)	**7.5**	(2.3)	**7.6**	(2.2)	0.29	**0.60**

Cognitive processes: The mean number of perceived advantages of prenatal testing differed by group allocation (table 6:10). The women in the routine consultation group listed more advantages of prenatal testing than those in the decision analysis group. There was no difference in the number of disadvantages listed. The mean number of correct responses to the multiple-choice questions for knowledge of prenatal testing did not differ by study group allocation (table 6:10).

Table 6:10 differences in advantages, disadvantages and knowledge of prenatal testing by study group.

	Total (n = 106)		**Routine** (n = 56)		**Decision Analysis** (n = 50)		**MANCOVA** (d.f. = 1, 103)	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	**Sig.**
advantages testing	**1.6**	(0.9)	**1.8**	(0.9)	**1.4**	(0.8)	8.88	**0.004**
disadvantages testing	**1.5**	(1.0)	**1.6**	(1.0)	**1.5**	(1.0)	0.87	**0.35**
knowledge	**14.5**	(3.0)	**14.3**	(3.2)	**14.8**	(2.9)	0.19	**0.66**

There was no difference in the women's perception of their triple test risk by study group allocation (table 6:11).

Table 6:11 perception of triple test risk by study group.

	Total (n = 106)		**Routine** (n = 56)		**Decision Analysis** (n = 50)		**Chi^2** (d.f. = 2)	
	n	%	n	%	n	%	Chi^2	**Sig.**
low	13	**12%**	7	**13%**	6	**12%**		
medium	43	**43%**	18	**32%**	25	**50%**		
high	50	**50%**	31	**55%**	19	**38%**	3.77	**0.15**

Table 6:12 difference in EUV of terminating for Down's syndrome by study group.

	Total (n = 106)		**Routine** (n = 56)		**Decision Analysis** (n = 50)		**Chi^2** d.f. = 1	
	n	%	n	%	n	%	R	Sig.
EUT value <30	58	**55%**	38	**68%**	20	**40%**		
EUT value >29	48	**48%**	18	**32%**	30	**60%**	8.3	0.004

Of the EUV for the consequences of prenatal diagnosis, the figure associated with terminating for a child with Down's syndrome differed by study group allocation (tables

6:12 and 6:13). The women in the decision analysis consultation provided a higher expected-utility value for terminating a child with Down's syndrome. There was no difference in perceived social norm by study group allocation (table 6:13).

Table 6:13 difference in EUVs for the testing consequences and perceived social norm by study group.

	Total n = 106		Routine n = 56		Decision Analysis n = 50		Mann-Whitney		
	x	(s.d.)	x	(s.d.)	x	(s.d.)	U	z	Sig.
EUT1 -normal baby	**39.5**	(10.9)	**38.1**	(10.2)	**41.0**	(11.5)	1253.0	-0.9	**0.35**
EUT2 - misc. DS	**13.0**	(11.1)	**12.5**	(10.7)	**13.5**	(11.6)	1330.0	-0.4	**0.66**
EUT3 - term. DS	**28.7**	(21.7)	**24.1**	(21.2)	**33.8**	(21.4)	NA	NA	**NA***
EUT4 - baby DS	**3.5**	(6.6)	**3.7**	(6.1)	**3.3**	(7.2)	1374.0	-0.2	**0.87**
EUT5 - misc. baby	**1.2**	(2.1)	**1.1**	(2.1)	**1.3**	(2.2)	1324.0	-0.5	**0.63**
Should have test	**1.5**	(1.6)	**1.6**	(1.4)	**1.4**	(1.6)	1306.0	-0.6	**0.54**

**NA = no analysis because bi-modal distribution*

There was no difference in the ability to rank order the consequences of prenatal testing by study group allocation (table 6:14).

Table 6:14 differences in the ability to rank order the EUVs associated with the consequences of prenatal testing by study group.

	Total n = 106		Routine n = 50		Decision Analysis n = 56		Chi ^{2} d.f. (1)	
	n	%	n	%	n	%		
ranked	48	**45%**	23	**41%**	25	**50%**	0.85	**0.36**
not ranked	58	**55%**	33	**59%**	25	**50%**		

Table 6:15 differences in consultation length and perceived usefulness of information by study group.

	Total (n = 106)		Routine (n = 56)		Decision Analysis (n = 50)		MANCOVA (d.f. = 1, 103)	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	Sig.
consultation length	**29.0**	(12.2)	**26.2**	(11.2)	**32.2**	(12.6)	8.67	**0.004**
usefulness information	**5.2**	(1.1)	**5.4**	(1.0)	**4.9**	(1.2)	4.4	**0.04**

Clinical quality: Differences in consultation length and perceived usefulness of information were observed by study group allocation (table 6:15). The decision analysis consultation was significantly longer than the routine consultation. In addition, although both study groups rated the consultation information to be useful, the women in the

decision analysis group perceived the usefulness of the information to be lower than of those in the routine information group.

There was no difference in the perceived encouragement of information provided by either the study or other health professionals by study group allocation (tables 6:16 and 6:17). Most of the women in both groups perceived the provided information to be neutral or non-directive.

Table 6:16 perceived encouragement of health professional information by study group.

	Total (n = 106)		Routine (n = 56)		Decision Analysis (n = 50)		Chi ^{2} (d.f. = 2)	
	n	%	n	%	n	%		Sig.
not discussed	10	9%	5	9%	5	10%		
neutral	88	83%	48	86%	40	80%		
directive	8	8%	3	5%	5	10%	0.89	0.64

Table 6:17 perceived encouragement of other health professional information by study group.

	Total (n = 106)		Routine (n = 56)		Decision Analysis (n = 50)		Chi ^{2} (d.f. = 2)	
	n	%	n	%	n	%		Sig.
not discussed	54	51%	28	50%	26	52%		
neutral	43	41%	24	43%	19	38%		
encouraged	9	8%	4	7%	5	10%	0.43	0.81

6.2.5 Analysis of follow-up questionnaire (T2) by study group.

Six of the women were not sent follow-up questionnaires because they had received either a positive diagnostic test result or had miscarried the pregnancy in the intervening weeks (table 6:18).

Table 6:18 known number of miscarriages and Down's syndrome diagnoses by study group.

	Total (n = 106)		Routine (n = 56)		Decision Analysis (n = 50)	
	n	%	n	%	n	%
negative test result or no test	100	95%	52	93%	48	96%
positive test result	4	3%	3	5%	1	2%
miscarriage of pregnancy	2	2%	1	2%	1	2%

Of these six women, four received a true positive diagnostic test result. Three women with a diagnosis of Down's syndrome terminated the pregnancy, whilst one carried on with the pregnancy to term. The notes of the women with the diagnosis of Down's syndrome did not record a family history of abnormality, although one woman had previously miscarried and was being treated for infertility. The two women not sent a T2 questionnaire because the pregnancy had miscarried, had both had the amniocentesis test. One woman who miscarried did not record any predisposition to miscarriage or a family history of abnormality or Down's syndrome but did refer to a cousin with Down's syndrome during the consultation. The second woman who miscarried indicated one previous miscarriage, one child with a disability and, during the consultation, mentioned that she had a kidney disease which suggested that the pregnancy would not reach term.

In total, a hundred women did not receive a positive diagnostic test result, did not miscarry and had attended the clinic for their nineteen week fetal anomaly scan. These women were sent a follow-up questionnaire (T2) at about twenty weeks gestation. Follow-up questionnaires (T2) were returned by 68% (68/100) of the women.

Three sets of analyses are described below. The first explored any systematic differences in the responses to the post-decision questionnaire (T1) measures by those completing (T1&T2) or not completing (T1 only) the follow-up questionnaire (T2). The second set evaluated any long-term differences in the quality of decision measures by group allocation. The final analyses assessed the changes in the quality of decision measures over time.

6.2.5.1 Differences in women's consultation and post-decision questionnaire (T1) responses by completion of the follow-up questionnaire (T2).

The following section summarises the results of the analyses carried out to compare the profile characteristics, consultation information and T1 responses of the women who either did or did not complete the follow-up questionnaire (T2). The aim of these analyses was to establish the representativeness of the women that completed the follow-up questionnaire compared with those that only completed T1. The data and results of the analyses are reported in Appendix XV; the following text provides a verbal description of the findings. The same types of analyses were used for these comparisons as previously described for differences by group allocation: the five SLIWC items were entered into a MANCOVA with consultation length as the co-variate,

the Bartlett-box M test for homogeneity of variance was borderline suggesting that the results should be interpreted with caution (M = 41.0, Chi^2 = 38.3, d.f. = 15, p = 0.001); the remaining interval variables with normal distributions were entered into a MANOVA to reduce the likelihood of type I errors occurring, the Bartlett-box M test for homogeneity of variance was satisfactory (M = 211.7, Chi^2 = 168.8, d.f. = 136, p = 0.03); the interval variables with skewed distributions resistant to data transformations were entered into Mann-Whitney analyses; the categorical variables were subjected to Chi square analysis. The results are discussed in the following order: group allocation; socio-demographic characteristics, reproductive history and individual differences; behaviour; affect; cognitive processes; clinical quality.

Group allocation: The return rate of T2 questionnaires was 75% of the women in the routine consultation and 60% of those in the decision analysis consultation. Although fewer women in the decision analysis group returned their T2 questionnaires than the 'routine' women, these percentages were not significantly different (table 1: Appendix XV).

Socio-demographic characteristics, reproductive history and individual differences: There were no differences for the age, gestation, number of children, number of miscarriages, religious activity and need for cognition between those who did or did not return the T2 questionnaire (tables 2 and 3: Appendix XV). Significant differences were observed by completion of T2 for: family history of abnormality (p = 0.04; table 2: Appendix XV); level of education attained (p = 0.008; table 2: Appendix XV); optimism (p = 0.04; table 3: Appendix XV). The women were more likely to return the T2 questionnaire if they did not have a family history of an abnormality, had attained a higher than GCSE level of education and had an optimistic trait.

Behaviour: There was no difference in the return rate of T2 by the women's decision to have a diagnostic test or not (table 4: Appendix XV).

Affect: There were no differences in the pre-consultation measure of anxiety, expressed affect during the consultation (SLIWC), post-decision measure of anxiety and measures of decisional conflict between those who did and did not return the follow-up questionnaire (tables 5 and 6: Appendix XV).

Cognitive processes: No differences by T2 return rate were observed for any of the cognitive measures: informed decision making and remaining SLIWC variables

extracted from the consultation transcripts (tables 5 and 6: Appendix XV); perceived advantages and disadvantages of testing (table 6: Appendix XV); knowledge (table 6: Appendix XV); EUVs of prenatal testing consequences (table 7 and 8: Appendix XV); ability to rank order the prenatal testing consequences (table 9: Appendix XV); perception of their triple test risk (table 10: Appendix XV).

Clinical quality: No differences by T2 return rate were observed for usefulness of information (table 6: Appendix XV) and perceived encouragement of other health professionals' advice (table 11: Appendix XV). However, those that returned the T2 questionnaire had had shorter consultations ($p = 0.05$; table 6: Appendix XV) and perceived the information provided by the study health professional to be more neutral ($p = 0.03$; table 12: Appendix XV) than those that did not return the T2 questionnaire.

The differences observed for the profile characteristics and T1 measures between women who did and did not complete the T2 questionnaire may affect the interpretation of subsequent analyses assessing the association between study group allocation with T2 measures of decision and clinical quality. Preliminary correlations were carried out between the T2 measures with the five variables found to differ significantly by T2 return rate: level of education; family history of abnormality; optimism; perceived directiveness of the study health professional; length of consultation (tables 13 and 14: Appendix XV). In addition, preliminary correlations were carried out between the remaining profile characteristic found to differ by group allocation, gestation, with the T2 measures. As associations were observed between optimism, level of education and length of consultation with some of the T2 measures, these three variables were incorporated in subsequent analyses as co-variates to adjust for their effect on T2 measures.

6.2.5.2 Differences in follow-up questionnaire (T2) by study group.

This section describes the findings of analyses carried out to evaluate differences in the women's responses four to five weeks after the information giving consultation. All 68 women completing a follow-up questionnaire (T2) had received 'good' news either upon receipt of a negative *diagnostic* result and/or following a 'normal' nineteen week fetal anomaly scan. The measures of affect, cognitive processes and clinical quality repeated in the T2 questionnaire were: measures of anxiety and decisional conflict; perceived advantages and disadvantages of testing, expected utility values for the consequences of testing, ability to rank order the values associated with the consequences of testing, perception of the triple test risk and knowledge; usefulness and perceived directiveness

of information. Differences by study group were assessed using the following analyses: all the interval measures with normal distributions were entered into a MANCOVA to reduce the likelihood of type I errors with optimism, consultation length and level of education as the co-variates (Bartlett-box M = 41.7, Chi^2 = 36.2, d.f. = 36, p = 0.46); interval measures with skewed distributions resistant to data transformations were analysed using a Mann-Whitney test for independent groups; Chi square tests of significance were applied to the categorical variables. The findings described below are grouped by type of measure.

Table 6:19 differences in anxiety and decisional conflict by study group (T2).

	Total n = 68		Routine n = 39		Decision Analysis n = 29		MANCOVA d.f. = 1,63	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	Sig.
anxiety	**34.9**	(13.3)	**34.7**	(14.5)	**35.3**	(11.8)	1.2	**0.28**
conflict - uncertainty	**7.5**	(3.2)	**8.1**	(3.5)	**6.6**	(2.7)	6.8	**0.01**
conflict - informed	**7.0**	(2.1)	**7.5**	(2.2)	**6.4**	(1.8)	2.8	**0.10**
conflict - efficacy	**7.8**	(2.7)	**8.6**	(2.9)	**6.8**	(2.1)	6.2	**0.02**

Affect: There was no difference in anxiety by study group allocation at follow-up (T2). However, differences in two of the three sub-categories of decisional conflict were observed (table 6:19). Women in the decision analysis group were more certain of their decision and had greater decisional efficacy at follow-up than those in the routine group.

Cognitive processes: No differences by study group allocation were observed for the following measures of cognitive processes: perceived advantages and disadvantages of prenatal testing; knowledge about testing; perception of triple test risk; EUVs associated with having a healthy baby, miscarrying a healthy baby, having a baby with Down's syndrome and miscarrying a baby with Down's syndrome; the ability to rank order the consequences of prenatal testing (tables 20-23).

Table 6:20 knowledge, perceived advantages and disadvantages of testing by study group (T2).

	Total n = 68		Routine n = 39		Decision Analysis n = 29		MANCOVA d.f. = 1,63	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	Sig.
advantages testing	**1.9**	(1.0)	**2.1**	(1.0)	**1.7**	(0.9)	2.8	**0.10**
disadvantages testing	**1.6**	(1.0)	**1.7**	(1.1)	**1.4**	(0.9)	1.9	**0.17**
knowledge	**13.7**	(2.8)	**13.5**	(2.8)	**13.8**	(2.7)	0.1	**0.73**

Table 6:21 perception of triple test risk by study group (T2).

	Total (n = 68)		Routine (n = 39)		Decision Analysis (n = 29)		Chi^2 (d.f. = 2)	
	n	%	n	%	n	%	Chi^2	Sig.
low risk	5	7%	3	8%	2	7%		
medium risk	27	40%	14	36%	13	45%		
high risk	36	53%	22	56%	14	48%	0.56	0.76

Table 6:22 the ability to rank order the consequences of testing by study group (T2).

	Total (n = 68)		Routine (n = 39)		Decision Analysis (n = 29)		Chi^2 (d.f. = 1)	
	n	%	n	%	n	%	Chi^2	Sig.
ranked	38	56%	21	54%	17	59%		
not ranked	30	44%	18	46%	12	41%	0.15	0.70

Table 6:23 prenatal consequences and perception of social norm by study group (T2).

	Total (n = 68)		Routine (n = 39)		Decision Anal. (n = 29)		Mann-Whitney		
	x	(s.d.)	x	(s.d.)	x	(s.d.)	U	z	Sig.
EUT1- have healthy baby	**44.9**	(11.6)	**45.2**	(10.9)	**44.4**	(12.6)	557	-0.1	**0.9**
EUT2- misc. Downs baby	**4.9**	(8.2)	**5.7**	(8.9)	**3.8**	(7.1)	497	-0.9	**0.4**
EUT3- term. Downs baby	**19.9**	(20.3)	**12.7**	(15.5)	**29.6**	(22.1)	NA	NA	**NA***
EUT4- have Downs baby	**1.8**	(3.8)	**1.7**	(3.6)	**1.8**	(4.1)	524	-0.6	**0.6**
EUT5- misc. healthy baby	**0.8**	(1.4)	**0.9**	(1.6)	**0.7**	(1.1)	565	0.0	**1.0**
Perception norm (should)	**1.6**	(1.1)	**2.0**	(1.6)	**1.1**	(1.3)	384	-2.3	**0.02**

** NA = no analysis carried out, bi-modal distribution*

Table 6:24 EUV for terminating for Down's syndrome by study group (T2).

	Total (n = 68)		Routine (n = 39)		Decision Analysis (n = 29)		Chi^2 (d.f. = 1)	
	n	%	n	%	n	%	Chi^2	Sig.
EUT value <14	35	52%	26	67%	9	31%		
EUT value >13	33	49%	13	33%	20	69%	8.5	0.003

Two significant differences by group allocation were observed: the perception of social norms about prenatal testing (p = 0.02; table 6:23); the EUV associated with terminating a child with Down's syndrome (p = 0.003; table 6:24). The women in the decision analysis group were more likely to perceive other's attitude to be in favour of prenatal

testing and to have a greater expected-utility value associated with terminating for Down's syndrome than the women in the routine group. The mean figures by study group for perceived social norm at T1 were: routine group 1.6 (s.d. = 1.4); decision analysis group 1.4 (1.6). It is feasible to suggest that, in retrospect, women in the decision analysis group were more certain and women in the routine group less certain about their perception of the social norm regarding prenatal diagnosis. A similar pattern was observed for the EUV measure: women in the decision analysis group had a higher EUV score regarding termination than women in the routine group. However, the median split for this variable was a lower figure at T2 (13) than at T1 (29). One explanation for this finding is that women evaluated the likelihood of these consequences occurring lower upon receipt of the 'good news' than at T1.

Clinical quality: No significant differences were observed for measures of decision quality by study group allocation (tables 6:25, 6:26 and 6:27). The finding that there was no difference in perceived directiveness of the study health professional's information should be interpreted with caution as the women in the sample returning the T2 questionnaire differed from those that did not on this variable. That is to say, this sample of 68 women perceived the information to be more neutral than those not returning T2. It is feasible that significant differences may have been observed if all the women had returned the T2 questionnaire.

Table 6:25 perceived usefulness of information by study group (T2).

	Total (n = 68)		Routine (n = 39)		Decision Analysis (n = 29)		MANCOVA (1,63)	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	Sig.
useful information	**4.5**	(1.4)	**4.5**	(1.4)	**4.4**	(1.4)	0.09	**0.76**

Table 6:26 perceived encouragement of study health professional by study group (T2).

	Total (n = 68)		Routine (n = 39)		Decision Analysis (n = 29)		Chi^2 d.f. = 2)	
	n	%	n	%	n	%	Chi^2	Sig.
not discussed	2	**3%**	1	**3%**	1	**3%**		
neutral	61	**90%**	36	**92%**	25	**87%**		
encouraged	5	**7%**	2	**5%**	3	**10%**	0.73	**0.69**

Table 6:27 perceived encouragement of other health professionals advice by study group (T2).

	Total (n = 68)		Routine (n = 39)		Decision Analysis (n = 29)		Chi^2 (d.f. = 1)	
	n	%	n	%	n	%	Chi^2	Sig.
not discussed	29	**43%**	16	**41%**	13	**45%**		
neutral	39	**57%**	23	**59%**	16	**55%**	0.10	**0.75**

6.2.5.3 T1 and T2 questionnaires by study group: repeated measures analyses.

The previous sets of analyses have all used a between-subjects design to compare differences in measures as a consequence of the women's allocation to two different experimental groups. This final set of analyses used a repeated measures design to evaluate changes in the measures employed across two time-points. The main advantage of this analysis was the separation of individual variations in responses from random error, so resulting in a more powerful design (Howell, 1985). Ideally, a profile analysis of repeated measures design (Tabachnick and Fidell, 1989) would allow several dependent variables to be assessed within one analysis. Unfortunately, the variables used within this study had different ranges of scores and could not be included within a single analysis. Consequently, eight single repeated measure analyses were carried out for anxiety, the three decisional conflict sub-categories, the perceived advantages and disadvantages of prenatal testing, knowledge and the usefulness of information. As the study numbers for the repeated measures only represented the women who completed the follow-up questionnaire, the same co-variates were used in these analyses as those included within the analyses of the T2 measures: level of education, consultation length and optimism. The criteria assessing homogeneity of variance were met for each analysis (table 6:28).

Table 6:28 tests for homogeneity of variance (repeated measures).

	Bartlett-Box's M	d.f.	Chi^2	Sig.
anxiety	10.7	6	10.2	**0.12**
uncertainty - decisional conflict	4.5	3	4.0	**0.26**
informed - decisional conflict	1.5	3	1.4	**0.69**
efficacy - decisional conflict	14.4	3	13.9	**0.003**
advantages testing	2.7	3	2.6	**0.46**
disadvantages testing	1.1	3	1.1	**0.79**
knowledge (pro-rated to 20)	0.1	3	0.1	**0.99**
usefulness information	4.8	3	4.6	**0.20**

Table 6:29 repeated-measures analyses for decision quality by study group.

	Group Allocation (n = 68)		Time (n = 68)		Group x Time (n = 68)	
	F	Sig.	F	Sig.	F	Sig.
anxiety	0.2	**0.66**	97.2	**<0.0001**	0.3	**0.8**
uncertainty- decisional conflict	4.3	**0.04**	8.0	**0.006**	3.5	**0.07**
informed- decisional conflict	0.3	**0.60**	0.02	**0.89**	8.8	**0.004**
efficacy - decisional conflict	3.5	**0.07**	0.3	**0.60**	6.5	**0.01**
advantages testing	5.2	**0.03**	2.1	**0.15**	0.2	**0.63**
disadvantages testing	2.9	**0.10**	0.1	**0.71**	0.01	**0.94**
knowledge (pro-rated to 20)	0.3	**0.58**	1.3	**0.25**	0.03	**0.87**
usefulness information	1.2	**0.20**	11.0	**0.001**	1.5	**0.22**

Table 6:30 revised mean scores for decision quality by study group.

	Routine group scores (n = 39)			Decision analysis group scores (n = 29)		
	Before T1 x (s.d.)	T1 x (s.d.)	T2 x (s.d.)	Before T1 x (s.d.)	T1 x (s.d.)	T2 x (s.d.)
anxiety	62.3 (12.8)	59.4 (14.1)	34.7 (14.5)	60.3 (14.6)	57.6 (15.4)	35.3 (11.8)
decisional conflict- uncertainty		8.4 (3.0)	8.1 (3.5)		8.0 (2.8)	6.6 (2.7)
decisional conflict- informed		6.8 (1.7)	7.5 (2.2)		7.2 (1.7)	6.4 (2.1)
decisional conflict- efficacy		7.6 (2.4)	8.6 (2.9)		7.4 (1.7)	6.8 (2.1)
advantages of testing		1.9 (0.9)	2.1 (1.0)		1.5 (0.8)	1.7 (0.9)
disadvantages of testing		1.7 (1.0)	1.7 (1.1)		1.4 (0.9)	1.4 (0.9)
knowledge (pro-rated 0-20)		14.6 (3.5)	15.0 (3.2)		14.8 (3.6)	15.4 (3.0)
usefulness of information		5.2 (1.1)	4.5 (1.4)		4.8 (1.1)	4.4 (1.4)

Two significant main effects and one trend towards significance were observed by study group allocation for the decisional conflict sub-category 'uncertainty', perceived advantages of testing and the decisional conflict sub-category 'efficacy' (table 6:29). Those in the routine group perceived more advantages to testing, had less certainty with the decision and a tendency towards less efficacy with the decision than those in the decision analysis group (table 6:29). The scores for the following measures were found to differ significantly from before the results of the diagnostic test or fetal anomaly scan to after this information had been received: anxiety; the decisional conflict sub-category; perceived usefulness of information (table 6:30). The women rated their anxiety as lower, uncertainty with the decision as lower and the usefulness of the consultation information as lower after receiving the 'good news' (table 6:30). For all three sub-categories of the decisional conflict scale, interactions were observed between the time of completing the questionnaire and of the study group allocation (table 6:30). The mean decisional conflict scores of women in the decision analysis group decreased by over 1.5 points for each sub-category from T1 to T2, i.e. a reduction in decisional conflict over time (table 6:30). For the women in the routine group, only the uncertainty sub-category decreased by a smaller number of points (0.3) from T1 to T2. The mean scores of the remaining two categories increased by over 0.8 points each, i.e. a constant or increased level of decisional conflict. These findings suggest that, at follow-up, there was an increase in decisional certainty amongst women in the decision analysis consultation, whereas the evaluation of decisional certainty amongst women in the routine consultation either remained at the same level or decreased at follow-up.

6.3 Discussion.

The purpose of this study was to evaluate the effectiveness of decision analysis to facilitate women's informed decision making about prenatal diagnosis upon receipt of a screen positive triple test result. Few, if any, studies have compared the quality of decisions made in routine health care consultations with consultations structured using the decision analytic technique. Proponents of decision analysis maintain that the quality of decision making improves following application of the technique to real-world decisions, whereas critics argue the opposite. In essence, a better decision would include: a more accurate evaluation of the information; integration of probability and value information; more reasoned decisions; less confusion surrounding the decision; greater satisfaction with the final decision (see chapter six, introduction). In the study described in this chapter, four types of measures were used to evaluate whether or not decision analysis facilitated women's prenatal diagnosis decision making: behaviour;

affect; cognitive processes; clinical quality. Assessments were made during the decision making process, immediately after the decision was made (T1) and four weeks after the decision was made (T2). Measures were both questionnaire-based and derived from the transcripts of audio tape-recorded consultations.

All women participating in the study had received a screen positive triple test result and were offered the opportunity to have, or not have, a diagnostic test. Over the fifteen-month study period, 106 women participated in the study, a participation rate of 84%. Six women were not sent follow-up questionnaires because they had either miscarried the pregnancy or received a positive diagnostic test result for Down's syndrome. One hundred women with a negative diagnostic test result and/or 'normal' nineteen week fetal anomaly scan were sent follow-up questionnaires (T2). The return rate of the T2 questionnaire was 68% (68/100).

The following discussion of findings is divided into four sections: a summary of the main results by category of measure (behaviour, affect, cognitive processes and clinical quality); an assessment of the internal validity of the results; an evaluation of the external validity or generalisability of results; the effectiveness of decision analysis to facilitate women's decision making.

6.3.1 Summary of main findings.

Decision behaviour was not influenced by the differences between the information giving consultations. The women in the decision analysis consultation were as likely to have, or not have, further testing than those in the routine group (table 6:8). The moderate sample size meant the study was under-powered in order to draw conclusions about non-significant associations, for example, that there was no relationship between the decision to have or not have testing with the decision analytic technique. However, it is unlikely that a consultation structured by the decision analytic technique would have an immediate impact on behaviour. Evidence suggests a number of factors are predictive of prenatal testing behaviour such as attitudes, perception of social norms and past behaviour (see chapter one). It is more likely that the evaluation of information is associated with a change in attitude rather than a behaviour change (see Eagly and Chaiken, 1993; Baron, 1994; Garnham and Oakhill, 1994).

The three measures of affect were: general anxiety as assessed by the short-form STAI; the number of positively and negatively emotional-laden words utilised by the women during the prenatal diagnosis consultation and analysed using the SLIWC; the

measure of decisional affect assessed by the decisional conflict scale. The findings suggested that there was an association between the consultation group and the women's experience of decisional affect but no such association with general affect. To explain further, anxiety levels did reduce dramatically over time from scores comparable with clinical significance before and after the decision making consultation (T1) to those considered normal for pregnant women upon receipt of 'good news' (T2). No differences by study group were observed for levels of anxiety at any time point (tables 6:9, 6:19 and 6:29). However, differences in findings by study group were observed for measures of affect associated with the decision making process both during the consultation and at follow-up (T2). First, the number of positive and negative words expressed during the information consultation differed by group allocation (table 6:6). The women in the decision analysis group were less likely to use words associated with positive emotions and a trend towards significance suggested that they were more likely to use words associated with negative emotions than those in the routine information group. Second, the results of the repeated measures analyses found there to be differences in the decisional conflict scores by group allocation (table 6:29). Those in the decision analysis group were more confident about their prenatal choice than those women receiving the routine consultation. In addition, the results of the repeated measures analyses suggested that there was an interaction between study group allocation and time of completion of measures (table 6:29). The women in the routine group reported an increase in decisional conflict upon receipt of 'good news' (T2), whereas women in the decision analysis group reported a decrease in decisional conflict (table 6:30). The pattern of decisional affect illustrated by these findings is that the women in the decision analysis group expressed greater decisional affect whilst making their decision, about the same decisional affect immediately after the consultation (T1) and less decisional affect at follow-up (T2) than those in the routine consultation.

The measures assessing cognitive mechanisms fell broadly into two categories, informed decision making and perception of the decision information.

Informed decision making: The findings suggest that the women in the decision analysis group were making more informed decisions than those in the routine group (table 6:3). During the consultation, the women in the decision analysis consultation appeared to reason more with the decision information than the women in the routine consultation (table 6:3). That is to say, women in the decision analysis group were able to generate more reasons for and against an option than those in the routine group. In addition, the results of the SLIWC analysis showed that the women in the decision analysis

consultation made reference to more words associated with thinking than those in the routine group (table 6:5). However, neither information-seeking within the consultation (table 6:3) nor the questionnaire-based measure of knowledge (tables 6:10, 6:20 and 6:29) differed by study group allocation. It is feasible to suggest from these findings that (a) the decision analysis consultation was associated with the facilitation of informed decision making and not information-seeking and (b), as previously discussed in chapters one and two, knowledge is not an adequate measure of informed decision making.

Perception of decision making: The measures assessing differences in the perception of decision information by study group allocation were mixed. There was no difference in the perception of the triple test risk by group allocation at either T1 or T2 (tables 6:11 and 6:21). The perception of perceived social norm differed significantly by group allocation at follow-up (T2) only. At follow-up (T2), the women in the decision analysis group were more likely than the women in the routine consultation to perceive others as being in favour of prenatal testing (table 5.12, 5.24). This finding may lend support to the idea that the women in the decision analysis group were more confident with their testing choice than women in the routine group, as suggested by the decisional conflict results. In addition, the women in the routine group were likely to generate more perceived advantages to prenatal testing than those in the decision analysis group (tables 6:10 and 6:20). Although not significantly different, those in the routine group also generated more disadvantages to testing than those in the decision analysis group (tables 6:10 and 6:20). One interpretation of this finding is that those in the decision analysis group had a more neutral perception of the prenatal testing process and were cautious in labelling the consequences of testing as an advantage or disadvantage. An alternative interpretation is that the difference in completion of these items suggests that the women in the decision analysis group were less satisfied with the decision analysis consultation than the women in the routine group. In support of this latter explanation were the findings that the women in the decision analysis group expressed more negative emotions during the consultation (table 6:5) and perceived the usefulness of the consultation information to be lower immediately after the decision was made (table 6:15) than those in the routine group. These two explanations are not necessarily mutually exclusive and may indicate that those in the decision analysis consultation were less 'falsely reassured' by the consultation information and/or evaluated the consultation information more objectively than the women in the routine group.

The measures assessing rational or accurate decision making were not associated with any consistent pattern by study group allocation. That is to say, the decision analysis consultation did not encourage women to make decisions more rationally or in accord with EUT than women in the routine information group. To explain further, one of the axioms of EUT is the ability to rank order the consequences of prenatal testing. There was no association between study group allocation and the women's ability to rank order the five consequences of prenatal testing at either T1 or T2 (tables 6:14 and 6:24). However, on exploring differences in the women's allocation of EUVs to the consequences of prenatal diagnosis, one difference by study group was observed for the consequences of 'terminating for child with Down's syndrome'. The women in the decision analysis group had a greater expected-utility score than the routine consultation women at both T1 and T2 (tables 6:12 and 6:22). One explanation for this finding is that, within decision analysis, the lottery method for utility extraction encourages women to focus on their attitudes towards termination for Down's syndrome. As mentioned previously (see chapters four and five), the routine information giving consultation does not provide women with an adequate opportunity to discuss their attitudes towards termination. In essence, the decision analysis consultation in this health context appears to be associated with an increase in informed decision making but not an increase in accurate, rational or optimum decisions.

The three measures of clinical quality were perceived usefulness of information, perceived directiveness of information and length of consultation. There was a main effect of time on perceived usefulness of information following the repeated measures analyses (tables 6:29 and 6:30). That is to say, regardless of group allocation, the women found the consultation information to be useful but their perception of the usefulness of information was lower than upon receipt of good news (T2) than when having made a decision (T1). Although the repeated measures analysis found no association between the study group allocation and consultation usefulness, women in the decision analysis group were significantly less enthusiastic about the consultation information at T1 than women in the routine group. There was no association between group allocation and perceived directiveness of health professionals at either T1 or T2 (tables 6:16, 6:17; 6:26 and 6:27). However, a total of 8% of the women perceived the information about prenatal diagnosis to be directive. Only the measure of consultation length differed significantly by group allocation; the decision analysis consultation was approximately six minutes longer than the routine information consultation (table 6:15). Pauker and Pauker (1987) suggested that consultations structured by decision analysis would increase the consultation length by about ten minutes. It is likely that the

increased consultation length is mainly attributable to the 'lottery' question, i.e. the elicitation of a woman's utility about the termination of pregnancy for Down's syndrome.

6.3.2 Internal validity.

As there is little *a priori* evidence evaluating the decision analytic technique in this or other areas, it is difficult to assert that the findings of this study are valid. However, the measures used to evaluate this study suggest some confidence with the results and their subsequent interpretation. A number of measures were standardised: the SLIWC categories; the short-form STAI; the decisional conflict sub-categories. Most measures were developed from questionnaire items used in previous studies of prenatal and genetic testing: attitudes towards testing; perception of risk; knowledge; elicitation utilities; usefulness of information. The remaining measures, perceived directiveness of information (section 6.1.2) and the informed decision making variables (see chapter five), were developed with reference to the decision making literature and appropriately piloted. In addition, although few studies have employed a randomised control trial design to evaluate the effectiveness of decision analysis, there is a large body of empirical work describing the experience and decision making processes of women in the context of prenatal diagnosis (see chapter one). There were at least five findings within the current analyses that replicated some of the dominant themes observed in previous empirical research. These similarities in findings increase confidence in the interpretation of this chapter's results and in the validity of the study's measures. The findings are described in more detail below:

- The number of the women choosing to have the diagnostic test result was 84%, a figure consistent with observed clinical practice in this area (Ward, 1991; Marteau, Kidd, Cook, Michie, Johnston, Slack and Shaw, 1992).
- The pattern of anxiety from receipt of a screen positive triple test result to the four week follow-up was the same as in previous research: clinical levels of anxiety upon receipt of a screen positive reducing to levels considered normal for a female and pregnant population (Beeson and Golbus, 1979; Burton, Dillard and Clark, 1985; Marteau, Kidd, Cook, Michie, Johnston, Slack and Shaw, 1992: Thornton, Hewison, Lilford and Vail, 1997).
- The reasons provided by women for choosing an alternative, such as the risk of miscarriage, timing of the tests, beliefs about termination and evaluation of the consequence of the unselected option have been described previously in questionnaire and interview-based studies (Farrant, 1985; Lippman, Perry, Mandel and Cartier, 1985; McGovern, Goldberg and Desnick, 1986; Sjorgen and

Uddenberg, 1988; Julian-Reynier, MacQuart-Moulin, Moatte, Aurran, Chabal and Ayme, 1994).

- When listing the perceived advantages and disadvantages of testing in pregnancy (table 6:6), 37% of the women stated that making informed decisions was an advantage whereas 27% identified taking responsibility for making the decision as a disadvantage. This apparent ambiguity towards prenatal diagnosis is supportive of the evidence suggesting that women want to be informed about the decision alternatives but do not necessarily want to take responsibility for their final treatment choice (Fallowfield, 1997).
- Forty-five per cent of the women were able to rank order the utilities associated with the consequences of testing in this study. Of those studies that have assessed the degree to which women made prenatal diagnosis test choices consistent with the 'optimum' or 'accurate' choice, about half of the women were able to do so (Pauker and Pauker, 1978; Heckerling and Verp, 1994).

6.3.3 External validity.

The sample representativeness, participation rates and questionnaire response rate of this study suggest that the findings have good external validity and are generalisable to a wider field of interest (Bowling, 1997). To explain further, over the fifteen month study period, all the women who received a screen positive triple test result were assessed for their eligibility to participate in the trial. Sixteen per cent (28/178) of the women were not invited to participate because they did not fulfil the study criteria. That is to say, they did not speak English, the test result was a screen positive for Spina Bifida or Edward's syndrome, another health professional informed the woman of her triple test result or the woman was a private patient. A further ten per cent (18/178) were excluded because they made their test decision over the phone. As stated, controlling the amount of information these women received, defining the decision context and assessing the decision making processes was compromised in all of these situations. In an effort to assess the representativeness of the sample invited to participate, the demographic characteristics, diagnostic test decisions and reasons for inclusion or exclusion were analysed. Those women invited to participate did not differ from those excluded from participation on any demographic and reproductive characteristics (Appendix XIV). However, the women making their test decision over the telephone were more likely to chose to have no further diagnostic testing. In consequence, it is likely that the women invited to participate in the study were more inclined to consider having a diagnostic test, i.e. the study sample under-represented those choosing to have no further diagnostic testing. The remaining 132 women were invited to participate in the study

and asked to complete a consent form. Fifteen women declined to participate and a further seven women were excluded post-randomisation because their test result was recalculated as a screen negative. There was no difference in anxiety, reproductive variables and demographic characteristics between those agreeing or those declining to participate (Appendix XIV). It was beyond the boundaries of the study's resources to explore what impact those women not included in the study sample might have had on subsequent results. Suffice it to say that, as there were few differences between those included in the study from those not included, the final sample of women were fairly representative of women offered prenatal diagnosis upon receipt of a screen positive triple test result at the LGI.

The participation rate of the women invited to participate was excellent (84%) (Bowling, 1997). The simple randomisation technique provided an effective method for the allocation of the women to study groups. Apart from gestation, the demographic and reproductive characteristics were equally represented between groups (Appendix XIV). In addition, one of the measures for individual differences, optimism (LOT), was also found to differ significantly between study groups. One explanation for these errant findings is chance. That is to say, those in the routine group had a later gestation and were less optimistic than those in the decision analysis group. Certainly, this is the most likely explanation for the differences in gestation which was noted before the women had been contacted about their triple test result. However, there is a second explanation for the differences observed in the measure of optimism. Theoretically, there should have been no association between this trait, individual or personality measure with the experimental intervention. As this measure was completed after the information giving consultation (T1), it is feasible to suggest that the intervention did influence the perception of optimism and, perhaps that the measure assessed state rather than trait optimism. To explain further, there was evidence to suggest that those in the decision analysis group were less satisfied with the consultation information (table 6:15). In addition, fewer women in the decision analysis group did not return their T2 questionnaires than women in the routine group (table 1, Appendix XV). Although this difference did not reach significance, it may be a further indication that women in the decision analysis group were less satisfied with the information giving consultation than women in the routine group as increased satisfaction is associated with greater compliance (Ley, 1988; Ley and Llewelyn, 1995). In consequence, it is feasible to suggest that the optimism measure may have assessed a mood state associated with consultation satisfaction. However, it was beyond the scope of this study to pursue this assertion further, subsequent analyses included gestation and optimism as co-variates

to adjust for the systematic differences observed at this preliminary stage of randomisation. In essence, women in each study group were generally representative of those women agreeing to participate in the study.

The response rate from the follow-up questionnaire was satisfactory (68%). Analyses were carried out on all the T1 and information giving consultation measures to assess the representativeness of those women returning the T2 questionnaire compared with those that did not (Appendix XV). Five variables differed significantly between those that did and did not complete the T2 questionnaire: level of education; family history of abnormality; optimism; length of consultation; perceived directiveness of consultation. Those that completed the questionnaire were more likely to have 'A' level or degree qualifications, less likely to have had a family history of abnormality, to have had shorter information giving consultations, were less optimistic and perceived the information giving consultation to be less directive than those that did not return the T2 questionnaire. Again, explaining these significant differences between those returning and not returning the questionnaire would have been difficult to pursue without further investigation. Suffice it to say, the women completing the T2 questionnaire were not as representative of the Leeds screen positive triple test population as those completing T1. In consequence, statistical steps were taken to counteract the impact that this less representative sample may have had on the generalisability of findings. Preliminary correlations were carried out between the variables that differed by completion of the T2 with the quality of decision measures at T1. Variables that were significantly associated with the measures of decision quality were entered as co-variates in the subsequent analysis of the women's T2 responses. It is feasible to suggest that these statistical adjustments would be sufficient to ensure confidence in the findings evaluating the effectiveness of decision analysis at T2.

There were at least three factors beyond the control of the author that may be associated with the interpretation of the current findings. The first concerns the study sample size. As previously stated, power calculations using the anxiety score to estimate sample size suggested that approximately 52 women were required for each study group. After fifteen months, the final sample included 56 women allocated to the routine group and 50 women to the decision analysis consultation. Although just under the approximate sample size, this final figure was in part a compromise with the demands of the clinical setting. On recalculating the sample size using the observed differences on the anxiety score (two point difference, s.d. = 16), about 1000 women would be required in each study group to achieve an acceptable level of power. This re-

analysis suggests that the study may have been under-powered. However, in retrospect, it could be argued that anxiety was not an appropriate measure with which to evaluate the decision quality. Perhaps subsequent power calculations will use the measure of decisional conflict. As the study may have benefited from having a larger sample size, conservative estimates of significance were employed suggesting that some type II errors may have occurred.

Two other factors of note concern the generalisability of the study outside the Leeds health authority: the uptake of the triple test by women at the LGI and the offer of the triple test to women by the Leeds health authority. The figures supplied by the LGI for the uptake rate of the triple test two years before and during the study period was 58% (3600/6253), one of the lowest rates within the country. In addition, Leeds only offered the triple test to women aged 29+ years at estimated date of delivery (EDD); some health authorities offer the triple test to all pregnant women regardless of age. One study has documented factors associated with the uptake rate of the triple test within Leeds and Bradford (Thornton, Hewison, Lilford and Vail, 1997) and found that older, Caucasian women in higher social classes with healthy children were more likely to have the triple test. This set of profile characteristics suggests that, within the triple test and prenatal diagnosis context, these findings of the effectiveness of decision analysis are generalisable. However, this evaluation of the decision analytic technique has been carried out on a sample of female, predominantly white, moderately well-educated and 'older' reproductive-age population. In consequence, it remains an empirical question as to whether the same findings would be observed within a different clinical and socio-demographic population.

6.3.4 The effectiveness of the decision analytic technique.

As summarised earlier in the discussion, a number of differences were found for both consultation and questionnaire measures of decision quality by study group. These findings are discussed below with specific focus on the efficacy of decision analysis to facilitate the quality of individuals' decision making.

Decision analysis is a technique or aid derived from EUT. It has been well established that individuals do not make decisions in accord with EUT (chapter one). However, an aim of the application of decision analysis is to encourage individuals to make decisions in accord with EUT. There was little evidence to suggest that the women in the decision analysis group made more optimum decisions. Most of the women were not able to rank order the utilities associated with the consequences of prenatal testing. Furthermore,

the women in the decision analysis group were no more or less likely to be able to rank order the utilities than the women in the routine group (tables 6.14 and 6.24). Although it is possible that with a larger sample, decision analysis may be seen to facilitate optimum decision making, no such association was established in this study.

Although decision analysis has not been associated with an increase in the accuracy of decisions, the women in the decision analysis group made more informed decisions about prenatal diagnosis than the women in the routine group (table 6:3). To explain further, the results of the theme-based coding frame suggest that the women in the decision analysis group evaluated more reasons for and against alternatives than the women allocated to the routine consultation group (table 6:3). In addition, the women in the decision analysis group were more likely to use words associated with cognition than those in the routine group (table 6:5). It is fair to say that the women in the decision analysis group made a more reasoned evaluation of the information during the consultation than those in the routine consultation. It was beyond the scope of this chapter's analysis to explore other group allocation differences in the strategies employed by women when making the decision to have or not have prenatal diagnosis; for example, whether or not women in the routine group employed more 'heuristic' information processing strategies than those in the decision analysis consultation. However, the frequency table summarising the information utilised by the women during the consultation indicates that the women in both consultations appeared to refer equally to the themes associated with a more heuristic appraisal of information (themes 13-20, table 6:1). In other words, it seems feasible to suggest that decision analysis was associated with facilitating the utilisation of the decision specific information rather than all the information referred to in the consultation.

Another advantage of applying a decision aid to a real-world decision is to reduce the degree of decisional conflict experienced by an individual when making a difficult decision (O'Connor, 1995). The women in the decision analysis consultation reported less decisional conflict at follow-up than those in the routine group (tables 6:29 and 6:30). In other words, decision analysis was associated with greater confidence in the decision made. However, during the consultation, the women in the decision analysis group were less likely to use positive emotion words and more likely to use negative emotion words than those in the routine group, i.e. greater expressed affect (tables 6:1 and 6:4). This finding supports the observation that the decision analysis technique may encourage women to confront difficult issues during the consultation, as suggested by Pauker and Pauker (1977). In particular, the decision analysis consultation may

provide women with a more effective forum to express how they feel about the diagnostic test decision than the routine consultation. After the decision was made, there were no differences in measures of anxiety by study group (tables 6:9 and 6:19). This finding may support the latter assertion. Alternatively, this lack of association between anxiety and study group allocation may reflect the idea that anxiety is not an appropriate measure of decisional affect. It is possible that a larger sample or more sensitive assessment of anxiety might have been able to establish a link between decision analysis and a global measure of affect.

Some of the concerns regarding the application of decision analysis to the real-world health care setting are those pertaining to clinical quality. The following measures were used to assess changes in the perceived clinical quality of the information giving consultation: satisfaction with the consultation information; length of consultation; perceived directiveness of information. The women did not perceive the decision analysis consultation to be any more or less directive than those in the routine group (tables 6:16, 6:17, 6:26 and 6:27). After making the decision (T1), the women in the routine group rated the usefulness of information as higher than those in the decision analysis group (table 6:15). However, the repeated measures analysis using the smaller sample size did not replicate this finding (tables 6:29 and 6:30). The decision analysis consultation did take, on average, six minutes longer than the routine consultation. Although this has implications for health care resources and return rates of follow-up questionnaires, it is unlikely that the women were unduly affected by the consultation length. On the contrary, as individuals are more likely to apply heuristic processes in time-pressured situations, it is likely that longer consultation lengths may be associated with more informed decisions.

As discussed, the main purpose of this study was to evaluate the effectiveness of decision analysis to facilitate women's informed decision making in the context of prenatal diagnosis. The randomised control trial compared decisions made following the application of decision analysis to the consultation with those made unaided. The findings suggested that (a) women's informed decision making can be facilitated and that (b) women in the decision analysis consultation made more informed decisions than women receiving routine information. However, the decision analytic technique requires the application of both information and decision aids to structure the decision information, elicit utilities, integrate utilities with appraisals of risk and proffer a decision based on expected-utility (see section 6.1). It is unclear from the evidence reported in this chapter what aspect of the decision analytic technique was associated with the

facilitation of informed decision making. For example, it is feasible to suggest that women's informed decision making was facilitated with reference to the decision tree representation of the prenatal diagnosis decision alone. In essence, the decision tree component of the decision analytic technique is an information aid. To explain further, the decision tree may have acted as a visual prompt for both the health professional and woman decision maker, thus enabling the health professional to provide sufficient information about the decision alternatives and/or encouraging woman to appraise equally all the decision information. Alternatively, it is as likely that the ensuing discussions of the lottery question, the threshold graph and/or the proffered decision between the health professional and woman were the stages of decision analytic technique that were associated with the facilitation of informed decision making. Although a more in-depth analysis of the health professional' and women's transcripts would provide some evidence to address these issues, further randomised control trials comparing decisions made with reference to information aids, decision aids or unaided would be required to ascertain what aspect of the decision analysis consultation was associated with women's increase in the utilisation of decision information.

6.3.5 Chapter summary.

The study discussed in this chapter was one of the first to evaluate the effectiveness of decision analysis to facilitate individuals' decision making. The study employed both information-tracing techniques and questionnaire measures to assess the quality of the prenatal diagnosis decision made following a routine information giving or decision analysis consultation. The study sample was representative of women having the triple test at Leeds General Infirmary. The decision analysis consultation was associated with the making of a more informed decision, with lower decisional conflict about the final decision and with greater confidence in others attitude to undergoing prenatal testing. Those in the decision analysis group were less likely to report the perceived advantages of prenatal testing and the usefulness of information following the making of the prenatal diagnosis decision. These findings may indicate that the women in the decision analysis consultation were less likely than those in the routine group to be falsely reassured by the diagnostic test information. During the consultation women in the routine group expressed more positive words and those in the decision analysis group expressed more negative words suggesting that either the decision analysis consultation was more confrontational than the routine information or that the decision analysis consultation provided a better forum for women to express how they felt about the consequences and alternatives of the prenatal diagnosis decision. The women did not perceive the decision analysis consultation to be more directive or to engender more

anxiety than the women in the routine consultation. The decision analytic technique was not associated with the making of more optimum decisions, with differences in the decision made, nor with knowledge and perception of individual risk.

As one of the primary goals of prenatal testing is to ensure informed decision making, implementing a technique demonstrated to facilitate informed decision making within routine clinical practice would seem appropriate. The decision analytic technique was associated with positive changes in informed decision making and with the experience of being offered prenatal diagnosis. The nature of the technique ensures women are (a) exposed to the information considered necessary to enable reasoned or informed decisions and (b) provided with the opportunity to integrate this information with their beliefs and values concerning the consequences of prenatal diagnosis. The findings suggest that knowledge is an inadequate measure of informed decision making and that the assessment of anxiety is an unsatisfactory measure of decisional affect. It remains an empirical question as to whether or not informed decision making can be adequately assessed using a pre- or post-decision questionnaire. The decision analytic technique has been found to facilitate informed decision making within the prenatal diagnosis consultation. However, this decision context is well defined and lends itself to such a formal technique (Pauker and Pauker, 1987; Ubel and Loewenstein, 1997). It remains an empirical question whether or not decision analysis facilitates decision making in other decision contexts.

7. Predicting women's prenatal diagnosis decisions.

This final, empirical chapter focuses on identifying those psychological factors associated with the decision to have or not have a prenatal diagnostic test. Describing the psychological factors associated with women's prenatal diagnosis decisions has been one of the main research questions of empirical studies carried out in the prenatal testing context (see chapter one). The following factors have been identified as predictive of women's prenatal diagnosis behaviour: age (Sjorgen and Uddenberg, 1988; Evans, Pryde, Evans and Johnson, 1993; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994); parity (Sjorgen and Uddenberg, 1988; Halliday, Lumley and Watson, 1995); reproductive history (Sjorgen and Uddenberg, 1988); level of education (Sjorgen and Uddenberg, 1988; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994; Halliday, Lumley and Watson, 1995); attitudes towards abnormality (Sjorgen and Uddenberg, 1988; French, Kurczynski, Weaver and Pituch, 1992; Evans, Pryde, Evans and Johnson, 1993; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994); termination (Volodkevich and Huether, 1981; French, Kurczynski, Weaver and Pituch, 1992; Heckerling and Verp, 1994); prenatal testing (French, Kurczynski, Weaver and Pituch, 1992; Lippman, Perry, Mandel and Cartier, 1985; McGovern, Goldberg and Desnick, 1986) and pregnancy (Burke and Kolker, 1993; Heckerling and Verp, 1994); the perception of others' prenatal testing attitudes (Volodkevich and Huether, 1981; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994); perceptions of risk (Volodkevich and Huether, 1981; Lippman, Perry, Mandel and Cartier, 1985; McGovern, Goldberg and Desnick, 1986; Kolker and Burke, 1993; Evans, Pryde, Evans and Johnson, 1993); affect (Fava, Trombini, Michelacci, Linder, Pathak and Bovicelli, 1983; Evans, Pryde, Evans and Johnson, 1993).

As described in more detail in chapter one, there are some methodological concerns pertaining to these studies that may limit the interpretation of subsequent findings (section 1.3.2.2). First, most studies employed a pro- or retrospective study design and did not describe the process of decision making. Second, few studies assessed the factors associated with the choice between having or not having prenatal diagnosis. A number of studies included a sample of women who were representative of those who had prenatal diagnosis or those who had not had prenatal diagnosis. In other words, these study samples only enabled the assessment of the decision making processes associated with one of the decision alternatives. Finally, few studies were designed with reference to established models of behaviour from either the decision making or social

cognition literature. In consequence, it is difficult to ascertain whether or not the psychological processes underpinning women's prenatal decision making have been adequately assessed or described. However, understanding the factors associated with women's prenatal diagnosis decision making may be useful to either inform subsequent interventions aimed at facilitating decision making or to provide a more comprehensive explanation as to why the decision analysis intervention was not associated with behaviour change.

The purpose of this chapter is to carry out further analyses of the data collected from the randomised control trial (RCT) described in chapter six in order to identify the psychological factors associated with women's prenatal diagnosis decision making. In this sample of women, 17 chose to have no further diagnostic testing and 89 decided to have a diagnostic test. As no association was observed between the women's test behaviour and allocation to the RCT's study groups, the women randomised to each trial arm were integrated to form one data source.

7.1 Methodology.

7.1.1 Sample, Design, Measures and Procedure.

The data used in this chapter's analyses were collected from the 106 women who participated in the randomised control trial reported in chapter six. As this chapter is concerned with psychological factors associated with the decision to have or not have prenatal diagnosis, only the data from the information giving consultation and T1 questionnaire will be analysed in more detail. The study sample, design, measures and procedure have been fully described in chapter six.

7.1.2 Analysis.

The aim of the following analyses is to identify the psychological factors associated with women's decisions to have or not have prenatal diagnosis. The purpose of the RCT described in chapter six was to evaluate the effectiveness of decision analysis to facilitate women's decision making. The study was not designed to operationalise or test a particular model of decision making. However, the measures used to evaluate the RCT were informed by theories of and empirical research from both the social cognition and decision making literature. In consequence, data were obtained for the following measures[1]:

[1] Categorical variables were only included in the analyses if the sample distribution was at least 25:75% between levels (Tabachnick and Fidell, 1989).

- demographic characteristics and reproductive history - age, gestation, number of children, number of miscarriages, level of education and religious activity;
- individual differences - need for cognition and optimism (LOT);
- cognitive processes - informed decision making variables, cognitive content of women's utterances, perception of risk, perceived advantages and disadvantages of prenatal testing, expected-utility values for the five consequences of testing, rank ordering of the values of the consequences of testing and knowledge;
- affect - anxiety (STAI) before and after the consultation, decisional conflict variables and emotional content of women's utterances;
- clinical quality - consultation length, usefulness of information and perceived directiveness of study and other professionals.

To establish the factors associated with women's decisions to have or not have prenatal diagnosis, a discriminant function analysis was applied to the data set. In a suitably powered study, all of the above variables could be included in an analysis. However, sample size is a consideration when selecting the number of variables to be included in a discriminant function analysis (Tabachnick and Fidell, 1989). Fewer variables than cases in the smallest group should be selected. In a study with a small sample size (n = 20 in one group), only a few variables should be entered. There is no correct way to select variables for inclusion within a discriminant function analysis (Tabachnick and Fidell, 1989). Certainly, the aim of the research is a factor in selecting the type of variables for inclusion in the analysis. The study reported in this chapter was particularly concerned with identifying the psychological factors associated with women's decision making. In consequence, the cognitive mechanisms identified in previous research and theories of individual's decision behaviour received particular consideration (see chapter one). In addition, items can be omitted from the analysis if there are several measures assessing a similar construct. In this instance, preliminary univariate and multivariate analyses should be carried out to identify the item showing the greatest association with the dependent variable. Finally, if no specific model is being tested or there are no *a priori* reasons for including one variable over another, a statistical or stepwise discriminant function can be used to select the variables that best classify the pattern of responses between groups. In consequence, the analyses in this section were carried out in two stages:

- Chi square tests, Mann-Whitney tests and MANOVA were carried out to assess differences between the test decision and the following: demographic characteristics, reproductive history and individual differences; cognition and clinical outcomes; affect.

- a stepwise discriminant function analysis was carried out to find the variables that best predicted group membership. A more detailed explanation for the inclusion of the nine variables included within this discriminant function analysis is described below.

Nine items were selected from the available measures for further analysis, either because they were found to differ significantly by test decision in the preliminary analysis or because there was an indication from other research and theory that they would be predictors of health behaviour. Items that were selected following the preliminary analyses of variables were: anxiety before and after consultation; informed decision making, reasons for an option; the expected utility values for the consequences of having a healthy baby, a baby with Down's syndrome and terminating for Down's syndrome; perception of the triple test risk; perceived social norm. Most of these eight variables selected on the basis of the preliminary analyses would also have been included in the discriminate function analysis for prior empirical or theoretical reasons. An additional variable included in the discriminant function analysis on the basis of prior research was the measure of knowledge. One variable, usefulness of information, was found to be associated with the test decision following the preliminary analyses. However, usefulness of information was omitted from the discriminant function analysis because it did not assess a process measure of decision making. In essence, usefulness of the information received was an outcome measure, an assessment of satisfaction. As there was little *a priori* evidence to suggest which variables would discriminate between the two behaviours, a stepwise discriminant function analysis was used to statistically select the most appropriate items from this list. No variables were transformed for this analysis as the discriminant function analysis is considered robust enough to accommodate variables with skewed distributions.

7.2 Results.

As mentioned, complete data were available for 106 women, 17 of whom decided to have no further testing and 89 to have either amniocentesis or chorionic villus sampling. The following results are presented in four sections. The first three parts summarise the preliminary analyses carried out to ascertain whether there were significant differences in scores by decision behaviour for the following variables: demographic characteristics, reproductive history and individual differences; affect; cognitive processes and clinical outcomes. The final section of the results describes the stepwise discriminant function analysis carried out to identify the variables that were the best predictors of the women's decision to have or not have prenatal diagnosis.

7.2.1 Demographic characteristics, reproductive history and individual differences.

None of the analyses carried out between test decision and any of the demographic characteristics, reproductive variables or measures of individual differences for information processing had a significance level of less than 0.05 (tables 7:1, 7:2 and 7:3). There was a trend towards significance indicating that a higher level of education was attained by those choosing to have no further testing (table 7:3).

Table 7:1 demographic characteristics and individual differences by decision behaviour.

	Total n = 106		No Test n = 17		Test n = 89		MANOVA d.f. = 1, 104	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f value	sig.
age	**35.2**	(3.0)	**34.8**	(3.8)	**35.3**	(2.9)	0.3	**0.57**
gestation	**14.8**	(1.0)	**14.7**	(1.0)	**14.8**	(1.0)	0.04	**0.83**
life orientation test (LOT)	**21.0**	(6.5)	**19.8**	(7.3)	**21.3**	(6.3)	0.7	**0.41**
need for cognition (NFC)	**81.7**	(20.8)	**84.3**	(24.2)	**81.4**	(20.2)	0.3	**0.58**

Table 7:2 reproductive history and presence companion in consultation by test decision.

	Total n = 106		Test n = 17		No Test n = 89		Chi^2 Analysis d.f. = 1	
	n	%	n	%	n	%	Chi^2	Sig.
have children	68	**64%**	10	**59%**	58	**65%**	0.3	**0.61**
miscarried	27	**25%**	3	**18%**	24	**27%**	0.7	**0.41**
family history abnormality	24	**23%**	3	**18%**	21	**24%**	0.3	**0.59**
companion present	78	**74%**	10	**59%**	68	**76%**	2.3	**0.13**

Table 7:3 level of education and religious activity by test decision.

	Total n = 106		No Test n = 17		Test n = 89		Chi^2 Analysis d.f. = 2	
Level	n	%	n	%	n	%	Chi^2	Sig.
Education:								
up to GCSE	46	**44%**	6	**35%**	40	**45%**		
'A' level equiv.	33	**31%**	3	**18%**	30	**34%**		
degree +	27	**25%**	8	**47%**	19	**21%**	5.2	**0.07**
Religious activity:								
not active	66	**63%**	10	**59%**	56	**63%**		
occasional attendance	31	**29%**	5	**29%**	26	**29%**		
frequent attendance	9	**8%**	2	**12%**	7	**8%**	0.3	**0.86**

7.2.2 Affect.

Apart from the anxiety scores (see below table 7:4), all the affect measures from the consultation transcripts and post-decision questionnaire were entered into a MANOVA. There was no significant association between the test decision with the measures of decisional conflict or expressed emotion during the consultation (table 7:4).

Table 7:4 measures of affect by test decision.

	Total n = 106		No Test n = 17		Test n = 89		MANOVA (1, 104)	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	Sig.
anxiety before consultation	**62.2**	(13.6)	**53.9**	(13.5)	**63.8**	(13.1)	NA	**NA**
anxiety after consultation	**60.1**	(14.8)	**44.1**	(14.9)	**63.2**	(12.8)	NA	**NA**
conflict - uncertainty	**8.4**	(2.8)	**9.1**	(3.0)	**8.3**	(2.7)	1.3	**0.26**
conflict - informed	**6.8**	(1.7)	**6.7**	(1.7)	**6.8**	(1.7)	0.1	**0.78**
conflict - efficacy	**7.5**	(2.2)	**8.0**	(2.3)	**7.4**	(2.2)	0.9	**0.35**
positive emotion (SLIWC)	**1.9**	(0.7)	**2.1**	(0.7)	**1.9**	(0.7)	1.8	**0.19**
negative emotion (SLIWC)	**0.9**	(0.5)	**0.8**	(0.4)	**0.9**	(0.5)	0.9	**0.35**

Anxiety was the only measure of affect to be assessed before the consultation and at T1 (post-decision). In consequence, a separate, repeated measures analysis was applied to the two anxiety scores. The repeated measures analysis enabled both a between-subject and within-subject comparison of the data (table 7:5).

Table 7:5 repeated measures analysis for anxiety over time by test decision.

	f	Sig.
effect of test decision	21.1	< 0.0001
effect of time	12.3	0.001
effect of test decision by time	9.6	0.002

Significant differences in anxiety were observed by time, test decision and an interaction between time and test decision (table 7:5). Univariate analyses indicated that those women choosing to have no further testing had significantly lower anxiety both before and after the consultation than those choosing to have a diagnostic test (respectively: $t = -2.83$, d.f .= 104, $p = 0.006$; $t = -5.49$, d.f .= 104, $p < 0.0001$). In addition, the anxiety of the women choosing to have no further testing was significantly lower after the consultation than before with no change in the anxiety of women

choosing to have a test (respectively: t = -3.13 d.f. = 14, p = 0.007; t = -0.52, d.f. = 88, p = 0.60).

7.2.3 Cognitive processes and consultation quality.

The measures included in the consultation transcripts and T1 questionnaire (post-decision) to assess cognitive measures were: informed decision making variables, information seeking, reasons for and reasons against; expressed cognitions or 'thinking words' as measured by SLIWC; perceived advantages and disadvantages of testing; expected utility values for the consequences of testing; ability to rank order the consequences of testing; perceived social norm; perception of triple test risk. The variables assessing consultation quality were: directiveness of consultation information; length of consultation; usefulness of information. Only about half of these were interval measures with normal distributions, a few interval measures had skewed distributions and the remainder were essentially categorical measures (see chapter six). All the interval measures with normal distributions were included in a single MANOVA (tables 7:6 and 7:11), a non-parametric test applied to the interval measures with skewed distributions (table 7:7), and Chi square analyses to the remaining categorical variables (tables 7:8, 7:9, 7:10, 7:12, 7:13 and 7:14). The findings pertaining to the cognitive processes are discussed first, followed by a summary of clinical quality results.

Table 7:6 cognitive processes and consultation quality by test decision

	Total n = 106		**No Test** n = 17		**Test** n = 89		**MANOVA** d.f. = 1,104	
	x	(s.d.)	**x**	(s.d.)	**x**	(s.d.)	f	**sig.**
informed decision making - information seeking (IDM-I)	**2.8**	(1.1)	**3.0**	(0.9)	**2.8**	(1.1)	0.5	**0.48**
informed decision making - reasons for option (IDM-F)	**2.3**	(1.0)	**2.8**	(1.1)	**2.2**	(1.0)	5.8	**0.02**
informed decision making - reasons against (IDM-A)	**2.1**	(1.1)	**2.4**	(1.1)	**2.1**	(1.1)	1.3	**0.26**
cognitive mechanism (SLIWC)	**11.3**	(1.7)	**11.6**	(1.9)	**11.2**	(1.7)	0.5	**0.47**
advantages testing	**1.6**	(0.9)	**1.8**	(1.0)	**1.6**	(0.9)	0.9	**0.36**
disadvantages testing	**1.5**	(1.0)	**1.9**	(1.1)	**1.4**	(0.9)	3.0	**0.09**
knowledge	**14.5**	(3.0)	**14.5**	(4.1)	**14.5**	(2.8)	0.0	**0.98**

A number of findings suggested that the cognitive processes underlying the decision to have a diagnostic test were different from those associated with choosing to have no further testing. The 'test' women generated fewer reasons for choosing an option than 'no test' women (table 7:6), 'test' women had greater perceived social norm scores than the 'no test' women, i.e. more likely to agree with the statement 'people who are important to me think I should have a diagnostic test' (table 7:7), 'test' women had lower expected-utility values for having a baby without Down's syndrome and having a baby with Down's syndrome (table 7:7) and a greater expected-utility value for terminating a baby with Down's syndrome than 'no test' women (tables 7:7 and 7:8), and more 'test' women perceived their triple test risk figure to be a high risk than 'no test' women (table 7:10). In addition, there was a trend towards significance suggesting that the 'test' women were less likely to be able to rank order the consequences associated with the prenatal diagnosis decision than 'no test' women (table 7:9). However, the actual *ranked order* of each of the expected utility values remained the same for both those choosing to have and not have a diagnostic test: having a healthy baby; terminating for Down's syndrome; miscarrying a baby with Down's syndrome; having a baby with Down's syndrome; miscarrying a healthy baby (table 7:7).

Table 7:7 expected-utility values and perception social norm by test decision.

	Total n = 106		**No Test** n = 17		**Test** n = 89		**Mann-Whitney** NA = no analysis		
	x	(s.d.)	**x**	(s.d.)	**x**	(s.d.)	U	z	**Sig.**
EUT1 - healthy baby	**39.5**	(10.9)	**46.1**	(9.3)	**38.2**	(10.7)	451	-2.6	**0.008**
EUT2 - miscarry DS	**13.0**	(11.1)	**8.6**	(7.6)	**13.8**	(11.5)	558	-1.7	**0.09**
EUT3 - terminate DS	**28.7**	(21.7)	**17.7**	(19.9)	**30.8**	(21.6)	NA	NA	**NA ***
EUT4 - have baby DS	**3.5**	(6.6)	**6.5**	(8.2)	**2.9**	(6.1)	389	-3.2	**0.002**
EUT5 - miscarry baby	**1.2**	(2.1)	**1.6**	(3.3)	**1.1**	(1.8)	719	-0.3	**0.75**
perception social norm (should have test)	**1.5**	(1.7)	**3.7**	(1.7)	**1.1**	(1.3)	178	-5.1	**>0.001**

** means only reported as a bi-modal distribution (see chapter six).*

Table 7:8 expected-utility for termination Down's syndrome by test decision.

	Total n = 106		**No Test** n = 17		**Test** n = 89		**Chi^2 (d.f. = 1)** Fisher's Exact	
	n	%	n	%	n	%	Chi ^{2}	Sig.
EUT value <30	58	**55%**	13	**76%**	45	**51%**		
EUT value >30	48	**45%**	4	**24%**	44	**49%**	3.9	0.04

Table 7:9 rank ordering of expected utilities by test decision.

	Total n = 106		No Test n = 17		Test n = 89		Chi ^{2} (d.f. = 1) Fisher's Exact	
	n	%	n	%	n	%	Chi ^{2}	Sig.
ranked	48	**45%**	11	**65%**	37	**42%**	3.1	**0.07**
not ranked	58	**55%**	6	**35%**	52	**58%**		

Table 7:10 perception triple test risk by test decision.

	Total n = 106		No Test n = 17		Test n = 89		Mann-Whitney		
	n	%	n	%	n	%	U	z	Sig.
low	13	**12%**	4	**24%**	9	**10%**			
medium	43	**43%**	12	**70%**	31	**35%**			
high	50	**50%**	1	**6%**	49	**55%**	376.5	-3.6	**0.003**

With regard to the quality of consultation measures, the women choosing to have no further testing perceived the information to be useful but not as useful as those deciding to have a diagnostic test (table 7:11). There was a trend towards significance to suggest that the 'no test' women had longer consultations than 'test' women (table 7:11). A small percentage of the women (8%) found the information or advice provided by the researcher, other health professionals and their friends and family to be directive (tables 7:11, 7:12 and 7:13). This perceived directiveness did not differ significantly by test decision.

Table 7:11 clinical quality by test decision.

	Total n = 106		No Test n = 17		Test n = 89		MANOVA d.f. = 1,104	
length consultation	**29.0**	(12.2)	**34.2**	(11.9)	**28.1**	(12.1)	3.8	**0.06**
usefulness information	**5.2**	(1.1)	**4.7**	(1.7)	**5.3**	(0.9)	4.6	**0.04**

Table 7:12 directiveness of study health professional information by test decision.

	Total n = 106		No Test n = 17		Test n = 89		Chi ^{2} d.f. (2)	
	n	%	n	%	n	%	Chi ^{2}	Sig.
not discussed	10	**9%**	1	**6%**	9	**10%**		
neutral	88	**83%**	13	**76%**	75	**84%**		
directive	8	**8%**	3	**18%**	5	**6%**	3.1	**0.21**

Table 7:13 directiveness of other health professionals' information by test decision.

	Total n = 106		No Test n = 17		Test n = 89		Chi ^{2} d.f. (2)	
	n	%	n	%	n	%	Chi ^{2}	Sig.
not discussed	54	51%	11	64%	43	48%		
neutral	43	41%	4	24%	39	44%		
encouraged	9	8%	2	12%	7	8%	2.5	0.29

7.2.4 Predicting test behaviour from cognitive processes and affect.

A stepwise discriminant function analysis was carried out to establish which cognitive processes and affect best predicted testing behaviour. As mentioned in section 7.1.2, variables were selected for entry in the analysis because they were either important components of theories explaining behaviour and/or had been found to differ significantly by test decision during the preceding preliminary analyses. The following nine variables were entered into the stepwise discriminant function command: the expected utility values for the consequences of having a healthy baby, a baby with Down's syndrome, and terminating for Down's syndrome; perception of the triple test risk; perceived social norm; the informed decision making reasons for an alternative sub-category; knowledge of testing; anxiety before and after the consultation. The pattern of responses for four variables were selected by the stepwise discriminant function analysis for inclusion in the final equation as predictors of testing behaviour: perceived social norm; the expected utility value for the consequence of having a healthy baby; the expected utility value for the consequence of having a baby with Down's syndrome; post-consultation anxiety (table 7:14).

Table 7:14 discriminant function analysis for test decision.

	Between-group Corr. with Fctn.	Univariate F (1,104)	Wilks lamda	Sig.
post-consultation anxiety	-0.52	13.6	0.55	<0.001
perceived social norm: should have test	0.70	48.6	0.71	<0.001
expected utility value: healthy baby	0.27	8.0	0.52	<0.001
expected utility value: Down's baby	0.20	5.5	0.51	<0.001
Canonical *R* (R^2)	0.72	(52%)		
Eigenvalue	1.07			

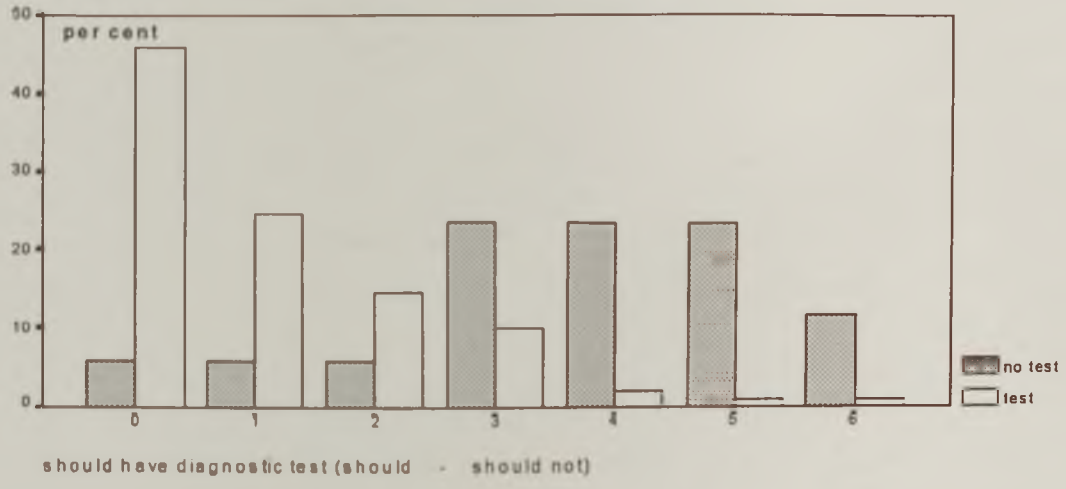
The discriminant function accounted for 52% of the variance in testing behaviour (X^2 = 74.4, d.f. = 4, $p < 0.00001$). The function correctly classified 94.3% of cases; 94.1% of women in the no test decision group and 94.4% in the test decision group. The Bartlett box M test for homogeneity of variance was not significant (Box's M = 19.4, d.f .= 10, 3583.1, p = 0.06) suggesting there was little threat to the interpretation of the multivariate analysis.

Bi-variate analyses showed no significant relationship between the cognitive processes found to predict test behaviour, i.e. the EUVs and perceived social norms (table 7:15). However, affect was significantly associated with both perceived social norm and the expected-utility value for the consequence of having a healthy baby (table 7:15). That is to say, women who had greater anxiety scores at T1 were more likely to perceive others as encouraging them to have a test and less likely to perceive themselves as having a healthy baby.

Table 7:15 correlation matrix between predictors of testing behaviour.

	Anxiety		Social Norm		EUT 1	
	r	Sig.	r	Sig.	r	Sig.
perceived social norm	-0.27	**0.006**				
expected utility for healthy baby (EUT1)	-0.30	**0.002**	-0.05	**0.60**		
expected utility for Down's baby (EUT4)	0.03	**0.71**	0.08	**0.40**	0.01	**0.93**

Figure 7:1 perceived social norm by test decision.



To provide a clearer understanding of the pattern of responses by group, bar charts of the responses to the perception of norm item (figure 7:1) and the two perception of risk components for the expected-utility values (figures 7:2 and 7:3) were constructed. The

pattern of responses suggests the following interpretation (tables 7:3 and 7:4; figures 7:1 - 7:3): the 'test' women had higher anxiety at T1 than the 'no test' women; the 'test' women perceived their likelihood of having a healthy child as slightly lower than the 'no test' women; the 'test' women perceived their likelihood of having a child with Down's syndrome as slightly higher than the 'no test' women; the 'test' women were more likely to perceive others as having expectations about them having prenatal diagnosis, whereas the 'no test' perceived others as having a more neutral attitude towards them having a diagnostic test.

Figure 7:2 perceived likelihood of healthy baby by test decision.

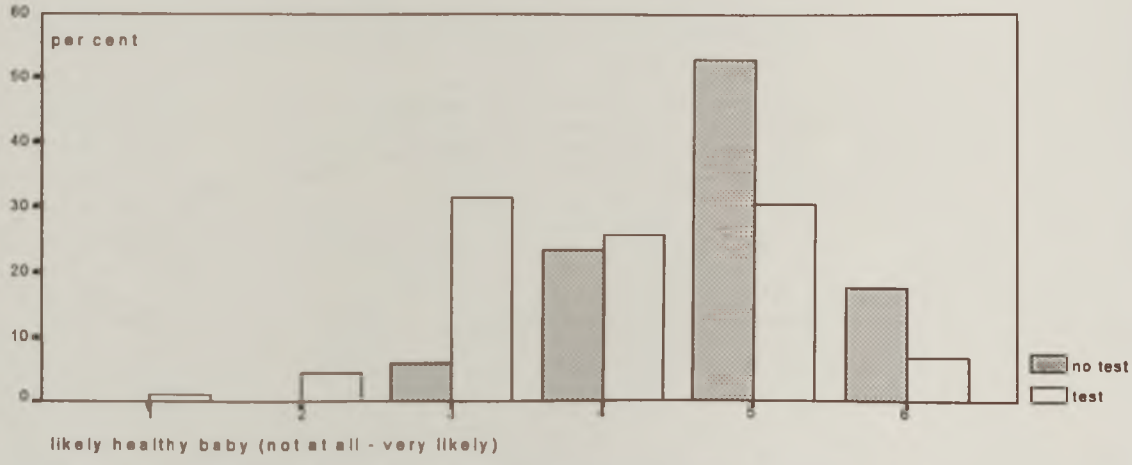
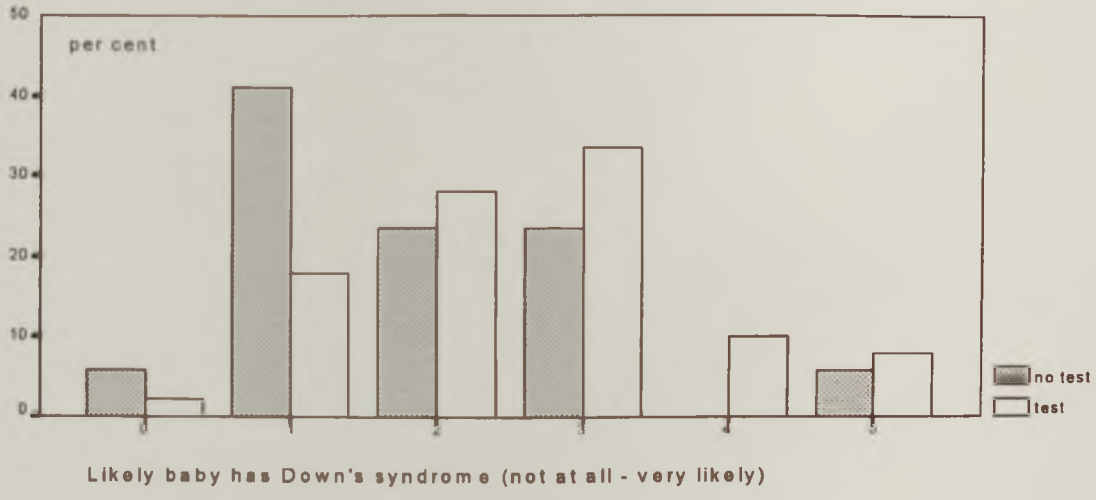


Figure 7:3 perceived likelihood of baby having Down's syndrome by test decision.



7.3 Discussion.

The purpose of these additional analyses was to describe the psychological mechanisms associated with women's decisions to have or not have prenatal diagnostic tests. To identify these mechanisms, a stepwise discriminant function was applied to the data. However, preliminary analyses were carried out on the following variables to select measures for inclusion in the discriminant function analysis: demographic characteristics, reproductive history and individual differences; affect; clinical quality and cognitive measures. The findings from these preliminary analyses are summarised briefly below:

- No significant differences by test decision were observed for the demographic characteristics, reproductive history or individual difference measures (tables 7:1, 7:2 and 7:3);
- The preliminary analyses assessed differences for three measures of affect by test decision: affect expressed during the consultation (SLIWC); decisional conflict measured after the decision was made (T1); anxiety measured before and after the decision making consultation (STAI - consent form and T1). Significant differences were observed for measures of anxiety only, both before and after the information giving consultation (tables 7:4 and 7:5). The women choosing to have no further testing had lower anxiety before and after the consultation than the women deciding to have a diagnostic test. Further, the anxiety of the 'no test' women was significantly lower after the consultation, whereas that of the 'test' women remained consistently high;
- The women deciding to have a diagnostic test had shorter consultations and perceived the information to be more useful than the 'no test' women (tables 7:11). Perceived directiveness of consultation information was not associated with test decision (tables 7:12 and 7:13);
- There were several measures assessing cognitive mechanisms: informed decision making assessed during the consultation; expressed cognitive or 'thinking' words during the consultation (SLIWC); knowledge; perceived advantages and disadvantages of testing; perception of risk; expected-utility values associated with the consequences of testing; ability to rank order the EUVs. The preliminary analyses suggested a number of significant differences in these measures by test decision (tables 7:6 - 7:10). The 'no test' women provided more reasons for choosing an alternative, were more likely to be able to rank order the EUVs, more likely to view their triple test risk as a medium risk, less likely to perceive others as having an expectation towards them having diagnostic test, more likely to perceive

that they would have a healthy baby, less likely to perceive that they would have a child with Down's syndrome and less likely to perceive that they would terminate for Down's syndrome than the 'test' women. No significant differences by test decision were observed for measures of knowledge.

Following these preliminary analyses, nine variables were selected for inclusion in the stepwise discriminant function analysis. From these variables, the discriminant function analysis selected four items for inclusion in the final equation predicting the women's testing behaviour, three assessing cognitive mechanisms and one measure assessing affect. The pattern of responses elicited from the following items contributed to the one function accounting for 52% of the variance in women's prenatal diagnostic testing behaviour: perception of others' attitude to testing; expected-utility of having a healthy baby; expected-utility of having a baby with Down's syndrome; post-consultation anxiety.

As previously discussed, the main advantages of this study were (a) the assessment of both the decision to have and the decision to not have prenatal diagnosis, (b) the inclusion of measures to assess cognitive mechanisms concurrently with and retrospectively to the making of the prenatal diagnosis decision, and (c) the use of decision making theories to inform the development of measures. In addition, the women participating in this study were making the prenatal diagnosis decision following receipt of a screen positive triple test result, i.e. the same reason for being offered prenatal diagnosis. The methodological rigour of this study is an important consideration when interpreting the findings. For example, although this study found no association between the test decision and demographic characteristics, reproductive history and individual differences, prior empirical research has observed such associations (Sjorgen and Uddenberg, 1988; Evans, Pryde, Evans and Johnson, 1993; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994; Halliday, Lumley and Watson, 1995). In addition, the discriminant function analysis applied to this data set accounted for about twice as much of the variance in women's behaviour as that of previous studies (Marteau, Johnston, Kidd, Michie and Cook, 1992). It is likely that methodological limitations, such as assessing the factors associated with only one of the decision alternatives and/or the reasons for being offered prenatal testing, contributed to discrepancies in the findings between prior research and the results of this chapter. However, the following section of this discussion focuses on describing the similarities between the findings of this study with those previously reported from empirical investigations of women's prenatal testing behaviour.

Most empirical research in this health context has reported the high levels of anxiety experienced by women when undergoing prenatal diagnosis (Beeson and Golbus, 1979; Verjaal, Leschot and Treffers, 1982; Burton, Dillard and Clark 1985; Marteau, Kidd, Cook, Michie, Johnston, Slack and Shaw, 1992; Green, Stratham and Snowdon, 1992; Evans, Pryde, Evans and Johnson, 1993). In addition, anxiety has been previously identified as a factor in predicting prenatal screening test behaviour: women refusing serum screening had lower anxiety levels than those accepting screening (Marteau, Johnston, Kidd, Michie and Cook, 1992). Anxiety has been identified as one of four predictor variables discriminating women's decision to have or not have prenatal diagnosis in this chapter's study (section 7.2.4). In particular, the discriminant function analysis identified post-decision anxiety (T1) as the only measure of affect to predict group membership for this data set (table 7:14). Further, the 'no test' women did have lower anxiety than those deciding to have a diagnostic test (table 7:4). However, the 'no test' women also had lower anxiety than the 'test' women before the information giving consultation. A number of explanations to account for these findings in anxiety are described in more detail below.

First, it is likely that anxiety assesses affect associated with the procedure rather than the difficulty in making the prenatal diagnosis decision (Marteau, Kidd, Cook, Michie, Johnstone, Slack and Shaw, 1992). This explanation is supported by (a) the finding that it was the post-decision measure of anxiety that distinguished the two test behaviours, i.e. after the decision was reached and before the test procedure was carried out, and (b) the finding that no measures of decisional affect differed significantly by test decision, i.e. both those choosing to have or not have the test found the decision making equally difficult (table 7:4). In other words, the 'no test' women's anxiety decreased after the receipt of a screen positive triple test result and information giving consultation because they were not going to have an invasive procedure. The 'test' women were as anxious after the information giving consultation as upon receipt of their screen positive triple test result because they were going to have an invasive procedure.

Second, there is some evidence to suggest that women may come to the information giving consultation with an idea of whether or not they want to have further testing (Verjaal, Leschot and Treffers, 1982; Sjorgen Uddenberg, 1988). Indeed, the findings reported in chapters five and six suggest that about a third of women discussed the prenatal diagnosis decision before the consultation. If anxiety is associated with the

diagnostic test procedure, then it is likely that the 'no test' women would have lower anxiety before the information giving consultation than the 'test' women. However, as the assessments of affect and cognitive mechanisms are cross-sectional, there is at least one more explanation for these findings.

The third explanation suggests that there is a relationship between affect, decision making processes and decision behaviour (Bell, 1982; Loomes and Sugden, 1982; Tymstra, 1989; Van der Pligt, 1998). The 'no test' women had a high level of anxiety but lower than that of the 'test' women (table 7:4). The relationship between anxiety and decision making is indirect, with anxiety being associated with differences in information processing; raised levels of anxiety result in a more systematic or vigilant evaluation of the information but high levels result in a more heuristic, less exhaustive appraisal of the information (Janis and Mann, 1977). It is feasible to suggest that anxiety was indirectly associated with the final decision through differences in the underlying cognitive processes. Certainly, the correlations carried out following the discriminant function analysis indicated an association between anxiety and measures of cognitive mechanisms (table 7:15). In addition, the findings from the preliminary analyses suggest that the 'no test' women tended to make more rational (table 7:9) and informed decisions (table 7:6) than the 'test' women; more 'no test' women were able to rank order the consequences of prenatal testing and provide reasons for alternatives than the 'test' women. It is likely that, in part, all the explanations provided for differences in anxiety by test decision are correct. However, the cross-sectional design of this study does not lend itself to a more comprehensive exploration of the nature of these relationships. In other words, it is beyond the scope of this thesis to provide a more detailed explanation of the association between anxiety and women's prenatal testing behaviour.

The remaining predictor variables selected by the discriminant function analysis were items assessing cognitive mechanisms: perceived social norm; EUV associated with having a healthy baby; EUV associated with having a baby with Down's syndrome (table 7:14). In previous empirical research, perceived importance of others' attitudes to serum screening has been associated with the women's test decisions (Sikkink, 1990). In this study, the 'no test' women had higher perceived social norm scores than the 'test' women. That is to say, the 'test' women were more inclined to agree with the statement 'those who are important to me think I should have prenatal diagnosis' (figure 7:1). There is some debate as to what the concept of perceived social norm is assessing (see Trafimow, 1998). Recurrent in a number of theories is the idea that the individual's

perception of others' attitudes to the behaviour and their evaluation of their own beliefs about the consequences of an action are conceptually distinct, with both concepts being independent predictors of the behaviour. The counter-argument suggests that perceived social norms and evaluations of individual beliefs are measures of the same underlying construct, both being measures of attitude. Although it was beyond the remit of this study design to adequately address this issue, the correlation matrix carried out after the discriminant function analysis suggests that perceived social norm was independent of the other cognitive mechanisms and, therefore, an independent predictor variable (table 7:16). More detailed explanations for the findings pertaining to the cognitive predictor variables following the discriminant function analysis are described below.

Previous empirical research provides at least one explanation for why women choosing to have a diagnostic test perceive others' attitudes to testing differently from those women deciding to have no further testing. It has been argued that women find it difficult to refuse a medical technology, such as prenatal diagnosis, when it is offered to them (Tymstra, 1989; Clark, 1990; Marteau, Johnston, Kidd, Michie and Cook, 1992; Gregg, 1993 Lippman, 1994). To explain further, it is feasible to suggest that health professionals are the 'experts' in the context of health care. Therefore, if a health professional offers prenatal diagnosis then this technology is endorsed by the act of offering it. In other words, having a prenatal test is perceived as the normative behaviour. There is some evidence to suggest that health professionals do perceive the 'correct' decision to be that of having a diagnostic test (Farrant, 1985; Marteau, Johnston, Kidd, Michie and Cook, 1992; Marteau, Plenicar and Kidd, 1993; findings in chapter four). There is also evidence to suggest that women are aware of this health professionals' attitude (Sjorgren and Uddenberg, 1988). In consequence, choosing to have no further testing requires a more proactive refusal or 'opting out' by the woman. The findings from this chapter's analyses support this idea of women pro-actively refusing prenatal diagnosis rather then choosing the no test alternative. The analysis of the transcripts described in this chapter found that the 'no test' women generated more reasons not to have testing than the 'test' women did to have a test.

The two final predictive variables identified by the discriminant function analysis were the EUVs associated with the consequences of having a healthy baby and having a baby with Down's syndrome (table 7:14). The EUVs associated with both having a baby with Down's syndrome or having a healthy baby were higher in the 'no test' women than those of the 'test' women (table 7:7). To provide a clearer understanding of these findings, the perception of risk component of these EUVs was looked at independently.

The pattern of responses described in figures 7:2 and 7:3 suggested that the 'no test' women perceived their risk of having a healthy child as higher and their risk of having a baby with Down's syndrome as lower than the 'test' women both the 'test' and 'no test' women. These results support prior empirical evidence describing differences in women's perception of risk by test decision (Volodkevich and Huether, 1981; Verjaal, Leschot and Treffers, 1982; French, Kurczynski, Weaver and Pituch, 1992; Marteau, Johnston, Kidd, Michie and Cook, 1992; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994). It is likely that on evaluating the decision information, the 'test' women minimised their likelihood of having a healthy baby and augmented the risk of having a baby with Down's syndrome (Croyle, Sun and Mart, 1997), i.e. augmenting the Down's syndrome risk and decreasing their likelihood of having a healthy baby. This interpretation is further supported by the finding that the 'test' women were more likely to rate their triple test risk as high, whereas the 'not test' women were more likely to perceive their risk as medium (table 7:10). These findings are consistent with the previous assertion that women choosing to have no further testing were making more informed decisions about the test decision. The 'no test' women were more likely to generate reasons for an alternative, rank order the EUVs associated with the consequences of testing and had a more 'accurate' perception of their triple test risk than 'test' women.

Finally, this chapter's results provide some empirical support for the explanation provided in chapter six to understand the lack of association between the decision analysis intervention and women's prenatal diagnosis decisions. As mentioned in chapter six, decision analysis is predominantly an information manipulation intervention. The aim of applying the decision analytic technique to a real-world decision is to encourage a more balanced appraisal of the decision information, a more accurate perception of the information and the opportunity to integrate this information with existing attitudes and beliefs. In the long-term, an information giving consultation structured by decision analysis may lead to a more accurate or complete representation of the decision information. The empirical findings described in this chapter suggest that the main predictors of women's testing behaviour identified by the discriminant function analysis were EUVs, i.e. a combination of attitudes and evaluations of risk. It is likely that the systematic evaluation of decision information may lead to the development of stable attitudes that are, in the long-term, associated with decision behaviour (see Eagly and Chaiken, 1993; Olson and Zanna, 1993; Stroebe and Stroebe, 1995). In other words, it is possible that decision analysis may be indirectly associated with decision behaviour through mechanisms associated with attitude change and development.

However, it is beyond the remit of this thesis to explore this ascertain and it remains, therefore, an empirical question.

7.4 Chapter summary.

In summary, the findings from this study support those of previous empirical research that suggest that perceptions of risk and attitudes were predictive of women's prenatal diagnosis decisions. More specifically, the discriminant function analysis applied to this data set identified four predictor variables of women's behaviour: post-decision anxiety (T2); perceived social norm; EUV associated with the consequence of having a healthy baby; EUV associated with having a baby with Down's syndrome. In general, the empirical evidence suggests that women choosing to have no further testing were making more informed decision about prenatal diagnosis than women choosing to have a diagnostic test. There was no association between demographic characteristics, reproductive history, individual difference measures, decisional affect and knowledge, and women's decision to have or not have prenatal diagnosis.

8. Discussion.

The focus of this thesis has been on understanding women's decision making in the context of prenatal diagnosis for Down's syndrome. One of the main objectives for those offering prenatal diagnosis is to ensure women make informed choices about testing (Royal College of Physicians, 1989). Over the last twenty years, empirical research has described women's prenatal diagnostic test experiences and the factors associated with the decision to have or not have testing (see chapter one). Few, if any, studies have defined and operationalised informed decision making (Marteau, 1995b; Bekker et al, 1999). Further, most studies in this health context have employed pro- or retrospective designs to address their research questions (section 1.3.2.2). Assessing informed decision making requires an evaluation of the decision making process, i.e. the information utilised when making the decision (section 1.1.5). In this health context, the most appropriate method to access the process of decision making is to use an information tracing technique (chapter five). In other words, studies assessing informed decision making should employ an observational design.

As mentioned, the previous empirical research has focused on describing the factors associated with women's prenatal testing behaviour (section 1.3.2.1). The findings suggest that women make decisions by weighing up the advantages and disadvantages of the testing alternatives. In particular, it has been observed that women's attitudes towards and risk perceptions of the testing consequences are associated with the decision to have or not have prenatal diagnosis. In addition, there was evidence that the final decision about testing was influenced by characteristics of the health professional (Verjaal and Treffers, 1982; Sjorgren and Uddenberg, 1988; Julian-Reynier, Macquart-Moulin, Moatti, Aurran, Chabal and Ayme, 1994). This finding and those describing women's varied and often incomplete knowledge of testing (Volodkevich and Huether, 1981; Farrant, 1985; French, Kurczynski, Weaver and Pituch, 1992) have been used to support the assertion that not all women were making fully informed decisions about prenatal diagnosis. To explain further, as women's knowledge of testing was poor and health professionals' characteristics were associated with the prenatal diagnostic choice, it was unlikely women had sufficient information to make an informed choice and likely that their autonomy to reach a decision was compromised. However, there were a number of methodological concerns that may restrict the interpretation of these previous findings (section 1.3.2.2): few studies assessed the decision making process; most studies employed a retrospective design, i.e. after women have made the prenatal

diagnosis decision; some study samples included women who had either had or not had the test, i.e. factors associated with the choice of one test alternative only; the study sample included women offered prenatal diagnosis for a number of different reasons such as increased maternal age, genetic predisposition, MSAFP screening or triple test screening, i.e. a heterogeneous sample. In essence, these studies may not have adequately defined the decision context, the responses of women are likely to be affected by issues of recall bias and the psychological processes described may not be representative of women choosing to have and not have prenatal diagnosis.

The empirical studies described in this thesis were designed to address some of the methodological limitations of prior research that assessed women's prenatal diagnosis decision making upon receipt of a screen positive triple test result. The research questions clustered around two main themes (chapter three), describing the conditions and criteria associated with women's informed decision making about prenatal diagnosis and understanding the psychological mechanisms associated with women's risky decision making. Both observational and questionnaire based designs were employed to assess decision making factors concurrently with and retrospectively to the women's testing choice. The empirical research was summarised in four chapters of this thesis (chapters four to seven). Within the following sections of this chapter, the findings from these studies are discussed with reference to the research questions outlined in chapter three. First, the conditions enabling women to make informed prenatal diagnosis decisions and the criteria of informed decision making are explored (originally mentioned in chapters four and five). Second, the effectiveness of decision analysis to facilitate informed decision making is assessed (chapter six). Third, the effectiveness of decision analysis to facilitate better decision making is discussed with reference to measures of behaviour, affect, cognitive mechanisms and clinical quality (chapter six). Fourth, the psychological strategies and factors associated with women's decision making are described (chapters five and seven). The final section of this thesis summarises the limitations of the analyses carried out in chapters four to seven, the directions for subsequent empirical work and the clinical application of these findings to the prenatal testing context.

8.1 Describing informed decision making: provision and utilisation.

The conditions to enable women to make informed prenatal decisions have been well documented in a number of clinical guidelines and texts (Emery, 1984; Royal College of Physicians, 1989; Royal College of Obstetricians and Gynaecologists, 1993; Clarke, 1994). In essence, it is the responsibility of the health professional to provide women

with sufficient information to be able to make the decision; this information should be neutrally communicated to encourage women's assimilation of the information without undue pressure from the health professional (section 1.2). As discussed in chapter one, there was no one definitive set of guidelines describing the information considered sufficient for women making the prenatal diagnosis decision upon receipt of a screen positive triple test result (section 1.3.1). In consequence, guidelines for both serum screening and prenatal and genetic testing were referred to when outlining the information considered sufficient for women's decision making in this health context. In brief, the consultation content should include (sections 1.3.1 and 4.1.3.2): a description of the abnormality being tested for; an explanation of the triple test result; an explanation of the consequences associated with having no further testing; an explanation of the consequences associated with having a diagnostic test; an exploration of the decision to terminate or not for Down's syndrome. This information should be imparted non-directively, i.e. without the health professional explicitly stating or subtly implying a desirable course of action. Few, if any, prior empirical studies had assessed whether or not the information provided during the prenatal diagnosis decision following a screen positive triple test result was sufficient and neutrally communicated.

A structured, observational design was employed to ascertain whether or not the conditions enabling women's informed decision had been met in the study described in chapter four. Twenty-two[1] routine information giving consultations between the health professional and woman receiving a screen positive triple test result were audio tape-recorded. The audio tape-recordings were transcribed by a third party at a later date. To assess whether or not sufficient information had been provided, the health professional's utterances of the transcribed consultations were coded. A checklist was developed to classify the health professional's utterances (section 4.1.3.2). The checklist was informed by the literature on the criteria for sufficient and neutrally communicated information. Each of the 22 health professional's transcripts was coded with reference to this checklist.

In brief, the findings were as follows. The information communicated during each consultation was fairly consistent. That is to say, the health professional provided

[1] Data from 128 prenatal diagnosis information giving consultations were used within this thesis. The study in chapter four audio tape-recorded 22 consultations between the health professional and women receiving a screen positive triple test result; the health professional's utterances were analysed. For the RCT reported in chapter six, 106 consultations were audio tape-recorded. Chapter five included the analyses of women's utterances; 22 from the chapter four study and 22 from the RCT. Chapters six and seven analysed the data from the 106 women who participated in the RCT.

different women with similar information. Most women received information about the triple test result, the risks and benefits associated with the diagnostic test alternatives, the option to have no further testing and miscarriage. However, as in associated research (Marteau et al, 1993; Bernhardt et al, 1998), this study observed that termination and Down's syndrome were not discussed with all women (table 4:1). in addition, the health professional provided some misinformation in most of the consultations (table 4:1). In other words, not all women received sufficient or accurate information to enable an informed choice between having and not having prenatal diagnosis. Further, there was evidence that some of the information provided was not neutrally communicated. It was suggested that this information may imply a correct or normative choice regarding the test decision. To explain further, from these directive statements it might be inferred that a woman having the triple test *should* have a diagnostic test upon receipt of a screen positive result which *should* be followed by a decision to terminate for Down's syndrome if it too was positive. In consequence, it is likely that for some women the autonomy to choose between having and not having a diagnostic test was compromised.

As mentioned, one of the main objectives of offering prenatal diagnosis is to ensure women make informed decisions to have or not have a diagnostic test. Within the applied medical literature, few, if any, studies have defined the criteria associated with an informed decision (Marteau 1995b; Bekker et al, 1999). In an attempt to address this dearth of research operationalising informed decision making, criteria for an informed decision were developed with reference to both reason-based models of decision making and medical guidelines for making informed decisions (section 1.2.2). Proponents of reason-based models of decision making are concerned with evaluating the quality of the process of reaching a decision rather than assessing the decision outcome (Zey, 1992; 1998). Two previously defined sets of criteria for making an 'ideal' decision were referred to when operationalising informed decision making for this thesis: Janis and Mann's (1977) seven stage approach to ensuring 'vigilant' decision making; Frisch and Clemen's (1994) three point standard forming the basis of making a 'good' decision (section 1.2.2 and chapter five). In essence, an informed decision requires an evaluation of the consequences of the different options, an accurate assessment of the consequences and evidence that the decision was made following a 'trade-off' between the consequences of the decision alternatives.

An observational study design was employed to assess whether or not women were making an informed decision to have or not have prenatal diagnosis (chapter five).

Forty-four routine information giving consultations between the health professional and woman receiving a screen positive triple test result were audio tape-recorded. The audio tape-recordings were transcribed by a third party at a later date. To assess whether or not women were making informed decisions, the information utilised by women during the information giving consultation was coded, i.e. an information tracing technique was employed. In consequence, all utterances made by women during the consultation were transcribed and classified. A coding frame was developed to categorise women's utterances (section 5.1.4). The coding frame was informed by theories of individual's decision making, findings from prior empirical research assessing women's prenatal diagnosis decision making and categories developed specifically to classify information generated during this health context. From the classification of information utilised by women during the information giving consultation, four measures of informed decision making were created (section 5.1.5): a global measure of informed decision making based on the utilisation of information about the two alternatives and three 'negative' consequences associated with prenatal diagnosis (IDM-T); a variable coding information-seeking utterances about the consequences of testing utterances (IDM-I); a variable coding the generation of reasons for selecting a decision alternative (IDM-F); a variable coding the generation of reasons against selecting a decision alternative (IDM-A).

In brief, the findings suggested that few women utilised all the information considered necessary to make an informed decision. Only 5% (2/44) of women utilised information pertaining to all five themes included within of the IDM-T measure; most women (63%) utilised information from just two or three of the themes (table 5:3). The pattern of results suggested that women were using the consultation to search for further information about the decision alternatives and to make trade-offs between the consequences (table 5:4). However, the findings indicated that not all the information considered necessary to make an informed decision was surveyed, evaluated and utilised during the consultation. In particular, a significant minority of women did not refer to the 'no test' alternative, miscarriage or the condition of Down's syndrome.

These findings lend support to the assertion from previous questionnaire-based studies that women were not making informed prenatal diagnosis decisions. Certainly, there was evidence to suggest that the conditions required to enable informed decision making were not met during every information giving consultation; not all women received sufficient information to enable informed decision making and aspects of the information provided were directively communicated. In addition, the process tracing

technique illustrated that not all women utilised the information considered sufficient to ensure the final decision was based on an evaluation of all the consequences of the decision alternatives. In consequence, it is feasible to suggest that the objective of ensuring women make informed decisions about prenatal diagnosis is not being fulfilled.

8.2 Facilitating informed decision making.

The evidence reported in both this thesis and previous empirical research indicated that some women were not making fully informed decisions about testing. In research published to date, few studies have adequately operationalised informed decision making. In consequence, whether or not informed decision making can be facilitated has remained an empirical question (O'Connor, Llewellyn-Thomas and Drake, 1995; Bekker et al, 1999). Decision analysis is a decision aid based on EUT that targets both the provision of information and the assimilation of information by the decision maker. Although decision analysis has been applied in the context of prenatal diagnosis decision making (Pauker and Pauker, 1977; 1978; 1987), there is little evidence to suggest that a decision structured by decision analysis leads to a better or more informed decision than one made unaided (O'Connor, Llewellyn-Thomas and Drake, 1995; Bekker et al, 1999).

A randomised control trial was carried out to assess the effectiveness of decision analysis to facilitate women's decision making. All women receiving a screen positive triple test result over a fifteen month period were considered for inclusion in the study (section 6.1). Upon opening a previously numbered, sealed, opaque envelope, women agreeing to participate in the study were randomised to one of two information giving consultations: an information giving consultation providing routine care for the LGI; an information consultation structured by decision analysis. In total, 106 women were randomised to either the routine consultation (n = 56) or the decision analysis consultation (n = 50). All women completed a consent form before the information giving consultation, the consultations were audio tape-recorded and transcribed by a third party at a later date and all women completed a post-decision questionnaire (T1) upon making their prenatal diagnosis decision. Women were sent a follow-up questionnaire (T2) if they received a negative diagnostic test result, a 'normal' scan result and did not miscarry within four weeks of the information giving consultation. In total, 100 women were sent a were sent a follow-up questionnaire (T2) and 68 were returned. Informed decision making was assessed by applying the coding frame developed in chapter five to the transcripts of women's utterances recorded during the information giving consultation. Three of the informed decision making variables

created from the categorisation of women's utterances were used in between group differences: IDM-I, information seeking; IDM-F, generation of reasons for alternatives; IDM-A, generation of reasons against alternatives.

The findings from the RCT suggest that women randomised to the information giving consultation structured by decision analysis did make more informed decisions than those receiving the routine information (section 6.2.3.2). To explain further, the process tracing technique and subsequent coding frame enabled the classification of information utilised by women when making the prenatal diagnosis decisions. The informed decision making measures were summaries of the frequencies of women's utterances categorised by the coding frame. The results of the MANCOVA (table 6:3) indicated that women in the decision analysis group were more likely to search for both reasons for ($f = 5.24$; d.f. = 1, 103; $p = 0.02$) and against ($f = 5.33$; d.f. = 1, 103; $p = 0.02$) the decision alternatives than women in the routine consultation. There was no difference by information giving consultation for the information searched during the decision making period ($f = 0.08$; d.f. = 1, 103; $p = 0.78$). In other words, women in both consultations were as likely to ask for additional information about the decision alternatives and consequences. In retrospect, this finding is not surprising as the two consultations were designed to provide the same information content. However, the utilisation of information when reasoning about the decision alternatives and consequences did differ by group allocation. That is to say, decision analysis was associated with a more reasoned or informed decision than women in the routine consultation. From the frequencies described in table 6:1, it is feasible to suggest that the decision analysis consultation provided women with more opportunity to discuss the consequences of miscarriage and termination than was available during the routine information giving consultations. In summary, these findings suggest that the decision analysis technique does enable women to make a more informed prenatal diagnosis decision.

8.3 Evaluating decision analysis.

Few studies have evaluated the quality of decisions made following the application of decision analysis compared with those made unaided (O'Connor et al, 1995; Bekker et al, 1999). In consequence, there is little evidence to indicate whether or not the decision analysis technique does facilitate decision making (section 1.2.1.3 and chapter six). Proponents of decision analysis suggest that the application of decision analysis to real-world decisions will influence the following aspects of decision quality (chapter six): facilitate rational decision making; encourage more accurate evaluations of the decision

information; reduce the amount of affect associated with making the decision; enable the individual to make decisions independent of the other's advice. However, critics suggest that decision analysis will have a detrimental affect on the quality of real-world decision making including: an increase in negative affect as the individual is confronted with unusual and difficult hypothetical questions to elicit utilities; greater decisional conflict as the individual is encouraged to make rational decisions rather than employ more 'natural' information processing strategies; a reduction in the 'personal' or 'human' aspect of the consultation; a needless increase in consultation time; a decrease in the individual's autonomy to make the decision.

There are numerous measures to evaluate the quality of decisions which can be broadly divided into those that assess behaviour, affect, cognitive mechanisms and clinical quality (Llewellyn-Thomas, 1995). In addition to measures of informed decision making, the randomised control trial described in the previous section incorporated measures to assess each of these aspects of decision quality (section 6.1.2). The findings from the RCT are summarised in brief below (see section 6.2 for a more in-depth account):

- ***behaviour*** - no difference was observed for the decision to have or not have prenatal diagnosis by group allocation. Eighteen per cent (10/56) of women in the routine group and fourteen per cent (7/50) of women in the decision analysis group had no further testing (table 6:8).
- ***affect*** - during the consultation women in the decision analysis group expressed fewer positive emotion words ($f = 4.7$; d.f. = 1, 102; $p = 0.03$) but tended to express more negative emotion words ($f = 2.8$; d.f. = 1, 102; $p = 0.09$) than women in the routine consultation group (table 6:5). A repeated measures analysis of decisional conflict indicated that women in the decision analysis group had decreased decisional conflict scores at follow-up, whereas women in the routine group had increased decisional conflict scores (tables 6:29 and 6:30). There was no difference in measures of anxiety by group allocation (tables 6;29 and 6:30).
- ***cognitive mechanisms*** - as mentioned in section 8.2, women in the decision analysis group made more informed prenatal diagnosis decisions. In addition, women in the decision analysis group referred to more 'cognitive' words during the consultation than women in the routine group (table 6:5). A repeated measures analysis found there to be little difference in knowledge scores by group allocation (tables 6:29 and 6:30). However, those in the routine group were likely to generate more advantages to prenatal testing than those in the decision analysis group. The expected utility values associated with the consequence of terminating for Down's syndrome were significantly different by group allocation at both T1 (table 6:12) and

T2 (table 6:24). Women in the decision analysis group had higher EUV scores associated with the termination consequence. There was no difference by group allocation in the ability of women to rank order the five consequences associated with the prenatal diagnosis decision (tables 6:14 and 6:25). There was no difference in women's perception of their triple test risk by group allocation (tables 6:11 and 6:21).

- ***clinical quality*** - the decision analysis consultation was approximately six minutes longer than the routine information giving consultation (table 6:15). Also, women in the decision analysis group were less satisfied with the decision information than women in the routine information giving consultation immediately after making the prenatal diagnosis decision (table 6:15). However, this association was not supported by the findings of the repeated measures analysis (tables 6:29 and 6:30). There was no difference in perceived directiveness of the consultation information by study group (tables 6;16 and 6:26).

One interpretation of these findings is that the application of decision analysis to real-world decisions is associated with positive changes in the quality of the decisions made. To explain further, women randomised to the decision analysis group had higher measures of informed decision making than women in the routine information giving consultation, i.e. the 'decision analysis' women utilised more of the decision information during the information giving consultation than the 'routine' women. In addition, the 'decision analysis' women expressed less decisional conflict about their diagnostic test choice than women in the routine information consultation. There were no differences in the experience of anxiety or women's perceived directiveness of the consultation information by group allocation. In other words, women's decision making experience was not adversely affected following the application of decision analysis. Indeed, the application of decision analysis may be associated with the facilitation of decision quality.

Alternatively, some of the findings from this study's evaluation of decision analysis may be interpreted as detrimental to women's experience of the prenatal diagnosis decision. Women allocated to the decision analysis consultation did have longer consultations, were less likely to use positive emotion words during the consultation, were less likely to generate advantages for prenatal testing and rated the usefulness of information as lower than that of women receiving the routine information. However, whether or not these particular differences by group allocation are negatively perceived is one of interpretation (section 6.3). For example, a health professional providing this

information may perceive these findings as illustrating the inadequacies of decision analysis; the consultation takes longer, women express fewer positive aspects to the process of prenatal testing and women appear to be less enthusiastic about the information offered than those receiving the routine hospital care. On the other hand, a health professional concerned with ensuring women make informed choices about prenatal diagnosis may infer that these findings support the use of decision analysis in this health context; the longer consultation length may indicate a more informed process of decision making and the less favourable expressions pertaining to the experience of prenatal testing experience may reflect a more accurate evaluation of the decision information. It is feasible to suggest that as informed decision was facilitated and affect associated with the making of a difficult decision reduced, the explanation for these findings is consistent with the latter interpretation.

There is little evidence from this study to suggest that the application of decision analysis to a real-world decision was associated with the making of a more accurate or correct decision. Women's appraisal of their triple test risk was not associated with group allocation. Most evaluations of the attitudes towards the decision consequences and likelihood of a consequence occurring did not differ by group allocation. Further, women in the decision analysis group were no more or less likely to rank order the consequences of the test alternatives than those in the decision analysis group. In addition, the structure of the decision analysis consultation was not associated with a greater recall in knowledge of prenatal testing. It is likely that as the study was under powered, type II errors may account for some of the 'none significant' findings. An alternative explanation may be that as this decision context is a particularly well structured risky-decision with serious consequences, women are likely to evaluate the decision information systematically (Ubel and Loewenstein, 1997). In consequence, decision analysis is unable to encourage women to employ a more accurate strategy with which to appraise the decision information. A final interpretation may be that the beneficial effects of the decision analytic technique are long term. That is to say, women's recall of information or evaluation of the decision consequences may be maintained for longer as a result of the elicitation of utilities and/or the structure of the consultation information. However, it was beyond the scope of this thesis to provide evidence to confirm or counteract these explanations.

8.4 Factors associated with women's decision making.

Most, if not all, published studies have assessed factors associated with women's prenatal diagnosis decision making pro- or retrospectively to women's decision making.

Few studies have employed a process tracing technique to describe the strategies employed by women concurrently with the making of the prenatal diagnosis decision. In consequence, it is unclear whether or not the strategies employed by women when making their prenatal diagnosis decisions have been accurately and comprehensively identified. As mentioned, chapter five summarises the findings from a study employing an information tracing technique to describe the information utilised by women when making their decision to have or not have prenatal diagnosis. The pattern of information utilised by women is an indirect measure of the psychological processes employed by women when making the decision (section 5.1.2). The purpose of summarising the information utilised by women during the consultation was to describe the type of information and psychological mechanisms employed by women when making the prenatal diagnosis decision. It was outside the remit of this thesis to operationalise a naturalistic or shared decision making model.

In brief, the results suggested that most (over 70%) women utilised information pertaining to the consequences and alternatives of the prenatal choice (table 5:1). In particular, most women weighed up the advantages and disadvantages of choosing between the two diagnostic tests. Both actual risks associated with the consequences of the alternatives and their evaluations were referred to by most women. Women tended not to verbalise attitudes towards the consequences of testing during the consultation but used reasons for and against alternatives when weighing up the decision information. These findings suggest that women employed systematic strategies when processing the decision information. However, most women also referred to information outside that provided by the health professional during the consultation. To explain further, most women mentioned their own and other's experience of prenatal testing, pregnancy and abnormality. As this information is peripheral to that of the prenatal diagnosis decision, these findings suggest that women also employed heuristic strategies to assimilate information during the consultation. In addition, about 40% of the sample appeared to *screen out* the 'no test' alternative so reducing the amount of information to be appraised. The previously reported finding that not all women were making informed decisions is probably attributable to women employing this screening technique.

The theoretical review described in chapter one summarised the findings from published empirical research investigating women's decision making in the context of prenatal diagnosis. A significant proportion of this prior research has been concerned with identifying the factors associated with the decision to have or not have prenatal

diagnosis. Understanding the psychological processes involved in women's decision making may be useful in either explaining women's responses to the testing experience or informing subsequent interventions aimed at the facilitation of informed decision making (Marteau, 1995a). As mentioned, there are a number of methodological issues pertaining to these studies that may adversely influence the interpretation of findings (section 1.3.2.2). Most of these methodological limitations were addressed by the study designs of the empirical research described in chapters four to seven of this thesis. For example, the study referred to in chapters six and seven included both concurrent and retrospective measures of factors associated with women's decision making, and a sample of women representative of those choosing to have a diagnostic test and those choosing to have no further testing. The findings from these analyses are summarised below.

Unlike findings from previous research, there was no significant association between the decision behaviour and demographic characteristics, reproductive history or individual differences (tables 7:1, 7:2 and 7:3). Of the three measures of affect, decisional conflict, emotional content consultation and anxiety, only anxiety differed significantly by test decision. Those women choosing not to have a diagnostic test had lower anxiety than those having a diagnostic test (table 7:4). The results from the application of the coding frame to the consultation transcripts suggested that women choosing to have no further testing were making more informed decisions than those having a diagnostic test (table 7:6). This result was observed in both analyses of the consultation transcripts described in chapters five and seven. From the frequency table summarising women's utterances during the consultation (table 5:2), it is feasible to suggest that women choosing to have a diagnostic test were more likely to *screen out* the information associated with the 'no test' alternative; whereas women deciding to have no further invasive testing assimilated information about both test alternatives. There was a trend towards significance for an item within the questionnaire (T1) assessing a similar construct ($f = 3.0$; d.f. = 1, 104; $p = 0.09$; table 7:6); 'no test' women tended to generate more perceived disadvantages to prenatal testing than 'test' women. Knowledge of testing was equally high in women choosing to have or not have testing (table 7:6). Apart from the consequence of miscarrying, all EUVs associated with the consequences of testing differed significantly by decision behaviour (tables 7:7 and 7:8). The 'no test' women had higher EUVs associated with having a healthy baby and a baby with Down's syndrome and lower EUVs associated with miscarrying or terminating a baby with Down's syndrome. In addition, there was a trend towards significance ($Chi^2 = 3.1$, d.f. = 1, $p = 0.07$; table 7:9) suggesting that the 'no test' women were more likely to rank order

the consequences of testing than the 'test' women, i.e. more likely to be making rational or accurate decisions. Women choosing to have a diagnostic test perceived their triple test risk as higher than the 'no test' women ($z = 3.6$, $p = 0.003$; table 7:10). The perceived social norm of others' attitude towards testing differed by decision behaviour ($z = 5.1$, $p < 0.001$; table 7:7); 'test' women perceived there to greater external pressure from others to have a diagnostic test, whereas the 'no test' women perceived there to be a more neutral attitude from others to have a diagnostic test. Women's perceived directiveness of the consultation information did not differ by group allocation (tables 7:12 and 7:13). Finally, 'no test' women tended to have longer consultations but found the information to be less useful than 'test' women (table 7:11). From those variables described above, a stepwise discriminant function analysis identified four predictor variables accounting for 52% of the variance in women's decision behaviour (table 7:14): the measure of perceived social norm; anxiety at T1; EUV associated with having a healthy baby; EUV associated with having a Down's syndrome baby.

A number of these results support those found in previous research (see chapters one and seven). First, anxiety has been observed to be higher in women undergoing prenatal diagnosis than those choosing to have no further testing. This finding supports the assertion that anxiety is experienced in anticipation of undergoing an invasive, diagnostic test rather than as a consequence of making a difficult decision (Marteau, Kidd, Cook, Michie, Johnston, Slack and Shaw, 1992). Second, previous empirical evidence described differences in the attitudes and perceptions of risk by decision behaviour (section 1.3.2.1). This pattern of results was observed in the analyses carried out in chapter seven; the 'no test' women were making more informed, rational and accurate decisions than those having a diagnostic test. Further, these differences in attitudes and perception of risk were predictive of women's decision behaviour (see section 7:3 for some explanations of these findings). Although the 'no test' women were making more informed and accurate decisions, they found the decision information less useful than the test women. As discussed in section 8.3, it is likely that the more informed decision is associated with a more accurate appraisal of the decision information. That is to say, women making more informed decisions appear to be satisfied with the information but less so than women employing a screening out information processing strategy. However, this finding may indicate that a more informed decision is a less falsely reassured one. Third, the findings pertaining to women's reasoning during the consultation support the assertion that the 'no test' women are 'opting out' of having a test rather than choosing to have no further testing. A final observation is that although knowledge has been found to differ by test decision

in previous research, the analysis reported in chapter seven found no such association. One explanation for this discrepant result is that this sample of women were well informed about prenatal diagnosis. Two results support this explanation: women's mean knowledge score in both groups was equally high (a mean score of 14.5 from a measure ranging between 0-20); the provision of test information was good if not sufficient to ensure a fully informed decision (chapter four).

8.5 Critique of methods.

The main strengths of the empirical studies described in this thesis were (chapters four to seven): the observational study design; the representativeness of the study sample; the well defined clinical context; the use of psychological models of decision making to enable the operationalisation of informed decision making and inform measures evaluating the decision quality. Below is a discussion outlining some of the methodological strengths of these studies with reference to the empirical research described in this thesis.

1. The application of an observational study design enabled the accurate appraisal of whether or not the conditions and criteria of informed decision making had been met. To explain further, from the information search techniques it was possible to describe the information provided by health professionals and utilised by women during the prenatal diagnosis decision consultation. Both the checklist (chapter 4.1.3.2) and coding frame (section 5.1.4) were informed by psychological theories of decision making and evidence from prior empirical research. It is likely that the good inter-rater reliability of both instruments was, in part, attributable to their thorough development and grounding within the literature.
2. The decision context was well-defined. The information provided to women was consistent between consultations. All women included within the analyses of this thesis were offered prenatal diagnosis as a consequence of receiving a screen positive triple test result. In other words, the reasons for being offered prenatal diagnosis were the same for each woman. All women invited to attend the information giving consultation were choosing either to have or not have prenatal diagnosis. To explain further, the final sample of women were representative of those making the choice between having or not having prenatal diagnosis.
3. The study evaluating decision analysis employed a randomised control trial design with a low risk of bias. The items included within the questionnaires were designed with reference to psychological models of decision making and standardised measures of women's prenatal testing experience (section 6.1.2). The

questionnaires were piloted before inclusion in the main RCT. It is feasible to suggest that this rigorous methodology and well-defined decision context contributed to the validity of the difference in findings observed between the decision analysis and routine care study groups (Bowling, 1997). In addition, it is feasible to suggest that the well-defined decision context and the theoretically driven measures were in part responsible for achieving a function accounting for 52% of the variance in women's decision behaviour (chapter seven). The four predictor variables identified by the stepwise discriminant function analysis (chapter seven) accounted for about twice as much of the variance in women's prenatal testing behaviour than previously published empirical research;

4. The study was carried out in a real-world setting and a concerted effort was made to integrate the research into the routine ante-natal care of the LGI. Women's rate of participation in the study was excellent (84%) and the return rate of follow-up questionnaires (T2) good (68%) (chapter six). These factors suggest that the study sample of women were representative of women making the triple test decision in Leeds.

The combination of study designs and measures employed in this thesis have provided some unique empirical findings for the area of women's decision making in the context of prenatal diagnosis. First, it is likely that informed decision making requires the assessment of the decision making process. Questionnaire-based measures assessing knowledge did not access the information utilised by women when making a decision. However, there was some evidence to suggest that an open-ended question encouraging women to identify perceived advantages and disadvantages of testing might be associated with the process of making the decision. Future empirical research of informed decision making may benefit from developing a questionnaire-based measure of informed decision making. Second, women's informed decision making was facilitated by application of the decision analytic technique to the information giving consultation. In other words, it is possible to operationalise and facilitate informed decision making. Third, women employed both systematic and heuristic strategies when assimilating information to make a decision about prenatal diagnosis. Fourth, women utilised both risk information and evaluations of risk during the prenatal diagnosis decision consultation. Retrospective assessments of the factors associated with women's decision making have implied that only risk perceptions were predictive of women's decision behaviour. Fifth, individual differences in preferences for information and perceived optimism did not appear to be associated with either women's informed decision making or factors predicting their decision behaviour. Sixth, measures of

decisional conflict and anxiety assessed different aspects of affect. It seems feasible to suggest that decisional conflict assessed the difficulty with making the decision, whereas anxiety measured the general affect associated with undergoing clinical procedures. Seventh, more informed decisions were associated with lower satisfaction scores. As the majority of women still perceived the information to be useful, it is possible that this score reflected a decrease in 'false reassurance'. Finally, women choosing to have or not have prenatal diagnosis had different perceptions of others' attitude to prenatal testing. The 'test' women perceived there to be more pressure from others to have a diagnostic test, whereas the 'no test' women's response to the perceived social norm question suggested that there was a more neutral attitude from others to have a diagnostic test. As perceived social norm was measured retrospectively to the decision behaviour, it is feasible to suggest that the response to this item was subject to a cognitive readjustment (section 1.3.2.2). That is to say, women's perceptions of risk and attitudes were, or were being, re-aligned in order to be congruent with their behaviour. To explain further, it might be adaptive for women undergoing the invasive procedure to perceive their choice as being the 'norm' and dependent on others' opinions; acknowledging that the invasive, risky choice was their responsibility alone may be a dissonant cognition. In contrast, it might be adaptive for women choosing the non-invasive alternative to acknowledge their responsbility in selecting the 'non-medical' alterniative. It is possible that knowingly 'going against a norm' is difficult, which might account for the lower perceived satisfaction scores of the 'no test' women.

There were a few limitations of the methods employed in this thesis. First, the randomised control trial mentioned in chapter six was under-powered. As the sample size of this study was small, it suggests that some type II errors may have occurred. For example, it is possible that decision analysis was associated with changes in women's decision behaviour or knowledge despite the analyses in this thesis being unable to establish such a relationship. Second, few studies have evaluated decision analysis to assess its efficacy in the facilitation of informed decision making. In consequence, a large number of measures were employed to evaluate changes in affect, behaviour, cognition and clinical quality. As several measures were analysed, it is possible that some type I errors may have occurred. For example, the results suggested that decision analysis was associated with a decrease in decisional conflict but this finding may have been attributable to chance. Third, although chapter six reported the results of a randomised control trial with a low risk of bias, the study compared only decision analysis with a routine information giving consultation. In consequence, it is unclear

whether or not the facilitation of informed decision making was attributable to decision analysis as a complete package or an attribute of the technique, such as the decision tree visual prompt. Finally, there are some issues concerning the generalisability of the findings described in the empirical chapters of this thesis and the analyses applied to the studies' data. These concerns are discussed in more detail below.

1. The study sample were representative of those women choosing to have a triple test at the LGI; the majority of women were Caucasian and had a moderate level of education (60% had 'A' level qualifications or higher). It is unclear whether or not the findings reported in this thesis would be replicated in a more demographically varied sample of women. In addition, the LGI triple test uptake rate is modest compared with rates of other hospitals nationally. In consequence, it is possible that the decision making processes of women at the LGI might differ from those of women at other UK hospitals.
2. The analyses applied to the data were consistent with the research aims of the thesis. In essence, to describe the factors associated with women's informed decision making about prenatal diagnosis. As mentioned, an observational study design generates a rich source of data. In consequence, more in-depth qualitative analyses might have been useful in trying to understand the relationship between the provision of consultation information by the health professional and women's subsequent utilisation of information when making the prenatal diagnosis decision. At present, it is unclear to what extent the woman discussed information pertaining to the health professional's agenda or vice versa.
3. Ensuring informed decision making is an objective of prenatal testing programmes. The implicit assumption, then, is that informed decision making is desirable or 'good'. In consequence, the analyses of both the consultation and questionnaire-based measures focused on assessing the quality of the decisions made, i.e. whether or not the decisions were informed. However, no analyses were carried out to establish the relationships between measures. For example, ascertaining whether or not informed decision making is always 'good' by assessing the relationship between informed decisions and affect or satisfaction. In addition, these analyses may be useful in addressing more specific research questions about factors associated with the decision making process, such as the relationship between raised anxiety and vigilant information processing (Janis and Mann, 1977). Although the evidence to test these associations exists within the current data set, it was beyond the remit of this thesis to carry out the appropriate analyses.

4. The randomised control trial study was carried out to evaluate the effectiveness of decision analysis to facilitate decision making. The findings suggested that women in the decision analysis consultation did make more informed decisions than women in the routine care consultation. However, the decision analysis consultation was not associated with changes in rational or accurate decision making. One interpretation of these findings was that decision analysis facilitates informed decision making but not rational decision making. Another explanation may be that the decision analytic technique was inappropriately carried out. To explain further, the lottery question that is used to elicit utilities within the prenatal testing context provides a global value or utility figure. It is possible that the use of an alternative elicitation method may provide individual utility values. An association between the application of the decision analytic technique and more accurate or rational decision making may be associated with this different elicitation of utilities.
5. As mentioned, informed decision making was facilitated in women randomised to the decision analysis consultation. However, the randomised control trial compared only the decision analysis consultation with a routine care consultation. It is unclear whether or not women's informed decision making would be facilitated by a consultation structured by an alternative information intervention. For example, genetic counselling research suggests that the use of scenarios to structure the decision information is a useful technique (Clarke, 1994). It remains an empirical question, then, whether or not a decision aid is more likely to facilitate informed decision making than an information aid.

Although some of these methodological concerns and new empirical questions may be addressed by secondary analyses of the data collected for this thesis, additional empirical research is required to adequately respond to its limitations. Further studies might focus on a number of issues raised from the findings in this thesis including: carrying out the research in a more varied sample of pregnant women; developing a questionnaire-based measure of informed decision making; evaluating decision analysis in a different health context; assessing whether or not informed decision making can be facilitated by the use of an information aid.

8.6 Clinical recommendations.

The empirical evidence reported in this thesis suggested that (a) the information provided by health professionals is not sufficient to enable informed decision making, (b) the women were not utilising sufficient information to ensure their final decision was informed, and (c) the application of decision analysis to the prenatal diagnosis

consultation was associated with an increase in informed decision making. In addition, the randomised control trial evaluating decision analysis found that the women in the decision analysis group experienced less decisional conflict about their prenatal diagnosis choice than those the routine group. There was also some evidence to suggest that the women in the decision analysis group were less falsely reassured by the consultation information than those in the routine consultation. The two adverse consequences of applying decision analysis to the prenatal diagnosis consultation were that the decision analysis consultations were approximately six minutes longer than routine consultations and the women were less satisfied with the consultation information than those in the routine group.

One of the main objectives of prenatal diagnosis programmes is to encourage women to make informed decisions. As decision analysis was associated with an increase in informed decision making, it is reasonable to suggest that the decision analytic technique be applied to the prenatal diagnosis information giving consultation. However, there are a number of issues that are likely to impede the implementation of this technique to the real-world context. First, health professionals would have to receive additional training to be able to structure a consultation by decision analysis. Second, the application of the decision analytic technique requires more time than routine information giving. Third, women are less satisfied with the decision analysis consultation. It is unlikely that health professionals would adopt a technique that requires re-training, is more time-consuming and is perceived as less useful by the woman, on the basis that it facilitates informed decision making. In consequence, further research is required to establish what aspect of the decision analysis 'package' facilitates informed decision making. To explain further, decision analysis may have increased informed decision making for the following reasons: the decision tree visual aid encouraged the health professional to provide sufficient information; the decision tree visual aid reduced the cognitive demands of the decision and/or prompted the woman to utilise sufficient information during the consultation; the lottery question provided women with an opportunity to discuss termination; the threshold graph enabled women to assimilate information about attitudes and evaluations of risk. The first three reasons suggest that an information aid is sufficient to enable informed decision making. The final reason suggests that a decision aid is necessary to facilitate informed decision making. Before recommendations for changes to clinical practice can be confidently asserted, further empirical investigation is required to assess which of these interventions or combinations of interventions might be associated with the facilitation of women's informed decision making.

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Zung, W.W.K. (1965) A Self-Rating Depression Scale. *Archives of General Psychiatry.* **12:** 63-70.

Appendix I

Informed decision making: an annotated bibliography and systematic review - executive summary.

Objectives

To provide an unbiased bibliography of controlled studies evaluating interventions that may affect informed patient decision making.

To classify studies by research design, decision-making theory, type of intervention and health setting.

To describe the measures of informed patient decision making and other outcomes reported.

To identify under-reported areas and direct future research.

Conducting the review

Study selection: Studies were included if they reported the results of a controlled study of any intervention using real patients making a health decision. Specifically, randomised controlled trials (RCT), non-randomised concurrent studies, historical studies, and same subject "before and after" studies were included. Health decisions were defined to include any reported health behaviour change as well as explicit decisions. Interventions were defined broadly to include any that could reasonably be expected to affect informed decision making, such as changes in information provision, cost, or service provision. Patients were defined as any individual making a decision about health care. Experimental studies on healthy student volunteers, studies of health professionals making decisions about another individual's care and articles not published in English were excluded.

Data classification: Articles were classified by study quality according to the hierarchy of evidence, underlying theory, the domain of health care, and the health decision. The comparison groups, other factors associated with the decision making process, reported measures, and a summary of the findings were recorded.

Data sources: The electronic databases Medline, BIDS (social science), and Psychlit were searched for 1991-1996. The journals Medical Decision Making, Patient Education and Counselling, and Preventive Medicine were hand searched for 1986-1996.

Article identification: Abstracts were assessed by the first author and articles retrieved if the review criteria were met. Final inclusion decisions was made by the first author and verified by another member of the project group.

Data extraction: This was performed by a member of the project group and checked by the first author with disagreements resolved by discussion.

Data synthesis: Descriptive summaries and qualitative analysis were performed. The health domains and decisions were too diverse for meaningful quantitative meta-analysis.

Review findings

Following hand-searching and abstract evaluation 825 articles were distributed to the project group members and 547 were subsequently included within the bibliography.

Study quality: There were 336 RCTs, 114 non-randomised concurrent studies, 34 historical, and 63 before and after same sample studies. Only 51 of the RCTs were classified as having a low risk of bias. 267 studies claimed to have approached a representative sample of participants, but only 243 reported the number invited to take part. Few studies provided adequate descriptions of the intervention materials.

Theoretical context: 206 studies referred to an underlying theory. Of these, 101 referred to theories explaining decision making such as expected utility theory, prospect theory or social cognition models.

Health domain and the decision: 251 studies were in general medicine, 114 cancer, 108 genitourinary medicine, 61 primary care, 31 paediatrics; 15 mental health, 10 dentistry, 11 surgery, 7 genetics, and 31 OBGYN and midwifery. The decision was classified as a life-style change in 357 studies, a screening decision in 114, a treatment decision in 107, a decision to participate in the consultation in 51, and as another type of decision in 26.

Interventions: 301 interventions were of information provision itself, 273 varied the delivery of information, 208 provided patient feedback, 94 manipulated information in some other way, 55 prompted active patient participation, and 89 of another intervention altogether.

Decision making factors: 512 studies assessed actual rather than hypothetical decisions, 476 involved decisions affecting the participant rather than a third party and in 525 studies the decision was made without time pressure. Only 26 studies explicitly made patients aware of their involvement in the decision making process.

Measures assessed: demographic details were recorded in 515 studies, knowledge in 181, decision making measures in 169, measures of affect in 69, satisfaction in 60, self efficacy in 75, personality trait in 20 and other variables in 111 studies.

Trajectory of knowledge: The annual number of included studies increased by approximately 50 percent over the five years 1991-96 but the proportion of better quality remained unchanged.

Summary result: Only five studies were *theory* driven, assessed measures associated with *informed* decision making **and** used a low risk of bias design. Although of disparate design these five studies suggest that information and education are relatively ineffective ways of facilitating informed decision-making, compared with the context and social influences. Studies reporting manipulation of information, and provision of feedback, were the most likely to report an effect.

Conclusions

There is a paucity of well designed, theoretically driven and adequately operationalised research assessing informed patient decision-making. Although additional information does not necessarily lead to more informed decisions, this was the most frequently coded intervention. Less than a fifth of studies assessed information manipulation or active patient participation. Informed patient decision making was under-evaluated with less than a third of studies assessing any measure associated with the process of informed decision making. No broad conclusions could be drawn from the five 'good quality' studies as the theoretical context, health domain, decision and measures were disparate. Given the small number of high quality studies and the relatively slow increase in research in this area there is no need for the NHS to revisit this topic as a review for five years. Resources should be concentrated on better primary research.

Recommendations

1. Future primary research should work under an explicit theory of decision making, record process measures to permit evaluation of whether the decision was informed and if evaluating experimental interventions use randomised trials with a low risk of bias.

2. The HTA programme should develop a booklet describing the main decision making theories, and an inventory of suitable outcome measures, to help clinical researchers design appropriate studies.

3. Three complementary systematic reviews should be commissioned.

The effect of interventions on patient preferences. At least 50 trials were excluded from the present bibliography because no behaviour change was recorded.

Observational studies of real patient decision-making. Studies using tape-recorded consultations, verbal "thinking aloud" protocols, and other written or computer based "process tracing" methods will predominate.

Assessing the effect of additional information, manipulation of information, provision of feedback, and group delivery of information on informed patient decision making.

4. Primary research is a priority such areas as genetics, prenatal diagnosis and where decisions are often made by proxy such as paediatrics and mental health.

5. Primary research is required to evaluate the following types of interventions

Decision aids, such as graphical and computer based devices.

Information manipulation, such as decision analysis, prompts, and feedback.

Appendix II

Coding form to extract article information: IPDM review.

Article Identification Details (reference details from electronic data-base - *idealist* - review database)

Field	*Details*
Article review number number studies project number number of duplicate	....
Authors	.....
Title	.....
Journal Details {name,year,vol., pages}	.....
Code 1-6 (reasons exclude)	[ ] .....
Database	medline ☐ psychlit ☐ BIDS ☐ handsearch ☐
Date downloaded Date sent to reviewer Date returned reviewer	_____ / _____ _____ / _____ _____ / _____
Initials Reviewer	MA ☐ HB ☐ JC ☐ JH ☐ JM ☐ MM ☐ SM ☐ AP ☐ MR ☐ JT ☐

Summary decision/health behaviour		
Location research: unknown ☐ UK ☐ North America ☐ S.Africa ☐ Australia/ NZ ☐ other ☐ (please write ....)	**Participants:** adult ☐ well ☐ pregnant ☐ physically impaired ☐ elderly (>65) ☐ mentally impaired ☐ child (<17) ☐ non-English speaking ☐ unknown ☐	**The nature of the decision is:** A) Real ☐ Intended ☐ Hypothetical ☐ B) Implicit ☐ Explicit ☐ C) affects - Participant ☐ other ☐ D) made at - time intervention ☐ later ☐
Domain of Health Care: Write in the type of illness or health domain that the research located within (eg. cervical cancer, breast cancer, asthma, general health promotion, cardiovascular risk factors, HIV/condom use, smoking cessation, drug use, tuberculoses, maleria, etc.)	**Defining type of health decision:** Write in the type of therapy decision below (eg. screening, diagnostic testing, medication/drug therapy, surgical treatment, behavioural therapy, willingness to pay for..., donating ..., taking part in research, cessation, etc.,)	**Authors Awareness theory:** ***Decision making theory:*** 0 = no theory referred to Write in the name of theory referred to in the introduction or methods section ....
Quality of Study: ***Design of study:*** randomised (RCT) a ☐ b ☐ c ☐ non randomised concurrent ☐ before/after different sample ☐ before/after same sample ☐ Other (eg. matched case-controlled,) ☐	**Quality of Study Continued...** ***Level of intervention:*** patient level ☐ other level ☐ not rec.☐ ***Sample invited to participate:*** total 'population' available ☐ stratified/ systematic sample ☐ volunteer/ non-systematic sample ☐ not adequately described ☐	**Quality of Study Continued...** ***Sample size:*** total number available _____ not rec. ☐ total number invited _____ not rec. ☐ number participated _____ not rec. ☐ number excluded _____ not rec. ☐ number in final analysis _____ not rec.☐
Quality study continued... ***Causes for concern:*** no/ not obvious causes for concern ☐ yes (please state any concerns including study poorly designed, sample not representative, bias, etc) ☐	**Quality study continued...** ***Development intervention materials:*** piloted some aspect of intervention: no ☐ yes ☐ in part ☐ piloted some aspect measures no ☐ yes ☐ in part ☐ applied a readability score: no ☐ yes ☐ in part ☐	**Reference to be accessed . . .**

Description Intervention Groups

Nature Intervention:	explicit information ☐ change in service ☐ other ☐ ....

Comparative Group	Description of Intervention (write . . .)	Code (see box 1)	number in group (n)
Group 1	....	[ ] [ ] [ ] [ ] [ ] [ ]	
Group 2	....	[ ] [ ] [ ] [ ] [ ] [ ]	
Group 3	....	[ ] [ ] [ ] [ ] [ ] [ ]	
Group 4	....	[ ] [ ] [ ] [ ] [ ] [ ]	

Other Independent, Moderator or Mediator Variables

Variable	Code	Descriptives	Main Effect Analysis	Interaction Analysis
age	101			
gender	102			
level education	103			
social class/ income	104			
marital status	105			
ethnicity	107			
anxiety	151			
depression	153			
other affect	158			
knowledge	159			
attitudes/ utilities	160			
perception risk/ severity	161			
other variables (write) . .				

Main Decision Making Outcomes Assessed

Description (write & code - box 3)	Continuous/ Categorical	Validated, Piloted or Study Specific	Description validated measure (write):	Code val. measure
		val ☐ pil ☐ study ☐		
		val ☐ pil ☐ study ☐		
		val ☐ pil ☐ study ☐		
		val ☐ pil ☐ study ☐		
		val ☐ pil ☐ study ☐		

Results of Intervention on *Main* Outcome variables

Code variable	Categorical only level / group (box 5) *eg adhere 1* *not adhere 2*	Group 1 *not available or* *'n' or* *mean (sd) n*	Group 2 *not available or* *'n' or* *mean (sd) n*	Group 3 *not available or* *'n' or* *mean (sd) n*	Group 4 *not available or* *'n' or* *mean (sd) n*	Group 5 *not available or* *'n' or* *mean (sd) n*

Summarise the results and main outcome measures of the study if different from the review outcome measures.

Appendix III

Summary extracted information from articles: IPDM review.

Table 1: Authors, title and source of decision aid or prenatal testing articles (IPDMRN is the informed patient decision making review number).

Binstock, M.A. and Wolde-Tsadik, G. (1995) Alternative prenatal care: impact of reduced visit frequency, focused visits and continuity of care. *The Journal of Reproductive Medicine*. **40**: 507-512. (IPDMRN: 83) *
Brennan, P.F., Moore, S.M. and Smyth, K.A. (1995) The effects of a special computer network on caregivers of persons with Alzheimer's disease. *Nursing Research*. **44**:166-172. (IPDMRN: 493)
Butow, P.N., Dunn, S.M., Tattersall, M.H.N. and Jones Q.J. (1994) Patient participation in the cancer consultation: evaluation of a question prompt sheet. *Annals of Oncology*. **5**:199-204. (IPDMRN: 700)
Katon, W., Von Korff, M.V., Lin, E., Walker, E., Simon, G.E., Bush, T., Robinson, P. and Russo, J. (1995) Collaborative management to achieve treatment guidelines: impact on depression in primary care. *JAMA*. **273**:1026-1031. (IPDMRN: 794)
Maisiak, R., Austin J. and Heck, L. (1996) Health outcomes of two telephone interventions for patients with rheumatoid arthritis or osteoarthritis. *Arthritis and Rheumatism*. **39**:1391-1399. (IPDMRN: 27)
McCann, S. and Weinman, J. (1996) Empowering the patient in the consultation: a pilot study. *Patient Education and Counseling*. **27**:227-234. (IPDMRN: 387)
Richardson, J.L., Mondros, G.T., Danley, K., Deapen, D. and Mack, T. (1996) Impact of a mailed intervention on annual mammography and physician breast examinations among women at high risk of breast cancer. *Cancer Epidemiology: Biomarkers and Prevention*. **5**:71-76. (IPDMRN: 154)
Rost, K.M., Flavin, K.S., Cole, K. and McGill, J.B. (1991) Change in metabolic control and functional status after hospitalisation: impact of patient activation intervention in diabetic patients. *Diabetes Care*. **14**:881-889. (IPDMRN: 472)
Rowley, M.J., Hensley, M.J., Brinsmead, M.W. and Wlodarczyk, J.H. (1995) Continuity of care by a midwife team versus routine care during pregnancy and birth: a randomised trial. *The medical Journal of Australia*. **163**:289-293. (IPDMRN: 678) *
Rutten, G.E.H.M., Beek, M.M.L. and van Eijk, J.T.M. (1993) Effects of systematic patient education about cough on the consulting behaviour of a general practice population. *Patient Education and Counseling*. **22**:127-132. (IPDMRN: 337)
Sachs, G.A., Stocking, C.B. and Miles, S.H. (1992) Empowerment of the older patient? A randomised, controlled trial to increase discussion and use of advance directives. *Journal of American Geriatrics Society*. **41**:928-930. (IPDMRN: 343)
Sander, R.W., Holloway, R.L., Eliason, C., Marbella, A.M., Murphy, B. and Yuen, S. (1996) Patient-initiated prevention discussions: two interventions to stimulate patients to initiate preventive discussions. *Journal of Family Practice*. **43**:468-474. (IPDMRN: 601)
Shepperd, S., Coulter, A. and Farmer, A. (1995) Using interactive videos in general practice to inform patients about treatment choices: a pilot study. *Family Practice*. **12**:443-447. (IPDMRN: 594)
Shiloh, S., Reznik, H., Bat-Miriam-Katznelson, M. and Goldman, B. (1995) Pre-marital genetic counselling to consanguineous couples: attitudes, beliefs and decisions among counselled, noncounselled and unrelated couples in Israel. *Social Science and Medicine*. **41**: 1301-1310. (IPDMRN: 430) *
Sikorski, J., Wilson, J., Clement, S., Das, S. and Smeton, N. (1996) A randomised controlled trial comparing two schedules of antenatal visits: the antenatal care project. *British Medical Journal*. **312**: 546-553. (IPDMRN: 425) *
Tabak, E.R. (1988) Encouraging patient question asking: a clinical trial. *Patient Education and Counseling*. **12:** 37-49. (IPDMRN: 506)
Thornton, J.G., Hewison, J., Lilford, R.J. and Vail, A. (1995) A randomised trial of three methods of giving information about prenatal testing. *British Medical Journal*. **311**: 1127-1130. (IPDMRN: 778) *
Whelan, T.J., Levine, M.N., Gafni, A., Lukka, H., Mohide, E.A., Patel, M. and Streiner, D.L. (1995) Breast irradiation postlumpectomy: development and evaluation of a decision instrument. *Journal of Clinical Oncology*. **13**: 847-853. (IPDMRN: 691)

** Articles selected for inclusion by health care context: obstetrics, gynaecology and midwifery; and, genetics.*

Table 2: Description of article's health context and type of decision.

First Author	Health Area	Decision
Binstock	obstetrics and midwifery	adherence with antenatal care schedule
Brennan	mental health - Alzheimer's	utilisation of health services
Butow	cancer	question asking
Katon	mental health - depression	adherence with medication levels
Maisiak	arthritis	utilisation of health services
McCann	primary care - general	question asking
Richardson	cancer	adherence with screening appointment
Rost	diabetes	question asking
Rowley	obstetrics and midwifery	utilisation of health services
Rutten	primary care - coughs	utilisation of health services
Sachs	geriatric	making of living wills
Sander	primary care - general	question asking / information seeking
Shepperd	primary care - hypertension	treatment decisions
Shiloh	genetics	reproductive decision making
Sikorski	obstetrics and midwifery	utilisation of health service
Tabak	primary care - general	question asking
Thornton	obstetrics and midwifery	attendance antenatal clinic
Whelan	cancer	treatment decision

Table 3: Design and intervention group descriptions by article.

First Author	Design	Intervention (sample size)
Binstock	RCTc	group 1 - assigned 13 visits (n=259) group 2 - assigned 8 visits (n=320)
Brennan	RCTb	group 1 - computer + training + decision & information aid programme (n=51) group 2 - questionnaire only (n=51)
Butow	RCTb	group 1 - leaflet prompting question asking group 2 - leaflet about cancer services (n=142 total)
Katon	RCTc	group 1- leaflet about depression & therapy techniques + video + prompt (n=108) group 2 - routine care by physician (n=109)
Maisiak	RCTb	group 1 - routine care + questionnaire (n=127) group 2 - symptom monitoring counseling sessions (n=124) group 3 - treatment counseling (n=128)
McCann	RCTa	group 1 - leaflet prompting question asking (n=59) group 2 - information about 'health eating' (n=61)

Table 3 continued.

First Author	Design	Intervention (sample size)
Richardson	RCTc	group 1 - routine letter of invitation (n=195) group 2 - leaflet + video information about breast cancer & breast self examination & reasons for attending mammography & procedure mammography (n=172)
Rost	RCTc	group 1 - nurse consultation before discharge + written self-completion exercise + information on skill acquisition (n=30) group 2 - routine care + questionnaire (n=31)
Rowley	RCTa	group 1 - midwife team planning continuity of care (n=405) group 2 - routine care (n=409)
Rutten	RCTc	group 1 - 'cough' education during consultation + leaflet on causes, symptoms & treatments for coughs (n=11120) group 2 - routine care for coughs (n=9511)
Sachs	RCTc	group 1 - information interview + copies of forms + reminder cards & patient prompts (n=52) group 2 - routine care (n=85)
Sander	RCTc	control1 - questionnaire before and after consultation (n=42) group 1 - patient prompt about health risk behaviours (n=87) control2 - questionnaire before and after consultation (n=59) group 2 - nurse talked about health risk behaviours (104)
Shepperd	same sample before & after	group 1 - interactive video: adjust patient risk estimates based on responses + lifestyle change + hypertension information + types of treatment (n=77)
Shiloh	comparative	group 1 - adult cousins + genetic counseling (n=58) group 2 - adult cousins (n=40) group 3 - unrelated adults (125)
Sikorski	RCTa	group 1 - routine care: thirteen visits (n=1416) group 2 - new style: six / seven visits (n=1378)
Tabak	RCTa	group 1 - 'readable' leaflet listing type of questions to ask (n=35) group 2 - 'readable' leaflet listing services available (n=32)
Thornton	RCTc	group 1 - routine booking in information (n=567) group 2 - group 1 + extra visit & information + leaflet (n=561) group 3 - group 1 + extra class + leaflet (n=563)
Whelan	RCTc	group 1 - routine + questionnaire (n=23) group 2 - physician prompt consultation (n=29) group 3 - decision board based consultation (n=30)

Table 4: Description theory and measures by article.

First Author	Theory	Measures
Binstock	no theory	reproductive history, number of antenatal visits, antenatal care required, patient satisfaction
Brennan	EUT	age, sex, utilisation health services by carer & patient, decision making confidence & skill, social support, burden of care, depression, patient functional status
Butow	Doctor-patient communication	age, sex, medical history, disease status, anxiety, tape-recorded consultation, satisfaction, information recall, information preferences, decision making involvement
Katon	no theory	age, sex, level education, marital status, employment, medical history, satisfaction, depression, neuroticism, adherence to medication
Maisiak	Doctor-patient communication	age, sex, ethnicity, education level, medical history, disease status, affect, pain, utilisation health services
McCann	Doctor-patient communication	age, sex, employment, readability leaflet, self-efficacy, health locus of control, satisfaction, consultation length, question asking, health professional measures
Richardson	no theory	age, level education, marital status, medical history, family history, attendance mammography, risk perception, perceived susceptibility & efficacy, screening
Rost	Doctor-patient communication	age, sex, level education, employment, medical history, disease status, question asking, information preference
Rowley	no theory	age, ethnicity, marital status, employment, reproductive history, preventive behaviours, pregnancy complications, breast feeding, attendance antenatal classes & appointments, satisfaction, consultation participation
Rutten	no theory	age, sex, health insurance, attendance
Sachs	no theory	age, sex, ethnicity, level education, marital status, health insurance, disease status, knowledge, living wills
Sander	Doctor-patient communication	age, sex, request for information, decision participation, behaviour change, recall information, sources of information, satisfaction
Shepperd	Doctor-patient communication	age, sex, level education, employment, disease status, decision preference and autonomy, usefulness video
Shiloh	no theory	age, sex, ethnicity, marital status, beliefs, counseling reasons, perception risk, reproductive behaviour
Sikorski	no theory	age, ethnicity, level education, marital status, accommodation, reproductive history, preventive behaviours, disease status, pregnancy complications, affect, attitudes, satisfaction, utilisation services, social support
Tabak	Doctor-patient communication	age, level education, employment, question asking, satisfaction with physician care
Thornton	no theory	age, ethnicity, employment, reproductive history, attendance antenatal classes, uptake tests, affect, satisfaction, knowledge
Whelan	Doctor-patient communication	age, level of education, marital status, diseases status, knowledge, treatment choice, usefulness information, decision autonomy, directiveness information

Table 5: Description main findings by article.

First Author	Results
Binstock	Attendance for antenatal visits, antenatal care and pregnancy outcomes did not differ by intervention group. Satisfaction was greater in group 1.
Brennan	There was no difference in use of health care services by group. Decision confidence but not decision skill increased in group 1.
Butow	There was no difference in the number of questions asked and duration of question asking by group. Those in group 1 were more likely to ask questions about disease prognosis. Results discuss differences in preference for information and involvement in decision making.
Katon	Adherence to medication was greater in group 1 than the routine care group (88% vs. 57% major depression; 76% vs. 50% minor depression).
Maisiak	There was no difference in service utilisation by group. However, when analysed by disease status, group 3 was associated with fewer visits amongst osteoarthritis patients but not rheumatoid arthritis patients.
McCann	Group 1 had longer consultations and a trend towards significance for asking more questions (p=0.07). There was no difference in satisfaction by group.
Richardson	Group 2 associated with greater attendance than routine care (40% vs. 30%). No differences between groups for perception risk, severity and perceived efficacy mammography.
Rost	Group 1 associated with greater question asking and longer consultations than routine care patients. There was no difference in preference for information seeking or satisfaction by group.
Rowley	Group 1 were less likely to attend antenatal classes and more likely to ask questions than the routine group. No difference for breast feeding was observed.
Rutten	Intervention associated with an increase in attendance for coughs.
Sachs	No association between number of living wills made and group.
Sander	Group 1 requested more information than control1, no other differences observed. Group 2 requested more information, recalled more, more likely to change behaviour than control2. No differences in perceived decision participation by intervention groups.
Shepperd	The video was associated with changes in choices of treatment and confidence with choice. There was no increase in consultation length.
Shiloh	Group 1 had fewer children, greater perceived risk and severity genetic diseases than group 2 or 3.
Sikorski	Group 1 received more ultrasounds, had more day admissions, and had more positive attitudes than group 2. There was no difference in the number of inpatient admissions, night admissions, and attending antenatal classes.
Tabak	No significant difference by group was observed for question asking. No association between question asking and satisfaction.
Thornton	Group 2 associated with a greater uptake Down syndrome screening; group 1 associated greater uptake cystic fibrosis carrier testing; and, group 3 associated lower attendance at extra class.
Whelan	There was no association between group and treatment decision. Group 3 associated with an increase in autonomy decision making and a reduction in perceived directiveness consultation.

Appendix IV

TEST REPORT

Check carefully and ☎ 0113 234 4013
with corrections or updated information

LEEDS
ANTENATAL
SCREENING
SERVICE

Consultant Not Known
c/o Sister, Antenatal Clinic
Leeds General Infirmary
Clarendon Wing
Belmont Grove
Leeds
LS2 9NS

22-Apr-99

PATIENT

Name :	Patient SAMPLE
DOB :	24-Aug-58
Sample # :	C,99.0000000.X

PREGNANCY DETAILS

Age @ EDD :	40 years 11 months (age-related Down's syndrome risk 1 in 100)
LMP :	04-Nov-98
Dating Scan Details :	14 weeks 3 days @ 15-Feb-99
Sample Date :	15-Feb-99
Gestation @ Sample :	14 weeks 5 days (by LMP); 14 weeks 3 days (by scan)
Weight :	75 Kg

MARKER LEVELS

AFP :	0.46 MoM
uE3 :	0.20 MoM
free-beta hCG :	1.38 MoM

INTERPRETATION

Down's Syndrome Result :	*** SCREEN POSITIVE *** (risk > or = 1 in 250)
Down's Syndrome Risk :	1 in 10
Edwards' Syndrome Result :	Screen Negative
Edwards' Syndrome Risk :	1 in 60
NTD & AWD Result :	Uninterpretable (AFP measured < 15 weeks gestation)

- *MoMs calculated using gestation by scan*
- *Risks calculated at term*

Appendix VI

Describing women's decision making process: item list.

P1	**Comments/ Questions about the triple test**
p1.1	understanding the cutoff (1 in 250) for a screen positive
p1.2	what causes a screen positive triple test result (age, family, previous test)
p1.3	what does a screen positive mean (not having a child with Down syndrome)
p1.4	what does a screen negative mean (having a child with Down syndrome)
p1.5	could have screened positive in previous pregnancies
p1.6	is the tt risk specific to this pregnancy/ screen positive in future pregnancies
p1.7	what is an average risk figure (is ours high/low)
p1.8	triple test is inaccurate/ is the risk figure accurate
p1.9	would I get a different triple test result if had it again
p1.11	does the triple test result affect health baby (high hormones, etc)
p1.12	does a screen positive mean there is something wrong with parents
p1.13	age risk not associated with triple test figure
p1.14	what does triple test test for/ not heard of Edward's
p1.15	why triple test called the triple test (hormones, scan/blood/urine)
p1.16	is gestation based on scan/ what is figure based on lmp
p1.17	how long has the triple test been around
p1.18	why does this figure differ so much from previous triple test figure/ lmp/ age
p1.19	is age risk more accurate
p1.20	edwards was mentioned
p1.21	is raised tt familial (sister had screen positive tt)
P2	**Comments/ questions about 'no diagnostic test' option**
p2.1	what would nuchal pad tell me
p2.2	what does a (nineteen week) scan tell me
p2.3	can more be known further on in pregnancy
p2.4	is there a non-invasive alternative
p2.5	can the child be tested at birth
p2.6	can the child be adopted
p2.7	triple test provides reassurance (can do/ has done)
P3	**Comments/ questions about diagnostic tests**
p3.1	are they diagnostic/ tell for certain
p3.2	are there limb defects with cvs
p3.3	are there limb defects with amnio
p3.4	what are tests (cvs/amnio) testing for
p3.5	which test is more reliable/ accurate
p3.6	which test is more common
p3.7	test might give a negative result
p3.8	how likely is mosaic Down's/ inconclusive test results
p3.9	
p3.10	who is offered amnio/ are tests offered to everyone
p4	**Planning for tests**
p4.1	when can the test be carried out (hospital limitations)
p4.2	what gestational age can the test be done (medical limitations)
p4.3	does the test hurt/ do they anaesthetise/ how test done
p4.4	do they scan the fetus (and needle) during the procedure
p4.5	what happens after the test (rest, antiobiotics, stay in hospital)
p4.6	how long does test take
p4.7	is the consultant proficient/ risk of miscarriage minimised
p4.8	how do you get results of diagnostic
p4.9	what happens if scan screens positive/ would they tell us after 19 week scan
p4.10	can test damage the baby in other ways
P5	**Comments/ questions about the consequences of testing/ not testing**
p5.1	what is the risk of miscarriage
p5.2	are the risk figures quoted for the hospital or national/has anyone miscarried

p5.4 what is the background risk of miscarriage/ could miscarry for other reasons
p5.5 when would a miscarriage occur
p5.6 why would a miscarry occur
p5.7 what would a miscarriage involve
p5.8 does a recent miscarriage affect test miscarriage
p5.9 what is the legal limit for termination
p5.11 what does termination involve
p5.12 is there counselling after termination
p5.13 when would termination take place
p5.14 if have a Down syndrome child, will it affect other pregnancies
p5.15 what information do you have about Down's syndrome
p5.16 what services do you provide for Down's syndrome
p5.17 how long do the diagnostic test results take/ can this time be reduced
p5.18 the diagnostic test result could be wrong/ inconclusive result
p5.19 differences in time between test result and scan are close
p5.20 would risk miscarriage reduce later in pregnancy
p5.21 what percentage of people have a diagnositic test and Down's syndrome kid
p5.22 even if scan +ve, still be normal

P6 Comments/ questions about Down Sydrome

p6.1 what are the causes Down syndrome
p6.2 there are different severities in Down syndrome/ can tests detect severity
p6.3 Down's syndrome child would be OK
p6.4 would want to hear parents view of Down's syndrome
p6.5 would want more information about Down's syndrome before made a decision to terminate
p6.6 what information do you have about Down's syndrome

P7 Comments/ questions about the decision

p7.1 do we have to decide now/ let you know later
p7.2 what do other people do
p7.3 what did you do
p7.4 what should we do/ what do you recommend
p7.5 difficult to take information in/ need to discuss it/ what does partner think
p7.6 need to go home and discuss my decision with family
p7.7 have all the information and know what want

P8 Use of risk

p8.1 used age-related risk when talking
p8.2 used triple test risk figure when talking
p8.3 used 'other' reference figure (previous triple test, cut-off figure)
p8.4 used amniocentesis risk figure when talking
p8.5 used CVS risk figure when talking
p8.6 used background miscarriage risk figure when talking
p8.7 used combined background and diagnostic miscarriage rates
p8.8 compared risk figures for triple test result and diagnostic
p8.9 compared risk figures for CVS and amniocentesis
p8.10 compared previous triple test figure with current risk
p8.11 compared population/age risk with own risk
p8.12 compared risk to cut-off limit
p8.13 misrepresented/ misunderstood risk figure

P9 Perception of risk

p9.1 triple test risk figure is a high risk
p9.2 triple test risk figure is an acceptable risk/ good odds
p9.3 diagnostic miscarriage rates are high
p9.4 diagnostic miscarriage rates are low
p9.5 I am more likely to get a screen positive
p9.6 I am more likely to have an Down syndrome child/ could be the one
p9.7 I am more likely to miscarry
p9.8 compared with other risks Down syndrome figure is low/ miscarriage high
p9.9 compared with other risks miscarriage figure is low/ others high
p9.10 I am unlikely to miscarry
p9.11 I am unlikely to have Down syndrome baby (baby moves)

p9.12 risk sounds better as a percentage
p9.13 risk Down's syndrome outwieghs risk miscarriage
p9.14 this risk is more certain because (previous result...)
p9.15 scan reduces risk

P10 Comments about triple test and result

p10.1 regret having triple test/ not have triple test again
p10.2 triple test result affects pregnancy/ worry not normal
p10.3 triple test creates uncertainty
p10.4 getting the result was a shock/ is upsetting
p10.5 tt might reassure/ thought I would get a negative result/ didn't cross my mind
p10.6 if I had a negative result/ was younger I would be none the wiser / happier
p10.7 didn't think hard about having triple test (HCP, family, had it before)
p10.8 thought hard about having triple test (reasons given for having it)
p10.9 had triple test because of age-ds link
p10.10 had the test and know I am at risk now
p10.11 wanted triple test for knowledge
p10.12 thought positive meant good
p10.13 not heard tt before
p10.14 concerned that it has come back positive again
p10.15 friend/ relative had a screen positive triple and everything OK
p10.16 worrying that the triple test is still being researched
p10.17 screen negative still at risk

P11 Reasons for not having a diagnostic test

p11.1 did not have triple test last time (everything OK)
p11.2 had triple test last time (everything OK)
p11.3 not had diagnostic tests before (other kids healthy)
p11.4 Down syndrome not in the family
p11.5 risk of miscarriage/ worry of losing the baby
p11.6 worried about the procedure (needles)
p11.7 this is the last pregnancy
p11.8 problems getting pregnant/ miscarried before
p11.9 would not terminate even if it had Down syndrome/ emoationally hard
p11.11 prenatal testing interfering with nature
p11.12 other conditions more severe/ DS not such a bad condition
p11.13 scan reassuring
p11.14 won't disrupt family/ other children
p11.15 practically a developed child (not terminate)
p11.16 want a baby no matter what
p11.17 want to take the Down's syndrome risk (not have a test)
p11.18 don't want to know whether Down's syndrome
p11.19 friend had amnio and lost baby

P12 reasons for having a diagnostic test

p12.1 decided to have the diagnostic test before consultation
p12.2 had the triple test to find out, therefore have diagnostic test
p12.3 had diagnostic test before
p12.4 friend/ HCP/ relative had diagnostic test (and baby was fine)
p12.5 do not want to regret not having a diagnostic test
p12.6 did not realise had a choice
p12.7 to have the scan, to check baby alive
p12.8 have to know whether an abnormality (set mind at rest)
p12.9 can't wait until term, live with uncertainty
p12.10
p12.11 do not want a Down syndrome child (can't take the risk)
p12.13 to be prepared (for the birth of a child with DS/ self & family)
p12.14 to terminate if result positive
p12.15 other children to consider/ we have one normal
p12.16 if had other children would be different
p12.17 woman/ partner would not cope with Down syndrome child
p12.18 woman would be the one to look after Down syndrome child
p12.19 have a family history of Down syndrome/ abnormality

p12.21 no problems getting pregnant
p12.22 miscarry for other reasons
p12.23 still an embryo, not a child
p12.24 heard of amniocentesis
p12.25 not heard of chorionic villus sampling
p12.26 heard of chorionic villus sampling
p12.27 get to know sex
p12.28 scan not a certain result
p12.29 19 weeks scan too far on in pregnancy
p12.30 friend had abnormal child (bad effect on her and family)
p12.31 friend had diagnostic and everything OK

P13 Reason to choose one diagnostic test over another

p13.1 want to have a test but don't know which one
p13.2 there is a risk with both tests/ might as well have CVS
p13.3 still have to go through labour no matter which test (amnio, time not adv.)
p13.4 CVS get a result in 24 hours
p13.5 worth the risk to get a result in 24 hours
p13.6 CVS have to wait three weeks for a follow up result
p13.7 amniocentesis takes three weeks to get a result (affect...hard)
p13.8 not worth the risk to get a result in 24 hours
p13.9 amniocentesis has a lower rate of miscarriage
p13.11 CVS has a higher rate of miscarriage
p13.12 CVS get limb defects
p13.13 CVS get more inconclusive results (md)
p13.14 cvs same risk as amnio
p13.15 find out sooner in pregnancy (before kicking)
p13.16 three weeks better than term
p13.17 have both tests

P14 Reference to Down Syndrome

p14.1 a child with Down syndrome never grows up/ child in adult body
p14.2 a child with Down syndrome will be unusual in the world/ freaks
p14.3 child with Down syndrome will not have a full life/ feel sorry for them
p14.4 Down syndrome is life-threatening
p14.5 it's for life/ what happens when we're not here
p14.6 condition 'x' less severe than Down syndrome
p14.7 had positive experiences of Down syndrome
p14.8
p14.9 more severe Down's syndrome picked up on scan
p14.10 knew someone with Down's syndrome
p14.11 Down's syndrome time consuming
p14.12 worked with Down's syndrome
p14.13 knowledge that Down's syndrome has physical problems
p14.14 if it is Down's syndrome, there now

P15 Causes of Down syndrome

p15.1 Down syndrome related to age
p15.2 there is a mental disability in the family/ inherited/ familial
p15.3 mental disability not in family

P16 Conflict in decision making

p16.1 it is a difficult decision/ don't know what to do/ don't want to make a decision
p16.2 want to make a decision today
p16.3 uncertain whether terminate, depends result/ know for sure/ one step at time
p16.4 will make a decision after the nineteen week scan
p16.5 if a positive result will seek more information about Down syndrome
p16.6 don't know how I would feel about a Down syndrome child until born
p16.7 my attitudes appear cruel/ heartless
p16.8 my decision is against the norm - most terminate
p16.9 my decision against norm - some people cope with Down syndrome
p16.11 decision to have diagnostic test goes against beliefs
p16.12 decision to terminate emotionally difficult/ against beliefs self/ family
p16.13 want test now

p16.14 don't want test now
p16.15 don't want to make a decision now/ need more time
p16.16 my decision against norm - should have test
p17 Health Professional Advice/ Comments
p17.1 's/he' said don't have test unless consider termination
p17.2 's/he' advice good/ talked through all alternatives
p17.3 's/he' expressed said triple test unaccurate/ unreliable
p17.4 's/he' encouraged me to have triple
p17.5 's/he' encouraged me to have diagnostic
p17.6 HCP said at scan baby healthy, active/ OK
p17.7 s/he discouraged me to have tt
p17.8 s/he discouraged me to have diagnostic
p17.9 HCP gave contrary advise to HB
p17.10 HCP said amnio was the better test
p17.11 HCP said chorionic villus sampling was the better test
p18 Experience
p18.1 had a recent bleed
p18.2 had a screen positive before
p18.3 had a diagnostic test before
p18.4 had a termination before/ know about termination (work)
p18.5 (related) baby ill/ abnormality
p18.6 experience infertility/ waited to have a baby
p18.7 miscarried before
p18.8 saw baby on scan and brings a new perspective
p18.9 genetic risk in family/ had genetic tests before/ know about risk

Appendix VII

Coding frame for women's decision making.

Woman ID: ***Group:*** ***Risk:*** ***Decision:***

Theme	Number Observations
1. No Test Option (non invasive/ scan)	
1.1 information seeking / planning	
1.2 reasons for	
1.3 reasons against	
2. Test Option	
2.1 information seeking / planning	
2.2 reasons for	
2.3 reasons against	
3. Choice between tests - neutral comment	
4. Chorionic villus sampling choice	
4.1 information seeking / planning	
4.2 reasons for	
4.3 reasons against	
5. Amniocentesis choice	
5.1 information seeking planning	
5.2 reasons for	
5.3 reasons against	
6. Down syndrome consequence	
6.1 information seeking/ planning	
6.2 neutral-positive reference	
6.3 negative reference	
7. Miscarriage consequence	
7.1 information seeking/ planning	
7.2 neutral - positive reference	
7.3 negative reference	
8. Termination consequence	
8.1 information seeking/ planning	
8.2 neutral - positive comment	
8.3 negative reference	
9. Risk figure reference	
9.1 single risk Down syndrome	
9.2 single risk miscarriage	
9.3 comparison risks	
10. Perception of risk reference	
10.1 minimised Down syndrome risk	
10.2 maximised Down syndrome risk	
10.3 minimised miscarriage risk	
10.4 maximised miscarriage risk	

Theme	Number Observations
11. Expressed Affect	
11.1 triple test (shock)	
11.2 diagnostic test (hard) - conflict	
11.3 termination (hard) - conflict	
11.4 reassurance testing	
11.5 anticipated regret	
12. Decision Making	
12.1 triple test	
12.2 diagnostic test made before consultation	
12.3 termination / stage at a time	
12.4 delay in making decision - uncertainty	
12.5 confidence with decision - certainty	
12.6 consultation / disucssion final decision	
13. Triple test - further explanation	
13.2 Nuchal pad	
14. Confidence with screening results	
15. Comparison norms (others / you do)	
15.1 what would you suggest	
15.2 what do others do / general percentages	
16. Personal experiences	
16.1 physical aspects pregnancy (scan/ move)	
16.2 complications with pregnancy (misc.)	
16.3 prior testing / pregnancies	
16.4 abnormalities / genetics	
16.5 not telling others about pregnancy	
17. Others experience	
17.1 prenatal testing	
17.2 abnormalities / genetics	
18. Preparation pregnancy (birth, telling others)	
19. Health professionals' advice	
19.1 neutral / positive comments	
19.2 dissatisfaction / directive	
19.3 inaccurate / conflicting consultation	
20. Other sources information	

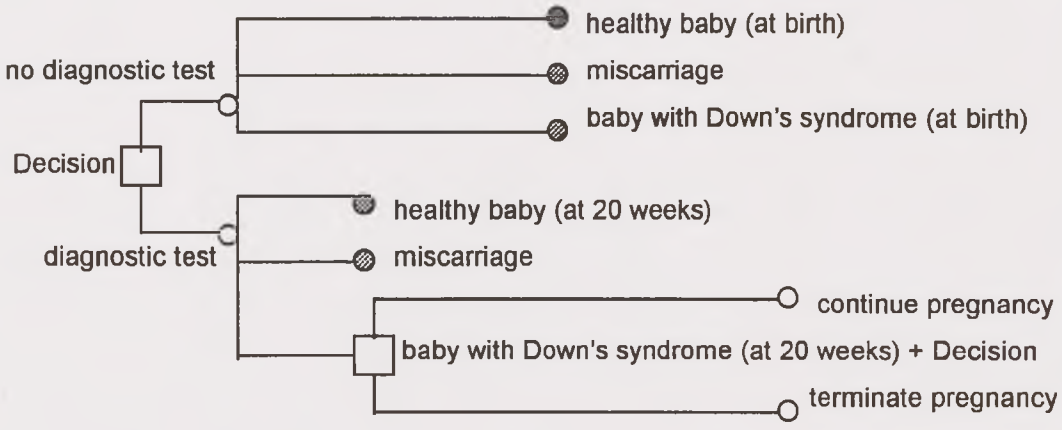
Modified coding frame categories aggregating themes.

Aggregated themes	women (n=44)	%
(2) All test options - calculated from themes 2, 3, 4 & 5.		
information seeking about testing (from 2.1, 3, 4.1 & 5.1)	43	98 %
reasons for having a test (from 2.2, 4.2 & 5.2)	41	93 %
reasons against having a test (from 2.3, 4.3 & 5.3)	37	84 %
(11) Total negative affect - calculated from 11.1 & 11.2	25	57 %
(16) Personal experiences - calculated from all sub groups	29	66 %
(17) Others experience - calculated from all sub groups	17	39 %
(19) Health professionals' advice - calculated from all sub groups	18	41 %

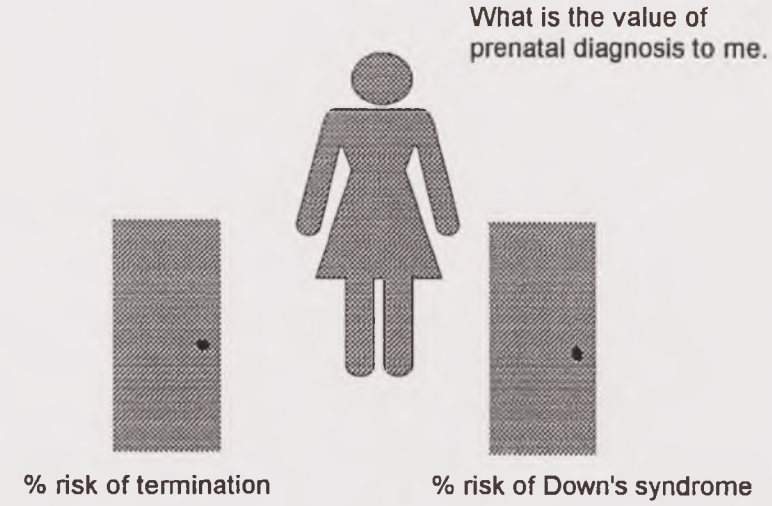
Appendix VIII

Visual prompts used during the decision analysis consultation: decision tree: lottery question: threshold graph.

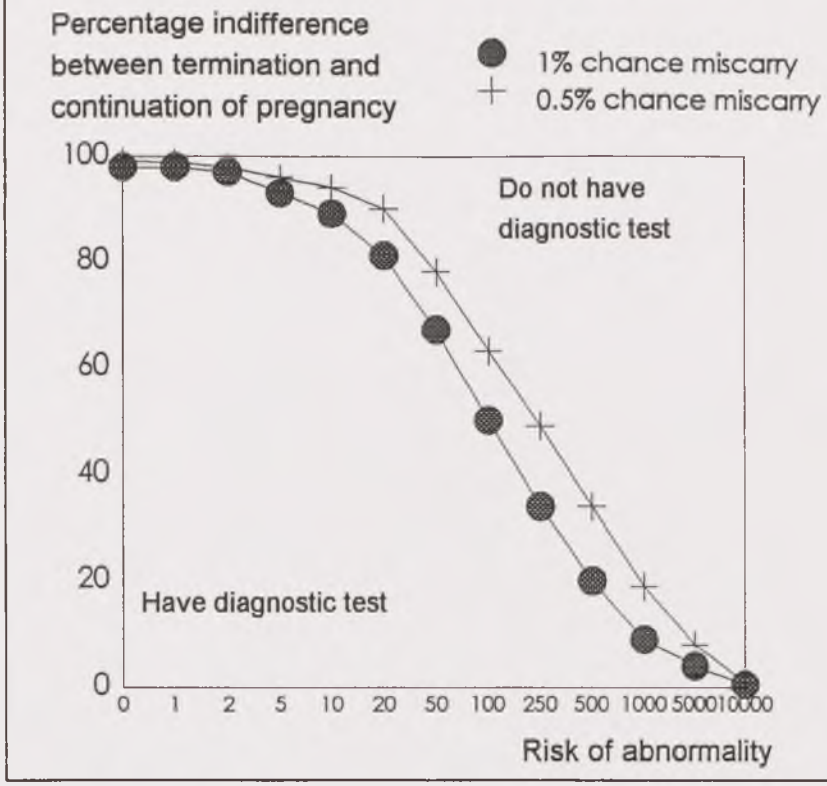
Decision tree prompt.



Prompt for extraction of utilities.



Prompt for the integration of utilities and triple test risk: threshold graph.



Appendix IX

Consent from for the randomised control trial.

LEEDS GENERAL INFIRMARY ANTE-NATAL STUDY

Making Decisions in Pregnancy

During pregnancy, women are asked to make many decisions that affect the well-being of either themselves or their fetus. For example, women can choose to have a test, or not, that will tell them the likelihood of their fetus having a disease or abnormality. Some decisions during pregnancy are more difficult to make than others.

This study is looking at two ways of giving information to women who are deciding to have further tests in pregnancy or not. Your views and experiences will further our understanding of what information women find most useful when making these decisions. For example, do women find the information easier to understand and more helpful if it is presented in one way rather than another.

Taking part in the study.

Hilary Bekker is the clinical researcher for this project. Hilary will be talking with you about your triple test result and giving you information about the options available at this stage in your pregnancy.

Agreeing to take part in the study involves being randomised to one of two information-giving groups. The only difference between the two groups is the *way* the information is presented. The consultations will be tape-recorded and you will be asked to complete a questionnaire after the consultation and in four weeks time. The questionnaire takes about fifteen minutes to complete.

Your responses during the consultation and from the questionnaires are **confidential**. You will be given a 'study number' so only Hilary can match your name with the study information. Hilary will be happy to answer any questions you may have about this study.

If you decide not to take part in the study, you will still receive the routine care provided by the ante-natal clinic at this hospital.

Consent

Name (in block letters) .....

Please answer yes or no

a) I have read the information sheet: **yes/ no** b) I agree to take part in the study: **yes/ no**

Signature:.....

We are interested about how you are feeling now. There are no right or wrong answers to these questions. Please circle one number for each question.

A number of statements which people have used to describe themselves are given below. Read each statement and then circle the most appropriate number to the right of the statement to indicate how you feel right now, at this moment. There are no right or wrong answers. Do not spend too much time on any one statement but give the answer which seems to describe your present feelings best.

		not at all	somewhat	moderately	very much
1.	I feel calm	1	2	3	4
2.	I feel tense	1	2	3	4
3.	I am upset	1	2	3	4
4.	I feel relaxed .	1	2	3	4
5.	I feel content .	1	2	3	4
6.	I am worried ...	1	2	3	4

7. How likely do you think you are to:	not at all likely	very likely
have a healthy child	0 -----1 -----2 -----3 -----4 -----5 -----6	
have a child with Down syndrome	0 -----1 -----2 -----3 -----4 -----5 -----6	
miscarry a fetus without Down syndrome	0 -----1 -----2 -----3 -----4 -----5 -----6	
miscarry a fetus with Down syndrome	0 -----1 -----2 -----3 -----4 -----5 -----6	

8. What concerns, if any, do you have at the moment about your:

a) **health** *(please list):*

b) **pregnancy** *(please list):*

c) **baby** *(please list):*

Thank you for answering all the questions.

Appendix X

Linguistic Inquiry and Word Count: II (SLIWC)

James W. Pennebaker and Martha E. Francis

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Dimension	Examples
1. STANDARD LINGUISTIC DIMENSIONS	
Word Count	
Words per sentence	
Sentences ending with ?	
Unique words (type/token ratio)	
% dictionary words captured	
% words longer than 6 letters	
Total pronouns	I, our, they, you're
1st person singular	I, my, me
1st person plural	we, our, us
Total first person	I, we, me
Total second person	you, you'll
Total third person	she, their, them
Negations	no, never, not
Assents	yes, OK, mmhmm
Articles	a, an, the
Prepositions	on, to, from
Numbers	one, thirty, million
II. PSYCHOLOGICAL PROCESSES	
Affect: Emotional Processes	happy, ugly, bitter
Positive Emotions	happy, pretty, good
Positive feelings	happy, joy, love
Optimism and energy	certainty, pride, win
Negative Emotions	hate, worthless, enemy
Anxiety or fear	nervous, afraid, tense
Anger	hate, kill, pissed
Sadness or depression	grief, cry, sad
Cognitive Processes	cause, know, ought
Causation	because, effect, hence
Insight	think, know, consider
Discrepancy	should, would, could
Inhibition	block, constrain
Tentative	maybe, perhaps, guess
Certainty	always, never
Sensory and Perceptual Processes	see, touch, listen
Seeing	view, saw, look
Hearing	heard, listen, sound
Feeling	touch, hold, felt

Dimension	Examples
III. PSYCHOLOGICAL PROCESSES	
Social Processes	talk, us, friend
Communication	talk, share, converse
Other references to people	1st pi, 2nd, 3rd per prns
Friends	pal, buddy, coworker
Family	mom, brother, cousin
Humans	boy, woman, group
Time	hour, day, oclock
Past tense verb	walked, were, had
Present tense verb	walk, is, be
Future tense verb	will, might, shall
Space	around, over, up
Up	up, above, over
Down	down, below, under
Inclusive	with, and, include
Exclusive	but, except, without
Motion	walk, move, go
IV. PERSONAL CONCERNS	
Occupation	work, class, boss
School	class, student, college
Job or work	employ, boss, career
Achievement	try, goal, win
Leisure activity	house, TV, music
Home	house, kitchen, lawn
Sports	football, game, play
Television / movies	TV, sitcom, cinema
Music	tunes, song, cd
Financial issues	cash, taxes, income
Metaphysical issues	God, heaven, coffin
Religion	God, church, rabbi
Death and dying	dead, burial, coffin
Physical functions	ache, breast, sleep
Body states	ache, heart, cough
Sex and sexuality	lust, penis, fuck
Eating / drinking	eat, swallow, taste
Sleeping / dreaming	asleep, bed, dreams
Grooming	wash, bath, clean
APPENDIX: EXPERIMENTAL TERMS	
Swear words	damn, fuck, piss
Nonfluencies	uh, rr*
Fillers	youknow, Imean

Appendix XI

Post-Decision Questionnaire (T1): facilitating informed decision making.

LEEDS GENERAL INFIRMARY ANTE-NATAL STUDY

Prenatal Testing Questionnaire (T1)

Study Number ..... **Group** **1 r / 2 (da) tt / age**

The following questionnaire is part of the project looking at how women make decisions during pregnancy. The questionnaire takes about fifteen minutes to complete. Filling in the questionnaire will not interfere with the care you receive from this clinic.

Your experiences and views of making decisions in pregnancy are important to this study. The results of this study will provide us with a better understanding of what information women find useful at this stage in their pregnancies.

If you have any questions about the study, or the questionnaire, please tell Hilary Bekker. The answers you give in this questionnaire are confidential and anonymous. Only Hilary can match your study number with your name.

Thank you for your time.

Hilary Bekker
Clinical Researcher,
University of Leeds.

Mr Jim Thornton
Consultant in Obstetrics and
Gynaecology, Leeds General Infirmary.

The following questions ask for details about you.

1. What is your marital status:

married ☐ living as married ☐ single ☐ other (.....)

2. What is the highest level of education you have received *(please tick the highest)*

no formal level of education ☐ GCSE equivalent ('O' level/ 'CSE') ☐
apprenticeship (Btec/ HND/ city & guilds) ☐ 'A' level equivalent (highers etc.) ☐
Degree or more ☐

3. Do you follow a religion: *yes, it is ..... / no*

If yes, how often do attend religious gatherings (please tick one box):

not at all ☐ a few times a year ☐ once a month ☐ once a week or more ☐

4. How would you describe your ethnic origin:

White ☐ Black - Caribbean ☐ Black - African ☐ Black - Other ☐
Indian ☐ Pakistani ☐ Bangladeshi ☐ Chinese ☐ Other ☐

5. Your increased risk figure for having a baby with Down syndrome is 1 in _____

Would you describe this risk as: low ☐ medium ☐ high ☐

6. Have you had any contact with people with Down syndrome?

no ☐ no, just seen someone in village/ street ☐ no, only on TV ☐
yes, family history ☐ yes, friend of the family ☐ yes, through work ☐

7. Was this pregnancy planned: yes/ no

8. What tests have you had or are about to have in:

a) this pregnancy and b) your last pregnancy:

	ultrasound scan	nuchal pad	triple test	amniocentesis	cvs
THIS pregnancy					
LAST pregnancy					

A number of statements which people have used to describe themselves are given below. Read each statement and then circle the most appropriate number to the right of the statement to indicate how you feel right now, at this moment. There are no right or wrong answers. Do not spend too much time on any one statement but give the answer which seems to describe your present feelings best.

		not at all	somewhat	moderately	very much
9.	**I feel calm .....**	1	2	3	4
10.	**I feel tense .....**	1	2	3	4
11.	**I am upset .....**	1	2	3	4
12.	**I feel relaxed .....**	1	2	3	4
13.	**I feel content .....**	1	2	3	4
14.	**I am worried .....**	1	2	3	4

The following question asks for your views and feelings about prenatal testing in general.

15. Some people think there are both advantages and disadvantages to having tests in pregnancy. In the box below, please list the advantages and disadvantages that are important to you and rate how much of an advantage or disadvantage it is (no more than six of each).

Advantages	
	small advantage 1 2 3 4 5 great advantage
	small advantage 1 2 3 4 5 great advantage
	small advantage 1 2 3 4 5 great advantage
	small advantage 1 2 3 4 5 great advantage
	small advantage 1 2 3 4 5 great advantage
	small advantage 1 2 3 4 5 great advantage
Disadvantages	
	small disadvantage 1 2 3 4 5 great disadvantage
	small disadvantage 1 2 3 4 5 great disadvantage
	small disadvantage 1 2 3 4 5 great disadvantage
	small disadvantage 1 2 3 4 5 great disadvantage
	small disadvantage 1 2 3 4 5 great disadvantage
	small disadvantage 1 2 3 4 5 great disadvantage

The following questions ask how you feel about the decision you made to have, or not have, amniocentesis or chorionic villus sampling (CVS).There are no wrong or right answers (please circle one number).

16. Before this consultation had you made a decision to have or not have a test:

yes ☐ no ☐ *If yes, please state which test* .....

17. Before this consultation did you intend to have a diagnostic test:

definitely did not						definitely did
0	1	2	3	4	5	6

18. How much time did you spend thinking about the decision to have/not have amniocentesis?

very little time						a lot of time
0	1	2	3	4	5	6

After reading the following statements, please circle a number that reflects how you much you agree with them

19. The decision to have/ not have a test was hard to make:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

20. I was unsure whether to have/ not have a test:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

21. It was clear what the best choice was for me:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

22. I was aware of the choices I had based on my triple test result:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

23. I feel I know what the benefits of having a test are:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

24. I feel I know what the risks of having a test are:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

25. I feel I have made an informed choice about whether to have a test or not:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

26. My decision shows what is most important for me:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

27. The decision to have or not have a test was entirely up to me

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

28. I am satisfied with my decision

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

29. I expect to stick with my decision

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

The following question is about the five consequences health professionals mention when talking about prenatal testing:

30. When health professionals talk about tests in pregnancy they mention five consequences of the tests (listed below). The consequences differ in importance for each person. Please place a mark along the dotted scale to show how 'bad' or 'good' each consequence is for you:

	bad	**good**
having a normal baby	.....	
miscarrying a baby with Down syndrome	.....	
terminating a fetus with Down syndrome	.....	
having a baby with Down syndrome	.....	
miscarrying a normal baby	.....	

31. Imagine a close friend has the same risk of having a baby with Down syndrome as you and she asks you for some advice about amniocentesis (CVS). Would you encourage her to have amniocentesis (CVS) *(please circle one response)*?

strongly encourage	encourage	neither encourage/ nor discourage	discourage	strongly discourage
1	2	3	4	5

The following questions ask you how useful the information about amniocentesis (CVS) was to you.

32. Was there anything you wanted more information about? yes/ no
if yes please state.....

33. Please write below what bits of information were helpful and unhelpful to you when making the decision to have, or not have, amniocentesis (CVS):

Helpful information	Unhelpful information

34. Overall, how useful was the information given during this consultation *(please circle one number)*: not at all useful ... very useful

0 ------ 1 ------ 2 ------ 3 ------ 4 ------ 5 ------ 6

35. Do you feel the following people encouraged you to have amniocentesis (CVS). Please circle one response for each person that you spoke to about amniocentesis (CVS).

		did not discuss	strongly encouraged	encouraged	neither encouraged nor discouraged	strongly discouraged	discouraged
a	midwives	☐	1	2	3	4	5
b	obstetrician	☐	1	2	3	4	5
c	general practitioner	☐	1	2	3	4	5
d	the researcher	☐	1	2	3	4	5
e	husband / partner	☐	1	2	3	4	5
f	other	☐	1	2	3	4	5

The following questions ask you about a previous decision you made: the decision to have the triple test. Again, these questions ask for your views so there are no right or wrong answers.

36. How much time did you spend thinking about the decision to have the triple test?

very little time ... a lot of time

0 ------ 1 ------ 2 ------ 3 ------ 4 ------ 5 ------ 6

37. There are many reasons why women have the triple test. What reason made up your mind to have the triple test?

38. Do you regret having had the triple test at all *(please circle one response)*:

no regret ... regret very much

0 ------ 1 ------ 2 ------ 3 ------ 4 ------ 5 ------ 6

39. Imagine a close friend the same age as you asks for advice about the triple test. Would you encourage her to have the triple test *(please circle one response)*?

strongly encourage	encourage	neither encourage/ nor discourage	discourage	strongly discourage
1	2	3	4	5

40. Do you feel the following people encouraged you to have the triple test. Please circle one response for each person that you spoke to about the triple test.

		did not discuss	strongly encouraged	encourage	neither encouraged nor discouraged	strongly discouraged	discouraged
a	midwives	☐	1	2	3	4	5
b	obstetrician	☐	1	2	3	4	5
c	general practitioner	☐	1	2	3	4	5
d	husband / partner	☐	1	2	3	4	5
e	other	☐	1	2	3	4	5

Some women find pregnancy a worrying time. The following questions ask you how worried you are .

41. How worried are you that the following harm the fetus *(please circle one number for each item)*:

		not at all worried						very worried
(a)	ultrasound scan	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(b)	emotional upset	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(c)	triple test	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(d)	amniocentesis	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(e)	taking medication	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(f)	chorionic villus sampling	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6

42. How worried are you about the baby having *(please circle one number for each item):*

		not at all worried						very worried
(a)	a physical disability	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(b)	a mental disability	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(c)	a serious health problem	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6

43. I need to know for certain whether or not this baby has Down syndrome

not at all very much

0 ------ 1 ------ 2 ------ 3 ------ 4 ------ 5 ------ 6

The following questions are about your views on disability and how likely you think the possible consequences of testing are to happen to you (please circle one number for each question)

44. Do you feel a child's life would be affected by the following: *(please circle a response for each item)*

Condition	*how affected*	**not at all affected**						**greatly affected**
cleft palate/lip	quality of life	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
	amount of care	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
Down syndrome	quality of life	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
	amount of care	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
spina bifida	quality of life	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
	amount of care	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6

45. How likely do you think you are to:	not at all likely					very likely
have a healthy child	0	1	2	3	4	5	6
have a child with Down syndrome	0	1	2	3	4	5	6
miscarry a fetus without Down syndrome	0	1	2	3	4	5	6
miscarry a fetus with Down syndrome	0	1	2	3	4	5	6
terminate a pregnancy for spina bifida	0	1	2	3	4	5	6
terminate a pregnancy for a cleft palate	0	1	2	3	4	5	6
terminate pregnancy for Down syndrome	0	1	2	3	4	5	6

46. Most people who are important to me think I should:	should					should not
have had the triple test for Down syndrome	0	1	2	3	4	5	6
have a diagnostic test	0	1	2	3	4	5	6
terminate a pregnancy for Down syndrome	0	1	2	3	4	5	6

47. Is there anything that you think may increase or decrease the likelihood of having a child with Down syndrome *(please list your response in the space below)*

48. Compared with women your age:	less likely					more likely
how healthy would you say you were	0	1	2	3	4	5	6
how likely to develop a health problem	0	1	2	3	4	5	6
how likely to have a healthy child	0	1	2	3	4	5	6
how likely have a child with Down syndrome	0	1	2	3	4	5	6
how likely to miscarry a pregnancy	0	1	2	3	4	5	6

The next part of the questionnaire asks your opinions about the purpose of tests in pregnancy. How much do you agree with the following statements (please circle one number for each question).

49. Tests during pregnancy are necessary for my well-being:

strongly disagree	disagree	neither agree nor disagree	agree	agree strongly
1	2	3	4	5

50. Tests during pregnancy improve my chance of having a healthy baby:

strongly disagree	disagree	neither agree nor disagree	agree	agree strongly
1	2	3	4	5

51. Tests during pregnancy are necessary for the well-being of the fetus:

strongly disagree	disagree	neither agree nor disagree	agree	agree strongly
1	2	3	4	5

52. Tests during pregnancy decrease my chance of miscarrying:

strongly disagree	disagree	neither agree nor disagree	agree	agree strongly
1	2	3	4	5

The following questions are about your knowledge of tests in pregnancy. Please answer the questions for all three tests. Do not worry if you do not know all the answers:

53. To whom are the following tests routinely offered, in this hospital *(please tick all relevant boxes)*?

	all women	women over 29	women over 35	family history Down's syndrome	screen positive triple test result	don't know
triple test						
amniocentesis						
CVS						

54. The following are tests for *(please tick all boxes that apply for each test)*?

	all health abnormalities	Down 's syndrome	all known chromosomal abnormalities	spina bifida	none of these	don't know
triple test						
amniocentesis						
CVS						

55. The following three tests use what type of sample *(please tick one box for each test)*?

	your blood	your urine	your baby's blood	fluid from around the baby	cells from the placenta	don't know
triple test						
amniocentesis						
CVS						

56. Are the following *screening procedures* (telling you whether you are more or less likely to have a baby with a disability), *diagnostic procedures* (give you as definite a 'yes' or 'no' answer as medicine can) or *neither* (e.g. routine checks) *(please tick one response for each test).*

(a) woman's age screening test ☐ diagnostic test ☐ neither ☐ don't know ☐
(b) triple test screening test ☐ diagnostic test ☐ neither ☐ don't know ☐
(c) amniocentesis screening test ☐ diagnostic test ☐ neither ☐ don't know ☐
(d) ultrasound scan screening test ☐ diagnostic test ☐ neither ☐ don't know ☐
(e) chorionic villus sampling: screening test ☐ diagnostic test ☐ neither ☐ don't know ☐

57. A negative test result for the following tests *usually* means (*please tick one item only for each test*)?

	the baby definitely does not have Down's	it is unlikely the baby has Down's	the baby might have Down's	the baby definitely does have Down's	don' know
triple test					
amniocentesis					
CVS					

58. A positive test result for the following tests *usually* means (*please tick one item only for each test*)?

	the baby definitely does not have Down's	it is unlikely the baby has Down's	the baby might have Down's	the baby definitely does have Down's	don' know
triple test					
amniocentesis					
CVS					

59. How long does it take for the results from each test to come back ***(please tick one box for <u>each</u> test)?***

	1 to 2 days	1 to 2 weeks	3 to 4 weeks	don't know
triple test				
amniocentesis				
CVS				

60. The extra risk of miscarriage after having the following three tests is ***(please tick one box for <u>each</u> test)?***

	no extra risk	greater than 1 in 50	1 in 50 (2%)	1 in 100 (1 %)	1 in 150 (0.7%)	1 in 200 (0.5%)	less than 1 in 200	don't know
triple test								
amniocentesis								
CVS								

61. The average population risk of a baby being born with Down syndrome is ***(please tick a box)?***

a) 1 in 5 (20%) ☐
b) 1 in 60 (2%) ☐
c) 1 in 100 (1%) ☐
d) 1 in 600 (0.002%) ☐
e) 1 in 1000 (0.001%) ☐
f) don't know ☐

62. The overall population risk of a woman miscarrying in pregnancy is ***(please tick one box)?***

a) 1 in 5 (20%) ☐
b) 1 in 60 (2%) ☐
c) 1 in 100 (1%) ☐
d) 1 in 600 (0.002%) ☐
e) 1 in 1000 (0.001%) ☐
f) don't know ☐

This is the final set of questions. Different people have different ways of thinking about problems. The following questions ask about how you feel towards solving problems. Please circle one number for each item.

63.	**I hardly ever expect things to go my way**	strongly agree	1	2	3	4	5	strongly disagree
64.	**Thinking is not my idea of fun**	strongly agree	1	2	3	4	5	strongly disagree
65.	**I would rather do something that requires little thought than something that will challenge my thinking abilities**	strongly agree	1	2	3	4	5	strongly disagree
66.	**The idea of relying on thought to make my way to the top appeals to me**	strongly agree	1	2	3	4	5	strongly disagree
67.	**The notion of thinking abstractly is appealing to me**	strongly agree	1	2	3	4	5	strongly disagree
68.	**I like to have the responsibility of handling a situation that requires a lot of thinking**	strongly agree	1	2	3	4	5	strongly disagree
69.	**I really enjoy a task that involves coming up with new solutions to problems**	strongly agree	1	2	3	4	5	strongly disagree
70.	**It's enough for me that something gets the job done: I don't care how or why it works**	strongly agree	1	2	3	4	5	strongly disagree
71.	**Learning new ways to think doesn't excite me very much**	strongly agree	1	2	3	4	5	strongly disagree
72.	**I always look on the bright side**	strongly agree	1	2	3	4	5	strongly disagree

Thank you for answering all these questions.

Appendix XII

Development of the short-form Need for Cognition Scale (NFC).

Background: "Need for cognition refers to an individual's tendency to engage in and enjoy effortful cognitive endeavours" (p.306; Cacioppo, Petty and Kao, 1984). The 'short-form' questionnaire developed by Cacioppo *et al* includes eighteen items to measure this one construct. As the eighteen items aim to measure one construct, participants comment on the repetitive nature of the items. Certainly, an eighteen item measure is long when a variety of constructs are being assessed. The following analysis was carried out to produce a shorter 'need for cognition' (NFC) scale on a British sample.

Sample: 160 psychology undergraduates at a university in the UK.

Analysis:
a) principal components analysis to extract items
b) Chronenbach's alpha correlation to assess internal reliability
c) Pearson's correlation to assess the relationship between the short form questionnaires with the eighteen item questionnaire

Results: eighteen items were entered into the principal components analysis. Factors with Eigen values of greater than 1.00 were included and items with loadings of .55 were considered for further analysis. Six factors were extracted, accounting for 62% of the total variance (factor 1, factor 2, factor 3, factor 4, factor 5, factor 6 accounted for 26%, 11%, 7%, 6%, 6%, 6% respectively of the variance). As the NFC has been developed to measure one construct, only the results of factor one are reported.

Table 1: loadings of items on factor 1 (>.55)

	Factor 1 26% variance
Thinking is not my idea of fun	.74
I would rather do something that requires little thought then something that is sure to challenge my thinking abilities	.73
The idea of relying on thought to make my way to the top appeals to me	.65
The notion of thinking abstractly is appealing to me	.61
It's enough for me that something gets the job done, I don't care how or why it works	.60
I try to anticipate and avoid situations where there is a likely chance I will have to think in depth about something	.59
I like to have the responsibility of handling a situation that requires a lot of thinking	.58
Learning new ways to think doesn't excite me very much	.58
I really enjoy a task that involves coming up with new solutions to problems	.55

Reverse scoring of items in italics.

The internal reliability of nine measures of the NFC scale were considered: 18 item, and nine to two item questionnaires. In order to select items for inclusion in the short-form questionnaires, individual items were correlated with the sum of the eighteen-item questionnaire. These correlations were ranked and those items with the lowest correlations were systematically removed to form the short-forms.

Table 2: items loading greater than .55 on factor 1 correlated with the sum of all eighteen items (reverse scoring of items in italics.)

	sum 18 item correlations
Thinking is not my idea of fun	0.70
I would rather do something that requires little thought then something that is sure to challenge my thinking abilities	0.66
The idea of relying on thought to make my way to the top appeals to me	0.60
The notion of thinking abstractly is appealing to me	0.57
I like to have the responsibility of handling a situation that requires a lot of thinking	0.56
I really enjoy a task that involves coming up with new solutions to problems	0.56
It's enough for me that something gets the job done, I don't care how or why it works	0.54
Learning new ways to think doesn't excite me very much	0.54
I try to anticipate and avoid situations where there is a likely chance I will have to think in depth about something	0.52

Table 3: Chronenbach's alpha coefficients for nine short-form questionnaires

	Alpha coefficient
18 items	0.81
9 items	0.83
8 items	0.81
7 items	0.79
6 items	0.77
5 items	0.76
4 items	0.76
3 items	0.74
2 items	0.72

Table 4: short-form and 18 item questionnaire correlations

	Eighteen item questionnaire
9 items	0.90
8 items	0.90
7 items	0.89
6 items	0.89
5 items	0.86
4 items	0.83
3 items	0.81
2 items	0.77

Summary: the first factor extracted by the principal component analysis accounted for 26% of the variance which is comparable with Cacioppo *et al's* original 34 item questionnaire. All the short-form questionnaires had satisfactory internal reliability

scores (>0.70). Following the correlation of the short-form questionnaires with the eighteen item scale, only two scales had correlation coefficients of greater than 0.90, the eight and nine item scales. In summary, the eight item short-form appears to be as reliable as the nine item and is comparable with the eighteen item.

Items to be included in the short-form need for cognition scale (reverse scoring of items in italics).

Thinking is not my idea of fun
I would rather do something that requires little thought then something that is sure to challenge my thinking abilities
The idea of relying on thought to make my way to the top appeals to me
The notion of thinking abstractly is appealing to me
I like to have the responsibility of handling a situation that requires a lot of thinking
I really enjoy a task that involves coming up with new solutions to problems
It's enough for me that something gets the job done, I don't care how or why it works
Learning new ways to think doesn't excite me very much

Hilary Bekker, Department of Psychology, University of Leeds, LS2 9JT
Mark Conner, Department of Psychology, University of Leeds, LS2 9JT
(November, 1995)

Appendix XIII

Follow-up Questionnaire (T2): facilitating informed decision making.

LEEDS GENERAL INFIRMARY ANTE-NATAL STUDY

Prenatal Testing Questionnaire (T2)

Study Number ..... **Group: 1 (r) / 2 (da) tt / age**

The following questionnaire is the final part of the project looking at how women make decisions during pregnancy. The questionnaire takes about fifteen minutes to complete. Filling in the questionnaire will not interfere with the care you receive from this clinic.

Your experiences and views of making decisions in pregnancy are important to this study. The results of this study will provide us with a better understanding of what information women find useful at this stage in their pregnancies.

If you have any questions about the study, or the questionnaire, please contact Hilary Bekker (0113 233 6696). The answers you give in this questionnaire are confidential and anonymous. Only Hilary can match your study number with your name.

Please return the questionnaire in the stamped-addressed envelope provided.

Thank you for your time.

Hilary Bekker
Clinical Researcher,
University of Leeds.

Mr Jim Thornton
Consultant in Obstetrics and
Gynaecology, Leeds General Infirmary.

Study number .....

The following questions ask you for some details about tests you had during this pregnancy.

1. Your increased risk figure for Down syndrome was 1 in _____
Would you describe this risk as a low ☐ medium☐ high ☐

A number of statements which people have used to describe themselves are given below. Read each statement and then circle the most appropriate number to the right of the statement to indicate how you feel right now, at this moment. There are no right or wrong answers. Do not spend too much time on any one statement but give the answer which seems to describe your present feelings best.

		not at all	somewhat	moderately	very much
2.	**I feel calm .....**	1	2	3	4
3.	**I feel tense .....**	1	2	3	4
4.	**I am upset .....**	1	2	3	4
5.	**I feel relaxed .....**	1	2	3	4
6.	**I feel content .....**	1	2	3	4
7.	**I am worried .....**	1	2	3	4

The following question asks for your views and feelings about prenatal testing in general.

8. Some people think there are both advantages and disadvantages to having tests in pregnancy. In the box below, please list the advantages and disadvantages that are important to you and rate how much of an advantage or disadvantage it is (no more than six of each).

Advantages	
	small advantage 1 2 3 4 5 great advantage
	small advantage 1 2 3 4 5 great advantage
	small advantage 1 2 3 4 5 great advantage
	small advantage 1 2 3 4 5 great advantage
	small advantage 1 2 3 4 5 great advantage
Disadvantages	
	small disadvantage 1 2 3 4 5 great disadvantage
	small disadvantage 1 2 3 4 5 great disadvantage
	small disadvantage 1 2 3 4 5 great disadvantage
	small disadvantage 1 2 3 4 5 great disadvantage
	small disadvantage 1 2 3 4 5 great disadvantage

The following questions are about how you feel about the decision to have or not to have amniocentesis or chorionic villus sampling (CVS).After reading the following statements, please circle a number that reflects how much you agree with them

9. The decision to have (not to have) a test was hard to make:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

10. I was unsure whether to have/ not have a test:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

11. It was clear what the best choice was for me:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

12. I was aware of the choices I had based on my triple test result:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

13. I feel I know what the benefits of having a test are:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

14. I feel I know what the risks of having a test are:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

15. I feel I made an informed choice about whether to have a test or not:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

16. My decision shows what is most important for me:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

17. The decision to have or not have a test was entirely up to me:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

18. I am satisfied with my decision:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

19. I expected to stick with my decision:

strongly agree	agree	neither agree nor disagree	disagree	strongly disagree
1	2	3	4	5

The following questions ask how useful the information about amniocentesis (CVS) was to you.

20. Was there anything you wanted more information about? **yes/ no**
if yes please state.

21. Please write below what bits of information were helpful and unhelpful to you when making the decision to have amniocentesis (chorionic villus sampling) or not:

Helpful information	Unhelpful information

22. Overall, how useful was the information given when making this decision:

not at all useful very useful

0 ------ 1 ------ 2 ------ 3 ------ 4 ------ 5 ------ 6

23. Since having the amniocentesis (CVS) test, or not, have you looked for information about the following topics *(please tick one response for each topic)*

		I looked for information:		***Someone gave me information:***	
(a)	triple test	no ☐	yes ☐	no ☐	yes ☐
(b)	amniocentesis	no ☐	yes ☐	no ☐	yes ☐
(c)	chorionic villus sampling	no ☐	yes ☐	no ☐	yes ☐
(d)	Down's syndrome	no ☐	yes ☐	no ☐	yes ☐
(e)	other diseases/ disabilities	no ☐	yes ☐	no ☐	yes ☐
(f)	termination of pregnancy	no ☐	yes ☐	no ☐	yes ☐
(g)	other (please state)			.....	

24. Do you regret having had the triple test at all *(please circle one response):*

no regret — regret very much

0 ------ 1 ------ 2 ------ 3 ------ 4 ------ 5 ------ 6

25. Do you regret the decision to have, or not have, amniocentesis (chorionic villus sampling) *(please circle one response):*

no regret — regret very much

0 ------ 1 ------ 2 ------ 3 ------ 4 ------ 5 ------ 6

26. Imagine a close friend the same risk of having a baby with Down syndrome as you and she asks you for advice about amniocentesis (chorionic villus sampling). Would you encourage her to have amniocentesis (chorionic villus sampling) *(please circle one response)*?

strongly encourage	encourage	neither encourage/ nor discourage	discourage	strongly discourage
1	2	3	4	5

27. Do you feel the following people encouraged you to have or not have amniocentesis (chorionic villus sampling). Please circle one response for each person.

		did not discuss	strongly encouraged	encouraged	neither encouraged nor discouraged	discouraged	strongly discourag
a	midwives	☐	1	2	3	4	5
b	obstetrician	☐	1	2	3	4	5
c	general practitioner	☐	1	2	3	4	5
d	the research	☐	1	2	3	4	5
e	husband / partner	☐	1	2	3	4	5
f	other	☐	1	2	3	4	5

Some women find pregnancy a worrying time, how worried you are about the following?

28. How worried are you that the following harm the fetus *(please circle one number for each item)*:

not at all worried — very worried

(a)	ultrasound scan	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------ 6
(b)	emotional upset	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------ 6
(c)	triple test	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------ 6
(d)	amniocentesis	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------ 6
(e)	taking medication	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------ 6
(f)	chorionic villus sampling	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------ 6

29. How worried are you about the baby having ***(please circle one number for each item):***

		not at all worried						very worried
(a)	a physical disability	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(b)	a mental disability	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(c)	a serious health problem	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6

30. How reassured were you about the health of the baby when you received the following test results ***(please circle one response for each test)*****:**

		not at all reassured						very reassured
(a)	ultrasound screening	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(b)	triple test	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
(c)	amniocentesis (CVS)	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6

The following questions are about your views on disability and how likely you think the possible consequences of testing are to happen to you (please circle one number for each question)

31. Do you feel a child's life would be affected by the following: ***(please circle a response for each item)***

Condition	*how affected*	not at all affected						greatly affected
cleft palate/lip	quality of life	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
	amount of care	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
Down syndrome	quality of life	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
	amount of care	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
spina bifida	quallty of life	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
	amount of care	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6

32. How likely do you think you are to:	**not at all likely**						**very likely**
have a healthy child	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
have a child with Down syndrome	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
miscarry a fetus without Down syndrome	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
miscarry a fetus with Down syndrome	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
terminate a pregnancy for spina bifida	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
terminate a pregnancy for a cleft palate	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
terminate a pregnancy for Down syndrome	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6

33. Most people who are important to me think I should:	**should**						**should not**
have had the triple test for Down syndrome	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
have had a diagnostic test	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
terminate a pregnancy if the fetus has Down syndrome	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6

34. Compared with other women your age:	**less likely**			**about the same**			**more likely**
how healthy would you say you were	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
how likely to develop a health problem	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
how likely to have a healthy child	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
how likely have a child with Down syndrome	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6
how likely to miscarry a pregnancy	0 ------	1 ------	2 ------	3 ------	4 ------	5 ------	6

The next part of the questionnaire asks about <u>your</u> opinions towards pregnancy and disease. How much do you agree with the following statements (please circle one number for each question).

35. Tests during pregnancy are necessary for my well-being:

strongly disagree	disagree	neither agree nor disagree	agree	agree strongly
1	2	3	4	5

36. Tests during pregnancy improve my chance of having a healthy baby:

strongly disagree	disagree	neither agree nor disagree	agree	agree strongly
1	2	3	4	5

37. Tests during pregnancy are necessary for the well-being of the fetus:

strongly disagree	disagree	neither agree nor disagree	agree	agree strongly
1	2	3	4	5

38. Tests during pregnancy decrease my chance of miscarrying:

strongly disagree	disagree	neither agree nor disagree	agree	agree strongly
1	2	3	4	5

The following questions are about your knowledge of tests in pregnancy. Do not worry if you do not know all the answers:

39. The following are tests for *(please tick all boxes that apply for <u>each</u> test)*?

	all health abnormalities	Down syndrome	all known chromosomal abnormalities	spina bifida	none of these	don't know
triple test						
amniocentesis						
CVS						

40. The following three tests use what type of sample *(please tick one box for <u>each</u> test)*?

	your blood	your urine	your baby's blood	fluid from around the baby	cells from the placenta	don't know
triple test						
amniocentesis						
CVS						

41. Are the following *screening procedures* (telling you whether you are <u>more</u> or <u>less likely</u> to have a baby with a disability), *diagnostic procedures* (give you as definite a 'yes' or 'no' answer as medicine can) or *neither* (e.g. routine checks) *(please tick <u>one</u> response for each test).*

(a) woman's age screening test ☐ diagnostic test ☐ neither ☐ don't know ☐
(b) triple test screening test ☐ diagnostic test ☐ neither ☐ don't know ☐
(c) amniocentesis screening test ☐ diagnostic test ☐ neither ☐ don't know ☐
(d) ultrasound scan screening test ☐ diagnostic test ☐ neither ☐ don't know ☐
(e) cvs: screening test ☐ diagnostic test ☐ neither ☐ don't know ☐

42. A negative test result for the following tests *usually* means (*please tick one item only for each test)*?

	the baby definitely does not have Down's	it is unlikely the baby has Down's	the baby might have Down's	the baby definitely does have Down's	don kno
triple test					
amniocentesis					
CVS					

43. A positive test result for the following tests *usually* means (*please tick one item only for each test)*?

	the baby definitely does not have Down's	it is unlikely the baby has Down's	the baby might have Down's	the baby definitely does have Down's	don kno
triple test					
amniocentesis					
CVS					

44. How long does it take for the results from each test to come back *(please tick one box for each test)*?

	1 to 2 days	1 to 2 weeks	3 to 4 weeks	don't know
triple test				
amniocentesis				
CVS				

45. The extra risk of miscarriage after having the following three tests is *(please tick one box for each test)*?

	no extra risk	greater than 1 in 50	1 in 50 (2%)	1 in 100 (1 %)	1 in 150 (0.7%)	1 in 200 (0.5%)	less than 1 in 200	don't know
triple test								
amniocentesis								
CVS								

Thank you for answering all the questions and returning the questionnaire in the stamped addressed envelope.

Appendix XIV

Study sample representativeness: facilitating informed decision making.

The data summarised in the following tables summarise the analyses carried out to assess the representativeness of the sample and comparability of women within the randomised control trial study groups (section 5.2.1 and 5.2.2).

Table 1: women receiving a screen positive triple test result by participation (n=178).

	Frequency	%
participated: invited, participated, completed T1 questionnaire	**106**	**60 %**
excluded random: invited, participated, did not complete T1	**4**	**2 %**
excluded random: invited, participated, screened negative	**7**	**4 %**
declined: invited, declined participation	**15**	**8 %**
excluded telephone: no invite, decision made before consultation by telephone	**18**	**10 %**
excluded criteria: no invite, study criteria not met	**28**	**16 %**

Table 2: Reproductive history by study participation (n=173; 5 missing values).

	Participated		Excluded: Random		Declined		Excluded: Telephone		Excluded: Criteria		Total	
	n=106	%	n=11	%	n=15	%	n=18	%	n=23	%	n=173	%
had children	68	**64**	7	**64**	9	**60**	13	**72**	14	**61**	111	**64**
miscarried	27	**25**	3	**27**	4	**27**	7	**39**	7	**30**	48	**28**
terminated	3	**3**	0	**0**	2	**13**	3	**17**	2	**9**	10	**6**
family history abnormality	24	**23**	2	**18**	2	**13**	2	**11**	7	**30**	37	**21**

Table 3: Age and gestation by study participation (n=173; 5 missing values).

	Participated n=106		Random: Excluded n=11		Declined n=15		Excluded: Telephone n=18		Excluded: Criteria n=26/25		Total n=176 /175	
	x	(sd)	x	(sd)	x	(sd)	x	(sd)	x	(sd)	x	(sd)
age	**35.2**	(3.0)	**35.3**	(3.1)	**35.3**	(4.3)	**35.6**	(2.8)	**35.7**	(3.5)	**35.3**	(3.2)
gestation	**14.8**	(1.0)	**14.8**	(1.2)	**15.0**	(1.4)	**14.9**	(1.1)	**14.5**	(1.2)	**14.7**	(1.1)

Table 4: Diagnostic test decision by study participation (n=169; 9 missing values).

	Participated		Random: Excluded		Declined		Telephone: Excluded		Criteria: Excluded		Total	
no test	17	**16%**	5	**45%**	1	**7%**	5	**28%**	6	**NA**	34	**NA**
test	89	**84%**	6	**55%**	14	**93%**	13	**72%**	13	**NA**	135	**NA**

NA: no analysis or percentages provided because there were 9 missing values.

Table 5: Differences in demographics and reproductive history by participation (n=178).

	Test	F or Chi^2 value	d.f.	Significance
had children	Chi^2	0.7	4	**0.95**
miscarried	Chi^2	1.5	4	**0.83**
terminated	NA	NA	NA	**NA***
family history abnormality	NA	NA	NA	**NA***
test decision	Chi^2	9.4	4	**0.05**
age	MANOVA	0.1	4,170	**0.97**
gestation	MANOVA	0.7	4,170	**0.60**

**NA (no analysis) too little variation in responses to carry out meaningful analyses.*

Table 6: Pre-consultation anxiety by participation (n=131; 1 missing value)

	Total n=131		Participated n=106		Excluded: -ve result n=5		Excluded: no T1 n=5		Declined n=15	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	x	(s.d.)	x	(s.d.)
pre-consultation anxiety	**62.6**	(13.9)	**62.1**	(13.6)	**53.3**	(14.3)	**60.7**	(23.4)	**69.6**	(10.2)

Table 7: Socio-demographic characteristics by study group (n=106)

	Total n=106		Routine n=56		Decision Analysis n=50		Pearson Coefficient		
Level	n	%	n	%	n	%	d.f.	Chi^2	**Sig.**
Ethnic origin:									
white	103	**97%**	54	**96%**	49	**98%**			
other	3	**3%**	2	**4%**	1	**2%**	NA	NA	**NA**
Marital status:									
married	81	**76%**	40	**71%**	41	**82%**			
living as married	22	**21%**	13	**23%**	9	**18%**			
single	3	**3%**	3	**6%**	0	**0%**	2	3.4	**0.18**
Education:									
up to GCSE	46	**44%**	25	**44%**	21	**42%**			
'A' level equiv.	33	**31%**	16	**29%**	17	**34%**			
degree +	27	**25%**	15	**27%**	12	**24%**	2	0.4	**0.83**
Religious Activity:									
not active	66	**63%**	35	**63%**	31	**62%**			
occasional	31	**29%**	17	**30%**	14	**28%**			
frequent	9	**8%**	4	**7%**	5	**10%**	2	0.3	**0.86**

**NA = no analysis carried out, too little variation responses*

Table 8: Experience of prenatal testing by study group (n=81)

	Total n=81		Routine n=40		Decision Analysis n=41		Pearson Coefficient		
	n	%	n	%	n	%	d.f.	Chi^2	**Sig.**
last pregnancy: scan	61	**75%**	27	**68%**	34	**83%**	1	2.6	**0.11**
last pregnancy: triple test	31	**38%**	13	**33%**	18	**44%**	1	1.1	**0.29**
last pregnancy: diagnostic	4	**5%**	1	**3%**	3	**7%**	NA	NA	**NA***

**NA = no analysis carried out too little variation in responses*

Table 9: Reproductive history by study group (n=106)

	Total n=106		Routine n=56		Decision Analysis n=50		Pearson Coefficient		
	n	%	n	%	n	%	d.f.	Chi^2	**Sig.**
have children	68	**64%**	32	**57%**	36	**72%**	1	2.5	**0.11**
miscarried	27	**25%**	15	**27%**	12	**24%**	1	0.1	**0.74**
terminated	3	**5%**	1	**2%**	2	**4%**	NA	NA	**NA***
family history abnormality	24	**23%**	16	**29%**	8	**16%**	1	2.4	**0.12**
companion present	78	**74%**	41	**73%**	37	**74%**	1	0.01	**0.93**

**NA = no analysis carried out, too little variation responses*

Table 10: Age, gestation, anxiety, optimism & need for cognition by study group (n=106).

	Total n=106		Routine n=56		Decision Analysis n=50		MANOVA d.f. 1, 104	
	x	(s.d.)	**x**	(s.d.)	**x**	(s.d.)	f value	**sig.**
age	**35.2**	(3.0)	**34.9**	(2.9)	**35.6**	(3.2)	1.5	**0.2**
gestation	**14.8**	(1.0)	**15.0**	(1.0)	**14.5**	(0.9)	6.0	**0.02**
pre-consultation anxiety	**62.2**	(13.6)	**62.3**	(12.9)	**62.0**	(14.4)	0.01	**0.9**
life orientation test (LOT)	**21.0**	(6.5)	**22.3**	(6.4)	**19.5**	(6.2)	5.2	**0.03**
need for cognition (NFC)	**81.7**	(20.8)	**81.2**	(19.1)	**82.3**	(22.7)	0.0	**0.8**

Appendix XV

Representativeness of sample returning follow-up questionnaires (T2): facilitating informed decision making.

The data summarised in the following tables summarises the analyses carried out to assess the representativeness of those women that returned the follow-up questionnaire (T2) (section 5.

Table 1: Study group allocation by completion T2.

	Total (100)	T1Q only (32)		T1&T2Q (68)		Chi^2 (d.f. = 1)	
	n	n	%	n	%	**Chi^2**	**Sig.**
routine	52	13	**25%**	39	**75%**		
decision analysis	48	19	**40%**	29	**60%**	**2.4**	**0.12**

Table 2: Socio-demographic characteristics and reproductive history by completion T2.

	Total (100)	T1Q only (32)		T1&T2Q (68)		Pearson Chi^2		
	n	n	%	n	%	d.f	Chi^2	**Sig.**
had children	63	24	**38%**	39	**62%**			
no children	37	8	**22%**	29	**78%**	1	2.9	**0.09**
had miscarried	25	7	**28%**	18	**72%**			
no miscarriage	75	25	**33%**	50	**67%**	1	0.2	**0.62**
had family history abnormality	22	11	**50%**	11	**50%**			
no family history abnormality	78	21	**27%**	57	**73%**	1	4.2	**0.04**
Education:								
up to GCSE	42	20	**48%**	22	**52%**			
'A' level equiv.	33	9	**27%**	24	**73%**			
degree +	25	3	**12%**	22	**88%**	2	9.6	**0.008**
Religious attendance:								
not active	64	21	**33%**	43	**67%**			
occasional	27	9	**33%**	18	**67%**			
frequent	9	2	**22%**	7	**88%**	2	0.4	**0.80**

Table 3: Socio-demographic characteristics and individual differences by completion T2.

	Total n=100		T1 only n=32		T1 & T2 n=68		MANOVA d.f. = 1,98	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	**Sig.**
age	**35.2**	(3.1)	**36.0**	(2.7)	**34.9**	(3.2)	3.0	**0.09**
gestation	**14.8**	(1.0)	**14.6**	(1.1)	**14.8**	(0.9)	1.7	**0.20**
optimism (LOT)	**21.2**	(6.5)	**23.1**	(6.7)	**20.3**	(6.3)	4.3	**0.04**
need for cognition	**81.6**	(21.2)	**82.8**	(23.3)	**81.1**	(20.4)	0.1	**0.72**

Table 4: Test decision by completion T2.

	Total (100)	T1Q only (32)		T1&T2Q (68)		Pearson Chi^2	
	n	n	%	n	%	**R**	**Sig.**
no test	17	4	**24%**	13	**76%**		
test	83	28	**34%**	55	**66%**	**0.67**	**0.41**

Table 5: Measures of affect, cognitive processes and clinical quality by completion T2.

	Total n=100		T1 only n=32		T1 & T2 n=68		MANOVA d.f. = 1,98	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	Sig.
anxiety-before consultation	**62.1**	(13.7)	**63.5**	(14.2)	**61.5**	(13.5)	0.49	**0.40**
anxiety (T1)	**59.7**	(15.0)	**61.9**	(16.0)	**58.6**	(14.6)	1.0	**0.32**
conflict - uncertainty	**8.5**	(2.8)	**9.0**	(2.5)	**8.2**	(2.9)	1.5	**0.22**
conflict - informed	**6.9**	(1.7)	**6.7**	(1.5)	**7.0**	(1.7)	0.5	**0.48**
conflict - efficacy	**7.6**	(2.3)	**7.8**	(2.3)	**7.5**	(2.2)	0.2	**0.63**
IDM: information seeking	**2.8**	(1.0)	**2.9**	(1.2)	**2.8**	(1.0)	0.3	**0.58**
IDM: reasons for	**2.3**	(1.0)	**2.5**	(1.1)	**2.2**	(1.0)	1.6	**0.20**
IDM: reasons against	**2.1**	(1.1)	**2.3**	(1.2)	**2.0**	(1.0)	1.5	**0.23**
advantages testing	**1.7**	(0.9)	**1.5**	(0.8)	**1.8**	(0.9)	2.3	**0.13**
disadvantages testing	**1.5**	(1.0)	**1.5**	(1.0)	**1.5**	(1.0)	0.004	**0.95**
knowledge	**14.6**	(3.1)	**14.3**	(2.0)	**14.7**	(3.5)	0.4	**0.54**
useful information	**5.1**	(1.1)	**5.3**	(1.1)	**5.1**	(1.1)	0.9	**0.36**
length consultation	**28.9**	(12.3)	**32.4**	(15.9)	**27.3**	(9.8)	3.9	**0.05**

Table 6: SLIWC variables of decision making process by completion T2 (covariate consultation length).

	Total n=100		T1Q n=32		T1&T2Q n=68		MANCOVA d.f. = 1,97	
	x	(s.d.)	x	(s.d.)	x	(s.d.)	f	Sig.
SLIWC: word count	**1121**	(916)	**1419**	(1310)	**982**	(618)	1.3	**0.26**
SLIWC: positive emotion	**1.0**	(0.7)	**2.1**	(0.8)	**1.9**	(0.7)	1.5	**0.23**
SLIWC: negative emotion	**0.9**	(0.5)	**1.0**	(0.5)	**0.8**	(0.5)	2.6	**0.11**
SLIWC: cognitive	**11.2**	(1.8)	**11.4**	(1.7)	**11.2**	(1.8)	0.2	**0.69**
SLIWC: social	**9.5**	(2.2)	**9.8**	(2.3)	**9.3**	(2.1)	0.2	**0.65**

Table 7: EUVs for the consequences of prenatal testing and perceived social norm by completion T2.

	Total (100)		T1Q only (32)		T1 &T2Q (68)		Mann-Whitney		
	x	(s.d.)	x	(s.d.)	x	(s.d.)	U	z	Sig.
EUT1 - have healthy baby	**39.5**	(10.7)	**40.6**	(11.9)	**39.0**	(10.1)	997.0	-0.7	**0.47**
EUT2 - misc. Downs baby	**12.7**	(10.5)	**12.5**	(10.8)	**12.8**	(10.5)	1073.5	-0.1	**0.88**
EUT3 - term. Downs baby	**27.8**	(21.4)	**27.8**	(22.2)	**27.9**	(21.2)	NA	NA	NA
EUT4 - have Downs baby	**3.6**	(6.7)	**3.9**	(8.8)	**3.4**	(5.4)	994.5	-0.7	**0.75**
EUT5 - misc. normal baby	**1.2**	(2.1)	**0.9**	(1.8)	**1.3**	(2.3)	1046.5	-0.3	**0.13**
perceived social norm	**1.5**	(1.7)	**1.3**	(1.6)	**1.6**	(1.7)	1017.5	-0.5	**0.59**

Table 8: EUV for termination Down syndrome by completion T2.

	Total (100)	T1Q only (32)		T1&T2Q (68)		Chi^2 (d.f.= 2)	
	n	n	%	n	%	**Chi^2**	Sig.
EUT value <30	56	19	**34%**	37	**66%**		
EUT value >30	44	13	**30%**	31	**70%**	0.2	0.64

Table 9: Ability to rank order the consequences prenatal testing by completion T2.

	Total (100)	T1Q only (32)		T1&T2Q (68)		Chi^2 (d.f.= 2)	
	n	n	%	n	%	**Chi^2**	**Sig.**
ranked	47	16	**34%**	31	**66%**		
not ranked	53	16	**30%**	37	**70%**	**0.17**	**0.68**

Table 10: Perception triple test risk by completion T2.

	Total (100)	T1Q only (32)		T1&T2Q (68)		Chi^2 (d.f.= 2)	
	n	n	%	n	%	**Chi^2**	**Sig.**
low risk	12	5	**42%**	7	**58%**		
medium risk	41	13	**32%**	28	**68%**		
high risk	47	14	**30%**	33	**70%**	0.6	**0.73**

Table 11: Perceived encouragement health professionals' information by completion T2.

	Total (100)	T1Q only (32)		T1&T2Q (68)		Chi^2 (d.f.= 2)	
	n	n	%	n	%	**Chi^2**	**Sig.**
not discussed	51	18	**35%**	33	**65%**		
neutral	40	9	**23%**	31	**77%**		
encouraged	9	5	**16%**	4	**6%**	**4.21**	**0.12**

Table 12: Perceived encouragement researcher information by completion T2.

	Total (100)	T1Q only (32)		T1&T2Q (68)		Chi^2 (d.f.= 2)	
	n	n	%	n	%	**Chi^2**	**Sig.**
not discussed	9	5	**56%**	4	**44%**		
neutral	83	22	**27%**	61	**73%**		
encouraged	8	5	**63%**	3	**37%**	**6.87**	**0.03**

Table 13: Correlations between level education, family history and optimism with T2 measures of decision quality (n=68).

	EDUCAT	NFAMHIS	TOPTIM	EUT1.T2	EUT2.T2	EUT4.T2
EDUCAT	1.0000					
	P= .					
NFAMHIS	-.0205	1.0000				
	P= .868	P= .				
TOPTIM	-.1749	-.0176	1.0000			
	P= .154	P= .887	P= .			
EUT1.T2	.0956	.0076	-.2621	1.0000		
	P= .438	P= .951	P= .031	P= .		
EUT2.T2	-.0273	-.0737	.1584	-.2423	1.0000	
	P= .825	P= .550	P= .197	P= .047	P= .	
EUT4.T2	-.1622	-.1054	.3848	-.0645	.1680	1.0000
	P= .186	P= .392	P= .001	P= .601	P= .171	P= .
EUT5.T2	.0604	.0895	-.0125	-.1417	.0959	.0582
	P= .625	P= .468	P= .920	P= .249	P= .437	P= .637
SHOLD2.2	-.0081	-.1198	.1429	-.0706	.1979	.3546
	P= .947	P= .331	P= .245	P= .568	P= .106	P= .003
TADVT2	.2300	.0876	-.1314	.4506	-.2846	-.2824
	P= .059	P= .478	P= .286	P= .000	P= .019	P= .020
TDIST2	.2256	.0592	-.0958	.3254	-.3750	-.2364
	P= .064	P= .632	P= .437	P= .007	P= .002	P= .052
TKNOWT2	.3296	.1116	-.3427	.2300	-.2150	-.1296
	P= .006	P= .365	**P= .004**	P= .059	P= .078	P= .292
USEFULT2	-.2033	.0293	-.1782	.2831	-.4098	-.2004
	P= .096	P= .813	P= .146	P= .019	P= .001	P= .101
T2DECEFF	.0501	.0165	.3246	.0648	.0489	.2641
	P= .685	P= .894	**P= .007**	P= .599	P= .692	P= .030
T2DECINF	-.0891	-.0469	.2983	-.0004	.2068	.2492
	P= .470	P= .704	**P= .013**	P= .997	P= .091	P= .040
T2DECUNC	-.0035	.0826	-.0016	.1071	-.0467	.0724
	P= .977	P= .503	P= .989	P= .385	P= .706	P= .557
TANXT2	-.0694	-.0629	.3784	-.1348	.2146	.2343
	P= .574	P= .610	**P= .001**	P= .273	P= .079	P= .055
RISKPER2	-.1444	-.0645	.0759	-.1226	-.1301	-.3434
	P= .240	P= .601	P= .539	P= .319	P= .290	P= .004

Table 14: Correlations between consultation length and T2 quality of decision measures.

	EUT1.T2	EUT2.T2	EUT3.T2	EUT4.T2	EUT5.T2	RISKPER2
LENCONS	.1323	-.0671	-.0552	.1550	-.0887	-.1009
	P= .282	P= .586	P= .655	P= .207	P= .472	P= .413

	SHOLD2.2	TADVT2	TDIST2	TANXT2	TKNOWT2	T2DECUNC
LENCONS	.2077	-.0474	.0363	.0156	.1369	.2651
	P= .089	P= .701	P= .769	P= .900	P= .266	**P= .029**

	T2DECINF	T2DECEFF	USEFULT2	LENCONS
LENCONS	.0738	.1368	-.2191	1.0000
	P= .550	P= .266	**P= .073**	P= .

Table 15: Correlations between researcher information and T2 quality of decision measures.

	DIRRES	EUT1.T2	EUT2.T2	EUT3.T2	EUT4.T2	EUT5.T2
DIRRES	1.000	-.0928	-.1562	-.0002	.1630	.0814
	P= .	P= .452	P= .203	P= .998	P= .184	P= .509

	RISKPER2	SHOLD2.2	TADVT2	TDIST2	TANXT2	TKNOWT2
DIRRES	-.1127	.0179	-.0992	-.0183	.0403	.0608
	P= .360	P= .885	P= .421	P= .882	P= .744	P= .622

	T2DECUNC	T2DECINF	T2DECEFF	USEFULT2
DIRRES	.0354	.0876	-.1239	-.1892
	P= .774	P= .477	P= .314	P= .122

Table 16: Correlations between gestation and T2 quality of decision measures.

	EUT1.T2	EUT2.T2	EUT3.T2	EUT4.T2	EUT5.T2	RISKPER2
GESTAT	-.1946	.2087	-.0026	.1812	.0181	.0826
	P= .112	P= .088	P= .983	P= .139	P= .883	P= .503

	SHOLD2.2	TADVT2	TDIST2	TANXT2	TKNOWT2	T2DECUNC
GESTAT	.1977	-.1975	-.0880	.0307	-.1330	.0510
	P= .106	P= .107	P= .475	P= .804	P= .280	P= .680

	T2DECINF	T2DECEFF	USEFULT2	GESTAT
GESTAT	.1598	.2272	-.1939	1.0000
	P= .193	P= .062	P= .113	P= .