

**Understandings of Down's syndrome and their
place in the prenatal testing context**

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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: There is a growing consensus that decisions about prenatal testing should a) be informed, and b) reflect the individual's attitudes and values. Most research has focused on information and attitudes in relation to the tests but there has been little attention given to these factors in relation to the target condition.

Aims: This thesis is concerned with informed choice in the context of prenatal testing for Down's syndrome. The main aims were to describe the written information that women receive about Down's syndrome prior to prenatal screening, characterise understandings of Down's syndrome that exist independently of the testing context, and identify the relationships between understandings of Down's syndrome, intentions towards using testing and termination, and actual screening choices.

Methods and Results:

Study 1 employed a content analysis of information about Down syndrome contained in 80 prenatal screening leaflets. Information about Down's syndrome was low in quantity (the median number of statements was one and 33% percent of leaflets contained no descriptive information on the nature of the condition). The majority of statements (63%) were rated as negative in tone, (25% were rated as neutral and 19% were rated as positive). 89% of the statements were of a medical, clinical or epidemiological nature and 11% concerned social, educational or psychosocial issues associated with Down's syndrome

Study 2 used Q methodology to characterise understandings of Down's syndrome. 76 people chosen as being likely to represent a diverse range of views Q sorted 50 beliefs about Down's syndrome. Five statistically independent understandings of the condition were extracted using Principal Components Analysis. There was a consensus across participants on the rights of existing people with Down's syndrome to a good quality of life, but there were significant differences in to how respondents believed they personally would cope with, and adjust to an affected child. Some tentative associations between these five understandings and attitudes towards testing and termination were identified.

Study 3 employed a self-completion questionnaire in 197 pregnant women to measure attitudes towards Down's syndrome and intentions to test and terminate for the condition. Serum screening uptake was collected later from patient records. Attitudes towards Down's syndrome were significantly associated with intentions to use screening, diagnostic tests, and termination, and also with actual screening uptake ($p < 0.05$). However, most women accepted screening tests (77% overall) regardless of whether their attitude towards Down's syndrome was favourable or not. Attitudes towards Down's syndrome were most strongly associated with intentions to terminate

a pregnancy for the condition. Women who were uncertain about terminating for Down's syndrome had significantly higher levels of ambivalence in their attitudes towards the condition than women whose behavioural intentions were either 'yes' to termination or 'no' to termination.

Discussion: The findings suggest that a) guidelines regarding informed choice are not being met in the case of written information provided about the target condition and b) screening choices might not always be directly informed either by attitudes towards Down's syndrome or towards termination for the condition. Further investigation into the psychological and situational factors associated with testing and termination choices is recommended.

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ABBREVIATIONS

AFP	- Alpha Fetoprotein
CF	- Cystic fibrosis
CVS	- Chorionic Villus Sampling
DS	- Down's syndrome
FISH	- Fluorescent <i>in situ</i> hybridisation
LD	- Learning difficulty
NTDs	- Neural Tube Defects
PQoL	- Parental quality of life
TRA	- Theory of Reasoned Action
TPB	- Theory of Planned Behaviour

CHAPTER 1 INTRODUCTION AND BACKGROUND

“I have never, ever, in my life come across anything as complicated as prenatal testing. Morally, psychologically, politically, socially – on every level, I have never come up against anything as difficult” (Rothman, 1997).

The views of Barbara Katz Rothman author of “The Tentative Pregnancy” (considered the seminal work on women’s experiences of prenatal testing in America (Rothman, 1986)) are echoed by many who have been involved in research in this area. Over the past twenty years or so researchers have tried to gain a better understanding of the psychological correlates and consequences of such tests, and the issues are indeed complex (Green *et al.*, 2002). Down’s syndrome (in the physiological sense) is at least as old as mankind itself¹, prenatal testing and selective termination for Down’s syndrome on the other hand have been around for less than 40 years. Thus understandings of the condition and attitudes towards affected individuals long preceded prenatal testing. However, while a number of studies have examined attitudes towards prenatal testing and termination for Down’s syndrome, there has been virtually no research that has specifically examined how women’s understandings and attitudes toward the condition relates to prenatal testing choices. Neither has there been much research emphasis on examining the information provided about the condition prior to testing choices being made. This is surprising, as a major factor in the decision to terminate a pregnancy for abnormality is known to be the perceived severity of the condition diagnosed (Abramsky, Hall, Levitan, and Marteau, 2001; Drugan *et al.*, 1990; Evans, Pryde, Evans, and Johnson, 1993; Evans *et al.*, 1996; Holmes-Siedle, Ryyänen, and Lindenbaum, 1987; Mansfield, Hopfer, and Marteau, 1999; Verp, Bombard, Simpson, and Elias, 1988).

The overall aim of this thesis is to address this gap in the literature and to inform further research and debate in the area of prenatal testing for Down’s syndrome. The remainder of this chapter will provide background and context to the rest of the thesis. The first section considers aspects of Down’s syndrome itself, what it is, how it affects the individual and their family, and some historical and socio-cultural background of attitudes towards the condition. The second section looks at aspects of prenatal testing for the condition, how and why testing was developed, and the

¹ Cases of trisomy 22 in chimpanzees and other apes show manifestations of Down’s syndrome including cognitive impairment. Trisomy 22 in apes is genetically equivalent to trisomy 21 in humans as the former have 24 rather than 23 pairs of chromosomes (Luke, Gandhi, and Verma, 1995).

psychological correlates and consequences of testing. Chapter 2 reviews and critically evaluates the existing literature relating to informed choice in the context of prenatal testing for Down's syndrome. It considers the literature on knowledge and information about Down's syndrome in the testing context, reviews studies that have considered attitudes towards testing for the condition and evaluates previous work that attempts to link understandings of Down's syndrome with attitudes towards using prenatal testing and termination. An overview of the literature reviewed in Chapters 1 and 2 is shown in Figure 1.1. At the end of Chapter 2 the aims of the thesis and the main research questions are set out, followed by an overview of the thesis and the empirical work conducted to answer the research questions.

1.1 DOWN'S SYNDROME

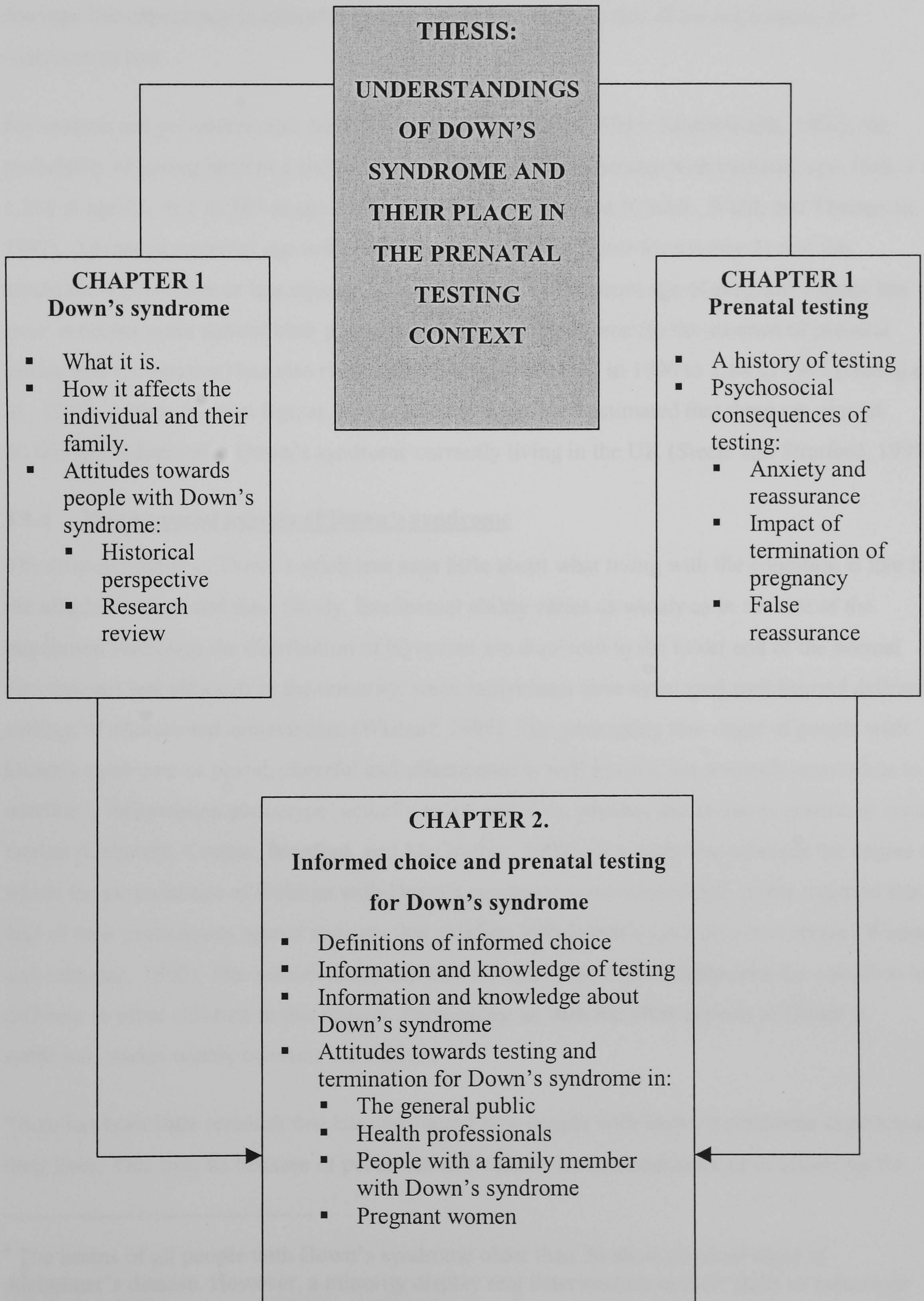
1.1.1 Epidemiology and clinical profile

Down's syndrome is caused by the presence of extra chromosome 21 material in a person's cells and occurs at conception. In about 94% of cases, an extra copy of the chromosome is carried by one of the parental gametes, hence the term trisomy 21. Around 4% of cases arise due to the Robertsonian translocation of chromosome 21 material to another chromosome and in some cases this translocation error can be inherited. The remaining 2% of cases are accounted for by mosaic Down's syndrome, where only some of the affected individual's cells contain an extra copy of chromosome 21. Chromosome 21 contains about 1% of the body's genes and unlike disorders caused by gene mutation (such as cystic fibrosis²), Down's syndrome is due to an alteration in gene quantity rather than quality (Kessling and Sawtell, 1996). However, not all the genes on chromosome 21 contribute to the Down's syndrome phenotype and research suggests that only one or two are responsible for the syndrome's most recognisable characteristics (Delabar *et al.*, 1993). The triple dose of genetic material is associated with between 120 and 300 features (Selikowitz, 1997). Although each individual will have only some of these, common phenotypic features include oblique eye fissures, a transverse palmar crease, slightly overlarge tongue, and short stature. In people with mosaic Down's syndrome the manifestations of the syndrome tend to be less marked. However, all people with Down's syndrome have some degree of intellectual impairment (learning difficulty³). The extra genetic material also brings some hidden effects, such as an increased risk of acute myeloid leukaemia (although in children with Down's syndrome the

² An inherited disorder causing excess production of sticky mucus that impairs respiratory and digestive function and usually leads to progressive respiratory disease. Severity varies and with early diagnosis and treatment life expectancy is now around 25 years but this expectancy continues to increase (Murray and Cuckle, 2001).

³ Learning difficulty is the preferred term of user-led organisations (Goodley, 2000). In this thesis it is used in preference to learning disability, intellectual disability, or mental handicap.

Figure 1.1. Chapters 1 and 2: overview of the literature reviewed



disease is more responsive to chemotherapy) and of an Alzheimer's like presenile dementia of which little is yet understood⁴ (Zaremba, 1996). In addition, approximately 50% of affected babies are born with heart defects, some serious enough to require surgery (Hallidie-Smith, 1996). Average life expectancy is around 60 years, but in line with the rest of the population this continues to rise.

For reasons not yet understood, but long recognised (Penrose, 1933; Shuttleworth, 1909), the probability of giving birth to a child with Down's syndrome increases with maternal age; from 1 in 1,350 at age 25, to 1 in 385 at age 35, to 1 in 30 in a 45 year old (Cuckle, Wald, and Thompson, 1987). Advanced maternal age is the only known predictive factor for trisomy 21 and the condition occurs more or less equally across all races. As the mean age of pregnant women has risen in recent years natural birth prevalence of Down's syndrome (in the absence of prenatal testing and termination) has also risen, from 1.44 per thousand in 1990 to 1.84 in 1997 (Huang *et al.*, 1997). Although exact figures are not known, it has been estimated that there are around 30,000 individuals with Down's syndrome currently living in the UK (Steele and Stratford, 1995).

1.1.2 Psycho-social aspects of Down's syndrome

The clinical profile of Down's syndrome says little about what living with the condition is like for the affected person and their family. Intellectual ability varies as widely as in the rest of the population (although the distribution of IQ scores are displaced to the lower end of the normal distribution) and although in the minority, some individuals have developed well beyond defined ceilings of educational achievement (Wishart, 1995). The personality stereotype of people with Down's syndrome as placid, cheerful and affectionate is well known, but research is mixed as to whether a 'behavioural phenotype' actually exists and if so, whether this is due to genetic or social factors (Collacott, Cooper, Branford, and McGrother, 1998). In a study that assessed the degree to which the personalities of children with Down's syndrome were stereotyped, it was reported that half of their participants agreed strongly that children with Down's syndrome love music (Wishart and Johnston, 1990). The authors noted that no evidence exists that children with the condition are different to other children in this respect. Personality, as with the other aspects of Down's syndrome, varies widely between individuals.

There has been little research that has considered how people with Down's syndrome experience their lives. This may be because of perceived difficulties in communication or in accessing the

⁴ The brains of all people with Down's syndrome older than 30 show physical signs of Alzheimer's disease. However, a minority display real deterioration in their skills or behaviour (Wishart, 1998).

population, a lack of awareness of (or belief in) the relevance of their views, or a belief that people with Down's syndrome do not have the ability to reflect on the quality of their lives. Nevertheless, a number of people with Down's syndrome have written or contributed to books about their life experiences (Williams, 1999) and as research participants, have discussed issues such as developing friendships and barriers to employment (Bottroff *et al.*, 2002). In the belief that the views of people with Down's syndrome must not be excluded from the debate on prenatal testing, three recent studies in the UK have obtained views on quality of life, and testing and termination from adults with the condition (Alderson, 2001a; Gow, 2000; Howarth and Rodgers, 2001; Rodgers and Howarth, 2001; Ward, Howarth, and Rodgers, 2002).

Alderson (2001a) interviewed five adults with Down's syndrome - four men and one woman, who ranged in age from 20 to 43 years. Three of the interviewees lived relatively independently and two lived with their parents. The article reported on "relationships, education or employment, leisure interests, hopes, aspects of themselves and society they would like to change, and their views on prenatal screening" (p. 627). All interviewees reported areas of their life with which they were very happy, including their families, friends, leisure activities, and their abilities. They also identified aspects with which they were unhappy. While a few instances of prejudice were described (being pushed in the street, and excluded from mainstream school) most frustrations were related to their restricted employment and social opportunities. Alderson notes how this view reflects the social model of disability, where disablement is largely attributed to barriers in society rather than to impairments caused by the condition.

Various papers from the study by Howarth and colleagues (Howarth and Rodgers, 2001; Rodgers and Howarth, 2001; Ward *et al.*, 2002) report on a workshop on prenatal testing for adults with learning difficulties, including one woman with Down's syndrome. During the workshop participants discussed various aspects of their lives; what they liked (for example, their achievements) and disliked (for example, experiencing prejudice due to their learning difficulty). As in the study by Alderson (2001a), all participants felt that other people's attitudes had the biggest impact on the quality of their lives. These two studies were conducted with a very small sample size and so generalisations of the findings to the wider population of people with Down's syndrome or learning difficulty cannot be safely made. Undoubtedly, there are people with Down's syndrome who could not contribute to a debate on their quality of life or prenatal testing because of cognitive impairment or communication difficulties. However, the studies demonstrate that some people with Down's syndrome can reflect meaningfully upon their lives, and that like all

of us find reward in some areas and not in others. Alderson suggests that assumptions about the relationship between perceived quality of life and degree of intellectual capacity are unwarranted.

The two studies above reported varied opinions towards prenatal testing in people who had Down's syndrome. In the interviews conducted by Alderson and colleagues, one person felt it was up to parents to decide what action to take, another was very uncertain about what he felt about testing and abortion for Down's syndrome, and three expressed some sadness and an unwillingness to talk about these issues. In the study by Howarth *et al.*, one workshop participant (who did not have Down's syndrome) said, "*the foetus should be aborted if a test shows it has a learning difficulty because I don't think it should be born into a cruel world*". Another participant said, "*I think babies with learning difficulties or disabled are good, very, very good. They should be born not aborted*" (Howarth and Rodgers, 2001, p. 36). The participant with Down's syndrome became distressed during the discussion on prenatal testing because she believed that her mother would have aborted her if testing had been available. She said she felt "lucky to be alive" (p. 37). Finally, as part of a doctoral thesis examining views towards prenatal testing in women with a congenital condition, five women with Down's syndrome were interviewed about their quality of life. The women were interviewed in the presence of a facilitator and although willing to talk about their lives the researcher had difficulty engaging the women in issues concerning testing and pregnancy. It was believed that this might be due to a lack of understanding of pregnancy on the part of the interviewees, the unwillingness of the interviewer to raise the issue of abortion, and also because the subject was felt to be too sensitive by the interviewees (and in one case the interviewee's mother) to discuss (Gow, 2000).

Work with people with Down's syndrome is valuable because it informs us of what otherwise we can only guess at, that is, what life is like for people born with the condition from their perspective. However, very little research has been conducted that allows people with learning difficulties to talk about their views on prenatal testing and termination. This might be because of perceived methodological and ethical difficulties, or it might be due to a belief that people with cognitive impairments cannot contribute meaningfully (or reliably) to such a discussion. In a comment on this view Rodgers and Howarth (2001) argued that it was not intellectual capacity that created a barrier to a discussion about genetics in their workshop, but the fact that many participants lacked (i.e. had not been given) basic knowledge about sex and reproduction. Nevertheless, it is an ethical requirement to avoid unnecessary distress in individuals involved in research, and researchers might reasonably fear that participants would be distressed by discussing issues around abortion of babies with the condition that affects them. Some distress (and perhaps anger) is probably inevitable, but it is argued that this is might be an appropriate response and not

necessarily a good enough reason on its own to exclude people with learning difficulties from the prenatal testing debate. Howarth and Rodgers (2001) reported that the woman with Down's syndrome who became upset in their discussion was well supported by her peers. They reported,

"She was clear later that she wanted [her distress] reported as she wanted people to know what it was like, hearing people talk about aborting disabled foetuses, when they were talking about people like her" (Howarth and Rodgers, 2001, p. 37).

It is argued that further work of this type with people with Down's syndrome is very important, although the methodological and ethical challenges it presents are substantial.

An issue of great concern to many potential parents is that having a child with Down's syndrome will have a negative impact on their family (Dimavicius, 1998b). A small number of studies have compared the parenting experiences and emotional adjustment of parents with and without a child with Down's syndrome (see review in Carr, 1995). A recent cross-sectional Nordic study (Hautamäki, 1997) compared 'stress, stressors, and strain' in mothers of children with and without Down's syndrome who ranged in age from 2 years to 17 years. It was reported that in contrast to the comparison group, mothers of children with Down's syndrome expressed less satisfaction with their leisure opportunities, experienced greater numbers of psychosomatic symptoms, and were less satisfied with their life situation generally. These factors were most prevalent in the mothers of adolescents and least noticeable in mothers of young children, women who worked outside the home and those living in rural rather than urban communities. The design employed by Hautamäki is typical of research in this area in that it started from the assumption that families automatically suffer as a consequence of having an affected child, and so only looked at negative outcomes - the so-called 'pathological model' (Cunningham, 1996). For example, the mother's view of her child was not assessed, and reasons why mothers felt their work and leisure opportunities were restricted were not recorded. In the report of an interview study of 17 mothers of children with Down's syndrome Bridle (2000) relates the negative aspects of mother's situation,

"The greater efforts and guilt, the medical appointments, fighting the system, dealing with insensitivity and rejection and feeling powerless to protect your child". However she adds, *"What these concerns did not add up to was the idea that their child was somehow less valuable or loved or that Down syndrome contradicts what is valuable in being a mother"* (Bridle, 2000, p. 10).

Two well-designed British cohort studies in Greater Manchester (Cunningham, 1996), and Surrey (Carr, 1995), have taken a more rounded approach to understanding the impact of a child with Down's syndrome on their family and vice-versa. These studies have each followed a sample of

over 100 families with a child with Down's syndrome, from shortly after their birth⁵ until early adulthood, applying a wide range of validated measures to all family members. Overall, the families in these studies did not have a higher incidence of psychological or physiological distress than matched comparison groups. On the contrary, Cunningham (1996) reported that children with Down's syndrome "*appeared to make a positive contribution*" to their family (p. 89), and Carr (1995) noted that the overall impression was of families' "*resilience and the ability to cope*" (p. 173). Studies taking a more qualitative approach have also reported that parenting a child with Down's syndrome appears to be more similar to than different from parenting other children, and thus includes many positive aspects as well as negative ones (Bridle, 1998; Craig, 2000; Elkins, Stovall, Wilroy, and Dacus, 1986; Felker, 1994; MacDonald-Smith, 1997). However, in support of the Nordic study, Cunningham (1996) also found that mothers of adolescent children with Down's syndrome showed a decrease in measures of life satisfaction, which was associated with a decline in perceived and actual social support for their family. There was also a trend for some mothers to feel that their adolescent child imposed restrictions on family life and their independence. Nevertheless, Cunningham (1996) noted that the strongest predictor of any measure in the teenage years was the score on the same measure five years previously. Thus patterns of adjustment and coping become established fairly early on. Carr (1995) noted that the mothers' work opportunities were substantially hampered by a lack of daycare facilities for children with disabilities.

In a recent American study of family adjustment the psychological functioning of 52 sets of parents whose birth child had Down's syndrome was compared with that of 53 sets who had adopted an infant with the condition (Flaherty and Glidden, 2000). The authors of this study argued that many measures used in comparison studies are almost certain to show that raising a child with a disability is more problematic than a non-disabled child. This is because regular attendance at paediatric clinics, for example, would automatically be scored as stressful and most children with a disability will be relatively frequent users of health services. They suggested that by comparing 'like with like' specific issues associated with being the birth parents of a child with Down's syndrome were more likely to be found. A range of validated measures, including the BDI (Beck Depression Inventory), were employed in the study along with a semi-structured interview. BDI scores were reported at two time points, the time of diagnosis or adoptive placement (retrospectively reported), and at the interview (an average of 5.6 years later). As expected, the results showed that birth parents experienced significantly higher levels of depressive symptoms than did adoptive parents at the time of diagnosis or adoption. However, there were no significant

⁵ The children in the Surrey cohort were born between 1964 and 1965. The children in the Manchester cohort were born between 1973 and 1980.

differences in the (low) levels of depression between the two groups at the time of the interview, although this was not a longitudinal study so the results have to be interpreted with some caution. Neither were there any significant differences in family functioning, child functioning, or marital harmony between the two parental groups at the time of the interview. Flaherty and Glidden argued that as adoptive parents were likely to have high levels of adjustment to their child, the findings indicated that most birth parents had also adjusted well. The only variable demonstrating a significant difference was that birth mothers (but not fathers) scored higher levels of 'personal burden'. The items measuring burden were not reported, but the discussion of the findings indicated they related to care-taking activities. This burden did not appear to have translated into adverse psychological effect.

Many women are concerned that having a child with Down's syndrome will be detrimental to their relationship with their partner and to the happiness of their other children. Although it has been suggested that the birth of a baby with Down's syndrome could hasten the break-up of some relationships (Gath, 1985), there is no greater rate of divorce or separation in couples who choose to raise their baby within their family (Carr, 1995; Cunningham, 1996; Cuskelly and Dadds, 1992; Hautamäki, 1997). In addition, early concerns about the effect of having a sibling with Down's syndrome have generally not been supported by research evidence (Cuskelly, Chant, and Hayes, 1998; Gath and McCarthy, 1996). While some siblings feel that they have suffered, most view their experience positively or as a combination of both loss and gain, and sibling relationships are generally good (Bryant, 1998; Carr, 1995; Cunningham, 1996; Fairbrother, 1988; Richardson, 1999). Cunningham (1996) reports that around 20% of siblings displayed signs of poorer adaptation, but that this appeared to be associated with general family functioning and personal psychological difficulties rather than with any characteristics of their affected sibling. Some families appear to be at particular risk of experiencing difficulties generally, and in the Manchester cohort up to one-third of families had some difficulties in coping with or adjusting to their child with Down's syndrome. Increased stress in parents was associated with behavioural problems in the index child, which in turn was associated with poorer physical health of the child (such as repeated infections) and a lower mental ability/higher physical dependency. This finding should however, be set in the context of the number of families who have difficulties coping with 'normal' children, and the knowledge that coping is related to the personal characteristics of parents as well as to the characteristics of the child.

Overall, the research suggests that having a child with Down's syndrome need not have, and generally does not have, significantly adverse long-term effects on parents or siblings, although a minority of families experience substantial difficulties in coping. Distress is almost unanimously

experienced at the time of diagnosis, but in most cases parents appear to adjust quickly and positively to having a child with Down's syndrome (Carr, 1995; Ryde-Brandt, 1988). However, it is important to note that in all the studies reviewed, only birth parents that had elected to keep their children were included. A number of studies have reported that parents of children with Down's syndrome in their samples are more likely to be married and be of higher socio-economic and educational status than average (Boon, 1986; Carr, 1995; Cunningham, 1996; Hautamäki, 1997). This is probably related to the fact that older women are more likely to have children with Down's syndrome. The importance of financial and social resources to the successful parenting of a child with Down's syndrome and other disabilities should not be underestimated (Cunningham, 1996; Knussen and Sloper, 1992). Some parents who relinquish their child for adoption might not have access to such resources. Furthermore, these studies undoubtedly represent a mixture of parents; those who might have terminated the affected pregnancy had testing been available to them, those who would have refused testing, those who had actually refused testing, those who had accepted screening but received a false-negative result, and those that had had testing and continued the pregnancy. These factors have not generally been considered in studies concerning parental adjustment to a child with Down's syndrome, however, families who opted out of prenatal testing or continued a pregnancy might find it easier to adjust to their child, than those who would have terminated an affected pregnancy had they had the choice (Hall, Bobrow, and Marteau, 2000).

Cunningham (1996) outlined key areas of social and emotional support for families who have a child with Down's syndrome, and concluded that the full potential of people with the condition would not be realised until families have such support. A supportive social context is therefore vital to the continued development and well being of people with Down's syndrome and their families. The following two sections provide a brief historical overview of this context and a review of the published research on public attitudes towards people with Down's syndrome.

1.1.3 Attitudes towards Down's syndrome: the historical and socio-cultural context

Down's syndrome has been described as "*the most common, the most easily recognised, and probably the most researched single condition causing learning disability*" (Carr, 1995, p. 1). However, it was only identified as a specific condition 140 years ago when John Langdon Down observed common features in some of the residents of the asylum where he was medical superintendent (Ward, 1998). Langdon Down's 'Ethnic Classification of Idiots' identified a group of patients 'Mongolian in appearance' and described common traits such as a 'lively sense of the ridiculous' along with their often 'feeble circulation' (Langdon Down, 1866). Prior to this, the literature suggests that there had been little awareness of people with Down's syndrome as a distinct group. This may have been due to the lack of scientific interest in learning difficulty, the

high proportion of affected individuals who died in childhood from complications of the syndrome, or to mortality rates in childbirth leading to there being fewer older mothers (Richards, 1968). It is also likely that the advent of institutionalised care for those with learning difficulties enabled sufficient numbers of affected individuals to be observed together. These, and other factors might explain why Down's syndrome is still not recognised as a distinct condition in all world cultures. In Southern Asia and sub-Saharan Africa, for example, no term for Down's syndrome exists (Chilaka *et al.*, 2001; Christianson, 1996).

The literature demonstrates that historically, children born with learning difficulties were often treated inhumanely by society generally, if not necessarily within individual families (see Cohen, (1995) Chapter 4, for review). Until the mid Victorian era, when a number of doctors including Langdon Down pioneered the specialised care of patients with learning difficulties, such people who were not cared for at home or boarded out to foster parents were placed in 'lunatic asylums' as distinctions were rarely made between intellectual impairment and psychiatric conditions. In recent history pre-Nazi Germany led the world in the progressive treatment of those with learning difficulties, advocating care in the home, providing financial support to parents, and setting up special school programmes (Rogow, 2001). However, when the Nazis came to power they closed down special schools and withdrew support for families on the basis that people with disabilities were an unacceptable burden to society. People with Down's syndrome, along with others with learning difficulties were officially classified as 'useless eaters' and were forcibly institutionalised, experimented upon, and subject to criminal euthanasia (Erdemir, 2001; Lifton, 1986; Rogow, 2002; Wolfensberger, 1981). Euthanasia of infants with Down's syndrome was also widely practiced by nurses and physicians on German neonatal wards during this time (Aly, 1994; Burleigh, 1994).

While the Nazi era was clearly an extreme example of how attitudes towards impairment can have appalling consequences, euthanasia of people with learning difficulties was openly advocated by some physicians in other Western countries until quite recently (Elks, 1993). The following extract is taken from a mainstream medical text of the 1950s, and refers to the '100,000 idiots and imbeciles in the [USA]':

"They cannot be employed to any advantage; moreover, many of these low-grade defectives are utterly helpless, deformed, repulsive, unlovable, and unloving. If they are capable of forming any relationship it is only on the basis of the simple egocentric dependence of a baby. ... Many clinicians believe that it would be an economical and humane procedure were their existence to be painlessly terminated, and that this would be welcomed by a very large proportion of parents" (Tredgold and Soddy, 1956).

Tredgold and Soddy continued to maintain this position up to the 11th edition of their text in 1970. While their insight into the views of contemporary clinicians might have been accurate, their assumption on behalf of parents was probably misguided. While instances of parental support for euthanasia of children with learning difficulties have been reported (Shepperdson, 1983), this was, and is, a minority view. Documentary evidence shows that many parents of murdered German children attempted to take legal action against the institutions where their children were taken (Rogow, 2002). Medical professionals have frequently made decisions on behalf of parents about the right-to-life of their children, some more openly than others (Hontela and Reddon, 1996).

It has been argued that Down's syndrome is as much a cultural creation as a biomedical condition (Lippman and Brunger, 1991). For example, labelling of the condition has changed over time to reflect the dominant scientific culture of the era. Langdon Down's term 'Mongolian Idiot' and the derivatives 'Mongoloid' and 'Mongolism' reflect 19th century interests in Darwinian theory and anthropological concerns to order races on hierarchical scales. Langdon Down's classification also included idiots of the 'Ethiopian', 'Malay', and 'Aztec' types, and he identified many patients in his care as belonging to one of these ethnic groupings. He postulated that 'ethnic idiots' were atavisms (throwbacks) to races whose intellectual evolution had arrested at an earlier stage than that of the Caucasian race. Thus, people with Down's syndrome were thought to be the intellectual equivalents of adults in Mongolia. Although Langdon Down lost faith in the ethnological approach to understanding learning difficulty (Ward, 1998), others took it up enthusiastically and developed it in a more obviously eugenic direction (Crookshank, 1931). Despite these associations, the terminology of Langdon Down was the accepted form in scientific circles until in 1965 a growing sensitivity to racial issues, complaints from the Mongolian People's Republic, and a deposition of geneticists led the World Health Organisation to rule that Down's syndrome should officially replace the term Mongolism. Down's syndrome (or Down syndrome) is now the term most usually encountered, although trisomy 21 is the preferred term in many medical journals reflecting the 'geneticization' of disease in the current scientific era (Lippman, 1991). The terms Mongol and Mongolism are still used by some medical professionals as well as being in relatively common usage in the general population (Rutter and Seyman, 1999).

A critical examination of some facts about Down's syndrome demonstrates that even medical aspects of the syndrome cannot be considered completely independently from their socio-cultural context. The presence of learning difficulty in people with Down's syndrome is indisputable although the upper level of their intellectual range has never been definitively agreed upon (Wishart, 1998). It has been suggested that an artificial ceiling of achievement has been ascribed to

people with Down's syndrome, and that many underachieve because of lowered expectations of these individuals in society (Alderson, 2001a; Borthwick, 1996). Again, while Down's syndrome is associated with certain physiological aspects that will reduce overall life expectancy, attitudes toward the care of people with the condition play an important role in morbidity and mortality rates. A study in Israel identified that many deaths of children with Down's syndrome from infections and other environmental causes were 'potentially preventable' (Sadetzki *et al.*, 1999). These researchers also recorded that children with Down's syndrome living in institutions had a significantly higher mortality rate than did children raised in a family, and that this was unexplained by differences in physical health at birth. In many world cultures learning difficulty is still associated with major stigma. The findings of another Israeli study demonstrate that Down's syndrome is generally viewed as an unattractive, embarrassing and even frightening condition, evoking feelings of pity, sadness and rejection (Shiloh and Berkenstadt, 1992). In Palestine, children with Down's syndrome have commonly been hidden, subjected to abuse, and even infanticide, especially when female (Fishman, 1994). Children with disabilities often suffer similar fates in China where cultural and political imperatives make parenting a child with a disabling condition especially difficult. It can be concluded that the increasing life expectancy of people with Down's syndrome is not entirely due to medical and surgical advances, but also to changes in the way individuals with disability are viewed in our society.

1.1.4 Attitudes towards people with Down's syndrome: research review

Studies that have measured attitudes towards people with Down's syndrome are rare perhaps because of the tendency to group all people with learning difficulties together (see Altman (1981) for a review of attitude research). Twenty years ago, MENCAP carried out a survey of public attitudes towards the 'mentally handicapped' but did not discriminate between different conditions (Mencap, 1982). However, their report notes the common perception that people with learning difficulty are often "*affectionate and happy (because of their child-like behaviour)*", and that "*Mongolism and Down's syndrome*" were cited as causes of learning difficulty (Mencap, 1982, pgs. iii and 3 respectively). This suggests that some respondents probably had people with Down's syndrome in mind when completing the survey. Homogenising disability in this way is common, but as Down's syndrome has such a central place in people's perceptions of learning difficulty it is surprising that so little work has focused on understandings of this condition specifically. Table 1.1 summarises the published attitude research that has considered Down's syndrome specifically.

Table 1.1. Studies measuring attitudes towards Down's syndrome (DS)

Study/country	Population	Measures used	Comments
Furnham and Pendred (1983), UK	Students and general public (N=96)	Survey. Attitudes Toward Disabled Persons (Yuker, Block, and Campbell, 1960). Degree of contact with disabled people (open-ended item).	Attitudes towards four conditions measured. N=24 randomised to DS condition.
Hall and Minnes (1999), USA	Undergraduates (N=92)	Survey. ATDP Form-0 (Yuker, Block, and Young, 1970). Feelings of Comfort Scale (Marcotte and Minnes, 1989). Volunteering Intentions Scale, Contact Questionnaire, Television Viewing Scale, Jackson Social Desirability Scale (Jackson, 1974).	Attitudes towards DS only. Measured the effect of television on attitudes.
Sinson (1985), UK	Mothers of pre-school children (N=100)	Structured interview and single-item attitude measure.	Attitudes towards DS only. Compared rural and urban groups.

The first two studies reviewed, (Furnham and Pendred, 1983; Hall and Minnes, 1999) used a version of the Attitudes Toward Disabled Persons scale to measure attitudes towards people with Down's syndrome. The ATDP (Yuker *et al.*, 1970) has been one of the most widely used measures of attitudes towards people with a disability. Other scales exist for measuring attitudes towards people with learning difficulty but they have usually been shown to be psychometrically weak (Antonak and Livneh, 1988). The ATDP scale conceptualizes attitude in terms of the degree to which people with disabilities are perceived as similar to non-disabled people. The type of disability is non-specific and so items have usually been modified to measure attitudes towards a specific condition. The ATDP requires participants to read 20 statements about the target group and then to score their response on a Likert-type 'agree-disagree' scale, for example, '*Down's syndrome people are as happy as other people.*' Furnham and Pendred (1983) found attitudes towards people with Down's syndrome to be significantly more unfavourable than those towards a non-observable 'mentally handicapping' condition and the physical disabilities of blindness and

deafness. In particular, the item *'Down's syndrome people are the same as anyone else'* received lowest agreement across the four conditions, and the items *'It is almost impossible for Down's syndrome people to lead a normal life'*, and *'Down's syndrome people cannot have a normal social life'* received the highest agreement. A resulting factor analysis of the responses showed that these items clustered on a dimension labeled 'normality', suggesting that for some people Down's syndrome is considered the antithesis of normality. However, this study represented the views of only 24 people, making generalisations unsafe.

In addition to measures of cognitive beliefs, 'comfort scales' have been used to assess the affective component of attitudes towards people with disability (Stoneman, 1997). These measures assess the degree to which respondents would feel at ease in a number of situations where a person with a disability was present. In the study by Hall and Minnes (1999) the responses to items such as, *'How comfortable would you feel sitting next to a young adult with Down syndrome on a bus?'* were recorded on a 5-point Likert-type scale of 'extremely uncomfortable' to 'extremely comfortable'. Hall and Minnes reported their sample of student participants to hold moderately favourable attitudes towards people with Down's syndrome. Generalisation from the results of this study is limited by the student sample, however, application of multiple regression revealed that favourable attitudes towards Down's syndrome were predicted by prior media exposure, favourably viewed contact with people with Down's syndrome, and social desirability scores. Comfort and intentions to volunteer were predicted by exposure to a documentary film about Down's syndrome and favourably viewed contact with affected persons. Of note was that the perceived favourability of the contact was more important than the frequency of contact in predicting attitudes. The value of measuring frequency of contact in studies of attitudes toward disability has been questioned previously, with authors suggesting that it is the perception of specific experiences that is important (Eayrs and Ellis, 1990; Finkelstein, 1980; Haddock, Zanna, and Esses, 1994). Hall and Minnes also suggested that appropriate media exposure was important in the development of more positive attitudes towards Down's syndrome.

Despite having good psychometric properties the ATDP and other similar scales, such as the Scale of Attitudes Toward Disabled Persons (Antonak, 1982), are criticised for determining that a particular response to an item indicates the direction of the participant's attitude. For example, a person who holds a favourable attitude toward people with Down's syndrome might believe that an affected person cannot have a normal social life because of barriers within society, although agreement with this item on the ATDP would be interpreted as a negative attitude. Thus, choosing which beliefs to include in a scale, and *a priori* determining the interpretation of the response

might reflect the subjectivity of the scale developer rather than that of the participant. Scales such as the ATDP have also been criticised for, “*regulating the range of possibilities that the person completing the questionnaire can perceive for disabled people*” (Finkelstein, 1980, p. 21). The comfort scales also display a limited conceptualisation of the emotions that are elicited by contact with people with disabilities as they measure only the negative feelings of fear and embarrassment, again perhaps revealing the subjectivity of the developers of these scales. Finally, the behavioural measures tend to situate people with a disability as recipients of care or charity, and do not take account of actual barriers to behaviour that may not reflect attitudes.

The study by Sinson (1985) used semi-structured interviews to compare attitudes towards Down’s syndrome in two groups of mothers of preschool children. Both groups lived near a hospital for people with learning difficulties but one group lived in an urban area, while the other lived in a rural community. Mothers were asked about their experiences with people with learning difficulties and their views on educational integration, prenatal testing, and adoption for babies with Down’s syndrome. They were also asked to rate themselves on a scale to denote their feelings “*about mentally handicapped people, and in particular Down’s syndrome*”. This scale was anchored as (1) they would like to have a child with Down’s syndrome living with them, to (5) they felt that all children with Down’s syndrome should be “*terminated either before or at birth*” (Sinson 1985, p. 45). The analysis showed that while social class and religion did not discriminate between attitudes, urban mothers ($n=50$) were significantly more likely to hold favourable attitudes towards Down’s syndrome than were rural mothers ($n=50$). Overall, less than half of all the mothers were in favour of educating children with Down’s syndrome in mainstream schools, although again, urban mothers held significantly more favourable views. Sinson concluded that the key to the difference in attitudes was contact with individuals with learning difficulties. In the town, hospital residents were relatively well integrated into their community and 88% of the mothers reported regular contact with them. In comparison, only 22% of the rural mothers reported that they had regular contact with the residents. Of particular interest to this thesis is that there was no difference between the two groups on attitude to termination for Down’s syndrome; 68% in both groups indicated they would terminate an affected pregnancy. Although the author did not comment on this, the finding again suggests that attitudes towards existing people with Down’s syndrome cannot be taken as a proxy for attitudes towards giving birth to a child with the condition. Although it is now 18 years old, this study perhaps gives a better insight into how women might view Down’s syndrome in relation to their own pregnancy. Unfortunately, the measure used confounded attitudes towards learning difficulty generally with attitudes towards Down’s syndrome specifically. In addition, the validity of using belief categories to represent the

attitude continuum is questionable, as respondents might have agreed with more than one belief or none at all. It is not possible from the report to know how the attitudes towards people with Down's syndrome related to attitudes towards prenatal testing and termination as no statistical analysis was performed and the data are presented in summary form.

1.1.5 Summary and conclusions

The studies reviewed suggest that attitudes towards Down's syndrome are multi-layered. There may be a relationship between attitudes towards existing people with the condition, those yet to be born, and towards having an affected child oneself, but each attitude is a construct in its own right. It is also likely that cognitive, emotional, and experiential factors play different roles in the informing and expression of these different attitudes. The perception of contact with people with Down's syndrome as favourable or unfavourable appears to be important to the expression of attitudes towards existing people with the condition, and it is likely that the media also has a role to play in this. The findings of Furnham and Pendred (1983) suggests that the degree to which people with Down's syndrome are considered 'normal' is also a critical discriminating factor between favourable and unfavourable attitudes, and this might interact with quality of contact.

It has been proposed that, "*the majority of children [with Down's syndrome] in this and future generations can be expected to live longer, healthier and happier lives than many of their predecessors*" (Wishart, 1995, p. 57). This is due both to medical technologies and the continued integration and acceptance of affected individuals into Western societies. Most significantly of all, changes in social policy mean that most children with Down's syndrome are raised within a family giving them the opportunity to develop the close relationships essential to the emotional well being of all humans. Paradoxically however, as the lives of people with Down's syndrome are improving, so are the techniques that enable them to be excluded from society permanently and before birth. It is within the prenatal testing context that attitudes towards people with Down's syndrome and towards having a child of one's own with Down's syndrome become salient for many women, and here that a fundamental tension between the two attitudes can exist (Pessione, 2001; Press *et al.*, 1998). The next section describes how prenatal testing for Down's syndrome has developed, and considers the role of attitudes towards the condition on testing choices.

1.2 PRENATAL TESTING AND TERMINATION FOR DOWN'S SYNDROME

Prenatal tests for Down's syndrome are now a routine part of antenatal care in many countries in the Western world. This section looks at how testing for Down's syndrome came into being, how and why it has become widely available, and the current testing situation in the UK.

1.2.1 A history of prenatal testing for Down's syndrome

As Rothman notes in the 'Tentative Pregnancy' (Rothman, 1986) there are many histories of prenatal testing, each providing a different perspective on its development, for example the medical, social, ethical, or feminist perspective. This section outlines the major technological milestones in the development of prenatal testing for Down's syndrome and considers this development from a psychosocial perspective.

Three main technological developments were essential to the evolution of prenatal testing for Down's syndrome. The first was the development of amniocentesis in the early 1950s, a technique that enabled the safe aspiration of amniotic fluid via the abdomen of a pregnant woman initially used to identify rhesus conditions in the fetus or to relieve hydramnios⁶ (Gadow, 1998). The second development was the identification of the trisomic chromosome 21 in people with Down's syndrome (Lejeune, Gauthier, and Torpin, 1959). The third was an advance in tissue and cell culturing techniques during the 1960s that enabled karyotyping⁷ of fetal cells found in amniotic fluid (Coventry and Pickstone, 1999). In 1967 two obstetricians in the USA carried out the first amniocentesis tests specifically to identify chromosomal abnormalities (Jacobson and Barter, 1967), and in 1968 the first termination following mid-trimester prenatal diagnosis of Down's syndrome was reported (Valenti, Schutta, and Kehaty, 1968). While technological advances were necessary for the development of prenatal testing procedures, social changes were necessary for the diffusion of testing into routine prenatal care (Schwartz-Cowan, 1993). Amendments to abortion laws in the USA and the UK allowed legal termination in cases where the fetus was considered to be at high risk of a serious disabling condition. In addition, in America a number of older women who had not been offered amniocentesis and who subsequently gave birth to a child with Down's syndrome successfully sued their obstetricians for compensation. By the late 1970s, amniocentesis was integrated into the routine antenatal care of women who were 35 years and older, first in the USA and Canada but shortly followed by Britain.

Amniocentesis is usually performed at around 16 weeks gestation, after which a period of two weeks is typically required for the karyotyping process to be conducted. A recent technological development called FISH (fluorescent *in situ* hybridisation) can produce a karyotype for trisomy 21 within 48 hours, although this technique is not yet widely available and it cannot be used for

⁶ Excess of amniotic fluid surrounding the fetus.

⁷ Karyotyping is a technique for systematically organising the chromosomes of a single cell as viewed through the microscope lens and presenting this in photographic form. The karyotype shows the number and arrangement of chromosomes within the cell nucleus.

identifying all chromosomal abnormalities (Cheong *et al.*, 2001; Toth *et al.*, 2001). Diagnosis of Down's syndrome via amniocentesis is accurate but the procedure is invasive, carrying a small risk of spontaneous abortion. In the literature aimed at pregnant women, the risk of miscarriage following amniocentesis is often cited as being between 0.5 % and 1% (Bounty, 2000; Health Education Authority, 1999). However, there is still a lack of consensus about the actual procedure-related loss rates (National Screening Committee, 2002), and recent studies report rates of between 0.03% and 3% dependent on a number of situational factors, for example, previous history of miscarriage and gestation. Risk of procedure related miscarriage appears to increase significantly after 18 weeks pregnancy (Antsaklis *et al.*, 2000; Roper *et al.*, 1999; Scott *et al.*, 2002). For social, psychological, medical, and procedural reasons physicians, pregnant women, and the population at large usually consider termination in the first trimester of pregnancy preferable to later termination (Kornman *et al.*, 1997; Norup, 1997). From the mid-1980s another prenatal diagnostic technique called chorionic villus sampling⁸ (CVS) became available that could be conducted at around ten weeks gestation. Schwartz-Cowan (1993) notes that cultural factors were especially crucial to the development of CVS as some religious groups consider second trimester abortion unacceptable (Modell, 1986). CVS collects placental material that is used to provide a karyotype within 24 - 48 hours of the procedure although in some cases the findings still need to be confirmed with amniocentesis. For various reasons including a higher rate of false-positive and false-negative results, a greater degree of technical difficulty, and higher procedure related miscarriage rate CVS remains less widely used than amniocentesis (Alfirevic, Gosden, and Neilson, 2003).

The risk of miscarriage along with the financial costs involved make amniocentesis and CVS unsuitable tests to offer to all pregnant women. However, the known association of advanced maternal age with birth prevalence of Down's syndrome has always provided a means to identify an 'at risk' group amongst an asymptomatic sub-population--in other words a screening test. The use of maternal age as a screening criterion for amniocentesis and CVS provided women who were *a priori* at higher risk of having a baby with Down's syndrome the opportunity to test for and terminate an affected pregnancy. However, the higher birth rate in younger women meant that around two-thirds of children with Down's syndrome were actually born to mothers under 35, with one-fifth born to women under 25. For this reason, researchers strove to develop a screening tool that could provide a pregnancy-related risk rather than a purely age-related one. Since the early

⁸ In CVS a biopsy of fetal membranes is conducted on placental material obtained by inserting a hollow tube into the uterus, by either transcervical or transabdominal means (Lilford, 1991).

1970s screening in pregnancy for neural tube defects (NTDs⁹) had been possible by means of measuring the level of the chemical alpha-fetoprotein (AFP) in a sample of maternal blood serum. A raised level of maternal AFP signified that the fetus was at higher risk of having an open NTD and amniocentesis was often recommended. In 1983 the relationship between abnormally low levels of AFP and chromosomal abnormalities was also noted (Merkatz, Nitowsky, Macri, and Johnson, 1984), and the first biomedical screening test for Down's syndrome based on AFP levels and maternal age was developed soon afterwards (Cuckle, Wald, and Lindenbaum, 1984). In the late 1980s two more chemical markers were identified¹⁰ which in combination with AFP levels increased the test's sensitivity for Down's syndrome. This combination of markers is often referred to as the 'triple test' and demonstration studies have shown that when performed between 15 to 22 weeks gestation, the triple test can identify around 60% of pregnancies affected by Down's syndrome for a 5% false positive rate¹¹. The probability that Down's syndrome affects a particular pregnancy is calculated¹² and if this figure is greater than a certain cut off figure (usually around 1 in 250), the woman is offered amniocentesis. Serum tests are now the most common methods of screening for Down's syndrome in the UK along with maternal age, and these tests also screen for NTDs and trisomy 18¹³ (National Screening Committee, 2002). In addition, a number of other serum markers for Down's syndrome have been identified, including ones measurable in the first trimester¹⁴ although debate continues as to which testing scenario is optimal (Cuckle, 1998; Wald, Watt, and Hackshaw, 1999; Wellesley, Boyle, Barber, and Howe, 2002).

Ultrasound scanning technology now also plays a major role in the identification of many chromosomal abnormalities as it is used to date pregnancies (necessary to calculate an accurate serum screening risk) and as a screening tool in its own right. A number of markers for Down's syndrome can be identified during a scan, such as the nuchal translucency measurement in the first

⁹ NTDs range in severity from anencephaly (a condition where there is no brain or spinal cord and the fetus is incompatible with life), through to varying degrees of spina bifida (a gap in the spinal column). In severe cases of spina bifida the spinal cord protrudes through the gap leading to degrees of paralysis and, in some cases, to learning difficulty.

¹⁰ Raised serum human chorionic gonadotrophin and lowered unconjugated oestrial.

¹¹ The false-positive rate is defined as the number of women identified as being at high risk who are subsequently found to have a pregnancy unaffected by Down's syndrome.

¹² In addition, probabilities for NTDs and trisomy 18 are also calculated.

¹³ Also known as Edwards syndrome. A condition caused by an extra chromosome 18 and characterized by severe intellectual impairment and a range of abnormalities of the skeleton and major organs. 90% of affected babies die before their first birthday (Barnes and Carey, 1998).

¹⁴ A 'quadruple test' was developed in the mid 1990s, although this is not yet widely available via the NHS. Developers claim that the test can identify over 70% of pregnancies affected by Down's syndrome for a 5% false positive rate (Wald, Kennard, Hackshaw, and McGuire, 1997).

trimester, and skeletal anomalies in the second¹⁵. Since this thesis was begun, it has been discovered that the nasal bone is missing in many fetuses with Down's syndrome at 11-14 weeks gestation, and that this can be seen via high definition ultrasound. This screening test for Down's syndrome has a claimed sensitivity of 85%, and a false-positive rate of only 1%¹⁶ and could have a major impact on the future of screening (Cicero *et al.*, 2001). In the UK today, the method of screening for Down's syndrome available to a pregnant women depends on her age and where she attends for antenatal care. Various combinations of age, serum screening tests and ultrasound technologies are used in different hospitals within each NHS trust (Department of Health, 2000; Gilbert *et al.*, 2001). In order to standardise this provision the Department of Health recently agreed that second-trimester serum screening should be offered nationally to all pregnant women by 2004¹⁷. However, because of the number of screening technologies available, there will undoubtedly still remain inequalities in the screening service provided to women dependent on their geographical location and whether or not they are willing to pay privately for certain tests (Gilbert *et al.*, 2001). Pre-implantation diagnosis of chromosomal abnormality is available for some couples using assisted conception methods, but for most potential parents, 'prevention' in the true sense is not an option.

While technological advances were necessary for the development of prenatal tests for Down's syndrome another essential factor in their history is often overlooked: certain life values and beliefs about disability and the disabled (Bridle, 2000; Felker, 1994; Lippman, 1994). At the time when prenatal testing for Down's syndrome was first considered, the common medical view (for example, as expressed by Tredgold and Soddy (1956)) was that the quality of life for people with Down's syndrome and their families was severely limited. To have a child with 'Mongolism' carried a severe social stigma, and many parents up until the early 1970s were encouraged to institutionalise their affected child and to 'try again'. In this context it is unsurprising that termination seemed an attractive option for many medical professionals as well as their patients. The 1967 Abortion Act in English law had made termination of pregnancy legal if 'there is substantial risk that if the child were born it would suffer from such physical or mental abnormalities as to be seriously handicapped'. Down's syndrome was considered to meet this

¹⁵ The thickness of the translucent area between the fetal skin and the tissue overlying the cervical vertebrae. There are at least 15 ultrasound markers for Down's syndrome (Ogle and Chitty, 1998).

¹⁶ In an ultrasound examination of the fetal profile of 701 fetuses at 11-14 weeks' gestation, the nasal bone was absent in 73% of fetuses subsequently found to have Down's syndrome, and in 0.5% of chromosomally normal fetuses.

¹⁷ Yvette Cooper, Minister for Public Health, 30 April 2001. As part of initiatives to modernise neonatal and antenatal screening in the NHS.

criterion, although not everyone believed that termination was appropriate. Perhaps ironically, Jerome Lejeune the discoverer of the chromosomal origin of Down's syndrome was strongly opposed to abortion. He called those involved in promoting prenatal testing,

“The National Institute of Death... a new facility for research and applied eugenics”, and asked, *“should we capitulate in the face of our own ignorance and propose to eliminate those we cannot help?”* (Lejeune, 1970, cited in Epstein, 2002, p. 309).

The debate as to whether prenatal testing for Down's syndrome equates to eugenics has continued, but this is an accusation that is strongly denied by medical researchers involved in this field (see (Cuckle, 2001a; Cuckle, 2001b; Lippman, 2001b; Lippman, 2001a; Parker, Forbes, and Findlay, 2002). However, financial analyses of screening programmes where ability to terminate is unproblematically classed as a benefit suggests that the financial costs of supporting certain members of society is considered undesirable by some (Cuckle, 2000; Fletcher, Hicks, Kay, and Boyd, 1995; Gilbert *et al.*, 2001; Wald *et al.*, 1992). This is clearly a personal view of disability whether or not it is eugenic in intention. However, the main reason why prenatal testing has developed is the view that disabled lives involve a substantial degree of suffering, either for the affected person, their family, or both (Shakespeare, 1998). An American paper in the early 1970s calling for wider availability of amniocentesis, acknowledged the financial elements of prenatal testing but argued,

“Is a detailed estimate of money costs required? The lifelong care of severely retarded persons is so burdensome in almost every human dimension that no preventative programme is likely to outweigh the burden” (Stein, Susser, and Guterman, 1973, p. 308).

It can be argued that a similar viewpoint continues to drive the development of prenatal testing for Down's syndrome along with financial considerations (including profit based ones) and the more recent emphasis on offering women an informed choice.

As long as women can choose not to use testing and termination for abnormality prenatal testing programmes are not eugenic in the same way as the extreme examples cited earlier. Ultimately, however, the result of widespread prenatal screening is likely to be a reduced population of people with Down's syndrome. It is interesting to consider that although a child born with Down's syndrome in the UK has the real opportunity to live a happy and healthy life in a society where acceptance of those with disabilities in society is generally improving, the availability and usage of prenatal testing for the condition is increasing. Despite the fact that over 900 conditions can now be diagnosed prenatally (Weaver, 1999) testing for Down's syndrome in pregnancy retains a central focus in terms of research effort and local and national policy decisions. The diffusion of prenatal testing has been so successful that testing for Down's syndrome is now a routine part of

the pregnancy experience for most women in the UK, USA and Western Europe. Inevitably, for some women their testing experience is a less than happy one. The next section summarises some of the major psychological aspects of prenatal testing.

1.2.2 Psychological correlates and consequences of prenatal testing

Anxiety

In pregnancy it is very common to experience a degree of anxiety that there is ‘something wrong’ with the baby (Green, Statham, and Snowdon, 1992; Statham, Green, and Kafetsios, 1997), and pregnant women are generally very alert to any indication that there might be something amiss. Women naturally seek assurance that their baby is healthy in common-sense ways by comparing their pregnancy with those of other women, stopping to check that the baby is still moving, and so on. This sensitivity might stem from a biological drive to have a healthy child in pay-off for the physical resources that mothers invest in their offspring (Buss, 1999). Some pregnant women are specifically anxious about having a baby with Down’s syndrome, particularly if they are considered to be ‘older mothers’, i.e. over the age of 35 (Berryman, Thorpe, and Windridge, 1995). Whether this concern is caused by (or at least supported by) a growing awareness of the availability of testing has been debated. For example, one commentator noted,

"In many ways these developments [in testing] are beneficial but anxiety may be heightened..... disability, or the fear of disability, can creep into each day throughout the pregnancy" (Gath, 1993), p 168).

This fear--natural or exacerbated--might make women more susceptible to the offer of procedures that seem to offer them the complete assurance that would usually have to wait until the baby was born, and the main reason that pregnant women give for having serum screening is for ‘reassurance’ (Browner and Press, 1995; Gokhale and Cietak, 2002; Kornman *et al.*, 1997; Roelofsen, Kamerbeek, and Tymstra, 1993; Santalahti *et al.*, 1998a). Most women do receive the reassuring test result they want, although the degree to which screening has a truly beneficial effect on underlying levels of anxiety is unknown.

In those women who undergo serum screening around five-percent will receive a positive result that identifies their pregnancy as being at higher risk for Down’s syndrome¹⁸. At this point (for the majority of women) any underlying anxiety about abnormality becomes acute. Studies using validated measures of anxiety such as the STAI (The State Trait Anxiety Inventory (Spielberger,

¹⁸ Some will also receive a positive screen result for NTD or trisomy 18, however, this thesis focuses on screening for Down’s syndrome only.

1983)¹⁹ report mean state anxiety scores of between 49 and 57 in pregnant women who have received a positive serum screening result. As a state score of 34 to 36 is considered 'normal', and a score of 48 an acute anxiety reaction to a stressful situation, this suggests that the receipt of a positive result is generally experienced as an extremely stressful event. Where narrative data have been collected, women consistently express feelings of shock, panic, distress, and fear for the baby and themselves. Studies that have considered the impact of a screen positive result on quality of life have reported disturbances in appetite and sleep, and negative thoughts about the pregnancy. These effects, feelings and thoughts can last for a month or more until (and if) a normal amniocentesis result is received (Jørgensen, 1995a; Roelofsen *et al.*, 1993; Santalahti, Hemminki, Latikka, and Ryyänänen, 1998b; Santalahti, Latikka, Ryyänänen, and Hemminki, 1996).

Anxiety in this context can be seen to be a natural response to the suggestion that one's unborn baby is at increased risk of a disabling condition and as a natural response might also serve a particular function. For example, it has been suggested that people act more appropriately in the face of a health threat if they have a moderate level of anxiety, as without anxiety, there is no motivation to engage and deal with the threat (Leventhal, Safer, and Panagis, 1983). However, there have been concerns that high anxiety might affect the ability to make informed choices regarding prenatal diagnosis, and these have not yet been adequately addressed (Green *et al.*, 2002). It is argued however, that while anxiety might be an appropriate response to a positive screening result for Down's syndrome it should not be considered unimportant because it is 'temporary'; one month is a substantial period of time in a nine-month-short pregnancy. Furthermore, the evidence suggests that a normal amniocentesis result does not always allay anxiety to the degree assumed and some women continue to experience residual anxiety about the health of their baby (Green *et al.*, 2002). The anxiety levels of women who decline the offer of amniocentesis following a positive screening result have also been little researched, reflecting not only the relatively small number of women who decline further testing, but also perhaps the interests of researchers.

Although testing related anxiety has been one of the most investigated aspects of prenatal testing, the cause of this anxiety, i.e. fear of the baby having an abnormality, is rarely explored. Alderson suggests there are two main fears (Alderson, 2001c). The first fear is, "*that having a child with learning difficulties will mean endless hard work, sadness and no fun. It will be a burden for the whole family*" (p. 64). This is a medically (and socially) acceptable concern as avoidance of

¹⁹ State anxiety is considered to be transitory and related to the event, whereas trait anxiety is considered to be of dispositional origin. Possible scores on the STAI range from 20 to 80.

parental burden and suffering has been one of the main justifications for the development of prenatal testing. The second fear, Alderson suggests, is perhaps less ‘acceptable’ to discuss;

“The other (and I’m guessing here) is so awful that hardly anyone talks about it, but (I think) it is the fear of carrying and giving birth to a monster. A baby too unlike you to feel like your own child. An ‘alien’ that you will never be able to talk and laugh with. Someone who looks so different that other people will point and stare; not like the pretty babies in the Mothercare books” (ibid.).

There is so little research on how pregnant women understand the conditions they are being tested for that evidence is lacking for this assumption. However, in the study of community attitudes towards Down’s syndrome by Sinson (1985, discussed in Section 1.1.4) one woman who was an experienced foster carer, spoke of how she had been quite willing to foster a baby with the condition but when pregnant at 37 was very concerned about having an affected child herself.

“I desperately didn’t want to have a Mongol baby myself – and all those reasons I gave out [i.e. effect on family].. were really excuses. Because really your child is a reflection of yourself isn’t it - and I didn’t want myself reflected as a Mongol” (Sinson 1985, p. 17).

In an ethnographic study of parents attending a genetic counselling clinic in the North of England, the researcher documented a great deal of fear surrounding the word ‘syndrome’ because of its immediate association with Down’s syndrome (Chapple, Campion, and May, 1997). This study demonstrated how common clinical terms like ‘syndrome’ and even ‘genetics’, which may be relatively neutral to health professionals, hold different connotations for parents.

“When they start talking about genetics we start thinking about little monsters”, and another said, “I mean syndrome! You think ‘God what have we got?’” (Chapple et al., 1997, p. 84).

Parents might feel very uncomfortable expressing their fears about Down’s syndrome to health professionals and researchers, and may not even acknowledge them to themselves. Perhaps for this reason the extent of the fear surrounding Down’s syndrome, and the beliefs behind it, has remained relatively unexplored in the prenatal testing context, despite the focus on anxiety generally.

Psychological consequences of termination for Down’s syndrome

For most of the women who receive a positive screening result, follow up testing will reveal that the fetus is unaffected by a chromosomal anomaly. However for around 2% of the women who undergo amniocentesis, a diagnosis of Down’s syndrome is made and termination of pregnancy almost inevitably follows. Termination of pregnancy following detection of Down’s syndrome is around 90% in the UK, Western Europe, and the USA (Alberman et al., 1995; Evans et al., 1993; Hook et al., 1995; Huang et al., 1998; Mansfield et al., 1999; Mutton, Ide, and Alberman, 1998). It is now recognised that the psychological sequelae of abortion for fetal abnormality are frequently

severe in the short-term and can extend for a number of years (Iles and Gath, 1993; Schaap *et al.*, 1997). These sequelae include the grief which accompanies severe loss of all kinds, but in addition, many couples experience reduced biological and moral self-esteem, a perception of social isolation, and fear of censure by others (Green *et al.*, 1992; Kolker and Burke, 1993; Statham, 1994; Suslak, Scherer, and Rodriguez, 1995). The distress experienced following termination for abnormality should not be underestimated. In one study, a number of standardised measures of mental health status were administered to women who had recently terminated a pregnancy following abnormality detection via ultrasound (Salvesen *et al.*, 1997). In the two-month period following the termination, women were reported as experiencing levels of intrusive thought about the event comparable to those seen in women following rape or a diagnosis of breast cancer. The authors concluded, “*termination because of fetal anomaly... represents a severe stressor for the woman*” (Salvesen, *et al.* p. 84). When it is considered that most such terminations occur after 20 weeks of pregnancy when the fetus is essentially fully formed, and that feticide and induction of labour are required, the level of distress is perhaps unsurprising. What may be more surprising is recent evidence confirming that termination for abnormality in the first trimester is often just as distressing in psychological terms (Statham, Solomou, and Green, 2001).

Deciding to terminate for a potentially lethal condition appears to be relatively less difficult than in cases where the prognosis is uncertain and the potential for a good quality of life exists (Davies and Doran, 1982; Garrett and Carlton, 1994; Herz, 1991). For this reason choosing to terminate a pregnancy for Down’s syndrome can be especially hard, even when the woman strongly believes that termination is the best option for her and that this is preferable to continuing an affected pregnancy (Green, 1992; Statham, 1994). Less is known of the psychological aspects of women who choose to continue pregnancies after a positive result, although shock and distress at the time of diagnosis is still the norm (Edwins, 2000; Helm, Miranda, and Chedd, 1998; Proud, 2000; Statham *et al.*, 2001). All pregnant women hope for a healthy baby, and any variation from this wished-for outcome requires adjustment²⁰. For many, the distress is largely resolved before the baby is born, but a diagnosis of a disabling condition is, along with miscarriage and stillbirth, one of the ‘worst case scenarios’ that pregnant women contemplate.

²⁰ It has been reported that within the Deaf culture some parents would prefer to have a child who is deaf than a hearing child (Middleton, Hewison, and Mueller, 1998). However, such parents would not consider a child who is deaf to be ‘unhealthy’.

The consequences of false reassurance

A very small number of women receive false reassurance from their screening test result. These are the women whose calculated probability of having a child with Down's syndrome was lower than the cut off point, but who were, for example, the 'one' in the one-in-a-thousand who had an affected child²¹. One recent study compared the psychological adjustment of three groups of parents of a child with Down's syndrome two to six years after the birth; parents who had received a false negative serum screening result, parents who had not been offered serum screening, and parents who had declined serum screening (Hall *et al.*, 2000). In general all parents were considered to have adjusted well to having a child with Down's syndrome. However, compared with mothers who had declined screening, mothers in the false negative group scored significantly higher on the parenting stress measure and held less favourable attitudes towards their affected child. Mothers who had received a false negative result were also most likely to blame others for the birth of their baby with Down's syndrome, and blaming others was associated with poorer adjustment. Couples in the false negative group were also most likely to have relinquished their child for adoption although this did not reach statistical significance. The researchers concluded that a false negative serum screening result appears to have a small adverse effect on parental adjustment. However, it might also be that parents who had chosen testing initially held less favourable views towards having a child with Down's syndrome and this might also have related to their adjustment difficulties. In addition, the parents of children given up for adoption were included in assessments of adjustment, which might have affected the results. Although this finding remains to be substantiated by further research, it is potentially important in light of the findings reported earlier that coping and adjustment patterns stabilise quite early on in families of children with Down's syndrome (Cunningham, 1996).

1.3 SUMMARY

It is understandable that pregnant women would like reassurance that their unborn child is healthy, however, no prenatal test can guarantee that a child will not be born with a disabling condition. Furthermore, there are adverse psychological consequences associated with prenatal testing and that reassurance cannot be the outcome for everyone. The termination of a pregnancy affected by Down's syndrome is traumatic even for couples at ease with their decision and the long-term consequences are potentially severe in some cases. For these reasons, it is desirable that women make decisions about the prenatal tests they are offered using good quality information and that

²¹ The number of false negative results depends both on the sensitivity of the test used and the risk cut off used as an indicator for diagnostic testing (Wald *et al.*, 1997).

they are given the opportunity to consider this information with relation to their own belief structures and personal circumstances.

Supporting women in making an informed choice about prenatal testing is a valid goal but a challenging one. Although the number of children born with Down's syndrome is small, the number of pregnant women offered prenatal testing for the condition is large. Following the implementation of the government's 'screening for all' policy in 2004 each year in England alone, over 600,000 women attending for antenatal care will be offered second trimester serum screening (Department of Health, 2001b). If projected serum screening uptake rates are met (Wald *et al.*, 1997) it is estimated that approximately 400 women per annum will receive a positive diagnosis of Down's syndrome via the serum screening route. When a positive diagnosis of Down's syndrome is given nearly all parents opt for an abortion (Alberman *et al.*, 1995; Huang *et al.*, 1998; Mansfield *et al.*, 1999). The latest available figures for the UK show the number of terminations for Down's syndrome to be approximately equivalent to the number of live births (Alberman, 2002). It is anticipated however, that the termination rate will begin to rise above the rate of live births as a result of a national serum screening policy and improvements in detection of Down's syndrome via ultrasound scanning. The next chapter summarises the literature relevant to informed choice within the prenatal testing context, and reviews studies that have considered information, knowledge, and attitudes in relation to prenatal testing for Down's syndrome.

CHAPTER 2 INFORMED CHOICE AND PRENATAL TESTING FOR DOWN'S SYNDROME. A LITERATURE REVIEW

For most of its history, the explicit goal of prenatal testing has been to reduce the incidence of disability in the population (Stein, Susser, and Guterman, 1973; Mikkelsen, 1988). In effect, therefore, Down's syndrome has been unproblematically viewed as a public health problem in much the same way as cancer or tuberculosis (Wilson and Jungner, 1968). It is only relatively recently that there have been concerns at an institutional level about promoting informed choice²² rather than maximising test uptake (Council of Europe, 1990; Raffle, 2001). In recognition of this shift away from paternalism in medicine the most recent report of the UK National Screening Committee states, "*There is a responsibility to ensure that people who accept an invitation [for screening] do so on the basis of informed choice*" (Department of Health, 2000, p. 1). This change in emphasis reflects, amongst other things, the general rise in consumerism in society (Charles, Whelan, and Gafni, 1999). Specifically in the context of prenatal testing, however, informed choice is also considered key in distancing the process from eugenic practices (Williams, Alderson, and Farsides, 2002c). Most of the research in the area of informed choice has focused on increasing patient knowledge about tests and their consequences, but there is a growing awareness that decisions should also reflect the individual's values (Bekker, 2003; Bekker *et al.*, 1999; Marteau, Dormandy, and Michie, 2001). A number of definitions of informed choice have been proposed that have differing emphases on either the behavioural outcome or the process depending on the theoretical perspective of the authors. In a series of recent papers concerned with the development of a measure of informed choice, Marteau and colleagues have used the following definition.

An informed choice is one that is based on relevant knowledge, consistent with the decision maker's values and behaviourally implemented" (Dormandy, Hooper, Michie, and Marteau, 2002a; Marteau *et al.*, 2001; Michie, Dormandy, and Marteau, 2002).

The authors argue that a decision to undergo testing is informed when,

"[A]n individual has a positive attitude towards undergoing a test, has relevant knowledge about the test and undergoes it. An informed decision to decline a test occurs when an individual holds a negative attitude towards undergoing a test, has relevant knowledge about the test and does not undergo it" (Marteau *et al.*, 2001, p. 100).

However it is argued that this definition of an informed choice does not account for the complex factors affecting prenatal testing decisions. Firstly, a number of attitudes and beliefs are relevant to

²² Informed choice is used in this thesis in preference to informed consent. 'Choosing' prenatal tests for Down's syndrome is not directly equivalent to consenting to recommended surgical procedures, for example, although many of the issues are related.

the decision of whether or not to undergo a screening test for Down's syndrome. Attitudes towards termination of pregnancy and attitudes towards people with Down's syndrome are relevant to the screening decision, and these might be related to, but independent of, attitudes towards undergoing a test. In addition, these different attitudes might not always be internally consistent but this should not preclude an informed choice. Secondly, this definition does not account for those individuals with neutral or ambivalent attitudes, whom, it has to be assumed can also make an informed choice. Thirdly, by setting behavioural implementation as one of the criteria for informed choice suggests that test uptake can be taken as a proxy for preference. This might not always be the case. For example, a woman could be well informed about screening and hold positive attitudes towards undergoing testing, but may be unable to attend the appointment that day. Alternatively, a woman equally knowledgeable about the test might view undergoing testing unfavourably, but go on to be tested to meet the wishes of a partner. She could even be tested without her awareness. Finally, the definition above does not account for the role that a woman's personal, financial, and social circumstances might play in her prenatal testing decisions.

In contrast to the definition by Marteau and colleagues, the following definition acknowledges the complexity of the information relevant to prenatal testing choices, and also the process by which informed decisions might be made.

An informed choice is based on (a) an accurate assessment of the information about the relative decision alternatives and their consequences, (b) an assessment of the desirability of these consequences in accord with individual beliefs, and (c) a 'trade-off' between these factors (Bekker, 2003).

This definition is preferable for a number of reasons. Firstly, it highlights the importance of assessing information about *all* alternatives and their consequences. In the case of serum screening, information about the target condition should be provided, along with information about the test process and its possible consequences. Such information is necessary if both continuing and terminating a pregnancy affected by Down's syndrome are to be promoted as equally acceptable choices. Secondly, the definition makes explicit the weighting of these alternatives in terms of their desirability within the framework of an individual's beliefs, and personal circumstances. The terms 'beliefs' encompasses a more inclusive range of information that individuals might consider than does the use of the high level construct of values. Finally, the definition recognises the trade-offs between knowledge and beliefs that people are seen to make in the real world when considering different options regarding prenatal testing (Carroll, Brown, Reid, and Pugh, 2000). The following two sections review the literature relating to women's knowledge and attitudes about prenatal testing for Down's syndrome and considers the degree to which it can be considered that women are currently making informed choices in this context.

2.1.1 Information and knowledge of prenatal tests

It is acknowledged that women need to receive four essential different types of information about prenatal tests; the purpose of the test, what the procedure for testing involves, any risks associated with the test, and the implications of the possible test results (Reid, 1988). This information is not only essential for informed choice but for understanding tests results and (potentially) for making future reproductive decisions. However, there is substantial evidence that significant numbers of women either do not receive this information, or if they do, do not fully understand it. In particular women struggle with concepts of population risk and probability and find this difficult to grasp in relation to their own pregnancy (Green *et al.*, 2002). For example, a French study of women being offered amniocentesis following a positive screening result noted a widespread lack of understanding of the accuracy of screening tests, the probability of having a baby with Down's syndrome, or of the miscarriage risk associated with amniocentesis (Gekas *et al.*, 1999). Examples of studies with similar findings come from Finland (Santalahti *et al.*, 1998a), the UK (Chilaka *et al.*, 2001; Green, Statham, and Snowden, 1993b; Grewal *et al.*, 1997; Hewison *et al.*, 2001; Smith and Marteau, 1995), the USA (Freda *et al.*, 1998) and Canada (Goel *et al.*, 1996; Glazier *et al.*, 1997). Perhaps more worrying still, is that a significant minority of women cannot say whether or not they have been offered serum screening testing for chromosomal abnormalities and NTDs, and some have inaccurate perceptions of whether or not they have actually had a test (Searle, 1997; Smith, Shaw, and Marteau, 1994). In a recent study in the UK it was reported that despite counselling only 48% of women who had undergone serum screening knew that they had had a blood test for Down's syndrome. This varied from 28% of Asian women who had been born outside the UK to 66% of Caucasian women born in the UK (Chilaka *et al.*, 2001). While worrying, women routinely have blood samples taken during pregnancy and so might not be aware, unless made so, that they did or did not have a blood sample taken for the purposes of screening for Down's syndrome. Factors that are associated with knowledge of prenatal testing are education, cultural background, the ability to read and speak English, social class, and previous experience of pregnancy (Chilaka *et al.*, 2001; Green *et al.*, 1993b; Grewal *et al.*, 1997).

A substantial amount of research effort has been put into improving the material provided prior to testing despite the fact that the role such information plays in making health decisions is not fully understood. Some studies suggest that information does not always have the impact on either knowledge, or on decision satisfaction that might be expected, but nevertheless, women want and value information about prenatal testing (Carroll *et al.*, 2000; Jepson, Forbes, Sowden, and Lewis, 2001; Michie, Marteau, and Bobrow, 1997a; Michie, Smith, McClennan, and Marteau, 1997b; Reid, 1988). Attempting to improve knowledge by providing more information prior to testing

does not appear to raise anxiety in pre- or post-test situations but its ability to significantly reduce it remains unclear (Green *et al.*, 2002). One study demonstrated some overall benefits of an intervention that provided women with basic information plus an additional one-to-one consultation with a health professional, but did not differentiate between the effects on screen-positives and negatives (Thornton, Hewison, Lilford, and Vail, 1995). High quality information provision is very important, but being better informed about the ‘four essentials’ might not protect women from the anxiety associated with a positive screening result. One study reported on a health professional familiar with serum-screening who received a positive screening result.

“Although because of her education she knew that few positive results indicated real abnormality, her first thought on learning of her positive result was ‘disaster’. That evening she was unable to sleep and felt like crying desperately. The next day she described herself as being ‘out of control’. Simply having technical knowledge did not prevent a negative emotional reaction” (Santalahti *et al.*, 1996, p.104).

This suggests that good information about the test itself is not sufficient to fully inform choices or to allay anxiety following an adverse result (Bekker *et al.*, in press). Informing women about what they might experience in emotional terms should they receive a positive result might be beneficial in setting any subsequent anxiety in some context and helping them to manage it more effectively. For some women, this knowledge might facilitate a more informed decision to decline screening. However, it can be argued that the anxiety expressed by women following a positive screening test is related to her understandings of disability and (in this context) Down’s syndrome. Therefore, even a perfect understanding of the tests is likely to have only a minimal impact on the feelings of women who are concerned about having a child with Down’s syndrome. The next section will consider the research related to information and knowledge about Down’s syndrome within the prenatal testing situation, and whether this relates to prenatal testing choices.

2.1.2 Information and knowledge about Down’s syndrome in the prenatal context

In order to facilitate informed decisions about prenatal testing, it is considered essential that women receive and understand information about the target condition(s) of that test (Advisory Committee on Genetic Testing, 2000; Marteau, 1995; Nuffield Council on Bioethics, 1993; Royal College of Obstetricians and Gynaecologists, 1996; Royal College of Physicians, 1989). However, as research has generally focused on issues around information about the tests and the testing process, what exactly constitutes the right information in the case of Down’s syndrome is still unclear. It has been suggested that, *“at the very minimum.... information about the seriousness of the condition needs to be conveyed”* (Figueiras, Price, and Marteau, 1999, p. 762). However, perception of the severity of Down’s syndrome has been demonstrated to be a subjective judgment rather than a piece of factual information. Others have argued that information containing a

balance of negative, positive, and neutral statements about the condition should be provided to enable couples to 'make their own minds up' about the severity of the condition (Loeben, Marteau, and Wilfond, 1998). While this sounds sensible, such balance in information about Down's syndrome is rarely found, partly because the domains of information covered are so limited. For example, a review article intended to help health professionals inform women about Down's syndrome prior to prenatal testing contains only information about the medical and clinical problems associated with the syndrome, thus portraying an essentially negative picture of Down's syndrome (Noble, 1998). A recent thesis concerned with decision making in the prenatal context lists the information that, "*has been identified within the literature as sufficient to enable informed decision making*" (Bekker, 1999, p.67). This list includes the prevalence of Down's syndrome, its chromosomal cause, associated life expectancy, increased risk of Alzheimer's disease, the range of learning difficulty, 'typical' facial features, and that such individuals are 'usually very loving and caring'. It is not clear why this particular information is considered sufficient to enable informed decision making, and in fact there is no consensus on what knowledge is sufficient in terms of the target condition.

Aside from the debate about which information to provide to pregnant women, the literature reveals that in many cases no information about Down's syndrome is given at all. One observational study of obstetricians presenting amniocentesis to pregnant women reported that descriptions of target conditions were not given, nor were existing understandings of the conditions determined (Marteau, Plenicar, and Kidd, 1993). Similar findings were reported in a study concerning information dissemination prior to the offer of serum screening (Marteau, Slack, Kidd, and Shaw, 1992b). Bekker (1999) audio-taped 44 instances of midwives counselling women who had received a positive serum screen for Down's syndrome and noted that information about the condition was given in only 23% of cases. Clearly the recommendations about information provision in relation to the condition being tested for are not always being followed, possibly because health professionals do not perceive that women desire such information. However, pregnant women themselves have raised the lack of information about Down's syndrome as an issue. In one French study, out of 200 women who were offered amniocentesis following a positive serum screening result, 58% reported that the information provided about Down's syndrome was insufficient, and a further 13% that no information had been given (Gekas *et al.*, 1999). Other studies report a similar dissatisfaction with this lack of information about the conditions being tested for (Carroll *et al.*, 2000; Edwins, 2000; Helm *et al.*, 1998; Levy, 1999; Moyer *et al.*, 1999; Roberts, Stough, and Parrish, 2002).

The research on pregnant women's knowledge of Down's syndrome is scant and suffers from a lack of consensus on what women should know about the condition. There are almost as many measures of knowledge about Down's syndrome as there are studies and these measures are generally poor in quality making it difficult to synthesise findings and draw conclusions (Green *et al.*, 2002). The following section summarises the studies of knowledge of Down's syndrome in the prenatal testing context.

- In a recent cross-cultural study in the UK (Chilaka *et al.*, 2001) knowledge of Down's syndrome was defined as 'good' if participants knew that the condition was associated with a 'significant mental disability', 'structural abnormality', and 'chromosomal abnormality'. It is not clear what is meant by structural abnormality, but it might be a reference to heart defects. Knowledge scores were calculated from importance ratings attached to each piece of information the women recalled. Awareness about chromosomal abnormality and learning difficulty were considered of most importance. Overall, only 33% of women were considered to have good knowledge of Down's syndrome, and this differed between 51% of Caucasian women born in the UK to 8% of Asian women born outside the UK. It is not clear from the study whether pilot work was conducted to ensure that the women understood the terminology used in the questionnaires, and thus the findings are only of limited use.
- In two Australian studies, pregnant women were interviewed at their first antenatal care appointment using a structured questionnaire that included the item, "*What do you know about Down syndrome?*" (Mulvey and Wallace, 2000; Mulvey and Wallace, 2001). In the first study 100 women were interviewed compared with 209 women in the second study. All women had been sent an information leaflet incorporating some material about Down's syndrome prior to attending the clinic. The following knowledge was reported: 90% in study 1 (2000) and 67% in study 2 (2001) were aware of Down's syndrome; 34% and 30% knew it was a chromosomal abnormality, 65% and 33% said it was associated with physical handicap (although this is technically an incorrect item of knowledge); 44% and 37% knew Down's syndrome was associated with 'intellectual handicap'. The knowledge scores vary markedly between the two different studies, although the samples were comparable in ethnicity, age, gestation, and education. The authors did not comment on this disparity, and the reasons for the difference are unknown, again making conclusions about the findings difficult.
- In an American study, knowledge of Down's syndrome was assessed by coding responses to an interview question about 'the problems caused by Down's syndrome' (Browner, Preloran, and Press, 1996). It was reported that 75% of pregnant women could correctly describe some of the problems associated with Down's syndrome, although 'only' 26% were aware of the chromosomal causes of Down's syndrome. A correct answer included mention of learning

difficulty and one other item, such as heart problems or a ‘particular appearance’. An incorrect answer did not include any reference to learning difficulty. Examples of partially correct answers included “they’re like vegetables”, “they all look the same”, or “it’s like mental retardation but not really” (Browner *et al.*, 1996, p. 143). It can be argued that these responses are actually incorrect, and that the coding reflects stereotyped views of the condition rather than knowledge.

- Another measure of knowledge developed by researchers in Canada (Glazier *et al.*, 1997; Goel *et al.*, 1996) aimed to cover the “*domains that under ideal circumstances, a fully informed woman should have knowledge of prior to [maternal serum screening]*” (Goel *et al.*, p. 426). This 14-item measure was piloted on a large sample of pregnant women and achieved acceptable levels of reliability and psychometric validity. Under the domain of ‘Target Condition’ however, only one item assessed knowledge of Down’s syndrome itself. Respondents were required to indicate their agreement/disagreement with the following statement on a 5-point Likert scale. “*All children born with Down syndrome have severe physical and mental disabilities which require lifelong care in an institution*” (ibid. p. 429). Although it is not specified, it is assumed that the correct answer to this item was ‘strongly disagree’ or ‘disagree’. Between 70% and 75% of pregnant women were reported as giving the correct answer depending on whether they had received a screening information leaflet intervention or not (Glazier *et al.*, 1997). However, even a correct response to this item signifies very little knowledge about Down’s syndrome, and cannot be considered to cover the information domain as the developers of this measure claim.

It is argued that the measures used in the reviewed studies provide a very crude gauge of knowledge of such a complex condition, and furthermore, that the validity of the response scoring is questionable. The levels of knowledge considered as ‘good’ are actually very basic, especially when the potential consequences of any prenatal testing decision are considered. The lack of a useful measure of knowledge about Down’s syndrome makes actual knowledge levels almost impossible to assess, which in turn hampers investigation of the factors associated with good or poor knowledge, and research concerned with improving this knowledge. It is not known, for example, the degree to which women have no knowledge of Down’s syndrome in comparison with ‘knowledge’ that is present but incorrect. For example, a study of pregnant women in Wales reported that 11 out of 20 women who accepted a screening test ‘did not know what Down’s syndrome was’. Unfortunately it is not clear whether this means they had never even heard of Down’s syndrome or whether they lacked basic information about the condition (Al-Jader, Parry-Langdon, and Smith, 2000). Based on the research reviewed so far, it is tentatively suggested that

many women, including both those who accept and those who decline prenatal tests, have relatively low levels of *accurate* knowledge of Down's syndrome. Factors that appear to be associated with knowledge of Down's syndrome appear to be the same as those associated with knowledge about tests, namely education, cultural background, the ability to read and speak English, and social class (Browner *et al.*, 1996; Chilaka *et al.*, 2001; Goel *et al.*, 1996; Mulvey and Wallace, 2001). Chilaka and colleagues (2001) also reported that 60% of women who knew an affected child had 'good' knowledge compared with 22% of those who did not have this experience.

To date, being 'knowledgeable' about Down's syndrome has essentially been defined as being aware of the clinical manifestations of Down's syndrome and its chromosomal origins. This is a view firmly situated within a medical model of disability that considers medical knowledge to be more valuable than social knowledge. Most research on informed choice has also been conducted from a medical perspective, within which there is little scope for investigating how information about the social aspects of having a child with Down's syndrome might impact on the testing choices of women (Asch, 1999). The message within the literature tends to be that if women were more knowledgeable about the condition, i.e. they knew about all the associated problems, they would be more likely to see the benefits of prenatal testing (Noble, 1998). This might be the case, and having knowledge of Down's syndrome (as measured by most studies) appears to be associated with higher levels of test uptake, although as has been noted previously, interest in testing might promote information seeking and the retention of test relevant information (Marteau *et al.*, 1992a). In addition, where there is already a high background test uptake rate, information provision tends to reduce uptake (Thornton *et al.*, 1995).

There is clearly a requirement to provide better quality information about Down's syndrome to pregnant women, but the content and format of this information provision has yet to be agreed upon. The recent review of the prenatal screening literature by Green *et al.* (2002) reported that women valued information provided in person by their midwife or obstetrician above that disseminated by indirect means such as a leaflet or video. This in itself raises a number of issues. While there is a general consensus that directive advice giving is inappropriate in the prenatal testing context (Clarke, 1994; Shakespeare, 1998; van Zuuren, 1997; Williams, Alderson, and Farsides, 2002a), there is also the view that as many parents ask for advice, refusing to give it leaves them to make testing decisions unsupported (Bernhardt *et al.*, 1998; Dimavicius, 1998b; Somer, Mustonen, and Norio, 1988). Unfortunately, health-professionals associated with prenatal screening appear too often to lack up-to-date knowledge of many of the aspects of Down's

syndrome, including medical factors (Brunger and Lippman, 1995; Marteau, 1995). It has been suggested that professionals lacking direct experience of Down's syndrome depend on medical textbooks for their information, and that as this is generally focused on the pathological aspects of the condition, a biased view is given to parents (Williams *et al.*, 2002c). For many situated within the prenatal testing context this direct experience might not be perceived as an issue however. A discussion article concerned with information provision in relation to screening tests noted that women should be given 'an explanation of Down's syndrome'. Later, the same article emphasised the importance of disseminating accurate information,

"Information needs to be conveyed by someone who has a sound knowledge of the tests"
(Kennard, Goodburn, Golightly, and Piggott, 1995, p. 209).

The need for someone who is knowledgeable about Down's syndrome was not mentioned.

2.1.3 Attitudes towards testing and termination for Down's syndrome: research review

As noted in the previous section, research has focused almost exclusively on the roles of knowledge and information in informing choices in the prenatal testing context. Far less interest has been shown in how values, beliefs, and attitudes inform testing choices. A number of studies have investigated attitudes towards prenatal testing and termination for disabling conditions, although only a handful have considered them in relation to Down's syndrome specifically, and even fewer in the context of informed choice. In terms of population samples these studies fall into four categories; 1) the general population, 2) health professionals, 3) people who have a family member with Down's syndrome, and 4) pregnant women. The following section reviews each of these study categories in turn.

2.1.3.1 Studies with a general population sample

The studies reviewed here were all conducted in North America and Western Europe. With the exception of one, none focused exclusively on attitudes towards testing and termination for Down's syndrome but covered a number of other disabling conditions as well (see Table 2.1). Where appropriate, attitudes towards termination for these other conditions have been reported.

The first study reviewed compared attitudes towards termination across lay populations and health professionals in three European countries (Germany, Portugal, the UK). It was reported that between 55% and 70% of the 'lay sample' (university employees) would terminate for Down's syndrome (Drake, Reid, and Marteau, 1996). This was just slightly more than the number who said they would terminate for spina bifida and cystic fibrosis and lower than the 80% who said they would terminate for anencephaly. Significant differences were found between countries, with German people being the least likely, and Portuguese people the most likely to indicate they would

terminate for disabling conditions. In a Gallup survey of the British general public, 65% agreed with the routine availability of testing and termination for Down's syndrome, compared to 41% who said they would use diagnostic tests with a view to a termination themselves (Marteau, Michie, Drake and Bobrow, 1995). A higher proportion of people here said they would terminate for anencephaly (63%), and a lower proportion for cystic fibrosis (32%). Similar findings were reported in a Danish study where 78% of the sample agreed with the availability of first trimester termination for Down's syndrome, but 50% said they would consider it themselves (Norup, 1997). A Belgian study reported that 67% of their sample would want to use prenatal diagnosis for Down's syndrome compared with the 38% who would terminate an affected pregnancy (Evers-Kiebooms, Denayer, Decruyenaere and Van den Berghe, 1993). This was a similar proportion to the number who would abort for a condition where the child would die soon after birth, but greater than for a condition which was related to physical handicap only (17%). Similar findings were seen in an American Gallup survey where, 65% of both women and men said they would want to use prenatal testing for serious genetic defects, but 41% said they would abort an affected pregnancy (Singer, 1993).

In the study by Singer (1993) attitudes towards testing and termination for Down's syndrome were not measured specifically, but it was the most frequently first-mentioned condition in responses to an item asking for a definition of 'serious genetic defect' (71% of respondents). Of note was that those people who mentioned Down's syndrome as a 'serious genetic defect' were more likely to be white than black, women than men, and of higher rather than lower income. This probably reflects the greater awareness of white middle-class women towards prenatal testing (and by association Down's syndrome) that has been noted elsewhere (Browner, Preloran, and Cozzarelli, 1999; Chilaka *et al.*, 2001; Ford *et al.*, 1998; Press and Browner, 1998; Rothman, 1986; Saridogan, Djahanbakhch, and Naftalin, 1996). However, those people who mentioned Down's syndrome specifically were also more likely to say that genetic screening would do 'more harm than good', and Singer noted that awareness of Down's syndrome did not necessarily equate to a willingness to test and terminate for it. In a replication of this study six years later, only two significant changes in the responses were found; a decline in the number of people who said they 'knew nothing' about genetic testing, and an increase in numbers who indicated that they would not choose termination (Singer, Corning, and Antonucci, 1999). The number of people who said they would choose termination remained the same, therefore fewer people reported uncertain attitudes. Singer and colleagues suggested that the greater knowledge levels might be related to the growing debate about the uses and abuses of reproductive technologies, and that attitudes were becoming more informed and more polarised as a result. They concluded that although use of

prenatal testing was becoming more widespread, there was no evidence that attitudes towards using it were becoming more favourable overall.

Table 2.1. Attitudes towards personal use of prenatal testing and termination for Down's syndrome: general population studies.

Study and country	N=	Attitude toward using testing	Attitude toward terminating	Comments
Drake et al. (1996). UK, Germany and Portugal.	600	Not measured	Approximately 55% (Germany), 60% (UK) and 70% (Portugal) would 'probably have' a termination for DS	Attitudes of over 1700 participants in 4 groups, including a 'lay sample' of 600 university employees.
Evers-Kiebooms et al. (1993). Belgium	385	67% would test for DS if "they were pregnant at 40 years".	39% believed they would abort for DS, 38% would not.	Convenience sample of adults attending further education.
Marteau et al (1995). UK	973	See next cell.	41% would test for DS with the "possibility of ending the pregnancy".	Stratified sample.
Norup, (1997) Denmark	731	Not measured	50% would terminate for DS in the first trimester.	Random sample
Singer (1993). USA	1000	63% would test for 'serious genetic defects' including DS. 28% would not choose testing.	41% would choose to abort for a 'serious genetic defect', 38% would not abort.	Probability sampling and a "nationally representative sample".
Singer et al (1999). USA	989	64% would test for 'serious genetic defects'. 29% would not choose testing.	42% would choose to abort for a 'serious genetic defect', 45% would not abort.	Replication of and comparison with Singer (1993)

In summary, a significant proportion of these general public samples reported favourable attitudes towards using prenatal testing with a view to terminating a pregnancy for Down's syndrome, although this varied somewhat across the studies. Three main conclusions can be drawn. One, that people hold more favourable attitudes towards the availability of these technologies than they do towards using them themselves. Two, that people hold more favourable attitudes towards using prenatal testing than they do towards terminating an affected pregnancy. Three, that on a hierarchy of appropriateness for termination, Down's syndrome tends to fall above physical disability or chronic illness, but below lethal conditions such as anencephaly. In those studies where factors associated with variation in attitudes were investigated, attitude toward abortion generally was predictive of attitudes towards using prenatal testing and termination for fetal abnormality (Evers-Kiebooms, Denayer, Decruyenaere, and van den Berghe, 1993; Norup, 1997). People who were Catholics were least likely to intend to use termination, and people who were Jewish were most likely (Evers-Kiebooms *et al.*, 1993; Singer, 1993; Singer *et al.*, 1999). However, religious observance was a stronger predictor of the target attitudes than religion (Drake *et al.*, 1996; Evers-Kiebooms *et al.*, 1993; Norup, 1997; Singer, 1993; Singer *et al.*, 1999). Increased age was related to more favourable attitudes towards abortion in some studies (for example, Norup, 1997) but evidence for the significance of this variable is mixed (Furr and Seger, 1998).

In the USA, abortion is politically more contentious than it is in Britain and Denmark. In Belgium and Portugal, where the populations are mainly Roman Catholic it would be expected that abortion would be considered less socially acceptable. Despite the different cultural contexts of the studies reviewed, the numbers willing to terminate for Down's syndrome were generally consistent. The higher rates of favourable attitudes towards termination for Down's syndrome seen in the study by Drake and colleagues (1996) might be due to the selection of university employees as the lay sample. Higher levels of education have been found elsewhere to be associated with more favourable attitudes to termination for abnormality (Green, Snowdon, and Statham, 1993a). Although understandings of Down's syndrome were not measured directly in any of the studies reviewed, Evers-Kiebooms and colleagues (1993) reported that the expected burden of caring for an affected child, the value placed on a successful life, and pleasure and relaxation were related to attitudes towards termination for all disabling conditions.

The final study reviewed in this section also measured attitudes towards termination for a number of disabling conditions, but also aimed to examine in more detail how such attitudes were associated with views about abortion generally, religious belief, and attitudes towards people with disabilities (Bell and Stoneman, 2000). At the start of the study the participants (American

undergraduates) completed three measures; the Attitudes Towards Abortion scale (Krishnan, 1991), the SATDP (Scale of Attitudes Towards Disabled Persons (Antonak, 1982)) and a 'comfort scale' to measure affective aspects of attitudes towards people with a disability (Stoneman, 1997). Following this, three videos were shown of a couple (played by actors) being told by their doctor that their unborn child had been diagnosed as having Down's syndrome, spina bifida, or haemophilia²³. Participants were then asked to imagine themselves in the position of the couple in the video and indicate on a five-point Likert scale how likely they were to abort the pregnancy. Respondents then selected one of five options to indicate their main reason for their view about termination for each condition. Overall, termination for Down's syndrome was considered more favourably than termination for the other two conditions. As expected, variables associated with an intention not to terminate were greater levels of religious observance, and having a generally unfavourable attitude toward abortion. In addition, a higher level of acceptance of people with disabilities (as measured using the SATDP) was significantly related to less favourable attitudes towards termination for Down's syndrome, as was degree of comfort with people with disabilities. However, these correlations were small and accounted for only 2 to 3% of the variation in scores. Of interest is that 'quality of the child's life' was given as the main reason for terminating the pregnancy, continuing it, or not being sure about continuing a pregnancy for all three conditions. This suggests that while an understanding of Down's syndrome was a key factor in the participants' intention to terminate, a scale measuring attitudes to *existing* individuals with a non-specific disability were not necessarily sensitive enough to capture this.

2.1.3.2 Studies with health professionals

Health professionals can be situated in the prenatal testing context in a number of ways. Firstly, they facilitate the testing process in antenatal practice, secondly, they can be involved in the development and implementation of prenatal testing policy, and thirdly, they can be consumers themselves. It has also been suggested that health professionals can influence the testing choices that women make, and for all these reasons their views are of interest when considering the issue of informed choice (Marteau *et al.*, 1993). A small number of studies have investigated the attitudes of health professionals towards prenatal testing and termination for Down's syndrome although most of these have considered views in terms of the acceptability of these technologies rather than attitudes towards personal usage.

²³ A hereditary disorder that results from an impairment of blood clotting function. The severity of the disorder varies greatly but can result in internal bleeding and early death.

Three studies have surveyed the views of midwives towards testing and termination for Down's syndrome. In one English study, 70% of respondents were in favour of serum screening for Down's syndrome, while 49% felt termination for the condition was justified - a higher proportion than for the chronic physical condition thalassaemia²⁴ (34%) but a lower proportion than for spina bifida (62%) (Khalid, Price, and Barrow, 1994). Another survey of midwives in a Northern English health authority reported that 95% were in favour of maternal serum screening for Down's syndrome, although attitudes towards personal usage or termination were not measured (Fairgrieve, Magnay, White, and Burn, 1997b). This health authority has been particularly active in establishing a prenatal screening service including a commitment to training health professionals, and this might partly explain the very favourable views reported by its midwifery staff. A recent Finnish study considered attitudes towards both the availability and personal usage of testing and termination in a large random sample of midwives and public health nurses (N=571) (Jallinoja, Santalahti, Toiviainen, and Hemminki, 1999). Overall, 80% of respondents agreed that serum screening should be available to all women, although the proportion that thought that they would use it themselves in a future pregnancy was somewhat lower at 66%. A smaller number again felt that termination for Down's syndrome was acceptable (54%) although personal views on having a termination were not collected. Midwives who were involved in the delivery of screening were most likely to find abortion for Down's syndrome acceptable, and those who felt that abortion was acceptable were most likely to agree that having a child with a disability is a 'disaster for the family'.

These studies demonstrate that, like the wider public, midwives hold a wide range of views on prenatal testing and termination for Down's syndrome and are more likely to be in favour of its availability than of using it themselves. The numbers supporting the availability of testing and termination for Down's syndrome are similar to those seen in the surveys of general public populations, with perhaps a greater favourability towards the availability of testing. However, the studies also reported that a number of midwives have concerns about prenatal testing and about the acceptability of termination for Down's syndrome. Concerns reported in these studies were related to lack of adequate counselling and information for women (Fairgrieve *et al.*, 1997b; Khalid *et al.*, 1994), anxiety caused by false positive results, and the emotional and ethical context of late abortion for themselves and their patients (Jallinoja *et al.*, 1999; Khalid *et al.*, 1994). Those who did not feel that termination for Down's syndrome was acceptable were most likely to agree that

²⁴ A recessively inherited condition in which red blood cells are destroyed, reducing the oxygen-carrying capacity of the blood. Toxic quantities of iron are released and in the most severe form this results in permanent organ damage and early death.

prenatal testing will lead to attitudes towards those with disabilities becoming more negative (Jallinoja *et al.*, 1999, p. 1019).

A number of studies have considered the views of obstetricians and other physicians towards prenatal testing and termination. Although obstetricians are less likely than midwives to be actively offering screening tests on a day-to-day basis they conduct prenatal diagnostic tests, counsel women at various stages of the process, and are more likely to be involved in decisions at a policy level. A Danish study compared the views of obstetricians, paediatricians, and 'other physicians' and found a number of differences in the acceptability of termination for disabling conditions between these groups (Norup, 1998). Ninety-three percent of obstetricians found termination for Down's syndrome acceptable until 21 weeks gestation compared with 76% of paediatricians and 60% of the 'others' group. The obstetricians were also more likely to find termination acceptable for a range of other conditions including relatively mild and late-onset disorders. A similar survey conducted in France and Canada compared the views of male and female doctors on prenatal testing and termination (Bouchard and Renaud, 1997). Female doctors were most in favour of increasing access to prenatal testing, and 59% of female physicians compared with 50% of male physicians considered termination for Down's syndrome acceptable. However, female doctors were less likely to agree that a physician should try and influence a patient's decision regarding use for these technologies. Bouchard and colleagues therefore suggested that the female doctors held more liberal views towards prenatal diagnosis and termination for disabling conditions because they were more committed to reproductive choice for their patients. Obstetricians and radiologists were more accepting of termination for Down's syndrome than were GPs and paediatricians.

A study that has also considered doctors' attitudes towards personal usage of prenatal testing and termination was the cross-European study by Drake *et al.* (1996) discussed previously. In this study the attitudes towards termination for Down's syndrome held by geneticists and obstetricians were compared with those of a lay sample and a sample of pregnant women (Drake *et al.*, 1996). The attitudes of the health professionals towards using termination for Down's syndrome were found to be significantly more favourable overall than the other two groups although they varied by country. In Germany 65% of geneticists said they would opt for termination for Down's syndrome, compared with around 80% of the geneticists and obstetricians in the UK and 85% of geneticists and obstetricians in Portugal. The authors proposed a number of reasons why health professionals might hold more favourable attitudes towards termination than lay populations. Firstly, they suggested that lay people might underestimate the likelihood that they would

terminate following a positive diagnosis, and secondly, that health professionals perceive Down's syndrome to be a more severe condition than the lay population.

Overall, health professionals in areas such as obstetrics, genetics, and fetal medicine appear to hold more positive attitudes towards prenatal testing and termination for Down's syndrome than do the general public, and the evidence suggests they are also more likely to want to use such technologies themselves. This is perhaps unsurprising as obstetric medicine is focused on the delivery of a healthy child, the development of prenatal testing was driven mainly by the medical model of disability, and termination is a medical 'answer' to the social problem of having a child with a disability. For some health professionals, parental acceptance of prenatal testing for Down's syndrome might also be viewed as a social responsibility. In a survey of the views of British obstetricians towards the availability of testing and termination for abnormality (Green, 1994; Green, 1995b) 13% of the respondents agreed that,

"The state should not be expected to pay for the specialised care of a child with a severe handicap in cases where the parents had declined the offer of prenatal diagnosis of the handicap" (Green, 1995, p. 229).

Agreement with this statement was strongly related to the obstetrician requiring an undertaking that an affected pregnancy would be terminated before proceeding with amniocentesis. It is of particular interest that in the 1980 survey on which the Green study was based (Farrant, 1985), 13% of obstetricians had also agreed with the statement suggesting that this view is a relatively stable one in a minority of health professionals. In one North American study, researchers made a training video with parents of children with Down's syndrome in which the parents talked about their own experiences and their attitudes towards prenatal testing (Cooley, Graham, Moeschler, and Graham, 1990). This video was then rated on a number of dimensions by three groups of women - mothers of affected children, nurses, and genetic counsellors. Eighty-nine percent of mothers compared with 41% of nurses, and 14% of genetic counsellors agreed that the film accurately portrayed the views of parents. Many of the genetic counsellors commented that the film was too positive to be of use in an educational or counselling role. When asked about aspects of parenting a child with Down's syndrome, 94% of mothers and 83% of nurses felt that the benefits outweighed the problem, while 48% of counsellors felt the problems outweighed the benefits.

Lippman and Brunger (1991) suggest that the tendency of medical texts to define people with Down's syndrome in terms of their condition, for example, a 'Down's syndrome child', and to emphasise pathological manifestations such as a 'protruding tongue' inevitably positions them as

“other”. They argue that this is not value-free information, and tends to shape the understandings of Down’s syndrome in many health professionals in a particular direction. This in turn may have an influence on their attitudes towards the value of prenatal testing, and ultimately on research funding and the diffusion of tests.

2.1.3.3 Studies with people who have a close family member with Down’s syndrome

In people who have a close family member with a disability it is reasonable to expect that their views about using prenatal testing and termination will be informed by their personal familial experience. Like health professionals, such individuals are more likely to be aware that the primary role of prenatal testing is the identification of abnormality rather than reassurance of normality, and to make their choices based on this awareness. While they might still seek reassurance that their child is not affected, they know that having a child with a disability is not something that ‘happens to other people’. Some researchers have therefore explored the testing choices of people who have a family member with a disability in order to understand the effects of such direct experience. For example, in one American study, 310 mothers of ‘congenitally impaired’ children were asked whether they thought abortion should be legally available where there was a ‘strong chance of a serious deformity of the baby’ (Breslau, 1987). Sixty eight percent agreed that it should, which was a virtually identical response to the control group of mothers whose children were ‘free from disability’. The strongest predictor of attitude toward abortion for fetal abnormality across both groups was attitude to abortion in general. Breslau stated, “*the majority of mothers of congenitally disabled children, like the majority of the general public does not defend the right-to-life of defective fetuses*” (p. 844). However, this conclusion can be challenged on two points. Firstly, the majority of the ‘impaired’ children were affected by chronic disease conditions such as cystic fibrosis, or had moderate learning difficulty. Therefore, it can be argued that the mothers did not see their own child as having a serious deformity (a phrase imbued with emotional connotations), and so the intended comparison is invalid. Secondly, as previously noted, people are more likely to support the termination rights of others than they are to consider it themselves; there is a world of difference between your own baby and a hypothetical fetus belonging to a hypothetical ‘other’.

Mothers of children with standard trisomy 21 have an overall probability of 1% that any subsequent pregnancy will be similarly affected and are thus automatically a ‘high risk’ population. A number of studies have been conducted with mothers of children with Down’s syndrome, regarding their attitudes towards using testing and termination in subsequent pregnancies.

- In an American study of 101 mothers of children with Down's syndrome, 40 women had gone on to have a subsequent pregnancy and 20 of these had used amniocentesis (Elkins *et al.*, 1986). Of these twenty, ten said they wanted time to prepare for the birth, and ten said they would consider termination; the one woman who received a diagnosis of Down's syndrome did abort the pregnancy. Of the 20 women who decided against testing, nine said they 'were not worried' about having another affected child and many quoted the risks of miscarriage as outweighing the benefits of knowledge. The sample represented approximately one-third of parents belonging to a parent support organisation in one region of America, and so the findings might be somewhat unrepresentative of the population of mothers as a whole.
- A different American study reported much higher rates of prenatal testing usage in women who had a child with Down's syndrome (Ekwo *et al.*, 1985; Ekwo, Kim, and Gosselink, 1987). In this study, of 25 pregnant mothers attending for genetic counselling at a hospital clinic, 21 (84%) had an amniocentesis. The numbers of those who intended to terminate following a positive diagnosis were not given. However, there may have been mothers who had subsequent pregnancies but who did not attend for genetic counselling so the testing uptake might be somewhat inflated.
- A Belgian study conducted in the early 1980s reported the testing choices of a group of 95 families with a child with Down's syndrome (Swerts, 1987). Of 35 who had a subsequent pregnancy, 30 used amniocentesis and two would have used testing but the pregnancy ended in miscarriage. However, these findings cannot be generalised because the sample was pre-selected to include equal numbers of those who had already used amniocentesis, those attending for genetic counselling, and those with no previous experience of counselling.
- A British survey conducted in 1978-1979 reported that out of 100 sets of parents of children with Down's syndrome, over two-thirds believed they would terminate any further affected pregnancies (Boon, 1986). In those women who subsequently went on to have more children, 74% used diagnostic testing when offered. Again, however this sample was largely drawn from a group of families who chose to attend for genetic counselling.
- Finally, a more recent American study (Felker, 1994) reports on mothers of children with Down's syndrome who had had at least one subsequent pregnancy. In this sample 11/20 (55%) 'did not' have amniocentesis and nine chose to have testing (it is unclear whether those women who did not have testing were offered it). Five women intended to abort the pregnancy if the result was positive and four said they had the test to be prepared. None of the women who intended to terminate said this was because they wished they hadn't had their child with Down's syndrome, instead financial concerns, and the desire for their affected child to have a typical sibling role model were reasons given. This sample of mothers were all 'middle to

upper class' and all but two were living with the child's father. As the author states this might mean that they had better than average resources for coping with a child with Down's syndrome.

These studies demonstrate that like other sections of the population, mothers of children with Down's syndrome hold a variety of views about the availability and personal usage of prenatal testing and termination for the condition; there is no consensus resulting from 'shared' experience. The reported usage of prenatal testing by the mothers also varied, reflecting the different study designs and the difficulty of obtaining a large representative sample in this population. In addition the studies did not generally report the views of women who chose not to have further children. However, in the study by Boon (1986) it was reported that out of 100 mothers, 36 had decided not to have more children either because they considered themselves 'too old' or because they 'could not cope on account of the affected child' (p. 156). Most of the studies were conducted prior to the availability of biochemical or ultrasound screening for Down's syndrome and it is likely that amniocentesis had not been offered to a high proportion of the women during their pregnancy affected by Down's syndrome. As noted in the review of studies of family adjustment, the mothers are likely to have been a heterogeneous group, including some who would have terminated their affected pregnancy had this been an option, and some who perhaps had used testing but continued the pregnancy following a positive diagnosis. In addition, the studies were conducted over a period of nearly twenty years during which time the social, educational, and medical outlook of people with Down's syndrome has altered. Thus it is not possible to draw definitive conclusions about the factors associated with testing and termination intentions in those women who already have a child with Down's syndrome. Nevertheless, this variation in outlook is likely to be related to the way in which individual mothers experience and adjust to their child with Down's syndrome and to their life situation generally as well as to the particular characteristics of the affected child.

Another group who have direct experience of a family member with Down's syndrome are their siblings. The majority of siblings will go on to have their own children, yet their attitudes regarding prenatal testing and abortion for this condition are under-researched. This is surprising considering that most will have experience of Down's syndrome over an extended period of time and will know a great deal about the impact of having an affected sibling on themselves and something about how their parents and family have been affected. They are likely to have experienced the attitudes of others towards people with the condition, and be aware of the social support and educational issues. In addition, many will have experience through adolescence and adulthood, and have thought about, if not dealt with, issues such as who will be responsible for

their sibling when their parents die or are no longer able to function as carers. These are the very issues raised by couples making decisions about prenatal diagnosis and termination for Down's syndrome. One unpublished study obtained the views of 78 women who had a sibling with Down's syndrome towards prenatal testing and termination for the condition (Bryant, 1998). Of these, 54% indicated that they would use diagnostic tests for Down syndrome in the future and 37% said they would not. While 76% said termination for Down's syndrome should be available for other women, 33% said they would consider termination themselves and 53% said they would not consider termination. A number of significant relationships were found between the target attitudes and other variables, but both attitudes were most strongly associated with the perceived burden of caring for a child with Down's syndrome and perceived family approval or disapproval if they should terminate an affected pregnancy. Of interest is that burden of care was not associated with the perceived degree of learning difficulty or the physical health of their sibling. It was, however, associated with the existence of mental health problems and behavioural difficulties in the sibling with Down's syndrome.

In summary, people who have a family member with Down's syndrome hold quite varied views. Some see prenatal testing for Down's syndrome as intrinsically offensive: an insult to themselves and their relative. Some welcome it because it allows them to have control over whether or not they have a(nother) child with Down's syndrome. Most appear to be accepting of the existence of testing even if they would not wish to terminate for Down's syndrome themselves, and in this they are similar to the other groups whose views have been reported. In a recent study of mothers of young children with Down's syndrome in Australia, the researcher reported that the mothers were tolerant of others' decision making on this issue (Bridle, 2000). She noted that,

“The overall impression was that participants were concerned, unsettled or saddened by the unquestioning enthusiasm for testing, but they did not condemn women who decide to use the technology or terminate an affected pregnancy”(p. 9).

Similar views have been expressed by siblings of people with Down's syndrome (Bryant, 1998).

Studies with people who have had intimate familial experience of people with Down's syndrome are important for emphasising that attitudes and beliefs in relation to prenatal testing choices for Down's syndrome are as important (perhaps more important) than factual knowledge of the condition. However, the studies described in the sections so far have used hypothetical scenarios to assess attitudes to prenatal testing and termination, the use of which has been criticised because they do not have a good track record of predicting behaviour (Green, 1995). As the relevance of prenatal diagnosis for the participants of these studies was unknown, the views reported may have

not reflected attitudes or behaviour in a real situation. The next section reviews studies that have examined the attitudes of pregnant women towards using prenatal testing and termination for Down's syndrome, and considers these attitudes in relation to actual testing choices.

2.1.3.4 Studies conducted with women during pregnancy or in the post-natal period

A substantial number of studies have investigated the choices that pregnant women make in terms of testing and termination for Down's syndrome, however in the majority, test uptake has been the dependent variable of interest. This makes it difficult to draw conclusions about attitude as distinct from behaviour. In addition, few studies have considered attitudes toward testing and termination for Down's syndrome specifically. This section will draw on the prenatal testing literature in an attempt to bring together what is currently known about the role of women's attitudes in the prenatal testing context. Attitudes towards screening tests, diagnostic tests, and termination will be considered separately as although there is clearly overlap there are also specific issues associated with each stage of the testing process. The review will focus mainly on studies conducted in the past ten years to reflect the period that screening for Down's syndrome has been routinely available.

Although ultrasound scans are very popular with expectant parents their screening capabilities are frequently not made explicit (McFadyen, Gledhill, Whitlow, and Economides, 1998). This is despite the fact that ultrasound markers are now the principle indicator leading to diagnosis of Down's syndrome (Mutton *et al.*, 1998). Women rarely decline a scan even when they have refused all other testing, suggesting that they are valued for other purposes than detection of abnormality (Baillie and Hewison, 1999; Crang-Svalenius, Dykes, and Jörgenson, 1998; Santalahti *et al.*, 1998a; Whynes, 2002). Attitudes towards using serum screening are perhaps less favourable than those towards ultrasound, although the availability of serum screening is generally viewed positively (Dormandy *et al.*, 2002a; Green *et al.*, 2002; Jørgensen, 1995a; Moyer *et al.*, 1999). In a Finnish study of women who had had serum screening for Down's syndrome during a recent pregnancy, 87% said they valued the test including most of those who had received a false-positive result and all of those whose baby had subsequently been prenatally diagnosed with Down's syndrome (Heikkilä, Ryyänen, Kirkinen, and Saarikoski, 1997). However, the views of women who did not participate in screening were not measured here and as the study was conducted retrospectively most participants had already delivered a healthy baby. In a recent prospective study, attitudes towards serum screening for Down's syndrome were measured by asking 1499 pregnant women to evaluate screening on Likert-types scales as 'good' or 'bad', beneficial or harmful, important or unimportant, and pleasant or unpleasant (Dormandy *et al.*,

2002a). This measurement took place after screening was offered but prior to it taking place. Fifty-seven percent of women overall were reported to hold favourable attitudes towards undergoing screening with attitudes considered positive if the attitude score was greater than the midpoint of the possible range. Seventy-three percent of those women with favourable attitudes towards serum screening went on to have the test some weeks later as did 19% of those with 'unfavourable' attitudes towards testing (overall uptake of 51%). The authors suggest that women whose behaviour was not consistent with their attitudes had not made an informed screening choice. It is argued however, that the measure used (the Multidimensional Measure of Informed Choice, (MMIC), Marteau *et al.*, 2001) might not be sensitive enough to draw conclusions about the role of attitudes towards testing in informing choice. A midpoint split is a rather crude way of defining favourable or unfavourable attitudes towards a complex issue such as prenatal screening, and, a person might view serum screening to be unpleasant because it requires a blood sample to be taken, yet also believe it to be important, and consider importance of greater weight than unpleasantness. Although the items were based on the Theory of Planned Behaviour (TPB) (Ajzen 1985, 1988, 1991) the full TPB model takes into account the weighting of variables in predicting behavioural intentions, which the MMIC does not.

Studies report a wide range of serum screening uptake figures that is at odds with the relatively consistent numbers of people who indicate that they would use testing in hypothetical situations. A review of screening demonstration projects reported uptake ranging from 67% to 92% (Wald *et al.*, 1997). A number of other recent studies report uptake rates from 25% to nearly 98% (Al-Jader *et al.*, 2000; Beaman and Goldie, 2001; Dormandy, Michie, Weinman, and Marteau, 2002b; O'Connell, Holding, Morgan, and Lindow, 2000; Saridogan *et al.*, 1996; Spencer *et al.*, 2000). The observed variation in screening uptake is likely to be related to a number of factors, including the ethnic and social mix of the district where testing is offered (Chilaka *et al.*, 2001; Ford *et al.*, 1998), whether screening is in the first or second trimester (Spencer *et al.*, 2000), and the accessibility of local testing services (Halliday, Lumley, and Watson, 1995). However, two recent studies by Dormandy and colleagues suggest that the organisation of screening services might also be an important factor in this variation. The first study looked at the differences in screening uptake across 29 hospitals in one English health region. In all the hospitals serum screening was the main method of testing for Down's syndrome yet uptake varied between 25% and 93% (Dormandy *et al.*, 2002b). The researchers found that where screening was offered as part of a routine antenatal appointment, screening uptake was on average 17% higher than where it took place at a separate appointment (73% vs. 56%). This wide variance in uptake was not explained by obvious differences in the way that women were informed about the screening as twelve hospitals

using the same leaflet had uptake rates varying from 25% to 76%. In the follow-up study (discussed previously) attitudes towards having screening for Down's syndrome were measured in pregnant women across two hospital sites; one where serum screening was conducted during a routine appointment and one where it was conducted at a separate appointment (Dormandy *et al.*, 2002a). Both sites used the same screening test and the same information leaflet. The proportion of women holding favourable attitudes towards using serum screening was 53% at the 'separate appointment hospital' and 61% at the 'routine appointment hospital'. Screening uptake at the 'separate appointment hospital' was 41% compared with 62% at the 'routine appointment hospital'. In addition, an interaction was found whereby the relationship between attitude and uptake depended on the hospital; where the test was carried out at a routine appointment, those with favourable attitudes were more likely to have the test. All these relationships were statistically significant even when values were adjusted for age, ethnicity and socio-economic status. This interaction was not seen with knowledge scores, and attitudes towards testing were found to be a better predictor of uptake than test knowledge.

As a result of their findings, Dormandy *et al.* (2002a) argued that routine visits facilitate informed choice by removing physical barriers to appointment attendance. However, the findings also suggest that organisation of screening services might impact on attitudes towards using screening. If screening is presented as a component of standard antenatal care rather than something requiring an optional extra appointment this might increase its perceived importance or its perception as the norm. Some have argued that this might impair a woman's ability to make an informed choice based on her own values (Alderson, Farsides, and Williams, 2001; Searle, 1997). Still others see routinisation as a subtle form of coercion (Bennett, 2001; Lippman, 1994). It can also be argued that when participants are women actually situated in the testing context, the attitudes being measured are attitudes towards testing *as it has been offered to them*. As Press and Browner noted (1997),

"Institutional support shapes not only rates of test acceptance, but also the way in which the [test] is understood by the patients" (p. 984).

In support of this, a retrospective study of over 3,500 pregnant women in two areas of Denmark reported that testing attitudes were strongly related to how prenatal screening was organised in the area where the women had attended for their antenatal care (Jørgensen, 1995a). In areas where ultrasound had been offered routinely, around 89% of women thought it should be offered to all women compared with 56% of women in an area where ultrasound was not a standard procedure. In an American study with women aged 35 years or older, it was reported that attitudes towards serum screening were related to whether or not the women had been offered the test as part of their antenatal care (Phillips *et al.*, 1998). Those women to whom screening was offered held

significantly more favourable attitudes towards using the test than those who had been offered diagnostic testing only. These findings are supported by a number of earlier studies of screening for NTDs, showing that the degree of provider support for screening tests are highly predictive of test uptake (Press and Browner, 1997). In conclusion, it is difficult to disentangle the value that women place on prenatal screening for detection purposes from their views on the care they receive and from the value that is placed on antenatal care in general (Browner and Press, 1991; Dowswell, Renfrew, Gregson, and Hewison, 2001; Porter and Macintyre, 1984; Press and Browner, 1997).

With the exception of women over 35 for whom the offer of amniocentesis for Down's syndrome might be standard procedure, a diagnostic test is not part of the routine antenatal care for most pregnant women. This is reflected in pregnant women's attitudes towards the availability and usage of amniocentesis and CVS when compared to those for screening tests. In the Danish study by Jørgensen (1995a) 26% of participants agreed that all women should be offered diagnostic tests regardless of age in comparison with 79% who believed serum screening should be generally available. Of note is that while 44% of women overall said they would accept a diagnostic test if it was offered to them, 85% said they would accept it if they were *advised* to do so, including 41% of those women who had already declined a serum screening test. In an American study of women who had received a positive serum screening result for Down's syndrome, those women who accepted an amniocentesis (57%) were more likely to say that their healthcare provider had recommended diagnostic testing than those women who had declined the test (Priest *et al.*, 1998). These findings emphasise the importance that many women accord to the views of health professionals responsible for their antenatal care.

Of those women considered to be at higher than normal risk of an affected pregnancy, due to advanced age or other screening indication, studies report uptake of amniocentesis to be between 45% and 90% with a typical uptake rate being around 80% (Johnson *et al.*, 1998; Marini, Sullivan, and Naeem, 2002; Marteau *et al.*, 1991; O'Connell *et al.*, 2000; Priest *et al.*, 1998; Roelofsen *et al.*, 1993; Tercyak, Johnson, Roberts, and Cruz, 2001; Wald *et al.*, 1997). A number of variables appear to be associated with favourable attitudes towards using prenatal diagnosis. In particular favourable attitudes towards abortion generally predict favourable attitudes towards prenatal diagnostic testing. Women from religious and/or cultural backgrounds that are generally less supportive of abortion are also less likely to undergo prenatal diagnosis (Halliday *et al.*, 1995; Moyer *et al.*, 1999). Another important factor associated with uptake of amniocentesis is the woman's perception of the risk of having an affected child. In one American study, women who

accepted amniocentesis following a positive serum screen had a higher overall risk value than did women who declined, suggesting that actual risk is an important variable. However, in this study acceptance of testing was best predicted in the situation where the screening generated risk was greater than *a priori* age, suggesting that the women did their own 'risk analysis' using information given to them by their care provider (Johnson *et al.*, 1998; Priest *et al.*, 1998). Similarly, a study comparing age related risk and perceived risk found that perception was more important in terms of amniocentesis uptake than was actual risk (Marteau *et al.*, 1991).

Other factors associated with a favourable attitude towards using diagnostic testing include advanced maternal age, the perceived burden of caring for a disabled individual, and the belief that an affected child would impact negatively on partner and other children (Jørgensen, 1995a; Marteau, 1991; Priest *et al.*, 1998). A number of studies have reported significant relationships between the perceived burden of caring for a child with Down's syndrome and attitudes towards diagnostic testing (Bryant, 1998; Marteau, 1992; Ekwo *et al.*, 1985; Evers-Kiebooms *et al.*, 1993). What constitutes burden has not been clearly defined however, nor has it been investigated within the prenatal context why some people perceive parenting a child with Down's syndrome as burdensome while others do not. In a qualitative study of Swedish couples who had elected for prenatal diagnosis, it was reported that half believed that a child with Down's syndrome would suffer (Sjögren and Uddenberg, 1987). One participant commented, "*Nobody looks after them. I would rather die than live in an institution*" (ibid. p.190). All of the interviewees believed the family suffered and wished to give preference to the concern of existing family members, for example, "*the whole family would have a terrible life, I have decided to avoid this*" (ibid., p. 192). Feelings of shame and of 'people staring' were also imagined. This study shows that when not obscured within the vague category 'burden' people can express a real fear of what having a child with a condition like Down's syndrome might mean. Reasons for declining testing include unfavourable attitudes towards abortion for religious or personal reasons, the risk of miscarriage associated with diagnostic tests, not wanting to receive 'bad news', and unfavourable attitudes towards increased medicalisation of pregnancy (Berne-Fromell, Josefson, and Kjessler, 1984; Gokhale and Cietak, 2002; Jørgensen, 1995b; Marteau *et al.*, 1992a; Faden *et al.*, 1987; Green *et al.*, 1993a; Press and Browner, 1998; Santalahti *et al.*, 1998b). However, one study demonstrated that women who accept screening tests share many of these reservations, and conversely, women who decline testing are not always rejecting the technology wholeheartedly. The authors argue that reasons for acceptance and refusal of screening are complex and linked to personal assessment of various types of 'risk' associated with prenatal testing generally (Markens, Browner, and Press, 1999). Personal positive experience of Down's syndrome has also been cited as a reason not to use

prenatal testing (Ekwo *et al.*, 1985; Marteau *et al.*, 1993) although experience with people with Down's syndrome *per se* does not appear to be a predictor of the direction of attitudes towards testing for the condition, as might be expected from the literature on women who have a family member with Down's syndrome (Carroll *et al.*, 2000; Felker, 1994; Marteau *et al.*, 1993).

Pregnant women generally hold more favourable attitudes towards abortion than those seen in general population studies. In a large sample study of pregnant women in the UK it was reported that two-thirds would consider abortion where there was a 'strong chance' that the baby would be 'handicapped' (Green *et al.*, 1993a). In the cross-European study discussed previously the views of nearly 400 pregnant women towards using abortion were assessed across a range of disabling conditions. Just over 60% of the women in Germany and the UK said they would terminate for Down's syndrome as did 80% of women in Portugal (Drake *et al.*, 1996). In line with the non-pregnant 'lay' sample in the same study, more women would consider termination for anencephaly than for Down's syndrome, but attitudes towards terminating for Down's syndrome were more favourable than for any of the other conditions. Pregnant women who have opted for diagnostic testing due to advanced maternal age also generally hold favourable attitudes towards termination for a pregnancy affected by Down's syndrome. Swedish researchers reported that prior to amniocentesis, 62% of their study sample had already decided they would terminate an affected pregnancy and a further 35% said they would consider it (Sjögren and Uddenberg, 1988). In a Canadian study of women undergoing amniocentesis 84% said they intended to terminate an affected pregnancy (Davies and Doran, 1982). Finally, 100% of 120 women undergoing diagnostic testing in one American study reported they would consider abortion for themselves where there was a strong chance of a 'serious birth defect' (Kolker, Burke, and Phillips, 1991). Variables associated with attitudes towards termination are the same as many of those noted in previous sections, i.e. religiousness, attitudes towards abortion generally, level of education, and country.

As noted previously, the role of attitudes towards the condition being tested for has not widely investigated in relation to testing choices. In particular, there has been little attempt to link attitudes towards Down's syndrome with prenatal screening decisions. Jørgenson (1995a) modelled over 15 likely influences on attitudes towards prenatal testing, however, no specific reference was made to knowledge or attitudes in relation to the condition being tested for. In a review chapter of the implications of prenatal diagnosis Marteau stressed the importance of providing information about the condition for informed choice, however, neither condition related knowledge or attitudes towards the condition were included in the list of factors that might influence testing choices (Marteau, 1991). In those studies attempting to statistically model the factors that might predict prenatal testing uptake, attitudes towards the condition have rarely been

included (Furr and Seger, 1998; Marteau *et al.*, 2001; Dormandy *et al.*, 2002a). However, in an early study by Marteau and colleagues (Marteau *et al.*, 1992a) the perceived burden attached to having an 'abnormal' baby was found to significantly predict serum screening uptake in an application of a number of psychological models including Subjective Expected Utility Theory (Edwards, 1961). This 'omission' might have occurred because attitudes towards Down's syndrome have not generally been considered as a distinct construct but rather as integral to attitudes towards testing. As noted previously, the concept of 'burden' is the exception to this rule but burden has seldom been viewed in the wider context of attitudes towards the target condition.

The findings of Bell and Stoneman (2000) discussed earlier suggest that those with more favourable attitudes towards Down's syndrome are less likely to intend to terminate for the condition in hypothetical scenarios, however the influence of these attitudes in real situations has been relatively unexplored. In an American study where attitudes towards prenatal testing, termination and Down's syndrome were examined in pregnant women from a diverse range of ethnic groups, the interaction of attitudes towards having a child with a disabling condition and attitudes towards prenatal testing technologies were shown to be somewhat complex (Moyer *et al.*, 1999). Overall, 60% of the sample held favourable attitudes towards the 'usefulness' of screening and diagnostic testing, however within this, European-American women were most likely and African-American women were least likely, to agree that testing was useful. Similarly, European-American women were most likely, and African-American women were least likely to say they would consider termination for Down's syndrome. Although the relationship between attitudes towards having an affected child and attitudes towards termination were not tested statistically, European-American women were most likely to say that having a child with Down's syndrome would be the 'worst possible outcome' for them (46%). None of the African-American women chose this response category, and the majority (84%) indicated that having a child with Down's syndrome would 'be difficult but I could adjust'. Of interest was that these two groups did not show any significant differences in the importance they attached to having an unaffected child; 73% of European-American women said that having a child without Down's syndrome was very important as did 74% of African-American women. The differences between the groups appeared to be in the willingness to take active steps to avoid having an affected child and in perceptions of ability to cope should the situation arise. Focus groups conducted with the women in this study revealed a wide range of beliefs about children with Down's syndrome; some felt the children were a strain and a burden to their families, others felt confident that they could cope with an affected child. The authors commented that some women held 'overly optimistic' views of the

effects of Down's syndrome and that some had never heard of the associated medical problems. It is of note that they did not comment that some women held overly pessimistic views.

Finally, a study by Press and colleagues investigated 'images and attitudes towards disability' along with willingness to terminate for a number of conditions (Press *et al.*, 1998). Researchers interviewed 140 pregnant women after the time they would have received their screening results (76% had had screening). It was noted that many women held quite positive attitudes towards people with disability but negative and 'fearful' attitudes towards having a child with a disabling condition themselves. However, only 13% of those who had screening said they would have definitely terminated following a diagnosis disability, and 40% said they would have definitely continued the pregnancy. Although the researchers did not specifically investigate views about Down's syndrome, they reported that most women spontaneously brought up the condition themselves and expressed opinions on it. They noted how Down's syndrome in particular was associated with ambivalence and conflicting beliefs. Positive images of the condition gained via television contrasted with personal views on the burdensome and stigmatising nature of learning difficulty. The authors commented that some women seem to have separated Down's syndrome out into a special category – not 'normal', but not quite 'disabled' either, and it was felt that views about Down's syndrome were somewhat 'romanticised'. Women were most concerned about having a child with a condition associated with severe learning difficulty, early death and quadriplegia, and were most likely to consider termination in these scenarios. However milder learning difficulty with an 'unusual appearance', was ranked tenth out of a list of seventeen conditions in terms of concern and ninth in terms of willingness to terminate. The report did not specify which category Down's syndrome was considered to fit into but noted that while health professionals associated Down's syndrome with severe learning difficulty most of the women they interviewed did not. Press and colleagues reflected on whose view would have persisted if a screening result had raised the issue of diagnostic testing and termination.

In summary, pregnant populations appear to use prenatal testing more readily and terminate more frequently than general population surveys would predict. The increased salience of the situation might make these choices a more accurate reflection of actual values than studies employing hypothetical scenarios, although it is unwise to draw definite conclusions about this because of the wide variance in the screening and diagnostic uptake rates reported. The dynamics of the testing process might be an important influence on testing behaviour as the action of offering a test carries the intrinsic message that the condition being tested for is serious enough to justify termination of pregnancy (Green and Statham, 1993). It has also been suggested that due to the shortage of time between receiving a screening result and the legal limit for abortion, a personal moral decision can

be recast as a 'medical emergency' in which there is little time for parental reflection (Press *et al.*, 1998). For this reason it is argued that it is inaccurate to assume that a woman's attitudes towards disability can be read from a decision about termination. In addition the method of service delivery appears to impact on choices, particularly in the case of screening tests, and it is suggested that method of service delivery might also inform attitudes towards testing. Further research in this area is warranted. The role of attitudes towards Down's syndrome in informing testing and termination choices has not been researched systematically, particularly in relation to screening tests, and there is a need to investigate the role of these attitudes to better understand influences on the choices that women make. The existing evidence suggests that the weakest relationship might be between attitudes towards Down's syndrome and attitudes towards using screening tests. Women offered diagnostic testing are already considered to be at higher risk of carrying a child with the target condition and these tests are associated with a risk of miscarriage and the risk of finding out unwelcome news during pregnancy. These factors might make values and attitudes towards Down's syndrome more salient when considering amniocentesis than when considering screening. The evidence suggests that most, if not all, women would prefer to have a child without a disabling condition. When anticipating decisions about terminating, women consider their current lives, their planned future, their existing relationships, and the potential relationship with their 'child-to-be' (Gilligan and Kitzinger, 1994). For many, a child with Down's syndrome does not fit well with this imagined future, yet there are also a substantial minority of women who appear willing to accept a child with Down's syndrome, and so decline prenatal testing.

2.2 SUMMARY

Studies which have examined attitudes towards Down's syndrome are few, of diverse methodology and populations, and some are 15 years old or more. It is not known which dimensions of these attitudes are also related to attitudes towards testing and termination for the condition. A number of factors appear to impact on the decisions women make in this situation but as yet, no psychological model has been shown to be a powerful predictor of test uptake. Prenatal tests are generally viewed positively by the general public and pregnant women, but conclusions about their value in terms of detecting abnormality are complicated by the different methods of testing available and the possible influences of 'normal' testing procedures in different antenatal clinics. Information provision about Down's syndrome has been rarely studied. Some women would like more information about the condition but the impact of 'missing' information on test uptake or decision-making is unknown. To date, research has focused mainly on information about the tests and testing procedure, rather than on information about the conditions being tested for. For this reason very little is known of the knowledge and understandings of these conditions that

women bring with them into the antenatal environment. The following section defines the aims and objectives of the thesis and outlines the research conducted in order to meet them.

2.3 RESEARCH QUESTIONS AND THESIS OVERVIEW

This thesis is concerned with understandings of Down's syndrome and their place within the prenatal testing context, including information provision, and beliefs and attitudes about the condition. Studies that have considered women's understandings of Down's syndrome have suffered from a lack of consensus about what information is needed and an overly medical focus. As a result little is known about how knowledge or attitudes towards Down's syndrome influence prenatal testing choices. The overall aim of the research presented in this thesis is to increase knowledge, inform further research, and contribute to the debate on informed choice. It is also hoped that the findings will contribute to the development of a sensitive prenatal testing service that respects the values of the individual woman thereby supporting the choices of all women.

2.3.1 Research questions

The research questions addressed by this thesis were driven by the gaps in the literature identified in the previous chapter. In particular the issues of interest were information and knowledge about Down's syndrome and attitudes towards the condition in relation to prenatal testing choices. The main research questions addressed by this thesis are:

1. What information do women receive about Down's syndrome to help inform their prenatal testing choices?
2. What understandings of Down's syndrome exist independently of the prenatal testing context?
3. What is the relationship between understandings of Down's syndrome and intentions towards personal use of prenatal testing and termination for the condition?
4. What role do understandings of Down's syndrome play in pregnant women's actual testing choices?

2.3.2 Overview of the research presented within this thesis

Research Question 1: Chapter 3. Descriptive information about Down syndrome: a content analysis of serum screening leaflets

The literature review revealed no published study that had analysed the information about Down's syndrome given to pregnant women prior to the offer of prenatal testing. The study presented in Chapter 3 took an existing collection of serum screening leaflets aimed at pregnant women, and described and critically evaluated the information about Down's syndrome that they contained in two ways. Firstly, a content analysis by information category was conducted to provide an overall picture of the type of information provided. Secondly, a more qualitative analysis of this content

was conducted to assess the comparative balance of positive, negative, and neutral information. The results of this second analysis were compared and contrasted with the findings of a similar study concerned with information from cystic fibrosis screening leaflets (Loeben *et al.*, 1998).

Research Questions 2 and 3. Chapter 4: Understandings of Down's syndrome: a Q-methodological investigation

The dominant discourse about Down's syndrome in the prenatal testing literature is situated within the medical model of disability. However, the literature as reviewed in Chapter 1 suggests that a number of other understandings of the condition exist within our society. The primary aim of this study was to access these diverse understandings of Down's syndrome and to identify where understandings are shared and where they are distinct. The secondary aim was to identify relationships between the understandings of Down's syndrome and attitudes towards the use of prenatal testing and termination for the condition. Q methodology (Stephenson, 1953) was the approach selected, as it is particularly suited to the investigation and modelling of complex views. Individuals from a diverse range of experiences and backgrounds were recruited, and their understandings of Down's syndrome extracted and characterised using factor analytic techniques.

Research Questions 3 and 4: Chapters 5, 6, and 7. Attitudes towards Down's syndrome in the prenatal testing situation

The results of the Q methodological study suggested that while understandings of Down's syndrome were related to intentions to use prenatal testing and termination, the relationships were not necessarily straightforward. The aim of this final study was to measure attitudes towards Down's syndrome in pregnant women and then to relate these attitudes to actual testing choices. Over a six-month period, women in the first trimester of pregnancy were asked to complete a questionnaire that incorporated open-ended measures of cognitive, emotional, and experiential aspects of attitudes towards Down's syndrome. An objective measure of serum screening uptake was then collected at a later date from patient records. Multivariate statistics were employed to assess the contribution of attitudes toward Down's syndrome in predicting prenatal screening choices and intentions regarding diagnostic testing and termination of pregnancy.

The final chapter of this thesis (Chapter 8) discusses the findings of the three studies in relation to the research questions and the issue of informed choice. The limitations of the studies are discussed there and suggestions for future research considered.

CHAPTER 3 DESCRIPTIVE INFORMATION ABOUT DOWN'S SYNDROME: A CONTENT ANALYSIS OF SERUM SCREENING LEAFLETS

3.1 INTRODUCTION

As noted in Chapter 2, it is recommended practice that prior to prenatal screening, parents should be provided with information about the target condition. Such information is considered necessary to facilitate autonomous and informed decision-making. Pre-screening information is usually disseminated via a combination of face-to-face contact (for example, with the midwife during the booking appointment) and an information leaflet, although some clinics also use educational videos (Fairgrieve, Magnay, White, and Burn, 1997a). Research in the area of informed choice has focused mainly on the information provided about the tests and procedures and little attention has been given to information about the target condition(s). One study that has specifically addressed this issue examined the presentation of information about cystic fibrosis in leaflets about carrier screening for the condition (Loeben *et al.*, 1998). Cystic fibrosis (CF) is a recessive genetic disorder²⁵ with an incidence of around one in 2400 births in the UK and a carrier prevalence of one in 24 of the general population (Murray and Cuckle, 2001). For a baby to be born with CF both parents must be a carrier of the faulty gene, therefore, unlike screening for Down's syndrome it is the parents who are initially tested rather than the pregnancy. This means screening for CF can be done prenatally or in a non-pregnant population. It is estimated that in one in every 600 couples in the UK both partner are carriers of CF (Murray and Cuckle, 2001). Screening for CF is offered as a routine part of care in a few NHS antenatal clinics or via private screening providers.

The study by Loeben and colleagues (1998) analysed the content of 28 leaflets about CF carrier screening that were obtained from obstetric providers in North America ($n=19$) and the UK ($n=9$). They found the amount of descriptive information provided about CF varied substantially across leaflets, ranging from one to 37 sentences per leaflet (median number 6.5). Most sentences were judged to be neutral in tone (65%), with 19% of sentences being considered positive, and 16% negative in tone. There was no significant difference in the length of the leaflets by country but positive statements about CF were more common in US than UK leaflets. Differences in the portrayal of CF were noted depending on the purpose of the leaflet, with positive sentences found

²⁵ A recessive (faulty) gene carries a trait that remains latent if there is a dominant 'healthy' gene at the same place on the matching chromosome. If genes on both chromosomes are recessive for the same trait, the trait (i.e. CF) is seen in the individual (Anderson, Anderson, and Glanze, 1998). In a pregnancy where both parents are carriers of the recessive gene, each fetus has a 25% chance of inheriting both recessive genes and so having CF, a 50% chance of being a carrier, and a 25% chance of not inheriting either recessive gene.

less frequently in prenatal leaflets than in population screening material aimed at non-pregnant groups. The authors suggested that this might reflect the beliefs of those involved in prenatal screening programmes as to the appropriateness of termination for the condition. An earlier commentary also observed that information about CF and Down's syndrome differed in tone depending on whether the material was intended for prospective parents or for parents who had already given birth to an affected child. The prenatal material was mostly negative in tone and focused on medical problems and limitations imposed by the condition, whereas the post-natal material was more positive, emphasising medical and social advances and some compensating aspects of the condition (Lippman and Wilfond, 1992).

It is not known to what extent information about the target condition affects prenatal screening behaviour, but there is some evidence suggesting that leaflets may influence intentions to test and terminate for Down's syndrome. A study in the UK evaluated the effects of textual information about Down's syndrome and photographs of children with the condition on expectations to test and terminate in a large sample of psychology undergraduates (N=814) (Figueiras, Price, and Marteau, 1999). Text, whether positive, negative or neutral, had no effects on the perceived severity of the condition, perceived likelihood of having an affected pregnancy, concern about having a child with Down's syndrome, or prenatal testing intentions; however negative text was associated with a greater intention to terminate. Most significant were the effects of the photographs on behavioural intentions. A photograph of a child with Down's syndrome, whether positive or negative (as judged by the researchers), increased concern about having an affected child and expectations of undergoing prenatal testing and termination compared with no photograph. In addition the negative photograph was associated with the most favourable intentions towards termination for Down's syndrome. The authors suggested that photographs might access different mental schemas than textual information and be more successful at eliciting pre-existing schemas of parenting a child with Down's syndrome. However, studies comparing the effects of videos and leaflets on women's screening decisions have found no significant effects of video on actual test uptake (Browner *et al.*, 1996; Hewison *et al.*, 2001; Michie *et al.*, 1997b). This suggests that the relationships between photographic representation of Down's syndrome and screening decisions are not straightforward even though visual information might be more memorable.

Despite prenatal screening for Down's syndrome being so widely available, no published study has analysed the descriptive information about the condition that is given to women in leaflet form prior to screening tests. The aim of this study was to describe and critically evaluate information

about Down's syndrome provided by such leaflets in the UK and to compare and contrast the findings with those reported by Loeben *et al.* (1998) in their study of CF screening leaflets.

3.2 METHOD

3.2.1 Study design

A cross-sectional survey of the descriptive information about Down's syndrome in prenatal serum screening leaflets was conducted. The independent variables were 1) the type of descriptive information about Down's syndrome, for example, information relating to health problems, and 2) the tone of this information (positive, negative, or neutral). The dependent variables measured were the number of sentences about Down's syndrome, the frequency of each type of information, and the frequency of sentences classed as positive, negative, or neutral.

3.2.2 Materials

The leaflets used in this analysis had originally been collected for a different study²⁶ that analysed the quality of information about screening tests for Down's syndrome in terms of factual content, accuracy, presentation, and reading ease (Murray *et al.*, 2001). This previous study had only considered the target condition in terms of the absence or presence of information about medical aspects of Down's syndrome. The collection of leaflets were re-visited originally as a source of material for the study using Q methodology (see Chapter 4), however, after the data were extracted it was believed that a detailed content analysis of the information provided about Down's syndrome would be a useful contribution to knowledge in this area.

The leaflets had originally been collected from laboratories enrolled with the United Kingdom National External Quality Assurance Scheme (NEQAS²⁷) who were asked to send in leaflets from each NHS maternity unit to which they provided biochemical tests. Out of 78 laboratories enrolled with NEQAS, 64 laboratories (82%) serving 149 maternity units returned 81 leaflets. A further two leaflets from private screening services were obtained. All the leaflets were intended for distribution to pregnant women and many were used in more than one maternity unit. Any leaflet that was specifically associated with diagnostic testing was excluded as screening information might have preceded it. A total of 80 leaflets formed the final sample.

²⁶ Not part of this PhD. The leaflets are held at Reproductive Epidemiology, University of Leeds.

²⁷ NEQAS offers a quality control service for biochemical analysis and risk calculation to antenatal care centres in the UK.

3.2.3 Content analysis

Content analysis can be broadly defined as “*any technique for making inferences by objectively and systematically identifying specific characteristics of messages*” (Holsti, 1969, p. 14). In this study content analysis was carried out to investigate the type of information that was provided about Down’s syndrome and the message that this information communicated. The analysis procedure was informed by a number of texts on content analysis that are suitable for research within the social sciences (Holsti, 1969; Krippendorff, 1980; Shaughnessy and Zechmeister, 1994). The leaflets were examined for any information that described Down’s syndrome.

Sentences related to the testing process or to maternal-age related risks were excluded, as this information is not descriptive of Down’s syndrome itself. Two types of content analyses were conducted and two units of analysis were employed. For the analysis by information type the unit of analysis was a statement or separate piece of information, and for the qualitative analysis of the message conveyed the unit of analysis was a full sentence. Thus it was possible for one sentence to contain several statements related to different aspects of Down’s syndrome but be judged to convey a positive, negative, or neutral message overall. Each sentence was transcribed into a coding frame, first in its entirety so that the overall message could be analysed and then broken down into component parts so that frequencies of information by type could be calculated (see Figure 3.1). The analysis was conducted in March 1999

Figure 3.1. Sample coding frame

Id*	Sentence	Code	Statements	Code
1.1	Down’s syndrome is the most common cause of severe learning disabilities and babies may also have heart defects.		Severe learning disabilities/ Babies may also have heart defects	
2.1	Down’s syndrome is caused by an extra copy of a chromosome and occurs in about 1 in 700 births.		Caused by an extra copy of a chromosome/ Occurs in about 1 in 700 births/	
2.2	Although they have varying degrees of mental disability the trend is to educate children in mainstream education.		Varying degrees of/ Mental disability/ The trend is to educate children in mainstream education/	

* Leaflet identifier and sentence number

Content analysis by information type: procedure

Each statement was coded as belonging to a mutually exclusive category based on the type of information it provided (see Table 3.1). It is recommended practice that researchers apply pre-existing categories where possible so that the findings can contribute to existing knowledge rather than 'reinvent the wheel' (Krippendorff, 1980). For this reason initial categories were based on the types of information about Down's syndrome as outlined by Noble (1998) as being important in the antenatal context. These were birth incidence, chromosomal origin, learning difficulty, medical problems, and life expectancy. However, other categories were created after initial coding revealed a wider range of material in the leaflets. For example, statements about the facial features of Down's syndrome were coded as 'Physical Appearance' as they did not fit any of the previously defined categories.

Table 3.1. Information categories with sample statements

Information Category	Sample Statement
Birth incidence	About 1 in 700 babies are born with Down's syndrome
Chromosomal or genetic origins	Down's syndrome is a genetic condition caused by the presence of an extra chromosome 21
Education and development issues	Most people with Down's syndrome will need special help with their education
Inability to predict severity before birth	There is no way to predict how serious any of the disabilities will be
Learning difficulty	The effect of this extra copy is mainly mental handicap
Life expectancy	Approximately 25% of children born with Down syndrome will not survive longer than 10 years
Medical problems	30% may develop some form of thyroid disease
Non-availability of treatment	It is not a disease and it cannot be treated
Physical appearance	The eyes are upslanting and the face is rather flat
Psychosocial/emotional aspects for affected person	Many of them are nevertheless happy children
Psychosocial/emotional aspects of parenting	Some parents find their experience is not what they hoped for but it is still positive
Social factors (independence, employment)	Generally as they grow older they will always require supported help and accommodation
Variation in disability or ability	The ability of adults with Down's syndrome varies considerably

Qualitative analysis of the message conveyed: procedure

The extracted full sentences were classified as positive, negative, or neutral in terms of the tone of message they conveyed. As in the study by Loeben and colleagues (1998), the classifications of positive and negative were used to “*capture both the content of the sentence and the sentence’s tone or ‘slant’*” (p. 1182). For example, a sentence could be classed as negative either because it contained information about a negative aspect of Down’s syndrome such as the prevalence of heart defects, or because it framed information about the condition in a negative way, such as emphasizing infant mortality rates rather than survival rates.

To allow a direct comparison to be made between the Down’s syndrome leaflets and the CF leaflets study, classification criteria were selected to be as close as possible to those used by Loeben *et al.*, (1998). Sentences classed as ‘negative’ focused on one or more of the following; (1) the clinical complications associated with the condition, (2) the developmental problems, (3) the reduced life expectancy, (4) the reduced quality of life of the affected person, (5) that there is no treatment for Down’s syndrome, or (6) stigmatising descriptions of Down’s syndrome. The sixth category was added retrospectively having observed the frequency with which leaflets drew attention to the morphological characteristics of Down’s syndrome in a negative manner.

Sentences classified as positive were those that focused on (1) the fact that treatments for the clinical complications are improving, (2) educational support and outcomes are improving, (3) that life expectancy is improving, or (4) that people with Down’s syndrome can participate in important life activities. Where sentences did not obviously fit the negative or positive criteria or were a combination of both, they were classed as ‘neutral’. Table 3.2 gives a sample of sentences and their classifications. As in the CF study, sentences that focused on birth prevalence or the genetics of Down’s syndrome were excluded from this analysis because they were not considered descriptive of the condition.

Across all sentences, an initial inter-rater reliability of 80% (130 out of 162) was obtained between the author and two other researchers²⁸. This was comparable with an inter-rater reliability of 81% between the two raters in the CF comparison study (Loeben *et al.*, 1998). Discrepancies between ratings were discussed and a consensus decision reached on the disputed items.

²⁸ Jenni Murray and Indera Sehmi see (Bryant *et al.*, 2001)

Table 3.2. Sample sentences about Down's syndrome: neutral, positive, and negative

Neutral	Positive	Negative
Most babies will sit between 6-30 months, walk at 1-4 years and be toilet trained by 2-7 years.	This means that many children with Down's syndrome will accomplish more than ever before.	Down's syndrome is the single most common cause of severe mental handicap.
Some will die very young but many will have a normal length of life.	Many babies with Down's syndrome will survive into old age.	Some babies are affected by serious deformities that may ultimately be fatal.
Each person with Down's syndrome is different.	If they have heart disease it can often be treated.	About 40% are born with heart problems and of these 20% will require some form of heart surgery.

Of five disagreements between positive and neutral classifications, all sentences were finally classified as neutral. Of twenty-five disagreements between negative and neutral classifications, twenty-four sentences were finally classified as neutral and one as negative. These sentences were mainly those that included reference to learning difficulty but did not give any indication of the range. It was decided to class as negative those that mentioned only the 'serious' or 'severe' end of the intellectual spectrum. For example, the sentence, *"People born with Down's syndrome have learning disabilities"* was classified as neutral, whereas the sentence *"Down's syndrome is the most common cause of severe mental handicap"* was classified as negative. Two sentences were originally classified as positive by one of the raters and negative by the other two. After discussion, both were finally classified as negative. The first sentence, *"Many of them are nevertheless happy children"* was considered to stigmatise children with Down's syndrome by classifying the group as 'them', and use of the word 'nevertheless' was felt to negatively frame the overall sentence content. It was agreed that the second disputed sentence, *"Most people with Down's syndrome will live to 40-60 years of age and the great majority will require some form of help and support throughout their lives"*, emphasised disability and the perceived long-term dependence of people with Down's syndrome. The initial disagreement over the classification of these last two sentences highlights the difficulties inherent in this type of analysis, particularly where an item has inter-dependency with preceding or following sentences. It was not believed

that this compromised the overall integrity of the analysis however, given that such disagreements were only encountered twice in 162 instances.

3.3 RESULTS

3.3.1 Content analysis by information type

The number of pages of information about prenatal testing ranged from one to eighteen, with a median of three pages. In contrast to the findings of the CF study where all leaflets contained at least one sentence of information about the target condition, 21% of the leaflets in this study ($n = 17$) gave no information about Down's syndrome of any sort. Of these, one leaflet gave no descriptive information and failed to name Down's syndrome as the condition being screened for. In total, 307 descriptive statements about Down's syndrome were extracted (297 sentences about CF were extracted from 28 leaflets in the comparison study). The number of sentences describing Down's syndrome ranged from zero to twenty-four with a median of one per leaflet (the median number of sentences in the CF study was 6.5). Table 3.3. summarizes the analysis by giving the absolute frequencies of statements and the proportion of leaflets carrying information of each type.

The table shows that 89% of the statements were medical, clinical or epidemiological in nature (categories 1-7, 10 and 11). Eleven per cent referred to social, educational or psychosocial issues (categories 8 and 9, 12 and 13). The statements in these last four categories all came from the same sixteen leaflets (20% of the sample). In contrast the previous analysis of this sample of leaflets had shown that 79% reported some 'medical facts' about Down's syndrome (Murray *et al.*, 2001). In thirteen percent of the leaflets ($n = 10$), the only piece of information about Down's syndrome was that it was an abnormality or defect of genetic or chromosomal origin. Many items of information were rather vague. For example, people with Down's syndrome were described as having learning difficulty and other 'birth defects', 'special needs', 'physical handicaps', 'serious deformities', and 'medical problems'. No detail was provided as to what these other factors might entail. Seven of the leaflets contained the colloquial terms 'Mongolism' or 'Mongol' (for example, "*children with Down's syndrome are sometimes called Mongols*"). Four leaflets provided some contact details for the UK Down's Syndrome Association.

Table 3.3. Content analysis of information about Down's syndrome by information category in descending order of statement frequency.

	Information Category	Statements	Statements	Leaflets	Leaflets
		(n)	(%)	(n)	(%)
1	Medical problems	63	20.5	33	41
2	Learning difficulty	61	20	53	66
3	Chromosomal or genetic origins	56	18	50	63
4	Birth incidence	23	7.5	23	29
5	Physical appearance	22	7	19	24
6	Variation in disability or ability	17	5.5	15	19
7	Life expectancy	15	5	13	16
8	Education and development	15	5	10	13
9	Social factors	12	4	10	13
10	Inability to predict severity	11	3.5	11	14
11	Non-availability of treatment	6	2	6	8
12	Psychosocial/emotional aspects of parenting	4	1.5	4	5
13	Psychosocial/emotional aspects for affected person	2	0.5	2	3
Total (statements)		307	100		

3.3.2 Analysis of the message conveyed

In this analysis, information that referred only to the birth incidence or chromosomal origins of Down's syndrome was excluded. This reduced the number of leaflets in the analysis from 80 to 53 (17 leaflets had contained no information at all and 10 contained only chromosomal information). These 53 leaflets contained 162 sentences of descriptive information about Down's syndrome, had a median length of four pages, and a median number of two sentences of description per leaflet. Of the total number of sentences, 103 were classed as negative (63%), 40 as neutral (25%), and 19 as positive (12%). This contrasts noticeably with the proportions in the CF study where 65% of sentences were considered to be neutral, 19% positive and only 16% negative. More specifically, median values for the different classifications are provided in Table 3.4 with corresponding figures shown for the *prenatal* CF leaflets only ($n=10$) in the study by Loeben *et al.* (1998). While these figures have been included to allow comparison across the studies, the fact that two out of three

median values for the Down's syndrome leaflets were zero, reduces the usefulness of these statistics.

As the leaflet increased in length (defined as the number of pages), so did the number of descriptive sentences about Down's syndrome (Spearman's $\rho = +0.42$, $p < 0.05$). For this reason median ratios²⁹ were also calculated to control for the contributing relationship between number of classified sentences and the length of the leaflets as in the CF study.

Table 3.4. Number of descriptive sentences about Down's syndrome (DS) and cystic fibrosis (CF) in prenatal screening leaflets classified as neutral, positive, and negative. Values for CF leaflets taken from Loeben *et al.* (1998)

Classification	DS sentences:	CF sentences:	DS: median ratio	CF: median
	Median (range)	Median (range)	to total no. of sentences (range)	ratio to total no. of sentences (range)
Neutral	0 (0 - 4)	4.5 (1 - 15)	0 (0 - 1)	0.7 (0.2 - 1)
Positive	0 (0 - 3)	0.5 (0 - 3)	0 (0 - 0)	0.1 (0 - 0.3)
Negative	1.0 (0 - 19)	2.0 (0 - 3)	0.7 (0 - 1)	0.2 (0 - 0.6)

Note: Number of DS leaflets = 53, number of CF leaflets = 10

The numbers of negative, positive, and neutral sentences all correlated significantly with the total number of sentences that described Down's syndrome but to differing degrees (Spearman's $\rho = +0.8$ (negative sentences) and $+0.6$ (positive sentences), $p < 0.0001$, and $+0.3$ (neutral sentences) $p < 0.05$). Therefore, while leaflets with more description about Down's syndrome were also more likely to contain some positive and neutral sentences, most significantly, as description size increased the message conveyed became more negative in tone. In most cases more description typically equated to more details about medical problems or other clinical information. For example, the leaflet with most description about Down's syndrome contained nineteen negative, one positive, and four neutral sentences. Within these sentences, 46% of the statements provided information about the increased likelihood of certain medical conditions, and 17% focused specifically on the appearance of people with Down's syndrome.

²⁹ For each leaflet, the ratio of neutral, positive, or negative sentences to the total number of sentences was calculated and then the median ratio identified.

The study by Loeben *et al.* (1998) found that the leaflets provided by commercial screening services contained fewer positive sentences than those provided by non-commercial ones. However, it was not possible to do a similar comparison in this study as there were only two leaflets from private screening services. Both these leaflets were longer than the average (18 pages and five pages), but the first contained ten sentences about Down's syndrome (including two positive ones) while the other provided no information about the condition other than its chromosomal origins.

3.4 DISCUSSION OF THE FINDINGS

A major finding of this study was that 21% of the leaflets ($n=17$) contained no descriptive information about Down's syndrome and a further 12% ($n=10$) only reported that Down's syndrome is a chromosomal or genetic abnormality of some kind. There are a number of possible explanations for this lack of information. Firstly, constraints of leaflet size may mean that providing descriptions about the condition(s) being tested for is just not feasible. However, some single page leaflets included information about Down's syndrome, while other more lengthy leaflets did not. Pragmatic reasons alone, therefore, cannot sufficiently explain the lack of information about Down's syndrome. Secondly, health professionals may be concerned that too much information at the screening stage causes unnecessary anxiety in pregnant women (Brunger and Lippman, 1995; Oliver *et al.*, 1996) and that it detracts from pregnancy as a normal event (Alderson *et al.*, 2001). However, exactly what constitutes too much information has never been quantified, and the view that leaving out information reduces anxiety lacks empirical support (Ley, 1988). On the contrary, research suggests that providing information about the potentially bad outcomes of a clinical procedure does not significantly increase patient anxiety and actually facilitates informed decision making (Kerrigan *et al.*, 1993).

A third explanation is that information about the condition is considered unnecessary because women already have a sufficient knowledge of Down's syndrome. One leaflet's descriptive information about Down's syndrome consisted of the statement, "*Most people know something about Down's syndrome (Mongolism)..... it causes mental handicap and other problems*". One third of the leaflets described Down's syndrome as the most "common" form of learning difficulty or chromosomal abnormality. One quarter included statements that referred to the distinctive facial characteristics of people with Down's syndrome. It is argued that such material is intended to remind women about Down's syndrome rather than to inform them. It is also likely that the non-medical terms 'Mongol' and 'Mongolism' may also have been included in some leaflets to aid recall. However, as 'Down's syndrome' is itself so widely recognised, the continued usage of these colloquial terms may serve mainly to perpetuate outdated stereotypes of the condition. In the

CF study, all of the leaflets contained at least one sentence of descriptive information about the condition and the amount of description provided was generally greater than in the leaflets analysed in this study. This suggests that providing information about CF is considered more important than providing information about Down's syndrome, perhaps because CF is a less visible disability so prior knowledge is not assumed, or because there is no equivalent stereotypical image of people with CF. In a survey of public attitudes towards termination for a number of conditions including CF and Down's syndrome, descriptions or indications of the severity of these conditions were given with the exception of Down's syndrome (Marteau, Michie, Drake, and Bobrow, 1995). However, the assumption of existing knowledge about Down's syndrome may be misconceived. The research reviewed in Chapter 2 showed that many women feel that they have little real knowledge of either its effects or of affected persons (Moyer *et al.*, 1999; Gekas *et al.*, 1999; Carroll *et al.*, 2000; Al-Jader *et al.*, 2000). The implications that this holds for informed decision-making have yet to be explored, however, simply increasing the amount of information provided will not ensure informed choice unless careful consideration is given to what information is provided and how.

As in the CF study, the amount and type of information provided about Down's syndrome varied greatly and the proportion of sentences classed as positive was very small. Unlike the CF study, the majority of the descriptive information about Down's syndrome was judged to be negative in content and tone. This difference could be due to the subjective bias of the individuals who rated the statements, although, the use of the same criteria as the comparison study and the satisfactory inter-rater reliability suggests otherwise. Alternatively, the information may be more negative in tone because Down's syndrome is a condition with more negative features than CF. With reference to physical health, this is clearly not the case, as around half of all babies with Down's syndrome are born with no serious health problems and have a longer life expectancy than do infants with CF. Unlike CF, however, Down's syndrome is primarily associated with learning difficulty. Health professionals appear to view abortion for Down's syndrome more favourably than they do for CF, suggesting that learning difficulty is perceived as a more appropriate reason for termination than chronic ill health alone (Green, 1995b; Drake *et al.*, 1996).

The research by Figueiras and colleagues (1999) suggests that unfavourable textual information about Down's syndrome can be associated with a greater expectation to terminate for the condition, and so it is possible that such information could impact on actual testing and termination decisions in the clinical context. Negative information, whether presented via photograph or text had a greater influence on testing and termination intentions than did positive or neutral information. It is known that negative information has a greater impact on overall

evaluations than does positive information, and that people seem to have a better memory for negative items of information (Ajzen, 2001). This is of concern, considering the bias towards negative information in the screening leaflets analysed. However, there is also evidence that attitudes towards testing are resistant to change and that once an initial decision has been made it is not altered by providing additional information (Bernhardt *et al.*, 1998). This suggests that prior attitudes towards Down's syndrome might be more important in terms of testing choices than information provided by leaflets.

A recent systematic review of the informed decision making literature, concluded "*information and education are relatively ineffective ways of facilitating informed decision making, compared with the context and social influences*" (Bekker *et al.*, 1999, p. iv). This conclusion has been supported by two related studies that were conducted to evaluate the impact of evidence based leaflets on choices in maternity care (O'Cathain *et al.*, 2002; Stapleton, Kirkham, and Thomas, 2002). The first of these studies (O'Cathain *et al.*, 2002) evaluated the effects of ten evidence based MIDIRS (Midwives Information and Resource Service) leaflets, including one on screening for Down's syndrome on levels of informed decision making in antenatal and postnatal care. A cluster trial design was employed using a leaflets/no leaflets condition, with the leaflets being randomized to groups of maternity units. The researchers found no significant differences in self-reported informed choice levels, or levels of knowledge between groups, although there was a significant increase in satisfaction with information in the antenatal setting. The authors concluded that evidence based leaflets were not effective in promoting informed choice. In the second study Stapleton and colleagues (2002) interviewed women and health professionals at the intervention sites of the cluster trial about their views on the MIDIRS leaflets. They also observed antenatal consultation sessions. They found that the leaflets were frequently 'invisible' in that they had not been distributed as planned, or were hidden amongst a mass of other information and advertising material. Those women who received the leaflets felt that they did not get time for discussion with the midwives about issues that concerned them. The midwives reported time pressures and using leaflets as an alternative to discussion, and it was observed that the midwives often did not refer to the leaflets during consultations. The authors argue that the findings of O'Cathain *et al.* (2002) must not be considered in isolation, or conclusions drawn about the effectiveness of evidence based information leaflets. They highlight the difficulties of empowering freedom of choice in a culture that supports normative patterns, due in part to time pressures imposed on staff.

While it may appear that information leaflets have little direct influence on actual testing choices, the reasons for this apparent ineffectiveness remain to be researched fully. However, it is known that information is still highly valued in its own right by pregnant women (Green *et al.*, 2002). In

addition, a study of patients' expectations of genetic counselling reported that while 30% came wanting help with making decisions, 79% came expecting information (Michie *et al.*, 1997a). For this reason, it is important to provide accurate, balanced information about Down's syndrome at the earliest stage in the testing process it could be found useful – but how? In order to achieve a balanced portrayal of a condition, Loeben *et al.*, (1998) proposed that descriptive information should include “*sufficient positive statements to achieve balance with the neutral and negative ones*” (p. 1187). While it is relatively straightforward to define negative and positive information, defining information as neutral is more problematic. To a pregnant woman being offered a test for Down's syndrome, the information that 40% of affected children will have a heart defect is likely to be viewed negatively. Therefore, this ‘fact’ cannot be considered neutral in its applied context. The potential of information to generate an emotional response should be taken into consideration before providing parents with a long list of the medical problems associated with Down's syndrome. Much essential information could be presented in a way that not only remains factual but also gives a more accurate representation of the situation. For example, the following description balances the frequency and potential seriousness of heart defects with a more positive statement on the outlook for children with Down's syndrome.

“40% of children with Down's syndrome are born with heart defects, which can be serious and require surgery. However treatment for these conditions continues to advance and the degree to which they are-life-threatening or limit achievement and well-being is reducing” (Down's Syndrome Association, 1996).

To structure information in this way requires more thought and perhaps longer leaflets, but it is argued that both the time and the space are warranted if informed choices are to be supported.

In addition to balance in the *tone* of information about Down's syndrome, attention must also be paid to balancing the *types* of information provided. Potential parents should of course understand the possible health outlook for their children, and Down's syndrome is associated with certain characteristics and conditions that make it of interest to clinicians. However, Down's syndrome is as much an educational and social ‘condition’ as a medical one. While genetic counsellors apparently consider diagnostic and prognostic information about a condition as most important, their clients rate knowledge about how a child with the condition may affect family functioning more highly (Michie *et al.*, 1997b; Michie *et al.*, 1997a). In one study, pregnant women reported a lack of understanding about what living with a child with Down's syndrome might be like, and some remarked that this type of information was not made available in pre-test material (Moyer *et al.*, 1999). The screening leaflets analysed here also mostly failed to provide this type of information or suggest where it could be obtained.

The bias towards medical information in the leaflets may well be a better reflection of the knowledge and perspectives of the professionals writing them than of the needs of the women receiving them. Is, for example, the chromosomal origin of Down's syndrome so frequently included in leaflets because it is useful information for parents, or because it has become a default feature of the clinical description of the condition? This raises the question about who is best qualified to provide information about Down's syndrome to prospective parents. There is no dispute that health professionals who have accurate and up-to-date knowledge of the procedures must inform women about screening tests. However, these same individuals may not be most suited to providing accurate and up-to-date information about Down's syndrome, especially on issues such as early intervention programmes, family adjustment, or the opportunities available for an inclusive lifestyle. Prenatal testing might be delivered within a clinical context, but both the condition itself and prenatal testing have social and psychological implications as well as medical ones. One midwife, who was also the mother of a child with Down's syndrome noted, "*as midwives we know all the basic facts of Down's syndrome but we know little beyond the first 10 days*" (Davies, 2000, p.432). Studies indicate that between 30 and 40% of midwives involved in prenatal testing do not feel confident to give pre-test information about Down's syndrome (Fairgrieve *et al.*, 1997b; Khalid *et al.*, 1994). It has been suggested elsewhere that parent support groups, or Down's syndrome associations are more appropriate sources of information (Birke, Himmelweit, and Vines, 1990), and Carr (1995) has argued that there is a need for some professionals to put

"aside their armchair convictions about the burdens that they intuitively know are carried by families with a disabled member, and to give credence to findings, from properly conducted research, based on the views of the parents themselves" (Carr, 1995, p. 73).

Nevertheless, parents and the organisations that represent them also have a particular viewpoint that cannot be considered completely impartial. As Lippman and Wilfond (1992) point out, the 'storyteller' inevitably shapes summary information of this kind, as he or she must decide what to include and what to exclude from the vast amount of material available.

There is currently a need to improve on the general quality of information provided about serum screening for Down's syndrome³⁰. The findings from this study show that the information about Down's syndrome provided in such leaflets is especially in need of attention. In particular, the information needs to be more balanced in its construction, with thought given to the needs of the

³⁰ Since this thesis was begun, the Down Antenatal Information Screening and Implementation Group (a working party of the National Screening Committee's antenatal subgroup) have been developing a leaflet about screening for Down's syndrome that is intended to be provided when the new policy of screening for all women is implemented in 2004.

reader, and to the tone and the content of the message conveyed. This is not a simple task and it would be highly desirable for some objective and definitive guidelines to be produced. The dominant discourse about testing and termination for Down's syndrome portrays a rather negative and overly medical image of the condition, therefore, it is unlikely that all individual beliefs regarding Down's syndrome can be equally supported and valued. It is recommended that those responsible for commissioning or writing prenatal screening leaflets consider the following points. First, what information about Down's syndrome is most useful to those making prenatal testing choices? For example, parents appear to value information on the functioning of families with an affected child. Such material is readily available from the disability literature or from other reliable sources such as the Down's Syndrome Association of the UK and the Down Syndrome Educational Trust. Second, how should this information be structured to provide balance of tone and content? Authors should actively consider the tone of the message they are conveying and aim for a combination of *naturally* positive, negative, and neutral statements about the condition that accurately reflects its complexity. A content analysis of the kind conducted here might help assess whether the 'finished product' delivers balanced information. Leaflets should include information on social, educational, and developmental aspects of Down's syndrome as well as medical details. Third, which person(s) are best qualified to provide this information? A willingness to recognize a wider range of both professional and lay expertise is needed if information about Down's syndrome is to provide an accurate and up-to-date portrayal of the condition. Such expertise may include the Down's syndrome associations and groups that represent the views of people with Down's syndrome themselves, such as 'Down 2 Earth'³¹.

3.4.1 Further research

A number of other possible avenues for research are suggested. Firstly, the focus of this study was limited to information provided via leaflets only and so might not be a fair reflection of the information provided by health professionals. It would therefore be useful to conduct a similar type of detailed content analysis on the narrative information given during pre-screening counselling sessions with midwives, obstetricians, or GPs. This, in combination with the current study would give a clearer picture of the information women received about Down's syndrome prior to making their screening choices. It might also shed some light on the way women respond to information about the condition and whether they appear to welcome this information, understand it, or question it further. Secondly, although the focus of this study was on Down's

³¹ Down 2 Earth is a group of adults with Down's syndrome who, in association with the Down's Syndrome Association and the Nuffield Foundation, have produced an information pack and video about having the condition, mainly aimed at informing other people with Down's syndrome.

syndrome, serum screening can also test for neural tube defects (NTDs) and trisomy 18. It was noted during the course of data extraction that there was often very little information about NTDs and even less about trisomy 18. It is therefore suggested that a content analysis of the material about these conditions be conducted. This analysis could be compared and contrasted with the study presented here and with the study by Loeben *et al.* (1998) on CF, providing a view of the information provided about the target conditions of screening tests more generally. Such an analysis might provide some insights into the understandings of four very different kinds of condition in the prenatal context; one associated with multiple disability and early death (trisomy 18), ones associated mostly with physical handicap (NTDs), a chronic health condition (CF), and one associated mainly with learning difficulty (Down's syndrome). As the literature review revealed, people have different levels of concern, and show a greater or lesser inclination to terminate dependent on the perceived severity of the condition. It is hypothesised that most women will be even less familiar with the complexities of these other conditions than they are with Down's syndrome.

Finally the literature review also revealed significant differences in the knowledge of Down's syndrome in non-English speaking groups, particularly in women of South Asian origin where there is no equivalent concept of Down's syndrome. This study considered only information written in English, however, in a review of the social and ethical issues associated with genetic testing, Alderson and colleagues (2001) commented on the 'unequal understanding' of pregnant women and the difficulties inherent in addressing this problem.

"Information leaflets posed irresolvable problems of inequality in their compromises between encapsulating sufficient, essential information which was also clear and brief, particularly as a sizeable minority of women in the [antenatal] clinics spoke little or no English" (Alderson, Farsides & Williams, 2001, p 19).

A recent survey of screening services in England reported that even where leaflets were translated into other languages the number of languages was small (National Screening Committee, 2002). It is believed that information about Down's syndrome is very limited for non-English speaking women, and that improving the accessibility of accurate and appropriate information about the condition is an urgent issue.

3.5 CHAPTER SUMMARY

The findings reported here demonstrate that the information about Down's syndrome provided in prenatal screening leaflets in the UK is often inadequate in quantity and quality. They also suggest that this problem might not be confined to leaflets about screening for Down's syndrome but applicable also to information about other conditions that are the target of antenatal screening

programmes. While it is not known what impact this paucity of information might have on prenatal testing choices, it is argued that in view of the current emphasis on informed choice this situation is unsatisfactory. As there is a lack of current, accurate information about Down's syndrome in antenatal clinics it is likely that most women are informing their prenatal testing choices based on their own beliefs and experiences about the condition and what it entails. However, as highlighted earlier, little is known about the understandings of Down's syndrome that women bring with them to the testing situation. In the next chapter of this thesis, a study is reported that considered the diversity of understandings of Down's syndrome outside of the prenatal testing context and then related these understandings to intentions regarding testing and termination for the condition.

CHAPTER 4 UNDERSTANDINGS OF DOWN'S SYNDROME: A Q METHODOLOGICAL INVESTIGATION.

4.1 INTRODUCTION

Current definitions of informed choice in the prenatal testing context require that decisions made should be in accordance with the person's own values (Bekker, 2003; Bekker *et al.*, 1999; Marteau *et al.*, 2001). Such values in relation to the target condition have generally been overlooked, and as the findings reported in Chapter 3 demonstrate, information provided to women prior to testing can be short on content regarding Down's syndrome. The literature reviewed in Chapter 1 revealed that while a number of understandings of Down's syndrome might exist, within the prenatal testing context the emphasis has been on the (abnormal) organic aspects of the syndrome. Little consideration has been given to the ways in which people not viewing Down's syndrome from the biomedical perspective might understand this complex condition.

Previous research that has attempted to access knowledge or beliefs about Down's syndrome in the prenatal testing context has mainly been concerned with pregnant women, for example, those considering screening (e.g. Mulvey and Wallace, 2000; 2001) or electing to have amniocentesis (e.g. Sjögren and Uddenberg, 1987). Other studies have explored the views of women with a family member of Down's syndrome (e.g. Bryant 1998; Felker, 1994), and a few have considered attitudes towards testing and Down's syndrome in non-pregnant populations (e.g. Bell and Stoneman, 2000). Arguments from certain socio-political positions concerning prenatal testing and termination for abnormality have also been presented, for example, the disability rights critique (Asch, 1999; Bailey, 1996), the feminist critique (Ginsberg and Rapp, 1999; Lippman, 1992; Rothman, 1986), and the ethical critique (Alderson, 2001b; Glover and Glover, 1996; Pueschel, 1991; Williams, 1995). The focus of all these studies has been on attitudes towards personal use of testing, with knowledge or attitudes towards the target condition reported as secondary explanatory variables. Missing from the literature is a study that considers understandings of Down's syndrome as the primary variable of interest and attitudes toward testing second. It is argued that a detailed study on this topic was justified because Down's syndrome has been and continues to be a central focus of prenatal testing technology. Existing surveys of attitudes were not considered suitable for this study because of their associated problems as discussed in Chapter 1. Furthermore, it was felt that to design a new questionnaire specifically for this study might have resulted in a replication of these problems. For these reasons an exploratory approach was considered essential to study understandings of Down's syndrome, and a number of qualitative

methods were considered including in-depth interviews and Q methodology: Q methodology was the approach selected.

4.1.1 Background to Q methodology

Q methodology³² was devised by William Stephenson (Stephenson, 1935; Stephenson, 1953) who was a research assistant to Charles Spearman during the period when Spearman was developing factor analytic techniques. Using a version of factor analysis as its main statistical technique Q methodology provides a structured approach to investigating human subjectivity, which is defined as a person's communication of his or her viewpoint (McKeown and Thomas, 1988). The method is generally used in exploratory research as it is more suited to generating hypotheses than to testing them or making predictions. Q methodology starts from the assumption that for each social object of interest (for example, Down's syndrome) there is a discourse or 'flow of communicability', which in Q terminology is called the *concourse*, i.e. a coming together (Brown, 1993). A *concourse* consists of the things that are written or said about the object, and that can be "*socially contested, argued about and debated. In other words, matters of taste, values and beliefs*" (Stainton Rogers, 1995, p. 180). The aim of Q methodology is to access as many of these alternative views as possible and to describe them in order to gain an understanding of the subjective nature of the object. The method is also useful for highlighting 'inner discursive conflict' or ambivalence, and so is particularly suited to an investigation of complex issues and attitudes (Stainton Rogers, 1995).

The central research tool of Q methodology is the Q sort. Q sorting (the procedure that generates the Q sort) requires participants to read propositions or statements (items) related to the topic of interest and then rank or sort them along a series of dimensions similar to Likert scales so that each item is allocated a ranked score. After a number of individuals have completed their Q sorts, the resulting sets of ranked scores are correlated between each pair of participants. The resulting correlation matrix is subjected to factor analysis in order to identify clusters of Q sorts that resemble each other statistically. Each factor is then considered to represent a common understanding of those individuals whose sorts cluster on that factor (Smith, 2000). The factor analysis used in Q methodology is an inversion of usual techniques because participants are considered to be the variables rather than the items, i.e. the data matrix is analysed by rows instead of columns. It is part of the philosophy behind the method that no one understanding is considered

³² The use of the letter 'Q' is a means of distinguishing the method from traditional 'R' correlational methods, 'R' being a reference to Pearson's product moment correlation (*r*) (Febbraro, 1995). For a discussion of 'Q versus R' see Smith (2000).

to be superior, expert, or objective (Febbraro, 1995). Stephenson argued that the only difference between objectivity and subjectivity, is viewpoint: “what is subjective from my point of view is objective from yours” (Smith 2000, p. 323). For this reason using a Q sort to measure attitudes in relation to a researcher imposed definition is considered an anathema to proponents of the method (Brown, 1980; Kitzinger, 1999). Instead it is believed that Q should be used explicitly to explore and facilitate the articulation of all viewpoints including those that are less dominant in the concourse.

Q methodology has been used in some areas of research more than others, notably nursing, communication studies and political science. Within psychology a number of topics have been examined using the Q approach although not all researchers subscribe to the full methodology. For example, Q sorting in the form of the ‘California Q-set’ (CQS) has been widely used as a diagnostic technique in clinical settings³³ (Block, 1961). Research that has subscribed to the original approach includes investigations of experiences of pain and illness (Eccleston, Williams, and Stainton Rogers, 1997; Risdon, Eccleston, Crombez, and McCracken, 2003; Stainton Rogers, 1991), jealousy (Stenner and Stainton Rogers, 1998), children’s perception of self (Taylor, Delprato, and Knapp, 1994), attitudes toward a spouse with aphasia (Zraik and Boone, 1991), understandings of the cause of genetic conditions (Weil, 1991), and accounts of lesbianism (Kitzinger, 1986; Kitzinger, 1987). The aim of this study was to identify and characterise understandings of Down’s syndrome and to explore their relationship with attitudes towards prenatal testing and termination.

4.1.2 Using Q methodology

A number of texts provide comprehensive accounts of how to conduct a study using Q methodology (Brown, 1980; Curt, 1994; Kerlinger, 1986; Kitzinger, 1987; Kitzinger, 1999; Smith, 2000; Stainton Rogers, 1991; Stainton Rogers, 1995; Stephenson, 1953). A summary of the main points is provided here along with the rationale behind the selection of particular techniques or procedures where appropriate. In particular, the points where Q methodology diverges from more popular research methodologies are highlighted. As a number of novel terms are associated with Q methodology, a glossary of the essential terms is provided in Table 4.1.

³³ For a discussion on this usage of the Q sort see Smith (2000).

Table 4.1. Glossary of Q methodological terms

Term	Description
Concourse	The discourse or 'flow of communicability' about the topic of interest, i.e. what is said or written about Down's syndrome.
Exemplar	A Q sort that loads significantly on only one factor and thus exemplifies the view represented by that factor.
Factor	In Q studies, a factor represents one understanding of the topic under enquiry. The viewpoint is operationalised by merging the exemplar Q sorts for each factor to produce a synthetic Q sort called the factor array.
Factor array	A synthetic Q sort that is used as the physical representation of a particular understanding or factor. Generated by amalgamating the exemplar Q sorts and averaging the scores for each item it represents a 'best estimate' of the factor.
Item	A statement or proposition relating to the topic under enquiry.
P set	The 'person set' of study participants.
Q sample	The sample of items that are selected to represent the whole concourse.
Q set	The Q sample transcribed on to a set of cards and used in the Q sorting process.
Q sort	The results of the ranking procedure whereby each item is allocated a score. The Q sort represents the pattern of beliefs of the individual sorter.
Q sorting	The process by which items are ranked or sorted.
Sorter	The person conducting the Q sorting procedure

Sampling the concourse

The first practical stage in any Q study is the collection of statements from the concourse that comprises the raw material for Q sorting (Brown, 1993). The ultimate goal of this stage is to collect propositions that are representative of the hypothetical universe of propositions on the given subject (Stainton Rogers, 1995). Thus, unlike most research methods, it is the statements that form a representative sample of the population rather than the participants. In order to achieve a representative sample statements are collected from a diverse selection of resources that access as wide a range of views as possible. Resources include both verbal and written material. Verbal material can be obtained formally via individual or group interviews, or informally via general conversation or a discussion on a radio programme, for example. Sources of written materials include academic writing, research reports, and other literature on the subject, and popular sources such as newspapers and magazines. The researcher uses their cultural experience as a guide to the

sourcing of potential material for statement extraction (Stainton Rogers, 1995). Typically, several hundred statements are collected during this stage (Curt, 1994).

Once source materials have been selected, statements about the topic of interest are extracted and classified. Extracted statements are examples of subjective concourse about the subject rather than factual items. This is so participants can express their own evaluations of the statements during the Q sort. For example, the statement *'If a baby with Down's syndrome dies it might be a blessing'*, is subjective and implies value judgment, whereas the statement, *'Approximately 10% of babies born with Down's syndrome die in their first year'* is a factual item of information. The aim of a Q study is not to test knowledge but to capture subjective understanding and meaning. Once extracted, the statements are classified. Categories can be data-driven using a bottom-up approach similar to that used in Grounded Theory (Strauss and Corbin, 1990), or theory driven using a top-down approach. However, it is important to note that statements are never assumed to have only one meaning, i.e. the one ascribed to them by the researcher, and categorisation is simply an aid to item selection. From the source materials the iterative process of extracting and categorising statements continues until no new categories emerge. This suggests that the approximate limitations of the concourse have been reached. It is not assumed that every possible view has been accessed, but following the rule of diminishing returns a decision is made that it is no longer useful to seek further material.

Creation of the Q sample

Once the initial collection of statements has been classified a sub-set are selected to form the Q sample. This set of statements (items) will be sorted by participants and will form the building blocks that characterize the different understandings. When creating the Q sample the aim is to select a reduced group of statements that represents the initial collection and can capture the essence of the concourse. Selection of the items is a subjective process although devices such as Fisher's experimental design principles are sometimes employed to ensure comprehensiveness of coverage (Brown, 1970). Equal numbers of items from each category can be selected, or categories can be weighted according to the proportion of statements each contains. Items should be relatively balanced across the sample in terms of the number of negative and positive propositions, and they must be able to discriminate between respondents. However, in contrast to the usual psychometric practice where repetition of items is employed to reduce error, items that are essentially duplicates or reversals of another statement are not included. The discussion of the selection criteria with an independent observer (as well as the comments of pilot participants) helps to highlight bias in the Q sample, or gaps in the representation of the concourse.

While the aim of Q sample construction is to create a small group of items that represents the whole, debate continues about the actual number of items that should be retained. This debate has sometimes been about the number of items needed to ensure statistical robustness³⁴, but most usually it is concerned with ensuring adequate coverage of the concourse while keeping the number of items small enough to be managed by participants during the sorting procedure (Curt, 1994). Stephenson acknowledged the compromise that is sometimes necessary when selecting items for a Q sample, but noted that,

For practical purposes it is well to restrict experiment to a few salient segments rather than to become lost in details. It helps, for this reason, to work with Q samples of less than 100 so that the cream of feeling is at issue, rather than the whey” (Stephenson, 1980, p. 12).

Most research using Q methodology with adult populations have used Q samples of between 40 and 90 items. However, when working with certain groups, for example children, the elderly, or those with learning difficulties, it might be necessary to work with fewer items.

Once the Q sample has been finalised some of the items may need to be re-worded to make them suitable for research purposes by employing criteria identical to those used when constructing questionnaires. Items should be unambiguous, expressed in clear language, and appropriate to the level of sophistication of the participants. Statements are transcribed onto numbered cards to produce a pack of cards (the Q set) to be used in the sorting procedure.

Participant selection

In Q studies, participants are the variables of the study and are selected for their (assumed) opinions rather than their ability to represent a particular population. The aim is to identify and describe the alternative standpoints on a topic rather than to identify the prevalence of views, and in contrast to traditional methods of measuring attitudes, there is no requirement to recruit large numbers. The twin aims of recruitment are to be confident that as many views as possible have been accessed, and that there are sufficient individuals defining each factor to provide a clear reading of the understanding which that factor represents (Brown, 2000). As no more than five people are needed to clarify a particular view (but not usually less than two), the number of participants recruited depends on the expected number of different views within the concourse. A typical initial recruitment would consist of 30 to 40 individuals, termed the P set (Person set).

³⁴ Kerlinger (1986) recommended that for statistical stability Q sets should contain between 60 and 90 items, and no fewer than 40 items.

However, as this is a subjective judgment and dependent on the subject under enquiry, P set sizes differ widely across studies. In addition following an initial analysis, recruitment often continues if it is suspected that a particular viewpoint is not being accessed.

To get a range of understandings as diverse and comprehensive as possible it is common practice to employ a form of strategic sampling when recruiting study participants (Stainton Rogers, 1995). Some individuals who are known to have some expertise in the area (lay or professional), or who are known to hold a particular type of view are approached. In addition, individuals for whom there is no reason to expect particular knowledge or opinions are recruited in order to ‘hear the unexpected’ or to investigate whether certain understandings are uniquely expert (Stainton Rogers, 1995). In contrast to traditional methods it is common practice to include the researcher as a participant. This reveals their position in relation to other participants views, and gives the reader more information when evaluating the interpretation of the data (Curt, 1994).

Q sorting

Q sorting is an ipsative³⁵ technique that requires participants to read items and then to rank them in order to build a pattern that best represents their understanding of the object or issue. The outcome of the sorting procedure is a Q sort, i.e. a set of ranked items that is taken to represent the pattern of beliefs of the individual sorter. As ranking 50 or more items in one go would be a difficult task for most people, sorting proceeds in a series of steps (Brown, 1993). The method of Q sorting can be either ‘forced’ or ‘free’ in that either the sorting pattern is determined beforehand or not. Using the forced method, items are ranked and then placed onto a matrix constructed as a quasi-normal distribution with an attached scale ranging from, for example, +4 (strongly agree) to -4 (strongly disagree) (see Figure 4.1). The forced method determines how many items can be allocated to the positions of strongly agree, strongly disagree, neutral, and so on. In contrast, free sorting allows participants to allocate as many or as few items as they want to each point on the scale. Although there is some debate about which method of sorting is preferable, most evidence suggests that statistically there is little difference (Brown, 1980). While free sorting does not impose such obvious constraints on participants, forced sorting ensures that respondents will make discriminations between items. This is something they may not do unless it is a specific requirement, and as Brown has argued,

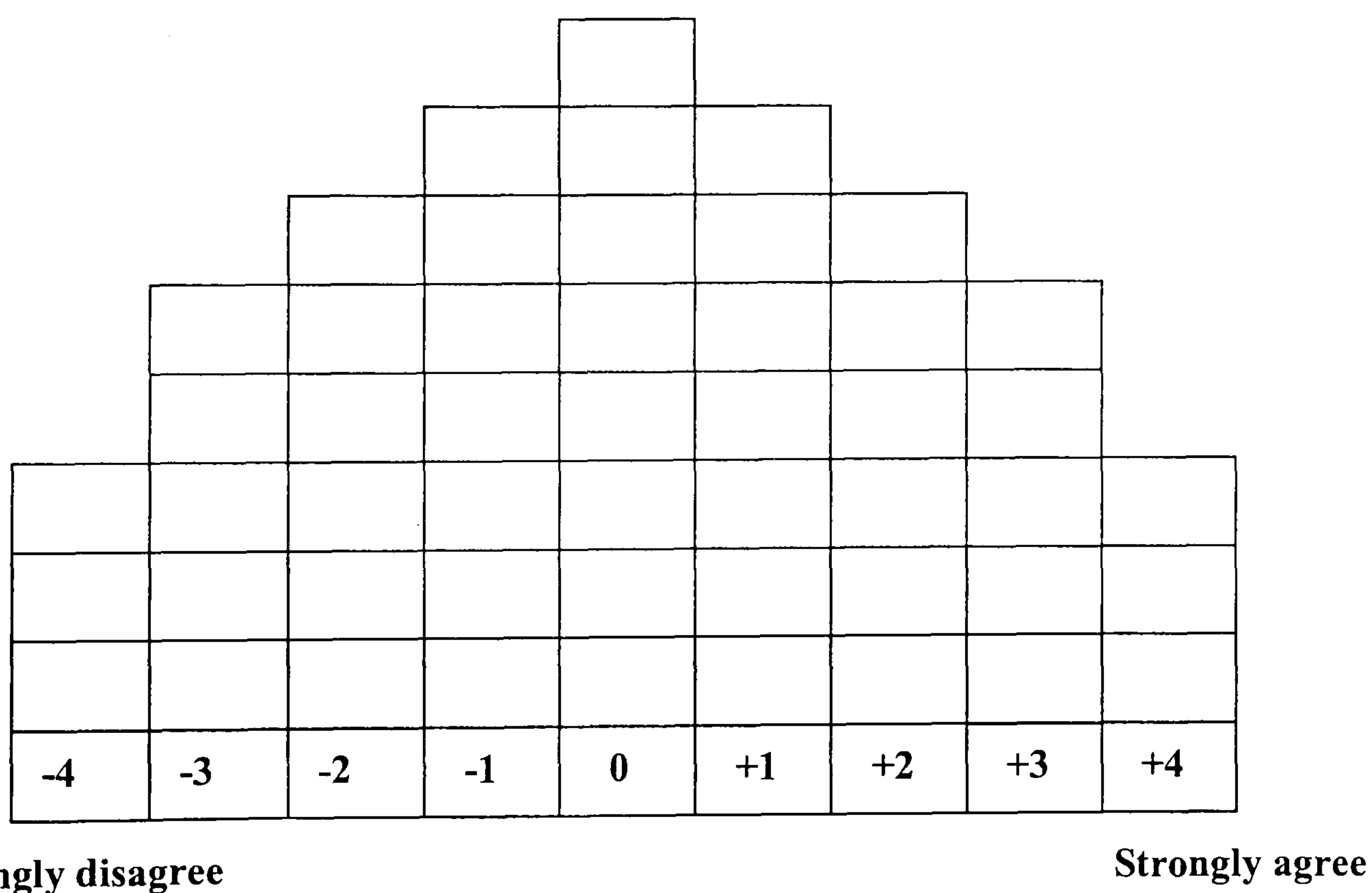
³⁵ Ipsative means measured against the self. An ipsative measure is one where the responder provides his or her own frame of reference to make judgements and comparisons (Reber, 1985). The outcome from an ipsative measure is observed in its own right rather than put in the context of an average or expected outcome, as is the case with ‘normative’ measures.

When people are compelled they have to make choices, and when they make choices they reveal [their] values (or attitudes, sentiments, viewpoints, etc.). When choices can be avoided, the values become obscure (Brown, 2002).

Forced sorting was the method selected for this study to help reduce socially desirable responding. Sorters cannot allocate the same position on the grid to more than a few items in contrast to the way they can allocate the same score to any number of items when completing a standard questionnaire. It was hoped that this would facilitate the expression of attitudes in an area that can be subject to perceptions of 'political correctness'.

During the sorting procedure, participants are encouraged to talk about the placing of the items and to make comments about the items and their perceived meaning. This helps the researcher interpret the factors and provides feedback on the quality and validity of the items chosen for the Q set. Once the participant is satisfied with their Q sort pattern, the item numbers in each cell on the matrix are transcribed onto a data collection grid and input for data analysis. A frequent addition to the Q sorting procedure is to take short biographical sketches from participants. It is hoped that these will give insight into the meaning structures of individuals and aid interpretation.

Figure 4.1: Response matrix



Factor analysis

Techniques other than factor analysis³⁶ have been used to analyse the data from Q sorts, however, the methodology's standard tool has always been factor analysis (Stephenson, 1953). In contrast to the conventional uses of factor analysis, Q methodological study is concerned with the relationships between people rather than between items or tests, and therefore participants are the variables that contribute to the factors, not the items in the Q set. The first stage in the analytic process is the calculation of pair-wise Spearman's *rho* correlations between all the item scores for all participants (or cases in conventional terminology). As the scores on the matrix are already ranked a separate ranking stage is not needed. There is no need for items to be fully ranked from 1 to *N* as it has been demonstrated that the use of the quasi-normal distribution produces an equally acceptable statistical result and is more 'user-friendly' (Stainton Rogers, 1995). The scores on the resulting correlation matrix reflect the degree of similarity and difference between the Q sorts. For example, if A had given item (1) a score of +1 and B had scored it -1 a difference of $D=2$ would exist and the squared difference would be 4. This is done for each item pairing. The squared differences are then summed. If the two Q sorts had been identical, each D would have been 0, each D^2 would have been 0, and the sum would have been 0, i.e. a perfect correlation ($rho = +1.00$). A negative *rho* value would indicate that many of the items that A agreed with, B had disagreed with.

The next stage in the procedure is the application of factor analytic techniques to determine how many alternative standpoints are represented in the P set by grouping together those sorts that are similar to each other, and distinct from the rest at a statistically significant level (Brown, 1993). There are a number of types of factor analysis, of which the main methods used in Q studies are Centroid analysis and Principal Components analysis (PCA). These techniques differ from each other mainly in the way that variance is dealt with (Tabachnick and Fidell, 1983). As usual, there is debate about which technique is best suited to Q methodological aims, however both are widely used and accepted within the Q methodological community. The most usual method is PCA with Varimax rotation, which creates factors that are orthogonal to each other, i.e. at right-angles. Thus the space between different understandings is maximised and the factors are considered to be independent. For each factor, exemplar sorts that load significantly on only one factor are

³⁶ Other analyses that have been used with Q generated data include cluster analysis (Eayrs and Ellis, 1990) and analysis of variance (Kerlinger, 1986). However, ANOVA should not be used with forced Q sorting as the scores are not independent. Factor analysis is usually preferable to cluster analysis because it allows for 'mixed' views, i.e. Q sorts that load significantly on more than one factor, whereas cluster analysis places Q sorts in a single cluster (Brown, 2001).

flagged³⁷ and used to create a synthetic Q sort called the ‘factor array’. The factor array represents a statistical best estimate of the views of all those whose Q sorts clustered on the factor. The item scores in the factor arrays are calculated using a weighted average of all the exemplar sorts. The array acts as the physical model of the understanding as extracted from the concourse, and is used to interpret the factor along with biographical information provided by the participants and any comments made during the Q sorting.

Factor interpretation

Each model sort represents a distinct understanding of the topic; however, Q is not only interested in finding patterns across individuals but within individuals. Because Q sorting is an ipsative process of making comparisons between items it, “enables people to articulate relatedness explicitly and intentionally” (Stainton Rogers, 1987, p. 166). This pattern of relatedness is examined and interpreted in a ‘wholistic’ (sic) manner (Stainton Rogers, 1991). The items which respondents have agreed or disagreed with most strongly are first examined, although the choice of items placed in the more ‘neutral’ columns³⁸ (position +1, 0, and -1) can also be revealing. Kitzinger (1999) makes three other recommendations for a thorough interpretation of Q factors. Firstly an inspection of scores should take place across as well as within factors. Secondly, any apparent discrepancies in Q sort ranking should be discussed, for example where the placing of an item seems at odds with the rest of the sort pattern. A return to the sorter’s comments can often help to interpret the ‘discrepancy’. Linked to this, the third recommendation is to discuss any apparent differences in item interpretation across different factors. Statements may have multiple meanings depending on the view within which they sit, and Q methodology allows for these different meanings to be explored. This contrasts with more traditional methods where the researcher determines the meaning of questionnaire items beforehand. Interpretation can also be aided by the biographical details collected before sorting began. Finally, validation of the factors can be achieved by discussing the interpretations with those people whose sorts they represent.

4.1.3 Criticisms of Q methodology

Despite having being around in its present form since the 1950s Q methodology is still a relatively unfamiliar research method within psychology. Celia Kitzinger, a researcher in the areas of feminist and lesbian psychology, recently wrote that she no longer uses Q methodology because she found it frustrating to be continually called upon to explain and justify her choice of method at

³⁷ This is usually done using an algorithm integral to Q dedicated software.

³⁸ ‘Neutral’ represents the midpoint of the scale (marked as zero), but can also be the position where participants place items about which they are uncertain, have ambiguity towards, or perceive as non-salient (Curt, 1994).

the expense of discussing her results (Kitzinger, 1999). She concluded that this was not a fault of the method but of the 'inflexible' research community within psychology. Those who work with other qualitative methods often criticise Q methodology because it uses preselected items and apparently constraining procedures. However, Kitzinger points out that interviews and focus groups use predefined schedules that also constrain the interviewee, even if this is in a less obvious fashion. Some also regard Q method's reliance on factor analytic techniques with suspicion because it can appear to be a way of putting people into categories. This is a misapprehension of the method, as the aim of using Q is not to categorise people, but to explore discourse in a structured manner so as to facilitate understanding of how diverse views are constructed.

Criticism from those who use traditional quantitative methods is often directed at the way that Q uses factor analysis but does not subscribe to the psychometric standards set for constructing tests. In particular, purposively selected and relatively small samples are criticised for not being large randomly selected samples. The interpretation of factors exemplified by only a small number of Q sorts is also frowned upon. Some authors call for Q studies to be replicated with large samples using the items in a scale so as to validate the findings (Furnham, 1994; Kerlinger, 1986). Kitzinger comments that these criticisms are unjustified because Q methodology starts from a different set of assumptions than do R methodological techniques, and studies using Q have very different aims. No claims are ever made as to the representativeness of the findings, or that the factors identify 'types' of people in the way that factor analysis has been used to identify introverts and extroverts for example. Q studies have also been criticised because statistical tests have not been applied to the Q data to identify relationships between socio-demographic variables and factors (Furnham, 1994). This is not an appropriate criticism in light of the purposive sampling employed in Q, as spurious conclusions could be drawn from conducting this type of analysis on data gathered from a non-random sample (Stainton Rogers, 1987).

In summary, most of the criticism directed towards Q methodology is founded on misapprehensions about the purpose of the methodology and its application. Nevertheless, in common with all other research methods, Q methodology does have limitations. The forced distribution results in data that are not independent, and all participants have the same general mean score and general standard deviation (Kerlinger, 1986). This makes the procedure unsuitable for investigation requiring statistical comparisons of scores between groups. In addition, the Q sorting procedure can be complex and effortful for the participant, and it could be argued that this limits its use to those with a relatively high level of educational ability and concentration. However, simplified Q sets have been used or alternatives to text employed when working with children (e.g. Taylor *et al.*, 1994). More generally, the Q sample can be biased if the discourse is

not explored adequately, the items have been selected with an agenda in mind, or participant recruitment lacks diversity. If data items are badly or ambiguously worded the resulting data will suffer in the same way as data collected using a badly worded questionnaire item or interview question. Subjectivity in interpretation is inevitable if it is accepted (as it must be within the philosophy of Q) that there is no such thing as an objective view. However, the rigorous Q researcher should give a full explanation of how the method was applied thus enabling the reader to make up his or her own mind as to the appropriateness of the conclusions drawn.

4.1.4 Objectives of the study

The Q methodological investigation had three main objectives.

1. To explore diversity in subjective understandings of Down's syndrome and identify the important similarities and differences in these understandings.
2. To investigate the relationship between understandings of Down's syndrome and attitudes towards prenatal testing and termination for the condition.
3. To generate hypotheses about these relationships for further research.

The next section details how Q methodology was applied to the concourse of Down's syndrome in order to meet these objectives.

4.2 METHOD

4.2.1 Sampling the concourse of Down's syndrome

In this study, the social object of interest was Down's syndrome, and more precisely, what people understand about the syndrome in terms of its effect on society, on families with an affected child, and on the affected person. Verbal information was collected from three main sources, (1) a focus group of midwifery students, (2) one in-depth interview, and (3) informal conversation with people known to have some specific interest in Down's syndrome or prenatal testing. Sources of written material were; (1) qualitative data collected in a previous study of women who have a sibling with Down's syndrome (Bryant, 1998), (2) academic and non-academic publications on prenatal testing and Down's syndrome identified through a literature search, and (3) a collection of information leaflets given to women prior to prenatal screening (analysed in Chapter 3). Two of the verbal data collection methods are described in some detail below.

4.2.1.1 Focus group

During April 1999 a focus group with midwives was conducted, the objective being to collect material for the Q sample of Down's syndrome. This participant group was selected purposefully

in order to situate the discussion in a context where prenatal testing was likely to be raised spontaneously. A focus group can be defined as “*a carefully planned discussion designed to obtain perceptions on a defined area of interest in a permissive non-threatening environment*” (Krueger, 1994, p. 6). During a focus group diverse views are sought and themes of consensus and difference surrounding a topic can be identified in a relatively short space of time. Focus groups allow the researcher to capitalise on the communication between participants in a social context (Kitzinger, 1995), thus being a particularly appropriate method for accessing discourse.

Participants

As part of their research methods module, a cohort of ten female midwives agreed to participate in the focus group. They ranged in age from 24 to 42 years (mean 31.5 years). On average they had been in nursing/midwifery for 7.5 years. Three participants had children and the rest did not.

Materials

While the aim of the discussion was to access diversity of views about Down’s syndrome, there were a number of areas considered of particular relevance. A topic guide was devised to direct the discussion containing a number of prompt questions for those issues not raised spontaneously.

Topic 1: Previous experiences of Down’s syndrome.

- What (if any) experience have they (the group) had of people with Down’s syndrome?
- Can they remember how they felt about these encounters?
- What images might come to mind when they think of Down’s syndrome?

Topic 2: Social integration, prejudice and discrimination

- How are people with Down’s syndrome talked about? (Discuss use of the term ‘Mongol’.)
- How do they feel about meeting people socially with Down’s syndrome?
- What are their views on integrating children with Down’s syndrome into mainstream schools?

Topic 3: Down’s syndrome and quality of life

- What do they imagine it is like for the family of a child with Down’s syndrome?
- From an affected person’s point of view what do they think having Down’s syndrome is like?
- What do they think about cosmetic surgery for children/adults with Down’s syndrome?

Topic 4: Prenatal testing and termination for Down’s syndrome

- What do they think about prenatal testing for Down’s syndrome?
- What do they think about termination for Down’s syndrome?
- What about adoption of children with Down’s syndrome (is it better/worse than termination)?

Some additional prompts in the form of photographs were used that showed people with Down's syndrome (babies, children, adults, and older adults) in a variety of settings (family, education, and work) (see Appendix 1). It was hoped that the prompts would facilitate discussion between participants and so move the group on from a simple question and answer session.

Procedure

The students sat round a large table in a teaching room at the University of Leeds. The researcher who facilitated the discussion sat at the table with the students and an observer³⁹ sat outside the circle to make notes on the process. The researcher described the purpose of the focus group, emphasised that a diverse range of views were being sought, and then gave each participant a consent form to complete if they wished to continue. A short questionnaire was administered to collect brief biographical details (age, length of time in nursing, and number of children). The discussion itself lasted approximately one hour. The discussion was audiotaped with the participants' permission and then transcribed verbatim.

Analysis

The transcript was analysed with the single aim of generating statements for the Q sample. Statements that reflected beliefs, views, and experiences of Down's syndrome were identified and marked using coloured highlighter pens. Once the whole transcript had been read and checked a number of times all the highlighted statements were extracted and categorised by general theme. Collected statements reflected the varied experiences of the participants. Two students had been at the delivery of a baby with Down's syndrome, and others reported on childhood experiences, experiences through voluntary work and during nurse training. The prompts related to cosmetic surgery on people with Down's syndrome generated a particularly lively discussion. Some felt that surgery to 'improve' the appearance of people with Down's syndrome represented the failure of parents and society to accept disability, others felt it might help reduce the stigma of the condition for the affected person and their family. This led to a discussion about how medical science was striving to enable the production of the 'perfect child', although interestingly the midwives did not bring up the issue of prenatal testing at this point or raise it by themselves at any time during the discussion. Other subjects that generated extensive discussion were the perceived impact of having an affected child on parents and family, and how participants felt personally about having a future child with Down's syndrome. Issues of dependence and independence were raised, as was the perception of elderly parents as carers.

³⁹ Dr Josephine Green acted as the observer at the focus group session.

When the researcher raised the topic of prenatal testing a number of different views were expressed about whether termination for Down's syndrome was acceptable, and whether screening tests were useful. The general consensus (as might be expected from this group) was that it was up to individuals to decide about testing for themselves, and for midwives to support all choices equally. The participants were asked whether they felt women could make informed choices about testing for Down's syndrome. In general, they felt this was not always easy. In relation to information provision different views emerged, for example, one midwife said,

"People have their own perceptions of what Down's syndrome is and how it would affect them and their family, nobody gives them a talk or a leaflet or anything. Unless they know someone, or have worked with them or something like that, then they've just got their own perceptions. So it's not really fair is it. It's not an informed choice really."

However, another felt that having access to a lot of information could make choices more difficult.

"Well, sometimes I think that, knowing what we know, and as much information that we have, I think it would be a really hard decision. I'd feel very contradictory, I'd be like swinging totally one way and then the other."

Comment

The focus group generated useful material for the Q study, and a number of statements from the group were selected as statements in the final Q sample (see Appendix 2). The discussion highlighted how different perceptions of Down's syndrome and prenatal testing exist even within one small group of health professionals. Nevertheless, it was felt that the views expressed might have been limited. A problem with focus groups is that views that contradict the perceived group norm can be hidden or shouted down (Kitzinger, 1995). For example, all the student midwives expressed the belief that people with Down's syndrome should be integrated into their communities, including mainstream education and the workplace where possible. However, one participant also related the experience of a teacher friend at a mainstream school who had experienced a child with learning difficulties as a disruption to other pupils. Other participants tended to 'jump' on this view as discriminatory, leaving the student to explain defensively "*I'm not suggesting... I'm just explaining that's the experience I've got, y'know*". Another issue is that sensitive subjects are not always easy to raise in a group environment. While transcribing the tape, the researcher found it difficult to hear the conversation about the topic of prejudice, name-calling and the use of the term 'Mongol'. The students appeared to feel embarrassment when discussing this topic and referred only to other people using the words Mongol, or the derogatory terms 'mongy' and 'mong'. The general style of this part of the discussion was hesitant.

In conclusion it was felt that views perceived as less socially desirable might have been stifled by the group interview method. For this reason, a single in-depth interview was conducted with a health professional known to have strong positive views about prenatal testing. It was hoped that this interview might provide some useful alternative perceptions of Down's syndrome.

4.2.1.2 Interview

In May 1999 the interview with the health professional took place at a room at the hospital where he worked. The interview questions were based on the focus group topic guide. The interview lasted 45 minutes. Outside of work, the interviewee's specific experiences of people with Down's syndrome were limited to knowing a boy in his childhood. His main experiences were work related and mainly with parents whose child had been diagnosed with Down's syndrome prenatally or after the birth. He was also involved with the case of a young woman with Down's syndrome who was being considered for sterilisation on the request of her parents. His perception was that,

“Down's syndrome is a very serious handicap. I think it's ridiculous to say that they are just a 'bit different'. Imagine if some doctor caused a normal child to have Down's syndrome this would cause an outrage. It's a serious, serious problem.”

An in depth analysis of the interview was not conducted, however, statements relating to Down's syndrome were highlighted and extracted from the interview transcript in the same way as described in relation to the focus group. These statements added a particular viewpoint to the collection of material that might otherwise have been excluded. A number of the statements were used as items in the final Q sample (see Appendix 2).

4.2.2 Creating the Down's syndrome Q sample

Using a bottom-up, data-driven approach 400 statements were extracted from the collected source materials and categorised according to their content. General headings were selected, for example, 'burden of care', 'eugenics', and 'quality of life'. Out of the original 400 statements 50 were retained to form the final Q sample with approximately one statement from each category being selected (see Appendix 2 for statements, their category and their source). The selection process was conducted with a researcher familiar with Q methodology but who had little knowledge of Down's syndrome or prenatal testing (Zak Avery, The Open University). Each category of items was discussed, and the item(s) that seemed to best represent the overall theme of the categories were selected. In order to pilot the Q sample, the initial set of items was presented to five individuals to sort and as a result, some items were reworded in order to clarify the statement.

4.2.3 Recruitment of the P set

The aim of recruitment in a Q study is to access as diverse a range of views as possible. The recruitment was conducted in two phases. The first phase recruited 36 individuals, approximately half 'experts' and half 'non-experts' on the subjects of Down's syndrome and/or prenatal testing. However, after analysing the Q sorts of these individuals it was felt that certain viewpoints were not being represented, for example, very few men had been recruited, and there was little ethnic or religious diversity. A second purposive sampling period then took place and a further 42 people were recruited. Two of the sorts conducted by post were not used because of incorrect completion of the Q grid and so the Q sorts of 76 participants were included in the analysis. Of the 76 participants in the final P set, 47 were women and 29 were men. Ages ranged from 23 to 80 years (mean age 37.2, SD 10.3). Appendix 3 gives the age, sex and brief biographical details of all 76 participants. The first sub-group (N=38, 13 men, 25 women) were recruited for their particular interest in Down's syndrome or prenatal testing in either a personal or a professional capacity. Some individuals were invited to participate via telephone, email, or by letter, and others were targeted as a group, for example the staff working in a cytogenetics laboratory who conduct karyotyping of fetal cells following a prenatal diagnostic test. Table 4.2 provides details of the 'expert group' participants.

The second group of participants recruited (N=38, 16 men, 22 women) had no known special interest, opinion or experience in the areas of prenatal testing or Down's syndrome. Of this group, 40% were academics, researchers, or post-graduate students at the University of Leeds, and 60% came from a diverse range of backgrounds and occupations. Participants in this group were recruited via a direct approach from the researcher, an email to colleagues asking for participants, and snowballing from other participants. In selecting participants for the second group, an attempt was made to include some diversity of religious belief, and ethnic or cultural background.

The recruitment of 'experts' and 'non-experts' is commonly used in Q methodological studies in order to maximise the range of views obtained. However, there is no claim that the participants in this study were representative of the general population as this is not a recruitment aim within Q methodology. The majority of the participants were white British. Five participants were of Pakistani or Indian origin, two were from North America and one was from Central Europe. Most participants associated themselves with Christian religions (C of E, Catholic, Methodist, Baptist, Quaker), seven were Jewish, three were Muslims, and two were Hindus. Two had a physical disability. In terms of socio-economic status, most participants would be categorised as belonging to social classes three and above. Thirty-nine participants had children, 34 did not, and three did not provide this information.

Table 4.2. Details of participants selected for special interest or expertise.

	Professional interest or expertise	Lay interest or expertise
Down's syndrome	Two staff at a residential home for adults with a learning difficulty (LD), Two community support workers for adults with LD, Teacher at a school for children with LD, Parent support worker at a nursery for children with LD, Psychology Assistant specialising in LD, General Practitioner, Researcher/lecturer in disability studies	Four mothers (including an adoptive mother) of children/adults with Down's syndrome, Two siblings of a person with Down's syndrome ⁴⁰ . Aunt of a woman with Down's syndrome Father of an adult daughter with severe LD.
Prenatal testing	Eight staff at a cytogenetics laboratory. Two consultant obstetricians. Two midwives. Two genetic counsellors Researcher in sociology/disability issues. Researcher in reproductive psychology. Medical researcher/screening specialist.	Two women who chose not to have prenatal testing for Down's syndrome. Woman who terminated a pregnancy for Down's syndrome. Woman whose mother terminated a pregnancy for Down's syndrome.

4.2.4 Ethical considerations

This research was carried out within the guidelines of the British Psychological Society's 'Ethical Principles for Conducting Research with Human Participants' (British Psychological Society, 1995). Particular references to key areas of the code are made below. Ethical approval was sought and obtained from the School of Psychology, University of Leeds, and the Educational Research Ethics Committee, University of Leeds before data collection began.

Informed Consent. Prior to participation in the study, respondents were given an information sheet about the purpose of the research and what their participation would entail. Consent was recorded on a form based on the standard suggested by the Ethics Committee of the School of Psychology. All participants were invited to take part and no reward was offered for participation.

⁴⁰ One of these was the researcher.

The information sheet and consent form both clarified the participant's right to withdraw at any point from the study. No participant withdrew consent retrospectively.

Confidentiality. All data provided by participants were kept confidential by keeping consent forms separate from questionnaires. In compliance with the Data Protection Act, no data was stored on computer that could be associated with an individual.

Protection of Participants. In respect of the privacy of individuals, participants were informed that they were not obliged to respond to any questions if they did not wish to. As the topic under inquiry was a sensitive one, every attempt was made to ensure the questions were worded in a way to take account of this. However, there was a possibility that negative feelings or unhappy memories could be evoked by participation in the study, and so the information sheet included contact details of relevant support agencies (Down's Syndrome Association and ARC (Antenatal Results and Choices)).

Debriefing. Participants were offered a summary of the study findings.

4.2.5 Materials

The materials required for the administration of the Q sort were;

- An information sheet and consent form,
- A piece of A4 paper headed 'Participant Background Information' for recording biographical details using three open ended questions about family and background, experience of prenatal testing, and experience with people with Down's syndrome;
- The Q set of 50 statements printed on to small cards (6cm x 4cm).
- A large piece of paper printed with the matrix of fifty cells (6cm x 4cm) arranged in a quasi normal distribution (referred to as the Q grid).
- Three pieces of A4 paper printed with three boxes (6cm x 4cm) labelled 'AGREE', 'NEUTRAL' and 'DISAGREE', or 'STRONGLY AGREE', 'QUITE STRONGLY AGREE' and 'SLIGHTLY AGREE', or 'STRONGLY DISAGREE', 'QUITE STRONGLY DISAGREE' and 'SLIGHTLY DISAGREE'
- A response booklet with a copy of the Q grid on the front on to which the participant's sorting pattern was transcribed. The 50 Q sample items were printed in the booklet with space to write comments about the item wording or the placing of the item on the grid.

If the Q sort was being administered by post (see Procedure section) the materials also included instructions on how to complete the Q sort, and a prepaid envelope for return to the researcher.

In order to investigate the relationship between understandings of Down's syndrome and attitudes towards prenatal testing and termination for the condition, a short questionnaire was used (see Appendix 4). This questionnaire was based on one used in a Flemish study of community attitudes towards prenatal testing (Evers-Kiebooms *et al.*, 1993). Four seven-point Likert type scales were used to capture responses to whether (1) participants thought prenatal diagnostic testing for Down's syndrome should be freely available to all women, (2) they would use such testing personally, (3) termination for Down's syndrome should be freely available to all women, and (4) they would terminate a pregnancy affected by Down's syndrome. This 'two-step' approach was used because it is more sensitive than direct questioning of personal behavioural intentions (Green *et al.*, 1993a) and also because it reduces confounding of the participant's attitude to their own actions with their attitude towards the actions of others. An open-ended item asked respondents to explain their answers in relation to prenatal testing and termination.

4.3 PROCEDURE

Just over half of the Q sorts (57%) and questionnaires were completed in a one-to-one session with the participant, either in the University of Leeds, or at the participant's workplace or home. The remainder were completed by post and returned in a prepaid envelope by the participant. Detailed instructions on how to complete the Q sort were provided in verbal form for those seen on a one-to-one basis and in written form for the postal sorts. The instructions were as follows.

1. Using the sheet with the AGREE, NEUTRAL and DISAGREE boxes, read each statement and place it in the box that most suits your view. For example if you read a statement which says "All people with Down's syndrome like music" and you agreed with this, you would place the card in the AGREE box. However, if you disagreed with the statement, you would put it in the DISAGREE box. If you neither agreed nor disagreed with the statement, or had no view on this statement, you would put it in the NEUTRAL box.
2. After sorting all the cards into three piles, take the statements in the AGREE box. Using the sheet that has three boxes labelled STRONGLY AGREE, QUITE STRONGLY AGREE and SLIGHTLY AGREE, read each statement again and place it in the box that most suits the strength of your agreement with the statement.
3. Now place the Q grid on your working surface. Take the statements in your STRONGLY AGREE pile and choosing the **three statements you agree with most strongly**, place these in any order in the column labelled +4. If you have any STRONGLY AGREE statements left after doing this, put these in the +3 column. If you have any left over after filling this column, put these in the column labelled +2, and so on. When you have used up all the STRONGLY AGREE pile take the QUITE STRONGLY AGREE statements and sort these, placing the

ones you agree most strongly with in the column spaces following the STRONGLY AGREE statements. Continue placing the statements until you have used up all the AGREE statements. If you have filled all the columns from +4 to +1 and still have some statements left over, add these to the neutral pile.

4. Now take the statements in the DISAGREE box and repeat steps 3 and 4 using the sorting sheet with the three boxes labelled STRONGLY DISAGREE, QUITE STRONGLY DISAGREE and SLIGHTLY DISAGREE. Place the **three statements you disagree with most strongly** in the column labelled -4, and so on until you have used up all the statements or filled all the columns from -4 to -1.
5. Now take the statements in the NEUTRAL pile, and any left over from sorting your AGREE and DISAGREE piles. If you still have some spaces in the agree (+1 to +4) or the disagree (-1 to -4) columns, read through your NEUTRAL statements again to see if you slightly agree or disagree with any statements more than others. Use these statements to fill the (+) or (-) column spaces. Eventually you should be left with the **eight statements** about which you feel most 'neutral'. Place these in the column labelled '0'.
6. You should now have no statements left and no spaces on the Q grid. Copy each statement number on the Q grid into the appropriate column on the grid printed on the booklet.
7. Use the spaces provided in the booklet to write anything you like about the statements.

Analysis

Once all the Q sorts had been collected, the data was entered into a Q dedicated software package (PQMethod) for analysis. Although general statistical packages can be used to analyse data from Q sorts there are a number of dedicated packages available that are tailored to the requirements of Q studies and provide output to support factor interpretation. The software selected to conduct the analysis in this study was PQMethod (2.09) a package used widely within Q methodological research and designed to run on PCs in DOS (Schmolck and Atkinson, 1998). Support for software users is provided on-line by the developers and via an electronic mailing list of over 300 researchers who actively use the methodology. Output from PQMethod provides factor loadings for each sort and a range of reports including 'consensus items', i.e. those items that do not discriminate well between participants.

The data from the Q sorts and prenatal testing questionnaires were also input to SPSS for Windows 9.0, to allow some comparison statistics to be run and hypotheses to be generated.

4.4 RESULTS

4.4.1 Factor analysis and factor rotation

Using the PQMethod software Principle Component Analysis was run against the data collected from the 76 Q sorts. A decision was then made as to the number of factors to be retained for rotation. A number of techniques can be used to inform the decision about retaining factors, the most common being the use of the 'Kaiser criterion' where factors with eigenvalues greater than one are retained (Kaiser, 1960). However, it is recognised that the Kaiser criterion can sometimes retain too many factors, and is the recommended test only where the number of variables is 30 or less. This proved to be the case here where the number of variables (cases) was 76 and twelve factors had eigenvalues of one or more (ranging from 39.5 to 1.1). An alternative was to use a graphical technique called the scree test (Cattell, 1966). Here eigenvalues are plotted on a simple line graph and those factors falling to the right of the point where the smooth decrease of eigenvalues levels off are considered 'factorial scree' and are not retained. Rust and Golombok advise taking "*as many factors as can reasonably be interpreted*" by trying a number of solutions around the number indicated by the criterion selected (Rust and Golombok, 1989, p.123). Using the scree method, four, five, and six factor solutions were considered. The five-factor solution was found to produce the 'best fit' in terms of producing interpretable data and understandings of Down's syndrome that were recognisable from comments made during the sorting procedure or in the response booklets. The PQMethod software allows for two rotation methods; Centroid rotation and Varimax rotation. Varimax rotation, the default of the PQMethod program, generates orthogonal factors and this method was used as the aim was to maximally separate the understandings of Down's syndrome.

The output from the rotation procedure is a listing of all the sorts and their loadings against each of the retained factors. Those Q sorts that exemplify a particular factor are flagged with an 'x' by a program generated algorithm within PQMethod⁴¹. The algorithm is designed to flag 'pure' cases only, i.e. those that load significantly on only one factor. These exemplar sorts are then used to create the factor arrays for each factor using a weighting formula devised by Spearman (1927, cited in Stainton Rogers, 1995). The loadings of each Q sort against the five rotated factors are given in Table 4.3, with 'x' marking those sorts identified by PQ Method as exemplifying the factor.

⁴¹ The algorithm flags cases according to the following rules. Flag loading a if (1) $a^2 > h^2/2$ (where h^2 is the sum of the squared loading coefficients, i.e. the proportion of a sort's variance explained by the factors) and (2) $a > 1.96/\sqrt{n}$ items (loading significant at $p < 0.05$).

Table 4.3. Q sort loading by Factor

Sort #	Factor 1	Factor 2	Factor 3	Factor 4	Factor 5
1	.62	.26	.33	.31	.39
2	.81x	.18	.20	.09	.24
3	.09	.69x	.26	.04	.25
4	.42	.12	.66x	.33	-.01
5	.55	.17	.16	.08	.54
6	.79x	-.10	.17	.02	.00
7	.72x	.12	.21	.24	.33
8	.83x	.07	.22	.11	.10
9	.60	-.13	.48	.20	.28
10	.79x	.12	.32	.11	.19
11	.47	.34	-.19	.33	.51
12	.68x	.13	.06	.16	.16
13	.50	.07	.17	.55x	.12
14	.74x	.15	.24	.32	.09
15	.61	.21	.23	.46	.31
16	.81x	.02	.20	.21	.09
17	.44	-.05	.36	-.09	.44
18	.65x	.25	.39	.29	.20
19	.83x	.07	.22	.14	.10
20	-.23	.57x	.23	-.18	.26
21	.29	.61x	-.05	.08	-.02
22	.72x	.16	.30	.36	.27
23	.81x	.15	.10	.23	.13
24	-.24	.79x	.15	.14	.05
25	.63x	.26	.42	.14	.19
26	.65x	.34	.39	.07	.32
27	.82x	.21	.19	.28	.15
28	.74x	.17	.09	.28	.21
29	.73x	.31	.09	.20	.05
30	.12	.43	.38	.65x	.10
31	.71x	.09	.18	.25	.41
32	.68x	.18	-.00	.10	.20
33	.55	.38	.25	.28	.47
34	-.56x	.45	-.01	.07	.16
35	-.09	.67x	.44	.17	.05
36	.63x	.12	.03	.56	.09
37	.00	.40	.20	.58x	.22
38	.78x	.13	-.00	.13	.24
39	.59	.31	.45	.23	.18
40	.76x	.04	.42	-.02	.14
41	.33	.52x	-.10	.35	.10
42	.83x	.06	.15	.01	.28
43	.70x	.39	.30	.16	.20
44	.57	.17	.34	.14	.54

x = Q sort flagged as exemplar of the factor

Table 4.3 continued.

Sort #	Factor 1	Factor 2	Factor 3	Factor 4	Factor 5
45	.82x	-.02	.16	-.16	.09
46	.61	.11	.52	.22	.24
47	.54	.27	.39	.16	.28
48	.55	.61	.14	.08	.24
49	.72x	-.03	.37	.08	.25
50	.35	.45	.17	.39	-.02
51	.57	.49	.41	-.08	-.02
52	.37	.68x	.27	.07	-.10
53	.50	.08	.42	.15	.28
54	.12	.59x	-.03	.25	.40
55	.53x	.23	.39	-.05	.27
56	.62x	-.01	.34	.06	.51
57	.52	.41	.48	.31	-.10
58	.29	.25	.01	.30	.62x
59	.59x	.43	.14	.28	.14
60	.35	.21	.23	.09	.64x
61	.70x	.09	.28	.18	.36
62	.50	.03	.37	.04	.50
63	.71x	.16	.28	.12	.32
64	.71x	.23	.35	-.05	.20
65	.03	.71x	-.16	.08	-.08
66	.68x	.27	.23	.36	.21
67	.74x	.09	.10	.36	.24
68	.23	.68x	-.02	.08	.28
69	.03	.39	.07	.39	.35
70	.64x	.23	.10	-.01	.37
71	.46	-.07	.65x	.18	.18
72	.81x	.01	.31	.11	.21
73	.35	.38	.62x	.10	.13
74	.70x	.15	.46	.08	.12
75	.51	.35	.44	.06	.29
76	.49	.22	.43	.04	.31

x = Q sort flagged as exemplar of the factor

4.4.2 Factor description and interpretation

Following factor rotation, five factor arrays were created. Table 4.4 gives the scores against each item by factor. Eight statements (shaded in Table 4.4) were ‘consensus items’ in that they did not discriminate between factors. These items fell into two groups. The first group (items 27, 30, 43, 46 and 50) constituted the view that people who have Down’s syndrome must have the same rights as other individuals to healthcare, education, and inclusion in their community. Two items (26 and 41) related to the stereotype of people with Down’s syndrome as happy and affectionate. Item (36) “A child with Down’s syndrome must bring continual sorrow to its parents” was disagreed with by all participants. The use of the word ‘continual’ was possibly too strong here making an ‘agree’

response unlikely. Because these eight items did not discriminate between participants, they were only considered in the factor interpretations when they appeared to be discrepant with the placing of other items.

Table 4.4. Factor arrays: scores against each item by Factor

#	Statement	F1	F2	F3	F4	F5
1	Children with Down's syndrome can achieve a great deal.	+2	+1	+3	+4	0
2	You can be as proud of a child with Down's syndrome as you can be of any child.	+3	+2	+4	+2	+1
3	If you have a baby with Down's syndrome it may be better to have it adopted and try again.	-3	-3	+1	+1	-3
4	A child with Down's syndrome is a family tragedy.	-3	0	-4	+1	-3
5	The normal siblings of children with Down's syndrome suffer as well.	-1	+2	-2	+3	-3
6	A problem with children with Down's syndrome is that they will probably outlive their parents.	0	+1	+2	-1	-1
7	It's not right to submit a child with Down's syndrome to cosmetic surgery, they should be accepted the way they are.	+1	-1	+1	-2	+1
8	I find people with Down's syndrome rather unattractive.	-1	+2	-2	0	-2
9	If you have a child with Down's syndrome it is because God chose you.	-1	-4	-4	-4	-4
10	If a child with Down's syndrome died, it might be a blessing.	-3	-1	-1	0	-4
11	Children with Down's syndrome are a burden throughout their lives.	-3	0	-2	-1	0
12	Normal children are just as demanding as children with Down's syndrome.	0	-1	+2	+2	-1
13	Nobody would choose to have a child with Down's syndrome.	0	0	-2	+1	+2
14	Choosing to bring a child with Down's syndrome into the world is just selfish.	-4	-3	-4	-4	-1
15	People with Down's syndrome are a financial burden on the state.	-3	-1	0	0	0
16	A person with Down's syndrome will always be totally dependent on others.	-2	+2	-3	-3	-4
17	People with Down's syndrome remain like children all their lives.	-1	+1	-1	-3	-3
18	For people with Down's syndrome, the biggest obstacle is not their learning disability but the attitudes of others.	+3	+1	-2	0	+2
19	If I had a child with Down's syndrome I would be worried about people staring at us.	-1	-2	-3	0	+3
20	Knowing someone with Down's syndrome enriches our understanding of what it is to be human.	+2	0	0	-2	0
21	Down's syndrome is an abnormality and an error of nature. It makes sense to try and prevent it.	-2	+2	+2	0	+2
22	I think that euthanasia of babies with Down's syndrome is acceptable if that is what the parents want.	-4	-3	-3	-2	-1
23	People with Down's syndrome make me feel uncomfortable.	-2	0	-3	-2	-2
24	People with Down's syndrome have the same feelings as anybody else.	+4	+1	+1	+2	+1

Table 4.4 continued

#	Statement	F1	F2	F3	F4	F5
25	The world would be a worse place if no more babies with Down's syndrome were born.	+1	-4	-1	-2	0
26	People with Down's syndrome give as well as receive love.	+3	+4	+4	+3	+2
27	It is wrong to treat people with Down's syndrome as a group. They are all individuals.	+3	+3	+1	+1	+2
28	I would find it as easy to love a child with Down's syndrome as to love any other child.	+2	-2	+3	-1	-1
29	I think you are lucky if you have a person with Down's syndrome in your family.	0	-4	-2	-3	-2
30	People with Down's syndrome should have the same health care as any other person.	+4	+4	+3	+3	+4
31	I wouldn't call Down's syndrome a major health problem.	0	-2	0	+1	-2
32	The medical profession paints an overly gloomy picture of what it is like to have a child with Down's syndrome.	+1	-1	0	+1	0
33	Having to say 'Down's syndrome' instead of Mongol, is just another example of political correctness.	-2	-1	0	-1	-2
34	Saying that having a child with Down's syndrome is as good as a normal child is just denying reality.	-2	+4	-1	0	0
35	For me, having a child with Down's syndrome wouldn't be the end of the world.	+1	-3	+1	-3	+1
36	A child with Down's syndrome must bring continual sorrow to its parents.	-4	-2	-3	-2	-3
37	People with Down's syndrome shouldn't be called sufferers.	+2	0	+3	+3	-1
38	I feel so sorry for people who have a baby with Down's syndrome.	0	+3	-1	-1	+1
39	It must be awful to have Down's syndrome.	-1	0	-1	-1	0
40	You would get a lot of joy from having a child with Down's syndrome.	+1	-2	+2	0	+3
41	People with Down's syndrome can live very happy lives.	+3	+2	+3	+2	+3
42	People with Down's syndrome can have as good a quality of life as everyone else.	+1	-1	+1	+2	+1
43	People with Down's syndrome have a right to be heard within society, especially when it comes to decisions that affect them.	+4	+3	+3	+4	+4
44	A family with a child with Down's syndrome is just like any other family.	0	-3	+2	-4	+3
45	Looking after a child with Down's syndrome needs certain qualities I don't think I've got.	-1	+3	0	+1	+3
46	I think mixing children with Down's syndrome into ordinary schools is a good thing.	+2	+1	0	+2	+1
47	People with Down's syndrome are just a bit different from other people.	0	-2	+1	-1	-2
48	People with Down's syndrome are severely mentally disabled.	-2	0	-1	-3	-2
49	People with Down's syndrome should be allowed to have a normal sex life like everyone else.	+1	+1	0	+3	+2
50	People with Down's syndrome should mix together with other people as much as possible.	+2	+3	+2	+4	+3

The remainder of this section gives a factor-by-factor interpretation that was conducted using the recommendations of Kitzinger (1999) to examine; (1) the placing of the ‘strongly agree’ and ‘strongly disagree’ items, (2) the ‘neutral’ items, (3) apparent discrepancies within the factor arrays sort, and (4) apparent differences between item interpretations across factors. To aid interpretation the factor arrays were reconstructed on full size grids. This pictorial representation of the sort pattern enabled the overall understanding to emerge more clearly. Comments made by participants during or after the sorting procedure or written in the response booklets were also used to help interpretation, and these are used here to illustrate the views of participants in addition to Factor scores for individual items.

Factor 1. Down’s syndrome within the continuum of normality

Factor 1 accounted for 35% of the variance explained by the five factors. The Q sorts of 37 participants defined this factor (28 women and 9 men). Twenty-one of the sorts came from the ‘special interest’ group. This group included seven women who had close family member with Down’s syndrome, five people in caring or support professions relating to people with learning difficulties (four women, one man), two male researchers in disability studies, three staff from a cytogenetics laboratory (two women, one man), a female midwife, a male obstetrician, a male genetic counsellor, and one woman whose mother had terminated a pregnancy for Down’s syndrome. The 16 sorts from the ‘no special interest’ group belonged to three men (two computer professionals and a researcher) and 13 women (eight research students, two computer professionals, a probation officer, a teacher, and a youth support worker). In this group were six people who commented that they had a strong religious faith; four women (three Christian, one Hindu), and two men (one Christian, one Muslim). Twenty participants had children, 17 did not. Twenty six had had substantial personal experience of people with Down’s syndrome.

The participants whose sorts defined this factor were most likely to express the view that Down’s syndrome was an integral part of the ‘human condition’; for example, they strongly agreed that people with Down’s syndrome have the same feelings as anybody else. This group strongly agreed with the consensus items relating to rights of people with Down’s syndrome to be included in society, and felt that the biggest obstacle to a good quality of life was the attitudes of others and a society that did not adequately support people with disabilities and their families. One participant wrote;

“Prejudices of others are a huge barrier to achieving potential for any person perceived as different. With appropriate support [people with Down’s syndrome] can learn like anyone else to live independently”.

This group was most likely to disagree that looking after a child with Down's syndrome requires qualities they did not possess, and that affected people are a burden to their families and the state. For example, one woman wrote, "*Children with DS are our own children. How can they be a burden?*"

The item scores of this group indicated that they believed people with Down's syndrome actively contributed to society and their families, and that their presence in the world was a positive one.

Item		Factor 1 scores
20	Knowing someone with Down's syndrome enriches our understanding of what it is to be human.	+2
25	The world would be a worse place if no more babies with Down's syndrome were born.	+1
21	Down's syndrome is an abnormality and an error of nature. It makes sense to try and prevent it.	-2

They believed that being the parent of a child with Down's syndrome was essentially the same as being the parent of any child.

Item		Factor 1 score
34	Saying that having a child with Down's syndrome is as good as a normal child is just denying reality.	-2

In response to Item 29 ("I think you are lucky if you have a person with Down's syndrome in your family") mothers of children with Down's syndrome wrote,

- "*Yes, although it's hard work battling education authorities and social services. On balance though, yes.*"
- "*We as a family feel greatly blessed from having a Down's syndrome baby.*"

The statement that "normal children are just as demanding as children with Down's syndrome" was disagreed with by a number of participants. This may appear to be discrepant with the placing of other statements. However, for those with a close family member with Down's syndrome, disagreement with this item was related to a favourable attitude. For example, the mother of an 18 year-old with Down's syndrome placed this item in the -3 column and commented in her response booklet that, "*Actually, in my experience other siblings are more demanding.*"

Out of all the participants, this group were most likely to agree or remain neutral about the statement that "if you have a child with Down's syndrome it is because God chose you", although just over half disagreed with this. Written comments about this item demonstrated the complexity of religious belief and the attribution of significant life events to God.

- *“Rubbish. Ridiculous.”*
- *“A blessing. Children with learning disability are innocent and will go to heaven, as will their parents. Their parents are prayed for.”*
- *“This statement implies punishment from God and therefore is incorrect.”*
- *“I found this really hard to answer due to my Christian faith. I just don’t know if God chooses everything in life!”*

Summary

In this understanding, people with Down’s syndrome are valued for themselves and for what they can bring to those who know them. The concept of burden is seen as inappropriate, although a child with the condition may well bring challenges. It is felt that the difficulties associated with having a child with Down’s syndrome are mainly due to the attitudes of others, and a society that struggles to genuinely include those with learning difficulties. As one participant said after the sorting procedure,

“It’s a disability, and so I wouldn’t choose it for him and I’m not glad that [he] has Down’s syndrome, but I feel fortunate to have him as a brother.”

Factor 2. Down’s syndrome as a parental misfortune

Factor 2 accounted for 11% of the variance explained by the five factors. The Q sorts of ten participants defined this factor (six women and four men). Three participants came from the ‘special interest group’, a male medical researcher/specialist in prenatal screening, a female GP, and a female genetic counsellor. The group with no special interest in Down’s syndrome included two male research students, two computer professionals (one man, one woman), one housewife, one part-time mature student, and one woman who was a medical secretary. Out of the whole group, five had children, and five did not. Most participants had had little contact with people with Down’s syndrome. However, the genetic counsellor had experience of families of affected children, and she also had an adult cousin with the condition. The participants whose sorts defined this factor were the most likely to believe that people with Down’s syndrome remain totally dependent on others and childlike throughout their lives. Their items scores revealed that they viewed the birth of a child with Down’s syndrome as a very negative event for the parents.

Item	Factor 2 scores
29 I think you are lucky if you have a person with Down’s syndrome in your family.	-4
25 The world would be a worse place if no more babies with Down’s syndrome were born	-4
44 A family with a child with Down’s syndrome is just like any other family.	-3
38 I feel so sorry for people who have a baby with Down’s syndrome.	+3

For this group, having a child with Down's syndrome would be a very difficult thing to accept.

Item		Factor 2 scores
34	Saying that having a child with Down's syndrome is as good as a normal child is just denying reality.	+4
35	For me, having a child with Down's syndrome wouldn't be the end of the world.	-3
28	I would find it as easy to love a child with Down's syndrome as to love any other child.	-2
40	You would get a lot of joy from having a child with Down's syndrome.	-2

Despite this unfavourable view of being the parent of a child with Down's syndrome, the consensus within this group was that should an affected baby be born, adoption and euthanasia were unacceptable options. Two written examples of this view were;

- *"Once the baby is born I think it is important to keep it and deal with the situation"*.
- *"Euthanasia is not right even if the parents wish it. That's murder"*.

An apparent discrepancy between the unfavourable view of parenting a child with Down's syndrome and the favourable attitudes towards the rights and general inclusion in society of affected individuals was seen. However, this, along with the comments regarding adoption and euthanasia can be interpreted as the participants making a clear distinction between persons with Down's syndrome already born and those yet to be born. Most of the items relating to how a person with Down's syndrome might experience their life were placed in the 'neutral' range of scores (-1 to +1) suggesting that beliefs about parenting an affected child were most important to this understanding. However, this group was most likely to comment on how people with Down's syndrome can be subject to intolerance, for example, written comments included,

- *"At a normal school they would be teased and a magnet for bullies"*.
- *"They will get a lot of stick from other kids"*.

These beliefs might reflect the participants' own perceptions of people with Down's syndrome as different. This group was most likely to agree that they found people with Down's syndrome unattractive, and least likely to disagree that affected people made them feel uncomfortable. They were aware that such a view was not necessarily 'politically correct'. As one person wrote, *"I realise that this is ignorance on my part, but I'm just being honest"*.

Summary

In contrast with Factor 1, this understanding of the birth of a child with Down's syndrome is viewed with sadness and as a misfortune for parents. A child with Down's syndrome can never be considered as equivalent to a normal child, and for this reason it is seen as desirable to prevent affected individuals from being born. However, once a person with Down's syndrome is born they

are viewed to have the same rights in life as any other individual including being raised by their birth family.

Mixed factor loading: Factor 2 and flagged negative loading on Factor 1

The Q sort of one participant (an obstetrician, married with children) loaded significantly on Factor 2, and significantly but negatively on Factor 1. PQMethod flagged this sort as an exemplar of an inverted Factor 1. Although in many ways the distribution of statements was similar to that of the Factor 2 Q sorts, there were a number of important differences.

Item		Scores
13	Nobody would choose to have a child with Down' syndrome.	+4
14	Choosing to bring a child with Down's syndrome into the world is just selfish.	+4
11	Children with Down's syndrome are a burden throughout their lives.	+3
47	People with Down's syndrome are just a bit 'different' from other people.	-3
12	Normal children are just as demanding as children with Down's syndrome	-3

In this view, the birth of a child with Down's syndrome is seen as a very negative event indeed.

This obstetrician felt this might be due to his involvement with parents whose child was diagnosed prenatally with Down's syndrome. He said during his pre-sort interview,

"I have been struck by how appalled people are ... maybe I've just seen the patients who are most appalled, that's how I've got involved. You know they've been very angry, almost as upset as if they've had a stillbirth. Of course most of the people I see with a Down's child I see as a result of prenatal testing [who then terminate], not those who carry on."

Unlike the understanding exemplified by Factor 2, in this understanding the distinction between those already born with Down's syndrome and those yet to be born was not clearly made.

Item		Scores
10	If a child with Down's syndrome dies, it might be a blessing	+3
22	I think that euthanasia of babies with Down's syndrome is acceptable if that is what the parents want.	+2

Factor 3. Down's syndrome and the burden of care

Factor 3 accounted for 9% of the variance explained by the five factors. The Q sorts of three participants defined this factor. One, a female researcher in the area of prenatal testing, had some experiences of people with Down's syndrome, the other two (a female clinical cytogeneticist, and a male engineer) did not. All three were married and had children. Like those represented by Factor 1, these participants strongly agreed that they would find it easy to love and be proud of a child with Down's syndrome. They felt that the families of affected children were like any other, and that a child with Down's syndrome could bring much happiness to its parents. However, unlike those in Factor 1, they still felt that as an 'error of nature' it was sensible to try and prevent

children with Down's syndrome being born (while acknowledging that other parents might choose differently). They viewed Down's syndrome as an unchangeable organic condition (rather than a disability due to social reasons) and something that the parents were ultimately responsible for dealing with.

Item		Factor 3 scores
18	For people with Down's syndrome, the biggest obstacle is not their learning disability but the attitudes of others.	-2
6	A problem with children with Down's syndrome is that they will probably outlive their parents.	+2

Summary

In this understanding, participants believed they could parent a child with Down's syndrome well if they had to. However, they foresaw long-term problems for a person with learning difficulty, including the necessity for someone other than parents to care for their child in the future, for example, one participant said, "*Burden is a loaded word – but they do need looking after all their lives*". For this reason they felt it was sensible to avoid the birth of a child with Down's syndrome.

Factor 4. The 'handicapped family'

Factor 4 accounted for 6% of the variance explained by the five factors. The Q sorts of three participants defined this factor, a woman who had terminated a pregnancy for Down's syndrome some years previously, and two men (the manager of a housing scheme for adults with learning difficulties, and a health care researcher/academic, neither had children). All had had some direct contact with individuals with Down's syndrome. These participants did not believe that Down's syndrome was a severe learning difficulty or even a very bad thing for the individual concerned. They were most likely to agree that people with Down's syndrome could achieve, be independent, and potentially have a good quality of life.

Item		Factor 4 scores
37	People with Down's syndrome shouldn't be called 'sufferers'.	+3
49	People with Down's syndrome should be allowed to have a normal sex life like everyone else.	+3

However, these participants were also most likely to feel that being the family of a child with Down's syndrome was probably a 'bad thing'.

Item		Factor 4 scores
44	A family with a child with Down's syndrome is just like any other family.	-4
29	I think you are lucky if you have a person with Down's syndrome in your family.	-3
35	For me, having a child with Down's syndrome wouldn't be the end of the world.	-3

All three participants placed the statement “You would get a lot of joy from having a child with Down’s syndrome” in the zero column. One wrote, “*You get a lot of heartache as well as joy, therefore neutral*”. In particular, this group strongly agreed that the birth of a child with Down’s syndrome would be detrimental to the child’s unaffected siblings. This view appeared to be based on their personal experiences. Two written comments were,

- “[I] agree that normal sibs can suffer. The whole family can be hijacked to run around after the child. The ‘normal’ sibs resent this. I wonder how many?”
- “A couple of adult friends have sibs with DS (now 40-50ish) who found childhood difficult, especially the girl with DS sister”.

Summary

In this understanding, a person with Down’s syndrome could have a happy and fulfilled life, but unfortunately, this would only be at the expense of their family who would have to sacrifice their own quality of life. One person summarised this view in his response booklet; “*Not just a handicapped child but a handicapped family*”.

Factor 5. Special children need special parents

Factor 5 accounted for 8%⁴² of the variance explained by the five factors. The Q sorts of two participants defined this factor, a male scientist working in a cytogenetics laboratory and a female cardiac nurse. The nurse commented she had very little experience of people with Down’s syndrome (the scientist gave no biographical information). This factor was characterised by quite ambivalent views about Down’s syndrome. Participants strongly disagreed that people with Down’s syndrome remained childlike and dependent on others, and in contrast to Factor 4, they did not see that having a child with Down’s syndrome was a ‘bad thing’ for a family.

Item	Factor 5 scores
40 You would get a lot of joy from having a child with Down’s syndrome.	+3
44 A family with a child with Down’s syndrome is just like any other family.	+3
10 If a child with Down’s syndrome died, it might be a blessing.	-4
4 A child with Down’s syndrome is a family tragedy.	-3

However they did not see it as a ‘good thing’ either.

Item	Factor 5 scores
13 Nobody would choose to have a child with Down’s syndrome.	+2
29 I think you are lucky if you have a person with Down’s syndrome in your family.	-2

⁴² Although the previous factor (Factor 4) explained less of the variance than Factor 5, it represented the views of three people as opposed to two, and so is presented first.

Both participants rated as 'neutral' the statement that "The world would be a worse place if no more babies with Down's syndrome were born". The nurse wrote that it was "*hard to say*". However, what most distinguished this factor from others was the strong agreement with the statement that "If I had a child with Down's syndrome I would be worried about people staring at us". When considered with a number of other statements, it might be said that while the participants believed that some parents (perhaps with special qualities) could be happy with a child with Down's syndrome, they did not believe that they could.

Item	Factor 5 scores
1 Children with Down's syndrome can achieve a great deal.	0
2 You can be as proud of a child with Down's syndrome as you can be of any child.	+1
28 I would find it as easy to love a child with Down's syndrome as to love any other child.	-1
45 Looking after a child with Down's syndrome needs certain qualities I don't think I've got.	+3

Summary

In this understanding, participants believed that the birth of a child with Down's syndrome would deprive them of parental hopes and expectations. They anticipated that they might find it difficult to accept the child as they felt they ought (and others might), and therefore they were not 'good enough' to be parents of a child with Down's syndrome. A similar view was expressed by a student midwife in the focus group; "*I think parents that can care for their child with Down's syndrome are marvellous.*" A rather stereotypical view of mothers of children with disabilities as self-sacrificing 'super-mums' has been noted elsewhere (Brookes, 2001; Moyer *et al.*, 1999; Press *et al.*, 1998).

4.4.3 Stability of the Q sort data

It is not a requirement of Q methodology that the reliability of the Q set is formally tested. The view is that values and attitudes are fluid to some extent and that over time people might be expected to sort statements differently. In addition, the construction of the Q sample does not lend itself to the type of psychometric assessment usually applied to psychological tests and measures. Nevertheless, it was felt that some measure of the stability of the data was appropriate. Four months after the initial Q sorts had been administered six participants were asked to carry out another sort using identical materials but without the researcher being present. Correlation coefficients between the first and second sort ranged from +0.69 to +0.88 with an average coefficient value of +0.80. This value is considered an acceptable level of test-retest correspondence (Shaughnessy and Zechmeister, 1994) and is similar to values found in other Q study test-retest assessments (Frank, 1956; Kerlinger, 1986; Steller and Meurer, 1974).

4.4.4 Attitudes towards prenatal testing and termination for Down's syndrome

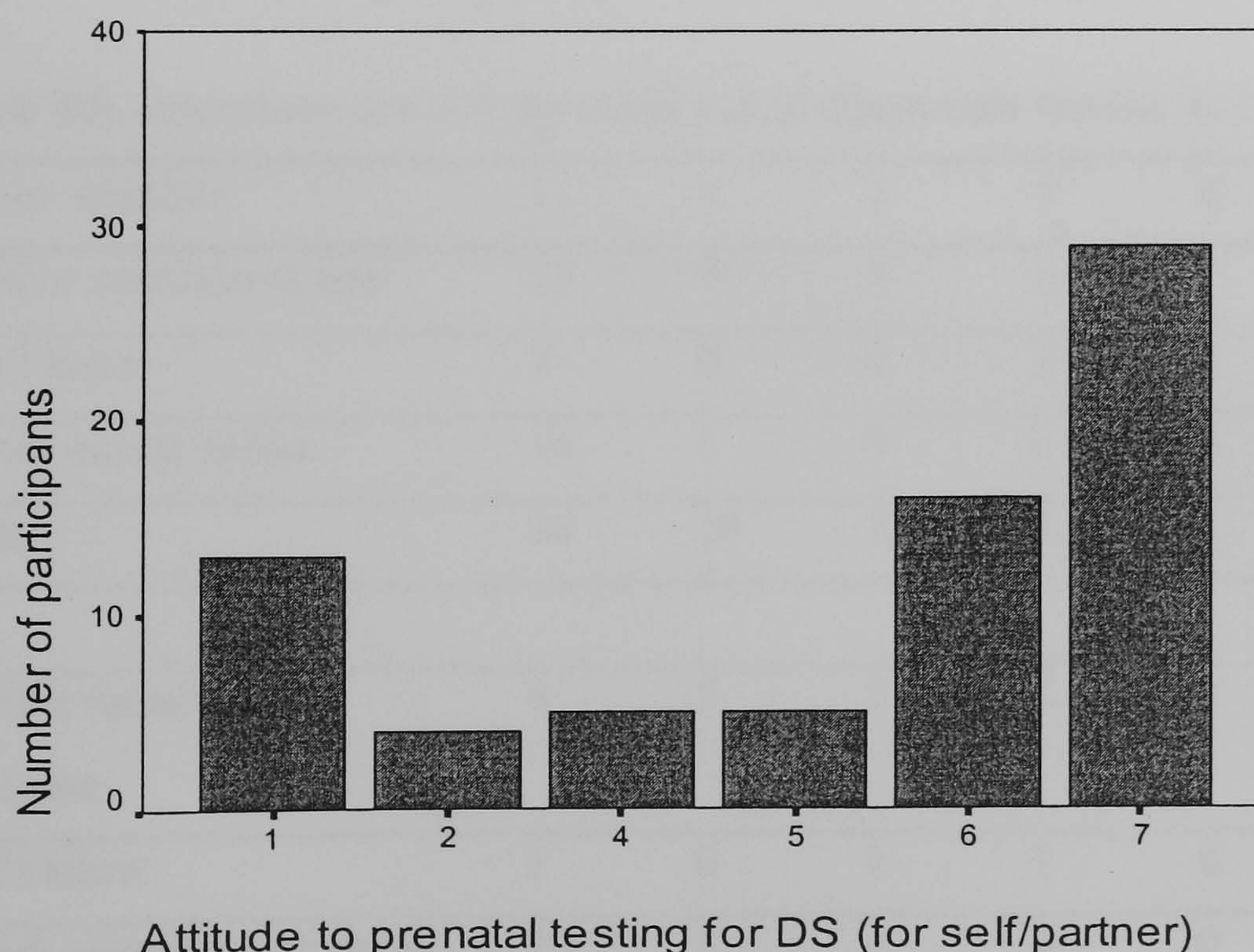
Of the 76 participants, 72 completed the questionnaire relating to attitudes towards prenatal testing and termination for Down's syndrome. Of these, 92% ($n=66$) agreed that prenatal diagnostic testing should be available for everyone who wants it. Those who disagreed generally made reference to the cost implications of 'free availability' rather than to the issue of free choice. However, one woman commented, "I think that people are taking advantage of technology to pursue their own happiness, forgetting their moral duties or standards". Ten individuals did not agree that termination for Down's syndrome should be freely available. Two written comments on this were,

- "Termination on grounds of disability alone devalues the lives of people with Down's syndrome."
- "I do not think of Down's syndrome as being anything like a serious enough problem to even contemplate abortion".

In general however, regardless of their own views most participants agreed that the choice of testing or termination for Down's syndrome should remain with the individual parents.

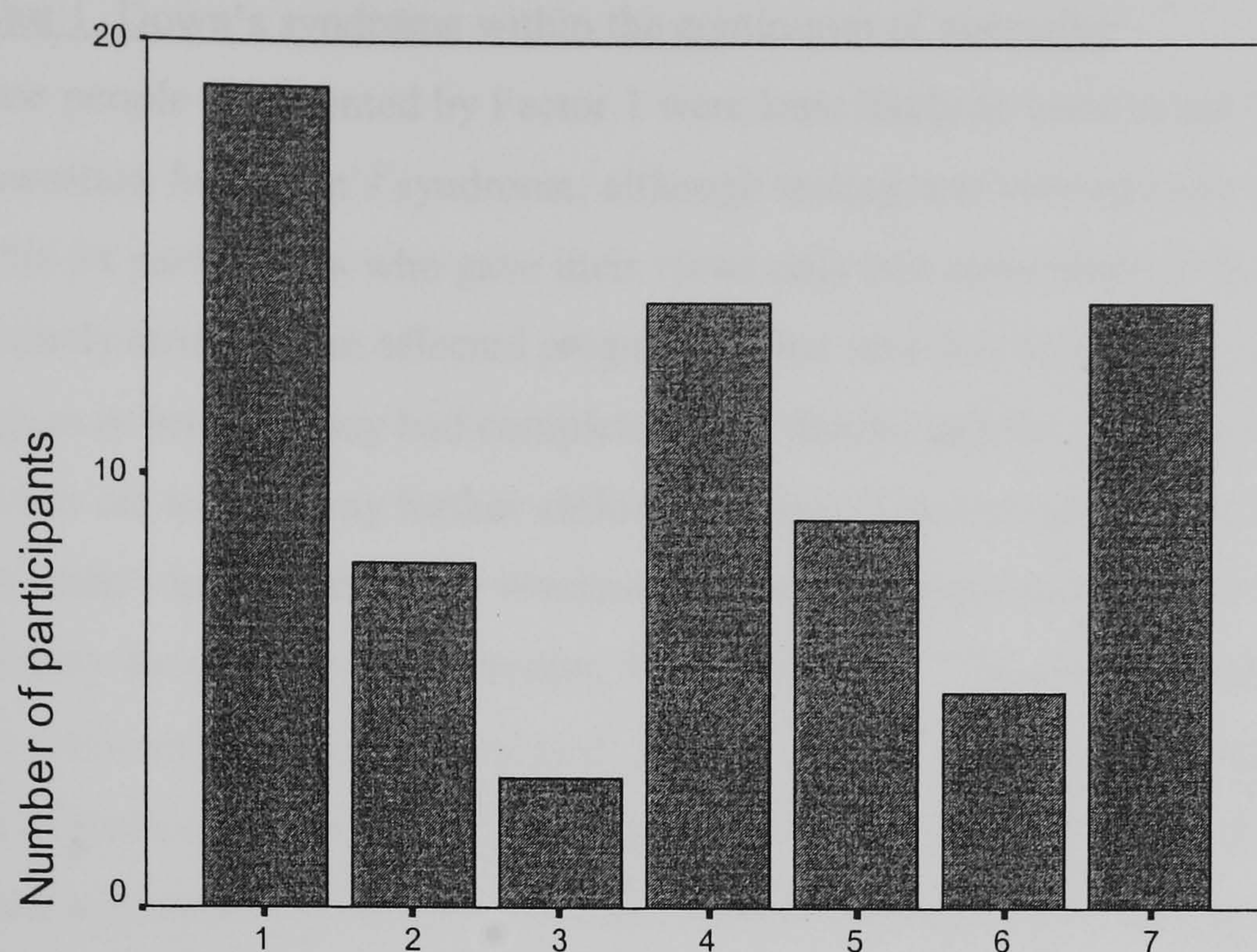
Figure 4.2 and Figure 4.3 give the distribution of scores for participants' intentions towards using testing and termination for Down's syndrome themselves (or their partner if they were male). The distributions are typical of those seen in other studies in that most participants held an intention towards using prenatal testing for Down's syndrome themselves, but intentions towards undergoing termination were more evenly split between unfavourable, favourable, and uncertain.

Figure 4.2: Intentions towards using prenatal testing for Down's syndrome (N=72)



Scale: 1 = definitely do not intend, 4= don't know, 7 = definitely intend (positive intention)

Figure 4.3. Intentions towards undergoing termination for Down's syndrome (N=72)



Attitude to termination for DS (for self/partner)

Scale: 1 = definitely do not intend, 4 = don't know, 7 = definitely intend (positive intention)

Participants were classified by their intentions towards using prenatal testing and termination for Down's syndrome into one of three categories; scores 1 to 3 = 'negative intention', scores 5 to 7 = 'positive intention', and score 4 as 'don't' know'. Table 4.5 gives these intentions by Factor.

Table 4.5. Intentions toward personal use of diagnostic testing and termination by Factor

Factor number	1	2	3	4	5	Mixed ^s	Total
Positive intention to test	17	9	3	2	2	17	50
Don't know	3	0	0	1	0	1	5
Do not intend to test	14	1	0	0	0	2	17
Total	34	10	3	3	2	20	72
Positive intention to terminate	6	9	2	2	2	7	28
Don't know	5	0	1	1	0	7	14
Do not intend to terminate	23	1	0	0	0	6	30
Total	34	10	3	3	2	20	72

^s Includes individual with mixed factor loading: Factor 2, and flagged negative loading on Factor 1

Factor 1. Down's syndrome within the continuum of normality

Those people represented by Factor 1 were least likely to want to use diagnostic testing or termination for Down's syndrome, although testing was viewed more favourably than termination. Of the 34 participants who gave their views only two individuals indicated that they would definitely terminate an affected pregnancy. One said that he and his wife would terminate any pregnancy because they had completed their family, and the other was a woman who at 53 felt she was too old to have any further children anyway. Twelve participants in Factor 1 indicated that they 'didn't know' what they would do regarding termination. One of these, a parent counsellor at a nursery for children with learning difficulties said, "*The decision would be very hard having seen what other parents have faced. Society's attitude is one of the biggest problems*". Just over half of participants in Factor 1 ($n=20$) indicated that they would not terminate a pregnancy for Down's syndrome. These included the mothers of people with Down's syndrome. One mother wrote, "*A DS child brings a lot of joy. Testing, decision-making, abortion: very little joy there*".

A number of other participants emphasised that it was the personal choice of each woman, bearing in mind her circumstances. One woman said she would not terminate for Down's syndrome in her first pregnancy, but might in subsequent ones depending on the needs of her existing children. Of the six participants who had commented that they had a strong religious faith, four indicated that they would not, or probably would not, terminate for Down's syndrome and two were uncertain. One woman wrote, "*I believe in taking things that come in life as a gift of God. We never know what will happen in the future*".

In summary, for those in Factor 1, the views regarding testing and termination for Down's syndrome reflected the understanding of Down's syndrome extracted from the Q sorts. People with Down's syndrome were of value as individuals, and an affected pregnancy was not considered an automatic candidate for termination. However, there was the acknowledgment that society could make life difficult for parents and individuals with Down's syndrome, and for that reason the termination decisions of others should be respected. A number of participants (including health professionals) commented that freedom of choice was an illusion in respect of the context within which Down's syndrome and testing is presented. Comments about 'biased' health professionals, lack of knowledge, lack of information given to women, and the level of support for disabled people in society were cited as reasons why informed choice was not possible in reality.

Factor 2. Down's syndrome as a parental misfortune

All but one of the participants who exemplified Factor 2 said they would use diagnostic testing for Down's syndrome if this was indicated and would terminate an affected pregnancy. For this group the point of having prenatal testing was to allow a termination to take place, and one person wrote *"There is little point in having the test if the information is not going to be used"*. They valued testing because it gave them the chance to avoid having a child with Down's syndrome, which was considered to outweigh the risks of miscarriage associated with a diagnostic test. For example, one participant wrote, *"I would not wish to risk having a Down's baby and should the test result in miscarriage, then I believe it would be for the best"*.

The one exception to this was a woman who had experienced three miscarriages at around 22 weeks of pregnancy. She would not use diagnostic testing partly because of the risk of miscarriage, but also because she felt she could not terminate "that late in pregnancy". However she had used screening tests in all her pregnancies and said, *"If a way could be found to eliminate the chance of conceiving a DS child in the first place, that would be wonderful"*. These attitudes tie in with the view that the birth of a child with Down's syndrome is a sad misfortune for parents because such a child is not considered equivalent to a 'normal' child. For this reason, participants would actively take steps to prevent the birth of an affected child.

Factor 3. Down's syndrome and the burden of care

All three participants who exemplified Factor 3 held positive attitudes to using diagnostic testing for Down's syndrome. Two specifically related prenatal testing to termination, and believed they would terminate an affected pregnancy because of the level of care required in parenting a child with Down's syndrome. For example, one woman wrote, *"I would prefer not to have a child who might make even more in-roads on other aspects of my life than average"*. These views again relate to the understanding that a child with Down's syndrome brings extra responsibilities that are burdensome to some degree. The third participant indicated that they were uncertain about termination but would use testing to "assist in the decision" of whether or not to continue with the pregnancy. This phrase suggests that the pregnancy might be discontinued for other reasons, but it might also reflect the view that testing doesn't require a decision because it is something that increases information and choice in its own right.

Factor 4. The 'handicapped family'

Two of the three participants who exemplified Factor 4 held positive attitudes to using diagnostic testing for Down's syndrome. They specifically related prenatal testing to termination, and

believed they would terminate an affected pregnancy. One wrote, *"I am well aware that people with Down's syndrome have a great variability of ability and potential [but] I personally would not want the responsibility of being the parent of such a person"*. This reflects the views as expressed in the Q sort that while people with Down's syndrome can achieve a great deal, this is down to the efforts of their parents. The third participant wrote that he was uncertain about whether or not he would use testing and termination, *"I think I might want to know, but only because it was available. What about other disabilities?"* This highlights a limitation of prenatal testing that is not always appreciated by consumers - most conditions associated with learning difficulty cannot be diagnosed prenatally. For this man, the benefits of testing for just one condition were uncertain.

Factor 5. Special children need special parents

The two participants who exemplified Factor 5 held quite positive attitudes towards diagnostic testing and termination, but neither was definite about whether they would use them. Both mentioned the risks associated with testing. A nurse, who had had nuchal translucency screening in a recent pregnancy (but had not realised at the time that the test was for Down's syndrome) wrote, *"It is very difficult to answer, but if I had a test I think I would then terminate. My dilemma would be whether to have the test or not"*. Such responses might reflect the ambivalent beliefs about Down's syndrome that these participants expressed in their Q sorts.

Mixed factor participants

It was considered important to consider the views toward diagnostic testing and termination in those participants (eleven men and nine women) whose sort loaded significantly on more than one factor and so were not flagged as exemplars. Five came from the special interest group (three laboratory staff, a midwife, and a teacher), the remainder consisted of six post-graduate research students, four computer staff, a doctor, a researcher, a clerical worker, and a warehouse manager. Eight of these participants had children and 12 did not. Most had had some close contact with people with Down's syndrome, but five said they had no experience and two gave no details. Half of those who had indicated they were uncertain about termination were in the mixed factor group. These seven participants (one with children, six without) all loaded significantly on Factor 1 and one other factor, suggesting that they held both negative and positive beliefs about Down's syndrome. For example, one such participant (female postgraduate student, no children) loaded significantly on Factors 1 and 5 (+0.55 and +0.54 respectively). She had grown to know a young Asian woman with Down's syndrome and her family quite well as part of a research project. Her views reflected her uncertainty about how she would feel about parenting a child with Down's

syndrome, which she felt was directly related to her not yet having had children. She said, *“I feel I can't connect on having a child at all, let alone one with DS”*. The ambivalence or uncertainty in her views is reflected by the following responses.

Item	Scores
20 Knowing someone with Down's syndrome enriches our understanding of what it is to be human.	+4
13 Nobody would choose to have a child with Down's syndrome.	+4
19 If I had a child with Down's syndrome I would be worried about people staring at us.	+2
28 I would find it as easy to love a child with Down's syndrome as to love any other child.	0
35 For me, having a child with Down's syndrome wouldn't be the end of the world.	0

This ambivalence was also reflected in some of the open-ended responses to the question about termination. For example, a scientist from the cytogenetics laboratory (sort loadings +0.49 on Factor 1 and +0.43 on Factor 3) gave a written explanation of his views about using testing (very positive) and termination (uncertain), from which the following statements were extracted in sequence.

“Being the parent of a normal child where both parents work is difficult enough and the burden placed by a Down's child can lead to stress on the whole family”. “Down's is one of the least severe chromosomal defects” “But the syndrome can be severe.” “However, whilst teaching these children, the majority were lovely people!”

The other uncertain (and perhaps ambivalent) participants generally expressed the view that a termination decision could only be made in consultation with their partner and using information available at the time. They felt they would use prenatal testing to provide this information.

It was of interest to note that while the understandings of Down's syndrome were closely tied in with attitudes towards termination, they were less clearly associated with attitudes towards prenatal diagnosis. Across all understandings diagnostic tests were viewed more favourably than termination in terms of personal usage. Those who did not intend to terminate for Down's syndrome but who indicated they would use testing said they valued it for information purposes or because they would want to be prepared for the birth of an affected child.

4.5 DISCUSSION OF THE FINDINGS

This Q methodological investigation had three objectives. Firstly, to explore diversity in subjective understandings of Down's syndrome and identify the important similarities and differences. Secondly, to investigate the relationship between understandings of Down's syndrome and attitudes towards prenatal testing and termination for the condition. Thirdly, to generate hypotheses about these relationships for further research. This section will discuss whether these

objectives were met, what conclusions can be drawn, the limitations of these conclusions, and make suggestions for further research.

In relation to the first objective of the study, five statistically independent understandings of Down's syndrome were identified⁴³. It is not claimed that the understandings identified represent all possible views. Applying the Q sort to a larger sample might reveal others with similar views to the 'mixed factor' participants and so form another independent factor. In addition, a completely new set of views might appear in samples from other cultures. Nevertheless, the study revealed a number of "*competing equivalent stories*" (Eccleston *et al.*, 1997, p. 699) about Down's syndrome, and showed that these stories are based as much on attitudes and values as they are on particular life experiences and factual knowledge. They also suggest that outside of the prenatal testing context the view of Down's syndrome as an abnormality to be eradicated does not necessarily predominate, although it is important to emphasise that conclusions should not be drawn about the actual proportions of people in the general population who might subscribe to the views identified. Participants were purposively selected and therefore over represented people with some expertise of Down's syndrome and health professionals. In addition, the consensus statements were all allied to a favourable attitude towards people with Down's syndrome and might have accounted for the high number of sorts clustering on Factor 1. A further analysis of the data excluding the consensus items (which is not possible using PQMethod) might result in less of the overall variance being explained by one factor.

The use of Q methodology enabled a structured approach to understanding where people's attitudes towards Down's syndrome might be shared and where they are most distinct. The first area of consensus was the stereotypic belief in the affectionate and loving 'Down's syndrome personality'. The consensus suggests that these beliefs are considered 'facts' in relation to people with Down's syndrome and so are unrelated to attitudes. The second area of consensus was belief in the rights of people with Down's syndrome to be included in society, and to receive equal access to medical care and education. While this consensus could be dismissed as a socially desirable attitude of tolerance and acceptance of those with disability and so of little interest, it could also be considered as evidence that people's views about Down's syndrome can be multi-faceted. The belief that people who have Down's syndrome have the right to be included in the community is not necessarily at odds with the wish to terminate an affected child. One belief is concerned with those people with Down's syndrome who are living, the other is concerned with

⁴³ Six views including that of the person whose Q sort loaded significantly but negatively on Factor 1.

those who are yet to be born. In a society where abortion for abnormality is legal until term, it is up to the individual to define when a fetus becomes a person for them. However, the findings demonstrate that to only include items about *existing* people with Down's syndrome in a Q sort (or indeed in a questionnaire or an interview), would be uninformative when trying to unravel the complexities of attitudes toward prenatal testing and termination.

There was greater distinction between the understandings in terms of beliefs about the quality of life of the person with Down's syndrome and in particular, of their parents and siblings. Beliefs about the gains and losses associated with the birth of an affected child were also seen to have greater or lesser salience depending on which factorial dimension the sort showed most commonality with; joy, love, and pride contrasting, and sometimes coexisting, with sadness, disappointment and burden. In Factor 1 and Factor 3 a person with Down's syndrome is seen as a potential or actual family member who, while having a disability, does not define their parents or their family by this disability. In contrast, the other understandings see a child with Down's syndrome as defining the family to some extent by sadness (Factor 2), sacrifice (Factor 4), and disappointment and guilt (Factor 5). In Factor 1, the similarities between people with Down's syndrome and the rest of population are emphasised, while in the others the differences are highlighted.

The second objective of the study was to examine how understandings of Down's syndrome might relate to views about prenatal testing and termination. More than any specific set of beliefs, the varying emphases on similarity and difference shed light on how views about Down's syndrome (which may also include views about disabilities in general or learning difficulty in particular) are linked to attitudes towards prenatal testing and termination for the condition. It might be argued that if a person believes a baby with Down's syndrome to be essentially the same as any other baby, then termination, and perhaps testing becomes less acceptable. In contrast, if a baby with Down's syndrome is believed to be essentially different from other babies, termination and testing are considered more acceptable. This perception of a disabled child as "*the 'other', neither normal, perfect, healthy nor the customary occurrence*" (Press *et al.*, 1998, p. 58) has been noted elsewhere. However, it is not suggested that to view a child with Down's syndrome as essentially different to other children means that the ultimate consequences of prenatal testing are less distressing. The decision to terminate a wanted pregnancy is never taken lightly, and, as the literature demonstrates can be the cause of severe distress even in women entirely at ease with their decision (Green, 1992). Conversely, believing that a child with Down's syndrome is essentially the same as any other child, does not necessarily protect parents from a experiencing loss and grief when they receive a positive diagnosis for the condition. As Press and colleagues

note, women are not generally seeking a perfect baby, but rather hoping for a 'perfectly normal' baby (Press et al., 1998). Strong reactions to both prenatal and neonatal diagnosis of conditions have been reported for even quite minor conditions (Green et al., 2002).

An overarching aim of this thesis is to contribute to the debate on informed choice in prenatal testing for Down's syndrome. It is argued that the findings of this study have implications for the issue of informed choice from two main perspectives - the women's and the health professionals'.

Understanding women's subjectivity

Multiple understandings of Down's syndrome exist within society. None of these views are objective but are based on the individuals' values, knowledge, culture, social position and current situation. These different understandings need to be accepted as equally valid if the choices of individual women are to be supported. When a woman is pregnant, depending on the testing path she takes, her views about Down's syndrome (and disability generally) will be called upon to a greater or lesser degree. The very fact that prenatal testing for Down's syndrome is considered a worthwhile use of health resources assumes that Down's syndrome is something people would wish to avoid. For someone whose understanding of Down's syndrome aligns them with this perception of difference the offer of testing does not challenge their attitudes or require them to act against the situational norms. If religious or moral beliefs about abortion, or concerns to avoid miscarriage override views of Down's syndrome, then these are considered acceptable reasons for opting out of testing and are unlikely to be challenged. However, if someone believes that a fetus with Down's syndrome is essentially the same as a fetus without Down's syndrome then their position could be considered to be at odds with the testing culture. In addition, refusing the offer of a test is not the norm within a paternalistic medical culture where the health professional's defined role is to provide appropriate care in the best interest of the patient, and the patient's role is to accept it. For some women it might be easier to cite beliefs about abortion or miscarriage as a reason to decline testing rather than discuss their views about Down's syndrome. In other cases some women might accept testing in the expectation that a reassuring result will not require any further thought on the issue. For those women who choose to have diagnostic testing because they would wish to prepare for an affected child, potential conflict with the norms of testing culture would only arise should they obtain a positive result yet wish to continue with an affected pregnancy. Prenatal testing is widely accepted in those situations where it is offered, but it is argued that taking this decision as a proxy for attitudes towards termination for Down's syndrome is not necessarily warranted.

Understanding health professional's subjectivity

The findings demonstrated that the midwives, obstetricians and genetic counsellors in this study were not experts on the topic of Down's syndrome: instead they were individuals with their own subjectivity regarding this condition. In addition they held personal views towards testing and termination for Down's syndrome that were generally (but not exclusively) favourable. Although this was not a representative sample, other research also suggests that clinicians hold more favourable attitudes towards testing and termination for Down's syndrome than the wider population (Drake *et al.*, 1996). While the views of the health professionals included in this study are most certainly valid, they are no more or less so than any other view. A strength of Q methodology is that it makes the subjectivity of all views transparent. The Royal College of Obstetricians and Gynaecologists (1996) acknowledge "*most obstetricians have limited experience of the effect of abnormality on children*" (p. 9). This lack of knowledge is not surprising and there is no reason why midwives, obstetricians, or counsellors should have privileged experience of Down's syndrome. It is argued that this is not in itself a problem, but it becomes an issue when a person offers (or is expected to offer) a subjective viewpoint in the guise of 'knowledge' or expert opinion. Bekker (1999) reports how Down's syndrome was described by a midwife in a counselling session; "*well, it's 20 or 30 years down the line when what you've got is essentially a baby still*". This is a point of view, not a fact. Should such subjectivity be allowed to pass for information? In a call for material that accurately reflects all aspects of the condition, Elkins *et al* note that without standardised guidelines counselling about Down's syndrome 'may express only opinions' (Elkins *et al.*, 1986). If many medical professionals think about Down's syndrome in a way that makes termination largely unproblematic, this will inevitably shape the way in which testing services are delivered (Ward, 2002).

The findings of this study demonstrate further that consideration must be given to the provision of information about Down's syndrome in the antenatal context. The main questions are, who should provide it, what should be provided and when and how such provision can be monitored? The findings also suggest that information about Down's syndrome may be of more use to some individuals than others. For those people with strong views on Down's syndrome and definite ideas about their desired testing path, further information about Down's syndrome might not be considered important, but accurate, useful and up-to-date material should be available should they wish to use it. However, should someone be uncertain in their views about Down's syndrome, and less definite about which testing path they would follow, information about Down's syndrome might help them resolve this uncertainty, or at least help them feel more satisfied with their decision process. For some pregnant women any information about Down's syndrome might be

new knowledge, and some women will know far more about Down's syndrome than any leaflet could be expected to cover. For some the information might be irrelevant, for some critical. It is not known in advance who might fall into which category. For this reason, quality information about Down's syndrome should be given equal status to information about the tests and procedures.

Further Research

This Q study was a simple implementation of the method and applied one Q set to one group of participants. However, a number of refinements could be employed should further understandings of Down's syndrome be merited in relation to prenatal testing or for other purposes. Removal of the consensus items might produce more sharply defined factors, or further diversify those in Factor 1. In addition, statements that allowed Factors 3, 4, and 5 to be articulated more clearly could be added. There were a number of items in the original collection of material that resonated with the views expressed in these factors. For example, one category of items labelled 'Saintly Parents' contained statements that reflected views about the specialness of parents of children with Down's syndrome similar to the views expressed in Factor 5. Facilitation of these views might have drawn some of the mixed-factor participants more strongly towards one factor in particular. An alternative approach would be to interview the participants who exemplified the 'smaller' factors to draw out their views about Down's syndrome directly and their understandings of burden, the 'handicapped' family, or the need for special parents. These interviews could have been fed back into a further Q study, or used to help validate and interpret the existing findings. Finally, the Q set could also be applied to a more specialised P set to identify differences within one population. In this study all those with a relative with Down's syndrome clustered on Factor 1. However, the literature shows that not all relatives hold favourable views about Down's syndrome (e.g. Bryant, 1998; Cunningham, 1996; Shepperdson, 1996). It is likely that applying a wider sample of family members to the Q set would reveal a wider diversity of views than was seen here.

The third objective of the study was to generate hypotheses about the relationships between understandings of Down's syndrome and attitudes towards using prenatal testing and termination for the condition. In particular two important question areas emerged. The first related to ambivalent beliefs about Down's syndrome and how these might relate to views about prenatal testing and termination. A number of the views expressed about Down's syndrome contained an element of ambivalence as positive and negative beliefs about the condition coexisted in many individuals. In those with quite high levels of mixed beliefs this was (tentatively) related to a more uncertain attitude towards testing, and to termination in particular. Proportionately more of those in the mixed factor group and the 'uncertain to termination' group had no children compared to

those whose views clustered on one Factor or were more certain about termination. This suggested that ambivalent attitudes to Down's syndrome might be related to some sociodemographic factors and their relationship with different types of experience. It was decided to take this issue forward into another study, and to investigate the role of attitudinal ambivalence towards Down's syndrome in the relationships between testing and termination intentions. It was anticipated that higher levels of ambivalence would be associated with greater degrees of uncertainty about testing and termination. It was also thought that youth and having no children might be associated with higher levels of ambivalence towards Down's syndrome. The second hypothesis related to the finding that understandings about Down's syndrome were more consistently related to views about termination than they were about diagnostic testing. The issue of termination is often not raised with pregnant women before they make their screening test decision, and so the two acts are often not explicitly related in the minds of many women (Press and Browner, 1997). For this reason, it was anticipated that views about Down's syndrome would also be less consistently associated with attitudes towards screening tests than with views about termination, as in temporal terms this test is even more 'removed' from the termination process than is diagnostic testing.

The next empirical chapters (Chapters 5, 6 and 7) describe how these hypotheses were operationalised in a study that examined the attitudes of pregnant women towards Down's syndrome and their relationship with actual prenatal testing choices.

CHAPTER 5 ATTITUDES TOWARDS DOWN'S SYNDROME IN THE PRENATAL TESTING SITUATION: BACKGROUND AND METHODS

5.1 BACKGROUND

The main aim of this thesis is to examine how understandings of Down's syndrome influence prenatal testing choices. The Q study reported in the previous chapter demonstrated that differences in belief patterns exist and that some association with these patterns can be seen with attitudes and intentions towards using prenatal diagnosis and termination for Down's syndrome in hypothetical situations. The next stage was to study how understandings of Down's syndrome relate to actual testing choices in a clinical situation with pregnant women. Q sorting is a labour intensive methodology suited to exploratory research and the generating of hypotheses. It was felt that a different approach was more appropriate to this study where it was important to access a larger sample that was more representative of pregnant women generally. The following three chapters report on a study that measured attitudes towards Down's syndrome and assessed associations between these attitudes, screening uptake, and intentions to use diagnostic testing and termination. This chapter (**Chapter 5**) describes the **theory** and rationale behind the **design** of the study, the study's **objectives** and the **methods** employed to meet these objectives. The following two chapters (**Chapters 6 and 7**) present the **results** of the study and **discuss** their implications.

5.1.1 Attitudes and behaviour

Debate about the structure of attitudes, their purpose, and their consequences for both the individual and society has been a central feature of social psychology for many decades (Ajzen, 2001). There are many different definitions of the concept, but generally an attitude can be said to be an evaluation of some 'object' (such as a person, a group, an event, or a social issue) as favourable or unfavourable (Eagly and Chaiken, 1993). Within this, attitudes are commonly thought to consist of multiple items of information about the object and an evaluation of these items as favourable or unfavourable. The types of attitude information most usually defined are cognitions (thoughts and beliefs about the object), affect (emotions elicited by the object), and behaviour (past behaviours or intentions regarding the object). In recent years this 'tripartite' model has been criticised by a number of authors particularly regarding the inclusion of behaviour as an inherent component of attitudes (Augoustinos and Walker, 1995; Greenwald, 1989a; Greenwald, 1989b). Nevertheless, the idea that attitudes are *informed* by beliefs, emotions and behaviour continues to provide a useful way in which to examine understandings of social objects (Eagly and Chaiken, 1993).

Attitude objects may be very broadly split into two categories: **targets** and **behaviours**. For example, a person might hold a favourable or an unfavourable attitude towards people with Down's syndrome, and they might also hold a favourable or an unfavourable attitude towards terminating a pregnancy for Down's syndrome. While the two attitudes may well be linked they are conceptually distinct. The former would be an attitude towards a **target**, and the latter would be an attitude towards **behaviour directed** at a target. Intuitively, it might be expected that the two attitudes would be relatively consistent: that someone with an unfavourable attitude towards people with Down's syndrome might be expected to use prenatal tests for the condition and to terminate an affected pregnancy. In contrast it might be expected that a person with a favourable attitude towards people with Down's syndrome would not use tests and would not wish to terminate an affected pregnancy. However, it has been demonstrated that this intuitive consistency is not always apparent in real life and that attitudes towards targets do not always accurately predict seemingly related behaviours (Wicker, 1969). Research to identify the factors associated with attitude-behaviour consistency has generally been in one of two areas: (1) the prediction of behaviour from attitudes towards that behaviour, and (2) the prediction of behaviour from attitudes towards the target.

The first approach of predicting behaviour from attitudes towards that behaviour is exemplified by the expectancy-value models of the Theory of Reasoned Action (TRA) (Ajzen and Fishbein, 1980; Fishbein and Ajzen, 1975; Ajzen, 1991) and the Theory of Planned Behaviour (TPB) (Ajzen, 1985, 1988, 1991). In the TRA/PB it is assumed that at some level of conscious thought an intention to act in a certain way precedes actual behaviour, thus the proximal cause of behaviour is considered to be the behavioural intention. Behavioural intentions represent "*a person's motivation in the sense of his or her conscious plan or decision to exert effort to perform the behaviour*" (Conner and Sparks, 1995, p. 122). Attitudes towards engaging in the behaviour are only one of the factors considered to influence behavioural intentions. The other factors are subjective norms (beliefs that important others think you should or should not engage in the behaviour) and, in the TPB, perceived behavioural control (perception of how easy or difficult it is to engage in the behaviour). Expectancy-values attached to each factor determine their relative contribution towards predicting behaviour via the behavioural intention (see Conner and Sparks 1995 for a full explanation of the model). Attitude towards the behaviour is considered to be a function of the perceived consequences of engaging in the behaviour, and the probability that engaging in the behaviour will result in this outcome. We can thus consider how the TPB might explain the behaviour of a woman who is being offered a screening test for Down's syndrome during a booking appointment. The woman may have a positive attitude towards undergoing the

screening test because it appears to offer her a valued outcome (the reassurance of a healthy baby), and she believes that this outcome (a negative screening result) is also highly likely to occur should she have the test. She may also believe that as the midwife is offering her the test, the midwife thinks she should have the test, and that the view of the midwife (as an 'expert') is important to her. Finally, if she is offered the opportunity to be tested there and then, it is very easy to actually have the test. Given these factors the TPB model, for example, would predict that it is highly likely that the woman would accept the screening test for Down's syndrome.

The TRA/PB models have been found to be reasonable predictors of a range of health related behaviours (Ajzen, 2001; Armitage and Conner, 2002; Conner and Sparks, 1995). However, the models have been criticised for excluding factors known to significantly influence action. These include past behaviour, personal moral beliefs, and anticipated decision regret. Prior behaviour is of relevance in the prenatal testing context as women who have a serum screening test in a previous pregnancy appear more likely to choose it again in a subsequent one (Rausch, Lambert-Messerlian, and Canick, 2000). As noted in Chapter 1 personal beliefs about the morality of abortion are also highly relevant to the testing choices of pregnant women. Finally, anticipated decision regret might also influence prenatal testing behaviour. Some women foresee that they might regret having declined testing if their baby is later found to have a disabling condition that could have been identified prenatally, and to prevent these feelings they accept testing (Tymstra, 1989; Tymstra, 1991). Alternatively a person might believe that if they decided to have screening this might result in them having an amniocentesis and they expect that they would regret this if the procedure then caused a miscarriage. This anticipation of regret might override a favourable attitude towards screening as a means of reassurance. Much effort has been directed at evaluating the added impact of these (and other) factors on behaviour prediction in the context of the TRA/PB (Conner and Abraham, 2001; Richard, van der Plicht, and de Vries, 1995)⁴⁴. On a more general level, the TRA/PB has also been criticised because it does not take account of actual barriers to behaviour, or of the broader social and cultural context within which the person and their behavioural options are situated (Conner and Sparks, 1995). A criticism of particular relevance to this thesis is that attitudes towards targets have no formal place in the model and are not seen as important proximal determinants of behaviour (Eagly and Chaiken, 1993). The TRA/PB models thus give no formal explanatory power to the beliefs and emotions that a woman might associate with Down's syndrome and what having a baby with the condition might mean for her, her family, and her unborn child. Yet the perceived importance of these beliefs and feelings,

⁴⁴ A detailed critique the TRA and TPB is outside the scope of this thesis, however see Ajzen, 2001, Conner and Sparks (1995, Chapter 5), and Eagly and Chaiken (1993, Chapter 4).

and an assumption as to their direction, formed the rationale for developing and offering prenatal tests in the first place.

The second approach to understanding the attitude-behaviour relationship has been to focus on the circumstances in which attitudes towards the target best predict related behaviour. The most influential work in this area has been by Fazio and colleagues. Fazio has argued that not all attitudes are equally powerful in their influence on behaviour and that this explains the inability of some attitudes to predict behaviour in seemingly related situations (Fazio, 1989). In particular, attitudes towards a target and a related behaviour are most consistent when the attitudes are based on direct behavioural experience with the object (Fazio, Chen, McDonel, and Sherman, 1982; Fazio and Zanna, 1981; Regan and Fazio, 1977). Attitudes based on direct experience are said to be strong in that the learned association between the attitude object and the stored information and evaluations of that object are more definitively formed. Attitudes based on direct experiences appear to be held more clearly and confidently and to be more readily accessible from memory than attitudes based on indirect experience. Only accessible attitudes are said to exert direct control over behaviour.

Accessibility is usually measured using response-time latency, that is the time taken between presenting someone with the attitude object and them responding with evaluations of that object. The argument follows that the more strongly an attitude is held, the more quickly the attitude comes to mind, and the more likely it is to influence related behaviour and to influence behaviour in an attitude consistent manner. Thus, if a person had a favourable attitude towards Down's syndrome based on direct experience with a close family member with the condition, this is likely to influence prenatal testing behaviour in an attitude consistent way, i.e. they would choose not to have prenatal tests. However, according to this theory, it would be more difficult to accurately predict the prenatal choices of someone who has a favourable attitude towards Down's syndrome based only on indirect experience, say via a television programme. Attitudes based on direct experience also appear to be more stable and less easily changed than those based on indirect experience (Ajzen and Fishbein, 1980). Other factors associated with attitude-behaviour consistency are having substantial knowledge about the attitude object (Fazio and Zanna, 1981) and having had the opportunity to express the attitude repeatedly (Fazio *et al.*, 1982). Both of these factors would be most likely to occur in people who have had direct experience with the attitude object.

Fazio proposed a model of the link between attitudes towards targets and target related behaviour, which was presented as an alternative to the expectancy-value TRA/PB models (Fazio, 1986). In this model, an attitude towards a target (for example, an unfavourable attitude towards Down's syndrome) is automatically activated in the presence of cues related to the attitude object (for example, the offer of a screening test). Once activated, the attitude biases the processing of information relating to the target, which in turn guides how the event is defined (for example, seeing the offer of testing as a positive event). From this definition of the event, the behaviour (in this case acceptance of testing) 'simply follows' on (Fazio, 1986, p. 237, cited in Eagly and Chaiken, 1993). The only other formal input to this process is the perception of norms regarding the situation in which the person is placed. In persons whose attitudes are too weak to be automatically activated, or the cues associated with the target are absent (for example, it is not made clear that a test is for Down's syndrome) normative factors would be the main guide to behaviour. Over a period of twenty years or so, Fazio and colleagues have produced evidence to support the importance of attitude accessibility and direct experience in attitude-behaviour consistency⁴⁵. Eagly and Chaiken (1993) suggest that the key might lie in the volume and range of information that direct experience makes available during attitude construction. In addition to cognitive information, behavioural and emotional information are likely to inform the development of such attitudes, especially if the direct experience takes place over a considerable period of time and in a number of contexts. Attitudes based on more input are less likely to be dramatically changed by a new piece of information, be more stable over time, and so relate more strongly to attitude relevant behaviours. However, Fazio's model has also been criticised on a number of points. In contrast to the TRA/PB models, attitudes towards the target are considered proximal determinants of behaviour. No differentiation is made between action and intention to act. Yet a number of factors might act as barriers to action regardless of the strength of the related attitude. Fazio's contention that only accessible attitudes are activated automatically has also been questioned (Ajzen, 2001; Eagly and Chaiken, 1993). In addition, it is argued that Fazio's model places too great an emphasis on attitudes and subsumes many other variables under vaguely defined 'norms'. Finally, research using Fazio's model has been laboratory based or confined to predicting simple volitional behaviours such as voting in elections. As such, the model can be considered as providing an important but limited contribution to understanding the attitude-behaviour relationship.

⁴⁵ For a review, see Eagly and Chaiken (1993), and for recent developments see Ajzen (2001).

Eagly and Chaiken (1993) argue that while both the expectancy-value approaches and Fazio's model contribute towards understanding the attitude-behaviour relationship, neither approach offers a complete explanation. Expectancy-value models help to understand the proximal causes of behaviours, particularly in situations where choice is more explicit and under conscious control. The work of Fazio and colleagues reaffirms the importance of attitudes towards targets as well as attitudes towards behaviour and helps explain why there is such variation in attitude-behaviour consistency across individuals. The two approaches could be considered complementary: for individuals who have no direct experience of the social object or have limited knowledge on which to base their behavioural choice, attitudes towards the behaviour, subjective norms, and other situational and personality variables might have a proportionately greater influence on behaviour. Eagly and Chaiken have suggested that each theory actually explains a separate stage within one process, and they have proposed a **composite model** that brings together the essential components of each theory (Eagly and Chaiken, 1993, Chapter 4). This model sets out a causal sequence whereby the attitude toward the target affects behaviour by influencing attitude toward the behaviour. Attitude toward the behaviour is then proximally related to behaviour via behavioural intention. In addition, habit, rewards and punishments associated with the behaviour, social norms, and impact on self-identity all have formal predictive roles. This model has not yet undergone empirical testing (to this researcher's knowledge) but is offered by Eagly and Chaiken as an integrative framework in which to consider attitudes and related behaviours. This model will be considered further when the role of attitudes towards Down's syndrome in pregnant women in the prenatal testing situation are discussed in Chapter 8.

Summary

Attitudes towards people with Down's syndrome and/or having a baby with the condition would be expected to have some influence on prenatal testing and termination intentions. However, the influence may not appear to be consistent in an intuitive 'common sense' manner. The research by Fazio and colleagues suggests that for some people the attitude-behaviour consistency will be greater than in others, and that direct experience related to Down's syndrome might influence this. In relation to prenatal testing, research has generally considered behaviour within a binary framework, i.e. using or not using screening or amniocentesis. It can be argued that this approach oversimplifies complex decisions and effectively ignores the relationship between attitudes and behaviour in women who are perhaps uncertain about what testing choices to make. In recent years there has been a move away from conceptualising attitudes as simply favourable or unfavourable evaluations and a greater acceptance that some attitudes have more complex evaluative structures. In particular the concept of ambivalence is currently emerging as an important one, not only for

understanding attitude structure but also for understanding the links between attitudes and behaviour, the way in which attitude relevant information is processed, and for persuasion and attitude change (Ajzen, 2001). Work that highlights the complexity of attitudes towards disability (including the findings of the Q study presented in this thesis) along with the dilemmas that this can cause for pregnant women (Pessione, 2001; Press *et al.*, 1998) suggests that the concept of ambivalence has relevance to understanding the relationship between attitudes towards Down's syndrome and prenatal testing choices. This concept will now be considered in more depth.

5.1.2 Attitudinal ambivalence

It has frequently been assumed that once they are formed, attitudes exist as an entity ready to be summoned up when needed. However, an alternative approach sees attitudes as dynamic and temporary constructions that are not independent of the external context (Wilson and Hodges, 1992). In this view, a particular situation will trigger recall of accessible information that is salient to the individual within that context. The nature of the information accessed will determine the type of attitude. If the information is mainly positive or negative in valence then a favourable or unfavourable attitude is generated. If the information is more mixed in valence then the attitude is said to be 'ambivalent' (Eagly and Chaiken, 1993; Wegener, Downing, Krosnick, and Petty, 1995). Ambivalent information may be most likely to be generated in situations that cause conflict between internal motives and external social norms, or when two different internal motives conflict. For example, someone with an ambivalent attitude towards Down's syndrome may not experience this ambivalence when asked to attend a fund-raising event for people with learning difficulties. Their motives may be to help others and to maintain an altruistic self-image. These motives are compatible and so their positive beliefs about people with the condition remain internally uncontested. However, the same person faced with a decision regarding amniocentesis following a positive screening result might experience a conflict between the motive to be altruistic towards those with a disability and the motive to bear a healthy child. At this point, more negative beliefs and feelings about Down's syndrome may also be accessed, generating an ambivalent attitude towards the condition, and hence towards having an affected child. In a series of interviews with pregnant women, Press and colleagues (Press *et al.*, 1998) noted that the women's views towards disability were "*compartmentalized, self-contradictory, and very much in flux*" (p. 50). In another study it was noted that prior to undergoing amniocentesis, some women 'oscillated' between wanting to accept a child with a disability and wanting to "get rid of the dilemma altogether" (Sjögren and Uddenberg, 1987, p. 192). This might be due to social or personal norms of tolerance towards those with disabilities competing with personal beliefs and

feelings about parenting a child with a disability. Alternatively, it could be due to intra-personal ambivalence towards a disabling condition, perhaps based on direct but conflicting experiences.

When ambivalence is experienced it can act to influence the attitude-behaviour relationship, (Conner and Sparks, 2002) although the direction this influence takes is not always easy to predict (Katz and Glass, 1979). In general, attitudes low in ambivalence appear to be better predictors of attitude consistent behaviour. Ambivalence also appears to play a role in attitude change and information processing and the evidence suggests that ambivalent attitudes tend to be more pliable and susceptible to change in the face of persuasive messages (Armitage and Conner, 2000). In one Canadian study it was found that people with high levels of ambivalence towards Oriental people were most likely to display changes in their attitudes towards a policy for immigration from Hong Kong after reading a persuasive message. They were also more likely to systematically process the message than were people low in ambivalence (Maio, Bell, and Esses, 1996). Most importantly, the research found that ambivalent individuals were influenced by the information they were given about immigration, whereas non-ambivalent people were only influenced by their prior attitudes towards Oriental people. The authors put forward a number of explanations for this finding. Firstly, ambivalent individuals devote more attention to new information in order to reduce psychological tension caused by conflicting beliefs and emotions, and limited evidence was found that systematic message processing did lessen ambivalence. Secondly, they proposed that the ambivalent individuals held more complex schemas of Oriental people and this enabled them to process information more deeply. This is consistent with research showing that ambivalence promotes complexity of attitude relevant thinking (See Eagly and Chaiken, (1993) for review of work by Tetlock and colleagues (Tetlock, 1989). Thirdly, they suggested that ambivalent attitudes reflected beliefs that are less confidently held and so individuals high in ambivalence attended closely to new information in order to attain confidence in their views. The use that people who are ambivalent might have for other types of input has also been investigated in a study examining the effect of 'consensus information' on the attitudes of ambivalent individuals (Hodson, Maio, and Esses, 2001). Consensus information is defined as "*socially derived information concerning the attitudes or behaviour of relevant others*" (Hodson *et al.*, 2001, p. 198). The authors hypothesised that holding ambivalent attitudes towards an object makes individuals more likely to be influenced by this socially derived information. In this study they measured attitudes towards Canadian social welfare policy prior to showing a televised debate between two people on this issue. Beliefs about the outcome of the debate were then gathered. Following exposure to information about other people's views on the outcome of this debate, ambivalent individuals were shown to significantly alter their opinions in line with the direction of the consensus.

The studies reviewed above suggest that ambivalent individuals are more likely to be influenced by factors external to their own attitudes and in particular may seek and use information in order to resolve their ambivalence. This might have implications for the information needs and choices of those women who hold ambivalent attitudes towards testing for Down's syndrome.

5.2 AIMS OF THE STUDY

A systematic and in-depth understanding of the role of attitudes towards Down's syndrome in predicting prenatal testing and termination behaviour is missing from the literature at present. The main aim of this study was to measure and describe attitudes towards Down's syndrome in pregnant women prior to any prenatal tests being carried out and then to investigate how these attitudes related to intentions towards testing and termination for the condition. In addition screening uptake would be measured to assess the degree of correspondence between attitudes, intention, and screening behaviour. Specifically the study had four objectives:

1. To describe attitudes towards Down's syndrome in women in the first trimester of pregnancy.
2. To investigate the relationships between testing and termination intentions, serum screening uptake and attitudes towards Down's syndrome.
3. To investigate the role of attitudinal ambivalence in the relationships between testing and termination intentions, serum screening uptake and attitudes towards Down's syndrome.
4. To identify the variables uniquely contributing to predicting testing and termination behavioural intentions for Down's syndrome, and screening test uptake.

The **conceptual framework** of this study was based on the theory and research as summarised in the previous two sections. A number of assumptions within this framework informed the selection of the attitude measures and the interpretation and discussion of the data collected:

- Attitudes are considered to be evaluations of objects based on multiple sources of information, in particular, beliefs, emotions, and experiences associated with the attitude object.
- It is assumed that while behaviour may inform attitudes, behaviour is an independent construct and not a *component* of the attitude structure as such.

- It is assumed that attitudes towards targets are conceptually different to attitudes towards behaviours. The focus of this study was primarily the attitude towards the target, in that beliefs, emotions, and experiences that a participant associated with the target condition of Down's syndrome were measured. It was believed that these items would inform attitudes toward having a child with Down's syndrome, and therefore contribute towards understanding the choices that women make regarding prenatal testing for Down's syndrome.
- The attitudes of interest in this study were towards having a baby with Down's syndrome, and attitudes towards the condition of Down's syndrome. Within the context of prenatal testing this means an evaluation in terms of stereotypical characteristics of the condition, feelings generated by seeing or meeting individuals with Down's syndrome, and their experiences with affected people. This is a slightly different approach to looking at attitudes towards people with Down's syndrome although clearly there is substantial overlap. The issue of interest is not how attitudes affect a participant's behaviour towards existing people with the syndrome, but how attitudes affect potential behaviour toward the participant's unborn child. Therefore it is considered that the attitudinal information accessed would be constructed within the context of considering having an affected child. Although it could be argued that an attitude towards having a baby with Down's syndrome is an attitude towards a behaviour, the sources of information that inform this evaluation are likely to be attitudes towards the condition.
- In line with the expectancy-value models of the attitude-behaviour relationship, it is assumed that behavioural intentions regarding an attitude object generally precede the behaviour at some level. In the context of this study, behavioural intentions regarding serum screening, amniocentesis and termination for Down's syndrome are therefore assumed to be measurable constructs in their own right. In addition, most women in this study would not have to make actual behavioural choices regarding amniocentesis and termination, and therefore, only their hypothetical behavioural intentions regarding these situations could be measured.
- Although attitudinal ambivalence may be a contributing factor towards attitude strength, ambivalence is assumed to be an independent construct with potential for moderating the attitude-behaviour relationship and influencing information processing.

Finally, psychology has been criticised for emphasising the individual aspects of attitudes at the expense of the social (Augoustinos and Walker, 1995). While the empirical aspect of this study measures the individual aspects of attitudes, it is also assumed that attitudes are social, inter-personal constructions as well as intra-personal ones. It is accepted that attitudes are developed and acted upon in a particular social and cultural context, and as such have social consequences.

The following section details the rationale for selecting the measure of attitudes toward Down's syndrome within the aims of the study and the conceptual framework as outlined above.

5.3 ATTITUDE MEASURES

Attitudes are hypothetical constructs and thus cannot be directly observed but only inferred from other responses (Ajzen, 1988). The most commonly measured expressions of attitude are verbally expressed cognitive responses (beliefs) and affective responses (emotions). These have been shown to reliably relate to overall evaluations of an attitude object. In addition behavioural acts and intentions towards the attitude object or experiences with the attitude object are sometimes measured. There are many ways to measure attitudes and a large literature within social psychology is devoted to this topic⁴⁶, however, the most commonly used method of accessing attitudes has been the use of self-report measures or questionnaires.

Within self-report measures, questions can be very broadly classed as 'closed' and 'open-ended'. Closed questions provide a range of responses from which the participant selects the option most appropriate to their own views and feelings. Open-ended or free-response questions allow the participant to select their own choice of words to express their views and feelings. The questioning methods that have been dominant in the measurement of attitudes are closed-ended – particularly semantic differential scales (Osgood, Suci, and Tannenbaum, 1957) and Likert scaling (Likert, 1932). Semantic differential measures consist of sets of bipolar adjectives anchored at either end of a rating scale, for example, 'easy-going' and 'difficult'. Respondents are asked to rate the attitude object (for example, people with Down's syndrome) by selecting the point between the anchors that most closely represents their own belief. The meaning of the response is assumed in advance, i.e., someone with a positive attitude towards people with Down's syndrome should be more likely to select a point close to the 'easy-going' end of the scale. With Likert scales, participants are presented with words or phrases pertaining to the attitude object, for example "People with Down's syndrome should have the right to marry if they wish". Respondents are then asked to express the degree to which they agree or disagree with the statement on a scale usually anchored with 'strongly agree' and 'strongly disagree' with a neutral option at the midpoint. Again the meaning of each end of the scale is pre-determined. The advantage of closed measures is that they are easy to score objectively and they allow direct comparison across participants on individual questions. The disadvantage is that the researcher defines what are the most appropriate terms or statements to use. This type of measure has been used most frequently in studies

⁴⁶ A detailed consideration of these methods is outside the scope of this thesis, however, see Eagly and Chaiken, 1993, Chapter 2 for a review.

assessing attitudes towards disability (as discussed in Chapter 1). By contrast, open-ended measures are often more difficult to score, but they allow for more complex and idiosyncratic responding and are not subject to the researchers definitions of what is important or relevant.

While a number of existing measures with closed items were considered for this study, for the methodological reasons given in Chapter 1, none were considered appropriate. However, it was felt that the development of another scale with closed items (even one developed using items from the Q study) would only replicate the known methodological problems. Despite this, it was considered important to use measures of attitudes that were psychometrically acceptable as well as valid in terms of capturing subjectivity. Open-ended measures of attitudes previously used in studies of intergroup attitudes were selected as most appropriate to the aims of the study as well as having properties of reliability and validity.

Open-ended measures of attitudes

The attitude measures selected for use in this study were originally developed by a group of Canadian researchers in order to examine prejudice towards, and stereotyping of, minority groups (Esses and Zanna, 1989; Esses, Haddock, and Zanna, 1993). Studies using these measures have been used to assess the differing contributions of cognitive, affective, and behavioral information in predicting attitudes toward people grouped by gender and political affiliation (Eagly, Mladinic, and Otto, 1994), sexual orientation (Haddock, Zanna, and Esses, 1993), race (Donakowski and Esses, 1996; Eagly *et al.*, 1994; Esses *et al.*, 1993; Haddock *et al.*, 1994), and disability (Esses and Beaufoy, 1994; Kiger, 1997). The measures assess each class of attitudinal information using a structured free-response format. The following sections describe how the measures are used and then go on to summarise their psychometric and methodological properties.

Measuring the cognitive aspect of intergroup attitudes. Open-ended measures of beliefs have a historical grounding in the ‘thought-listing’ procedure that been used elsewhere to assess the cognitive component of attitudes (see Esses and Maio, 2002). For example, the use of free-response measures to determine the ‘modal salient beliefs’ associated with an attitude object was advocated by the developers of the Theory of Reasoned Action (Ajzen and Fishbein, 1980). The assessment of stereotypic beliefs has generally been central in the assessment of the cognitive component of attitudes towards social groups. Stereotypic beliefs relate to the characteristics attributed to typical members of a target group (e.g. the belief that group members are friendly or lazy). To measure stereotypic beliefs using the open-ended format, respondents are asked to list the characteristics they would use to describe a member of the target group and then to allocate a

'valence' to each response on a measurement scale. The valence is the degree of favourability with which the response is viewed and ranges from the very positive to the very negative (for example, from +2 through to -2). A 'stereotypic belief score' is then computed by summing the allocated valences and dividing this value by the number of responses.⁴⁷

Measuring the affective aspect of intergroup attitudes. The affective component of the measures considers the emotions that members of the social group elicit in respondents. For example, a typical group member might elicit fear, admiration or both. Research shows that emotions contribute to the prediction of attitudes over and above the amount predicted by cognitive measures alone (Haddock and Zanna, 1998). To measure affective responses using an open ended format, respondents are asked to list the feelings and emotions they experience when they see, meet, or think about a member of a social group, and to list as many responses as they can. They are then asked to give each feeling an evaluative rating from 'very positive' to 'very negative' and an emotion score is computed as described previously.

Measuring the behavioural aspect of intergroup attitudes. Behavioural attitude information in terms of past experiences with the target group has been found to uniquely contribute to the prediction of inter-group attitudes over and above cognitive and affective information (Haddock *et al.*, 1993; Haddock *et al.*, 1994). Haddock and Zanna (1998) note that "*given the individualized nature of human behavioural experiences, the behavioural component of attitude is particularly well suited for the use of open-ended measurement strategies*" (p. 140). The open-ended measures are concerned with the evaluations of experiences, that is their 'quality' rather than the frequency of their occurrence. Quality of contact is measured by asking respondents to list salient experiences with the target group and then to evaluate each experience on a scale of 'very positive' to 'very negative' and an experience score is computed as described previously.

Psychometric properties of the open-ended measures. The open-ended measures described above have been shown to be reliable and valid measures of attitudinal components. In a study measuring attitudes toward fourteen different targets, Haddock compared the psychometric properties of open-ended measures of cognition and affect with those of semantic differential scales (reported in Haddock and Zanna, 1998). Using the split-half method of reliability, Haddock found the two different measures had comparable internal consistency. The open-ended measures were also shown to display both convergent and discriminant validity, and a higher level of

⁴⁷ The equation is written as $(\sum v)/N$, where v = valence and N = the number of responses.

discriminant validity than the semantic differential scales. In addition, Eagly, Mladanic, and Otto (1994) computed Cronbach's alpha coefficients for open-ended measures and semantic differential scales when measuring attitudes toward four different target groups. They reported satisfactory alpha values for the open-ended measures, which were comparable to those of the semantic differential scales. Good internal consistency values for both cognitive and affective open-ended measures were derived. Despite the concerns of some that open-ended measures may be less reliable than traditional measures, a review of the survey research method reports that the reliability of open-ended questions might actually exceed those of the closed type (Krosnick, 1999).

Methodological aspects of the open-ended measures. The advantages to using free-response measures have been reviewed in detail elsewhere (Eagly *et al.*, 1994; Esses and Maio, 2002; Haddock and Zanna, 1998) and the following section highlights only the main points relevant to the context of the current study. As noted previously, closed items measure beliefs, feelings and behaviours using pre-determined values deemed by the researcher as important and appropriate to the subject under examination. Thus they do not capture the idiosyncratic responses that help to understand a participant's evaluations of the attitude object. Open-ended measures however 'free the researcher' from having to choose the items *a priori*, thus avoiding the selection of ones that do not validly represent participants' attitudes (Stangor and Lange, 1994). This is particularly important in the area of attitudes towards disability where frequent changes in the terms and labels used means that pre-defined measures can quickly become outdated and even offensive. The responses given to open-ended measures are also believed to be more likely to express attitudinal information that is salient to the current context. This is true for both the descriptive response and the associated valence. For example, a person might consider someone with Down's syndrome to be 'trusting'. They might consider this to be a neutral or a positive attribute in general, but when considering their potential child they might associate 'trusting' with vulnerability and evaluate this unfavourably. It has also been argued that freely-responding more accurately reflects the degree of association between objects and their attributes in terms of attitude accessibility and information stored in memory (Eagly *et al.*, 1994; Stangor and Lange, 1994). Closed measures do not require the respondent to have considered in advance the issue to which the question refers, yet they also effectively remove the option of non-responding. Where participants have no evaluation of a particular item they may therefore construct a response that is consistent with their others. In contrast, when using open-ended measures Eagly and colleagues suggest,

"There is no need for respondents to invent new beliefs and affects if they have stored beliefs and affects that they can readily access, and they are free to list as few (or many) beliefs and affects as come to mind" (Eagly *et al.*, 1994, p. 118).

While it has been proposed that open-ended measures are too effortful for many participants (Krosnick, 1999), Esses and colleagues claim the measures are easy to use and that less than 10% fail to complete the measures correctly (Haddock and Zanna, 1998).

While there is no strong evidence that open-ended measures are better predictors of attitudes or behaviour than more traditional measures, in some circumstances they may be more suited to the objectives of the study (Esses and Maio, 2002; Stangor and Lange, 1994). However, in studies where the aim is to make direct comparisons between participants on the same content, closed measures would be more suitable. In this case, the aim was to capture the attitudes towards Down's syndrome that women brought with them to the testing situation, in order to see how individual attitudes related to prenatal testing choices. Should a participant provide none or very few responses using the open-ended measures this in itself could be important data within the context of informed choice. Open-ended measures allow for both quantitative and qualitative content analysis which is highly desirable when the attitudes under examination have not been explored previously in a particular context (Esses and Maio, 2002). An added advantage was that these measures have also been used successfully to measure attitudinal ambivalence towards social groups.

Using open-ended measures to measure attitudinal ambivalence. In order to minimise forced choice responding, it is common practice in attitude research to include a 'don't know' response option, or a neutral midpoint on a Likert scale, for example. However, selecting this midpoint or 'don't know' option might indicate a number of psychological states - a neutral view, an indifferent view, a desire to keep one's view or intention private, uncertainty due to a lack of knowledge, or an ambivalent view towards the item in question (Breckler, 1994; Gilljam and Granberg, 1993). Using traditional measures of attitudes it is not possible to indicate both agreement *and* disagreement with the item, thus attitudinal ambivalence is effectively obscured. A number of tools designed specifically to measure attitudinal ambivalence have been developed but generally using closed responses (Riketta, 2000; Thompson, Zanna, and Griffin, 1995). However, researchers working with Esses and colleagues have developed a formula to calculate ambivalence using the open-ended measures described above, and this formula was used here.

Using the open-ended measures, ambivalence towards an attitude object is calculated from the valence ratings assigned to each response. For each attitude component (i.e. stereotypic beliefs, emotions) the frequency of the positive valences (*P*) and the negative valences (*N*) are collected.

These are then input to the formula $(P + N) - 2 |P - N| + C$, where C is a constant added to preclude negative ambivalence scores (Bell, Esses, and Maio, 1996)^{48,49}. The ambivalence rating of each component is calculated separately and then a mean ambivalence score is computed. This score reflects the degree of conflict between the favourable (positive) and unfavourable (negative) dimensions of the attitude. The higher the score the greater the conflict or ambivalence is considered to be. In terms of validity, the score produced by the formula possesses the qualities identified as desirable in a measure of ambivalence (Thompson *et al.*, 1995). Firstly, as the difference between the negative and positive dimensions increases, the attitude becomes more polarised and the ambivalence score decreases, secondly as negative and positive dimensions become closer, ambivalence increases, and thirdly when dimension scores are equal, ambivalence increases as the dimension scores increase (see also Esses and Maio, 2002). This formula has been used to study ambivalence in attitudes towards minority groups (Bell and Esses, 1997; Bell and Esses, 2002; Bell *et al.*, 1996; Maio *et al.*, 1996) and towards social policy issues (Maio, Esses, and Bell, 2000).

Summary

This first section of this chapter provided the theoretical background to the study, described the conceptual framework within which it is set, and laid out the aims and objectives of the research. The rationale behind using open-ended measures of attitudes has also been presented along with a summary of the way in which the measures have been used previously. The next section (Method I) explains how the open-ended measures of attitudes were adapted to make them appropriate for both the aims of the study and the target population.

5.4 METHOD I: CREATING THE OPEN-ENDED MEASURES OF ATTITUDES TOWARDS DOWN'S SYNDROME

The measures were based on those used in previous studies investigating attitudes towards minority groups (Esses and Maio, 2002). They were designed to be as similar as possible to the ones used by Esses and colleagues but were adapted in a number of ways where it was considered necessary. The following section describes the measures of Down's syndrome used in this study and highlights where these measures differed from the originals.

⁴⁸ The straight brackets indicate that the absolute value of P-N is used in this part of the equation.

⁴⁹ This constant is calculated by multiplying the positive end of the score range, for example +2, by the maximum number of open-ended responses allowed, for example 12.

5.4.1 Open-ended measures of attitudes towards Down's syndrome

Measuring cognitions

In order to capture stereotypic beliefs associated with Down's syndrome respondents would be asked to write down words or phrases that 'came into their heads' when they thought about a person with the condition. They would then evaluate each characteristic using one of four options.

- If the characteristic was considered to be favourable, respondents would put a plus sign against their response. If the characteristic was considered to be very favourable they would put two pluses against the response. One plus sign was scored as +1 and two pluses as +2.
- If the characteristic was considered to be unfavourable respondents would put a minus sign against their response. If the characteristic was considered to be very unfavourable they would put two minuses against the response. One minus was scored as -1 and two minuses as -2.
- If the characteristic was considered to be neither favourable nor unfavourable, respondents would put a zero against their response.
- If the characteristic was considered to be both favourable and unfavourable, respondents would put both a minus and a plus sign against their response. This was scored as a zero. Use of this response would indicate some ambivalence, which is not taken into account in the attitude scoring, however, an ambivalence score was calculated separately. This 'mixed' evaluation was not part of the original measures but was added here as a result of feedback from the pilot study (see section 5.5.5).

In addition to cognitions based on stereotypes, Esses and colleagues have argued that measuring more abstract, value-based beliefs is also relevant in assessing the cognitive component of attitudes towards social groups. This argument is based on research into racism where perceived dissimilarity of intergroup values separately predicts attitudes (see Esses *et al*, 1993 for discussion). Value-based beliefs are said to represent the perceived relationship between the social group, and the values and norms esteemed by the respondent. An example would be the belief that as a group, people who are homosexual threaten family values. In the context of this study it was felt that pregnant women might find these abstract beliefs difficult to generate in relation to people with Down's syndrome and irrelevant in the context of prenatal testing. However, it was agreed that measuring only stereotypical beliefs may not access important cognitions relevant to an attitude towards having a baby with Down's syndrome. The Q-study (Chapter 4) had demonstrated that beliefs about how a child with Down's syndrome would affect their parents' lives discriminated between groups in terms of testing and termination intentions. Dorothy Wertz has similarly noted that women's decisions about prenatal testing and termination involve evaluations of the quality of life of themselves, their family, and their child were it to be born (Wertz, 1992).

Values placed on life-goals such as ‘success’, and ‘pleasure and relaxation’ have also been shown to significantly relate to hypothetical intentions to test and terminate for conditions associated with learning difficulty (Evers-Kiebooms *et al.*, 1993). For these reasons it was considered important to measure these cognitions within the open-ended measures of attitudes.

Respondents would be asked to write down the things that were most important to them in their life and to evaluate how each ‘valued life object’ would be affected by having a baby with Down’s syndrome using the options of positive, negative, neutral or mixed. These cognitions were labeled **Parental Quality of Life beliefs (PQoL)**. The term ‘quality of life’ referred to a subjective experience of well-being and life satisfaction, that encompasses physical well being, material well being, social well being, and productive well being (Felce and Perry, 1997). PQoL beliefs were defined as beliefs that having a child with Down’s syndrome would ‘promote or threaten’ valued aspects of the respondent’s life. For example, a person may value their relationship with their partner and believe that having a child with Down’s syndrome would ‘put pressure on’ (threaten) this relationship. Alternatively, a person might believe that having a child with Down’s syndrome would ‘strengthen’ (promote) their marital relationship.

Measuring emotions

In Chapter 1 the limitations of the ‘comfort scales’ typically used to assess the affective component of attitudes towards people with disability was discussed. It was anticipated that using an open-ended measure of the emotional content of attitudes towards Down’s syndrome would allow for a richer analysis of this component. Respondents would be asked to write down the feelings that they experienced when they saw, met or thought about people with Down’s syndrome and to evaluate each response as positive, negative, neutral or mixed as described above.

Measuring experiences

It is believed that experiences of people with Down’s syndrome can be very important to the construction of a person’s attitudes towards the condition. As such, a measure of experiences was considered essential to this study. Respondents would be asked to think about any experiences they had had of people with Down’s syndrome and to write down as many or as few experiences as they wanted to. They would be encouraged to include those experiences that were most salient, that is to say, those that ‘stood out most strongly’ in their minds and then to give each experience an evaluation of positive, negative, neutral or mixed as described above. In contrast to the way that the experiences measure had previously been operationalised (Haddock *et al.*, 1993; Haddock *et al.*, 1994) it was decided not to ask for recent experiences only, as it was felt that these might not necessarily be the most important experiences in terms of attitude formation.

5.4.2 Open-ended measures: creating some example responses

Open-ended measures of attitudes were selected with the aim of capturing the respondents' own representations of Down's syndrome, nevertheless, it was felt that examples of the type of response required were necessary. These measures are somewhat novel and participants were unlikely to have encountered them before, yet they had to understand the nature of the responses required for the measures to be useful. To minimise experimenter effect (Rosenthal, 1966) it was decided that these examples should not be generated by the researcher and so a data collection exercise was conducted with staff members at the School of Psychology and the Mother and Infant Research Unit at the University of Leeds.

Materials and procedure

A short questionnaire (see Appendix 5) based on the open-ended measures was sent via internal mail to staff members. Both academic and non-academic staff members were included and 82 questionnaires were sent out. Responses were anonymous. Respondents were asked to write down stereotypic beliefs about a 'typical' person with Down's syndrome, feelings that might be elicited by people with Down's syndrome, and beliefs about how having a child with Down syndrome might affect their life. It was explained that the responses were to be used to generate examples for an open-ended attitude survey, and that participants could also give responses that they thought other people might use. It was not thought useful to generate example 'experiences' due to the idiosyncratic nature of such encounters. Thirty-one questionnaires were returned (38%).

Results and example selection

Stereotypes. A total of 229 stereotypic beliefs were collected, a mean of 7.4 per respondent. The most common beliefs were that people with Down's syndrome are affectionate ($n=17$), have learning difficulties ($n=10$) are happy, and are small ($n=9$). The responses were classified into categories similar to those used in the leaflet analysis (Chapter 3). However some categories were modified and two new ones added to reflect the responses to the questionnaire. Categories are defined below (Table 5.1).

Table 5.1. Stereotypic belief categories.

Appearance. Responses relating to the physical phenotype of people with Down's syndrome, such as the epicanthal fold and short stature.

Care requirements. In the leaflet analysis this category was termed 'psychosocial aspects of parenting' but it was renamed to encompass responses relating to caring in general, which will frequently, but not exclusively be, the role of the parents of people with Down's syndrome

Table 5.1 continued.

Learning difficulty. Responses referring mainly to cognitive impairment, but also to positive cognitive abilities associated with people with Down's syndrome.
Medical aspects. Responses relating mainly to health problems, for example, heart defects, which are associated with Down's syndrome.
Personality/behavioural phenotype (P-B phenotype). Responses which related to the behaviours and personality traits that have historically been associated with the perceived personality/behavioural phenotype of people with Down's syndrome (Collacott <i>et al.</i> , 1998; Wishart and Johnston, 1990).
Psycho-social aspects. Responses relating to potential consequences of the interaction between the person with Down's syndrome and their physical and social environment (Shiloh and Berkenstadt, 1992). For example, 'they suffer prejudice'; 'they are dependent on others' or 'have a lower quality of life'
Differentness. Responses that referred to people with Down's syndrome as different from the rest of the population or as similar to each other. This category excluded responses that specifically referred to morphological difference or sameness, which were placed in the 'Appearance' category.

Table 5.2 shows frequencies and sample responses for each category.

Table 5.2. Stereotypic belief categories and sample responses

Category	Sample responses	No. (%)
Appearance	Chubby, slanty eyed, stunted	37 (16)
Care requirements	Demanding, hard work, needy	9 (4)
Learning difficulty	Learning difficulties, slow, intelligent	23 (10)
Medical aspects	Heart problems, short life	13 (6)
P-B phenotype	Happy, loving, moody, stubborn	126 (55)
Psycho-social aspects	Dependent, rejected, lonely, at risk	14 (6)
Differentness	Alien, different	7 (3)
<i>Total</i>		229 (100)

It can be seen that responses relating to the perceived personality/behavioural phenotype of people with Down's syndrome were most salient followed by physical appearance and learning difficulties. Thirty-two examples were selected for the open-ended measure of stereotypic beliefs (see Table 5.3). An attempt to balance positive, negative and neutral examples was made although

this classification was subjective on the behalf of the researcher. It was considered important to include 'negative' examples to facilitate honest responding, however terms that might have caused offence were avoided.

Table 5.3. Examples selected for the open-ended measure of stereotypic beliefs

Category	Examples selected	No. (%)
Appearance	*Attractive, Plump, Short, Unattractive	4 (13)
Care requirements	Demanding, Difficult	2 (6)
Learning difficulty	Learning problems, Slow	2 (6)
Medical aspects	*Healthy, Unhealthy	2 (6)
P-B phenotype	Aggressive, Capable, Childish, Clumsy, Emotional, Friendly, Fun, Good-natured, Happy, Huggy, Innocent, Kind, Loving, Noisy, Stubborn, Trusting, Uninhibited	17 (53)
Psycho-social aspects	Dependent, *Independent, Vulnerable	3 (10)
Differentness	Different, *Ordinary	2 (6)
<i>Total</i>		32 (100)

* These examples were not found in the responses of the pre-study group but were included to provide balance within the categories.

Emotions. A total of 147 emotion responses were collected, a mean of 4.7 per respondent. The most common responses were that people with Down's syndrome elicited feelings of curiosity ($n=9$), sadness ($n=9$), being uncomfortable ($n=9$), and of feeling sorry for the affected person ($n=8$). The responses were classified into categories using Plutchik's classification of primary and secondary emotions, which encompasses most common feeling states (Plutchik, 1994). The categories are given in Table 5.4 along with frequencies and sample responses from each category. A category of 'feeling fortunate' was added to the classification. Although not an emotion exactly, this was an 'emotion' response given by a number of respondents in this exercise, and also in a previous study of attitudes towards various disabled groups that used the open-ended measures (Esses and Beaufoy, 1994). Seven responses were considered unclassifiable, for example the response "Intellectually unable to communicate with them", and these unclassified responses are not included in the table.

Table 5.4. Emotion categories and sample responses

Category	Sample responses	No. (%)
Acceptance	Happy they are part of the community	2 (1)
Anger	Hatred, Irritation	4 (3)
Disgust	Disgust, Horror	4 (3)
Fear	Confused, Intimidated, Scared	18 (13)
Feeling fortunate	Lucky, Appreciative of own health	5 (4)
Guilt	Guilty, Obligated	6 (4)
Interest	Curious, what is their life like?	9 (6)
Joy	Amused, Happy	11 (8)
Love/concern	Friendly, Loving, Protective, Sympathetic	31 (22)
Sadness	Distressed, Pity, Sorry	22 (16)
Shame	Awkward, Embarrassed, Ashamed	28 (20)
<i>Total</i>		140 (100)

Twenty-four examples were selected for the measure of emotions (see Table 5.5). An attempt to balance positive, negative and neutral example emotions was made. No example was included for the 'Acceptance' category, due to lack of this type of response in the pre-study exercise. No 'Disgust' example was included as it was thought that this might be offensive to some participants.

Table 5.5. Examples selected for the open-ended measure of emotions

Category	Examples selected	No. (%)
Anger	Irritated	1 (4)
Fear	Confused, Nervous, Scared, Uncomfortable	4 (17)
Feeling fortunate	Lucky, Relief	2 (8)
Guilt	Guilty	1 (4)
Interest	Curious	1 (4)
Joy	Amused, Happy, Pleasure	3 (13)
Love/concern	Admiration, Caring, Concerned, Loving, Protective, Supportive, Sympathetic	7 (29)
Sadness	Sad, Sorry	2 (8)
Shame	Awkward, Embarrassed, Inadequate	3 (13)
<i>Total</i>		24 (100)

Parental Quality of Life beliefs. A total of 155 responses (i.e. a 'valued life object' plus an evaluation of how this object would be affected by having a child with Down's syndrome) were collected, a mean of 5.0 per respondent. The valued life objects (VLOs) most commonly listed were the respondents' family, including children and partner, ($n=34$), a career ($n=18$), social relationships ($n=11$), and independence ($n=8$). The VLOs were classed into six broad themes based on the 'life values' measured in the study of attitudes towards prenatal testing by Evers-Kiebooms *et al.*, (1993), that in turn were derived from a classification of values (Rokeach, 1973).

Valued Life Object categories:

- 1 **Health and well-being.** Responses relating to the importance of the physical and/or emotional health of self and/or family.
- 2 **Job/Career.** Responses relating to the importance of the respondent's current or future job or career.
- 3 **Material aspects.** Responses relating to the importance of money and/or the material items associated with having money.
- 4 **Pleasure and relaxation.** Responses relating to being free to live a lifestyle of choice, go on holidays, have leisure time etc.
- 5 **Relationships.** Responses relating to the importance of relationships with family members and friends or to the importance of feeling 'related to others' in general.
- 6 **Self-actualisation.** Responses relating to the need to develop one's potential or to achieve personal fulfillment (Maslow, 1959) cited in Rokeach, 1973).

Table 5.6 gives sample responses for each of the VLO categories together with the relative frequencies of each.

Table 5.6. Valued life object (VLO) categories and sample responses

VLO category	Sample responses	No. (%)
Health and well-being	Health (family), eyesight, my happiness	16 (10)
Job/career	Enjoying work, promotion	
Material aspects	Quality living conditions, financial security	10 (6)
Pleasure and relaxation	Travel, quality time for yourself	39 (25)
Relationships	Relationship with my husband, my existing children, spending time with kind people	50 (32)
Self-actualisation	Personal growth, spiritual development	20 (13.5)
<i>Total</i>		135 (100)

Twenty examples of valued life objects were selected for the open-ended measure of PQoL beliefs (see Table 5.7) using the percentages given in Table 5.6 as a guide. Where appropriate, the actual words of the participants were used as examples.

Table 5.7. Examples of valued life objects (VLO) selected for the measure of PQoL beliefs

VLO category	Examples selected	No. (%)
Health and well-being	My health, being happy myself	2 (10)
Job/career	My job or career	1 (5)
Material aspects	Having money to spend or save, my home	2 (10)
Pleasure and relaxation	Going out/social life, being able to relax, going on holiday, sports and leisure activities, being free to do what I want	5 (25)
Relationships	My family, my children, relationship with partner/husband, feeling happy with my baby, finding a partner in the future, my friends, love and affection, caring for people,	8 (40)
Self-actualisation	My religion, developing as a person	2 (10)
		20 (100)

Discussion

The findings demonstrated that beliefs and emotions about people with Down's syndrome could be generated using the open-ended measures in this sample, and that a certain amount of consensus in the responding was observed. It is not claimed that the responses were exhaustive or representative of the general population, nevertheless, the examples used for the measures were generated by a number of independent 'others' rather than by the researcher. One change in the potential wording of the measures resulted from this exercise. The measure of stereotypic beliefs followed the wording of the original measures and asked participants to describe a 'typical' person with Down's syndrome. A number of respondents commented that there was no such thing as a 'typical' person with Down's syndrome and that they were all individuals. The wording of the measures in the actual questionnaire took account of this view and participants were instead simply asked to describe "a person with Down' syndrome".

The next section (Method II) sets out the main elements of the study design, the study location and target population, and the way in which the study was operationalised.

5.5 METHOD II. DESIGN, POPULATION, MATERIALS AND PROCEDURE

5.5.1 Study design

A cross-sectional, prospective survey was conducted using a self-completion questionnaire as the method of data collection (see Appendix 6). The main variables related to **prenatal testing choices** were; 1) intention towards having serum screening (triple-test) in the second trimester, 2) intention to use amniocentesis following a positive screen result, 3) intention to terminate for Down's syndrome following a positive diagnosis, 4) serum screening test uptake.

The variables related to **attitudes towards Down's syndrome** were; 1) attitude towards having a baby with Down's syndrome, 2) stereotypical beliefs associated with people with Down's syndrome, 3) beliefs about how having a baby with Down's syndrome would affect parental quality of life (PQoL beliefs), 4) emotions associated with people with Down's syndrome, 5) experiences associated with people with Down's syndrome, and 6) attitudinal ambivalence towards Down's syndrome (comprised of ambivalence scores for the beliefs, emotions and experiences variables).

In addition variables known previously to relate to prenatal testing intentions and choices were collected; age, education, religiousness, anxiety about having a baby with Down's syndrome, perceived likelihood of this event, and attitude towards abortion generally.

5.5.2 Study location and target population

The study took place in an antenatal outpatient clinic in a maternity hospital in an urban area of Yorkshire. The hospital was selected because it was the only one in the region offering serum screening to all women regardless of age, thus avoiding potential bias in an age-restricted population⁵⁰. All other antenatal care providers in the region had age-related policies, for example offering serum screening only to women aged 30 and over. A sample of 'older mothers' could result in bias for three reasons. Firstly, perceived risk is known to affect testing intentions, and age-restricted policies automatically assigns those women offered screening to a higher risk group (Marteau *et al.*, 1991). Secondly, older women tend to hold more favourable attitudes towards abortion, and this has been shown to be predictive of attitudes towards termination for abnormality (Green *et al.*, 1993a). Thirdly, within an older population there would be fewer first-time mothers.

⁵⁰ Originally, the study was to have been run at two sites but early in the development phase the second hospital changed its screening policy to an age-restricted one.

This was considered important because previous testing behaviour may be associated with current testing intentions and behaviour. The population served by the hospital is largely white, with a substantial proportion being from social classes IV and V. In terms of educational attainment, the percentage of pupils achieving five or more GCSEs at grades A* to C is just over half the national average, and 42% complete their formal education at age 16, compared to around 30% nationally. The area has the highest teenage pregnancy rate in England and Wales, with around 10% of conceptions being in women aged under 20 years (Peach, Harris, and Bielby, 1996). The number of births at the hospital is approximately 2800 per annum.

Within the catchment area covered by the clinic, antenatal care is centrally organised and clinic-based midwives carry out the booking appointments. A small number of bookings take place in the community - mainly for geographical reasons - but these women were not included in this study. A multiple-marker serum-screening test ('triple test') is offered to all women who attend for booking prior to 19 weeks gestation. If the woman wishes to have the triple-test, an appointment is made for her to have blood taken at 15-18 weeks gestation. This appointment takes place at the antenatal clinic but is additional to the next routine check-up. The inclusion criteria for the sample were that participants must be at least 18 years old⁵¹ and of 14 weeks gestation or less at the time of booking. This was to give women time to consider any issues raised by the study and the opportunity to discuss them with their midwife. A recruitment target of 300 participants was set as this was considered to be large enough to conduct reliable statistical tests and small enough to be achievable within the time constraints available. Booking figures provided by the Clinical Midwifery Leader (CML) indicated that approximately 190 women booked for antenatal care at the clinic each month, and that over a six-month period around 800-1000 women would meet the inclusion criteria. Recruitment was carried out between August 2000 and the end of January 2001.

5.5.3 Questionnaire development

The development of the questionnaire was informed by a number of methodological discussions on the use of surveys in health-related research (Conner and Waterman, 1996; Fallowfield, 1995; Kirk-Smith and McKenna, 1998; McColl, 1993). There are many valid and reliable ways of capturing the responses to questionnaire items and in this questionnaire a number of different formats were used dependent on the type of response required. Categorical scales were used in some 'factual' items, for example, prenatal testing history and adjectival and Likert-type scales were used to capture beliefs, attitudes and behavioural intentions. Within these different response formats however, response conventions were used consistently.

⁵¹ Women younger than 18 were excluded as a condition of ethical approval by the NHS Trust.

An important consideration was to keep the items as simple and unambiguous as possible and to avoid jargon. However, the use of some technical terms (such as amniocentesis) was unavoidable in the study context. In addition, because of the sensitivity of the subject and the nature of the population, a certain amount of contextual cushioning was given to some items. This may have detracted slightly from the simplicity of item wording but was considered necessary to minimise the level of threat of sensitive questions. Fallowfield (1995) recommends that the reading age of a questionnaire should not be above 12 years. The average reading age of this questionnaire was eleven years (Flesch-Kincaid Grade Level 5.9) with a good Flesch Readability Ease score of 76%.

The questionnaire was divided into five self-contained sections (A to E), each containing items related to a particular area of interest. This reduced the task of questionnaire completion into manageable chunks. The sections were ordered so that less-sensitive items were placed first and the most sensitive ones, for example, participants' attitudes towards abortion, were placed in later sections. It is often recommended that sociodemographic items are placed first or last, but in this questionnaire they were situated after the items about abortion and before the final items measuring attitudes towards Down's syndrome, to act as a kind of buffer. It was considered that to have the items about termination followed directly by ones about people with Down's syndrome may imply a natural connection between the two. This might have offended some participants or implied a demand characteristic on behalf of the researcher. Within sections, very few filter items were included as they can increase question avoidance and unintentional data loss.

Questionnaire content

Section A

The first section of the questionnaire provided a basic explanation of screening and diagnostic tests. This information was included as an introduction to the study topic and to reduce the need for repeated explanation later on. To raise the saliency of the main issues and to act as a measure of basic knowledge, participants were then asked if they had heard of the screening tests, of amniocentesis and of Down's syndrome itself.

Section B

The second section contained factual items about the current pregnancy, behavioural items about any previous history of prenatal testing for Down's syndrome, and cognition items relating to anxiety, perceived risk and attitudes towards having a baby with Down's syndrome.

- Current gestation and number of previous pregnancies was included for background data and to allow between-group comparisons by parity.
- A previous history of prenatal testing for Down's syndrome was collected as previous testing behaviour might relate to current testing behaviour. Of interest also, was whether declining serum screening in a previous pregnancy predicted the same behaviour in a subsequent pregnancy. To the researcher's knowledge no published study has investigated this relationship.
- Anxiety about having a child with an abnormality has been shown to be related to uptake of testing (Marteau *et al.*, 1992a). The perceived likelihood (risk) of that event occurring has been found to be more predictive of the uptake of certain tests than actual age-related risks (Marteau *et al.*, 1991; Marteau *et al.*, 1992a). Items measuring anxiety and risk were adapted from a questionnaire used previously in a study of decision making in prenatal testing (Bekker, 1999).
- As in other studies that have used open-ended measures of attitudinal components, an overall attitude or evaluation of the target was included. This evaluation is used as a dependent variable towards which the component scores are used as predictors to establish the relative importance of the beliefs, affect and experiences in the expression of the overall attitude (Haddock *et al.*, 1994). In this study an evaluation of how 'good or bad' it would be to have a child with Down's syndrome was selected rather than an evaluation of people with Down's syndrome as a group. Based on the findings from the Q-study, it was felt that the latter evaluation would not discriminate between groups and might be subject to a high degree of socially desirable responding⁵². The difference between holding favourable attitudes towards people with a disability, and holding favourable attitudes towards having a child with a disability has also been noted elsewhere (Press *et al.*, 1998).

A single item evaluation measure was selected as they have been shown to be as reliable as multi-item evaluation measures when used in this context (Haddock *et al.*, 1994). Participants were asked to rate their evaluation on a scale of one to nine, with one being anchored as 'extremely good' and nine as 'extremely bad'. Similar items have been used previously (Figueiras *et al.*, 1999; Moyer *et al.*, 1999). However, both these constrained responses within the neutral to negative range and to avoid experimenter bias it was decided to allow for the full range of evaluations.

⁵² The Q-study demonstrated that while there was consensus about the rights of a person who is already living to a good quality of life, beliefs about a person with Down's syndrome who is yet to be born were not subject to consensual responding.

Section C

Items measuring attitudes and behavioural intentions towards testing and termination were based on ones taken from a survey study of community attitudes towards prenatal testing (Evers-Kiebooms *et al.*, 1993). Firstly, participants were asked whether they thought screening tests, diagnostic tests and termination for Down's syndrome should be available to all women. Secondly, respondents were asked about their intentions regarding these three behaviours in their current pregnancy. This two-step approach was also used in the Q-study (Chapter 4) because it is considered more sensitive towards people's feelings than direct questioning of personal behavioural intentions (Green, *et al.*, 1993a). This approach also reduces the chance of confounding the participant's attitude to their own actions with their attitude towards the actions of others. An open-ended item allowed respondents to explain further their answer in relation to each of the intentions. The use of hypothetical situations has been criticised because such scenarios do not have a good track record of predicting behaviour (Green, 1995a). While this limitation is accepted, what people *think* they would do was the object of interest as the aim of this study was not to predict actual behaviour.

To measure general attitude towards abortion participants were asked to indicate whether they believed they would terminate in five hypothetical scenarios; following rape, where their health was in severe danger, where they were on a low income, where they were not in a stable relationship, or were in a stable relationship but 'didn't want' the baby. Responses allowed were 'yes' 'no' and 'don't know'. These responses were coded so that a higher score indicated a more favourable general attitude to abortion, i.e. 'yes' = 3, 'don't know' = 2, and 'no' = 1, and the mean score of the five scenarios constituted a 'general attitude to abortion' score.

Section D

This section collected sociodemographic data including age, age of leaving full-time education, marital status, ethnic origin, religious affiliation and the extent to which religion affects important life decisions (religiousness). As well as providing a useful description of the sample, these variables have been shown to relate to prenatal testing and termination intentions and behaviour.

Section E

This section consisted of open-ended measures of attitudes towards Down's syndrome as described earlier. Four measures of attitudinal information were included: stereotypic beliefs about people with the condition, parental Quality of Life beliefs, emotions, and experiences with people with Down's syndrome. Each measure was a self-contained unit with instructions for completion

being kept consistent across the measures. The measure of experiences was placed first in Section E to take advantage of the priming effect of questionnaire items (Conner and Waterman, 1996). It was felt that making the participant's experiences of people with Down's syndrome mentally accessible would facilitate the retrieval of beliefs and emotions required in the following sections.

The questionnaire was produced as a double-sided A5 booklet to reduce a perception of bulk that may have been off-putting to some women. The main typeface used was Times New Roman as serif typefaces are considered the easiest to read (McColl, 1993). To comply with directives from the NHS trust, the questionnaire (as well as the information sheet and consent form) had the official logo of the Hospital trust marked clearly on the front. A space at the end of the questionnaire was allocated for participants' views or comments about the questionnaire or its subject matter.

5.5.4 Semi-structured interviews

Despite the assurances of the developers of the open-ended measures, there was some concern that the target population may have difficulty spontaneously generating and evaluating the responses required. For this reason it was decided to conduct some exploratory interviews with women prior to piloting the measures, to assess whether beliefs, feelings, and experiences in relation to Down's syndrome could be freely generated. A number of semi-structured interviews were conducted with pregnant women at the antenatal clinic. An interview schedule was drawn up but within each interview certain points of interest were explored further.

Interviewees and interview procedure

Over a two-week period, nine women attending for booking agreed to be interviewed after being invited by their midwife during the appointment. The women were given an information sheet about the purpose of the interviews and a consent form conforming to the standards of the Trust's ethics committee. One woman declined to be interviewed after reading the information. The eight women who gave interviews were white European, had a mean age of 27 years (range 18 to 36 years) and a mean gestation of 12.5 weeks. Four women were nulliparous (this was either their first pregnancy or previous pregnancies had miscarried or been terminated) and four were multiparous (defined here as having had at least one previous successful pregnancy). No details of social, religious or educational background were collected. The interviews were conducted in a private room away from the main waiting area. The interviews were tape-recorded with the permission of the interviewee and one interviewee asked not to be taped. No identifying details

were recorded on tape. The interviews lasted an average of 30 minutes and were all transcribed by the researcher.

Interview schedule

After a few questions about the current pregnancy, and number of existing children, the first set of items related to general awareness of prenatal testing for Down's syndrome. These questions were asked first for three reasons. Firstly to enhance the face validity of the interview by asking about issues salient to the interviewees. Secondly, to set the later items about Down's syndrome in some context. Thirdly, to leave potentially sensitive questions about attitudes until later in the interview.

The questions were:

- Has your midwife mentioned the triple test to you?
- Have you thought about the triple test?
- What conditions you would want to test for? Why would you want to test for them?
- Which conditions wouldn't you want to test for? Why wouldn't you want to test for them?

The second set of questions related to the interviewee's experiences of people with Down's syndrome, and the beliefs and emotions that they associated with affected individuals.

- Have you heard of Down's syndrome?
- Have you ever had any experiences of people with Down's syndrome? Were these experiences positive (good) or negative (bad)?
- What sort of words come to mind when you think about describing people with Down's syndrome? Are these words positive or negative ones?
- What sort of feelings do you have when you think about or see people with Down's syndrome? Are these feelings positive or negative ones?
- In what ways might having a baby with Down's syndrome affect your life? In what ways might it not affect your life?
- What good things might there be about having a child with Down's syndrome? What bad things might there be about having a child with Down's syndrome?

Interview findings

Awareness of prenatal testing for Down's syndrome.

All eight women said they had been told about the triple test during the booking interview and all definitely or probably intended to have it. All four women who had experienced a previous pregnancy said that they would have the triple-test because they had it 'last time', for example, "*I had the test last time and it was OK, so I'll have it again*". For these women the triple test was

considered a routine part of their antenatal care. When asked about the conditions that they would want to be tested for, none of the women could think of anything other than Down's syndrome. The leaflet provided at booking also informs readers that the triple test screens for 'spina bifida', but the women did not spontaneously mention this, suggesting that their midwife had not spoken about neural tube defects in this context. A 36 year old women in her sixth pregnancy commented,

"Umm... you don't really hear about the others. Down's syndrome is the first one you hear about. I don't suppose you really think about anything else – unless they actually tell you if there was something else."

None of the women could think of any condition they would not want to be tested for, and two commented on the importance of having 'all the tests available'. For example,

- *"Any test offered is a good thing – it is good to get the test done – if there is anything to detect."* [32 years, first pregnancy].
- *"Because my partner has Hepatitis C I'm having every test that's totally possible. I'm having all of them. I haven't thought specifically what I want to be tested for. I don't really know all the things that you can be tested for – but whatever they offer me I will have."* [21 years, fourth pregnancy].

For virtually all interviewees the triple test was perceived to have only benefits. One woman spoke of how she welcomed the offer of screening, *"The good thing is that you get a choice at this hospital – it is your decision. You are made aware here – it's not like that at other hospitals"*. Despite holding favourable attitudes towards having the triple-test, the women generally viewed amniocentesis more cautiously.

- *"I didn't really know there was a risk of miscarriage until [the midwife] told me today – so that's something to think about isn't it?"*
- *I wouldn't have an amnio because of the risk of miscarriage."*
- *"Yes I think I'll take it [the triple test]. If I was offered an amniocentesis I would be reluctant to have that – but the triple test – it's just a simple blood test isn't it?"*

Experiences, beliefs and emotions associated with Down's syndrome

Experiences. All the women had heard about Down's syndrome previously, and all could report some experience of people with the condition even if no actual contact, for example,

"I suppose you just see people don't you? I just know when I see somebody I just know, that yeah they have got [Down's syndrome] or they haven't. I've seen some documentaries on telly, things like that, but I've never come into contact with anybody who is...like that".

Five women had had personal contact with people with Down's syndrome through work or school/college. One woman's partner had an uncle with Down's syndrome. The other three women could only recollect television programmes about Down's syndrome or 'people on the street'. The interviewees did not appear to have difficulty evaluating their experiences in some way.

- *“I met a lady – had a daughter with Down’s syndrome.... A positive experience, I got to meet the child – it made a difference on my views about Down’s syndrome.”*
- *“I worked at a mental hospital. Some of the patients had Down’s syndrome... It was rewarding from a working point of view – both a positive and a negative experience.”*
- *“I’ve seen people out in the street, on TV, no nothing else – no contact. I don’t take much notice – I think it’s awful – that people stare.”* [Interviewer: Were your experiences positive or negative?] *“Don’t know really – neither positive or negative.”*

Stereotypic beliefs. When asked about the words that ‘came to mind’ to describe people with Down’s syndrome, all interviewees were able to express some beliefs. These fell into three main categories; the appearance of people with Down’s syndrome, their personalities, and their difference from others.

- *“[They] have that look – some more than others”.*
- *“Very loving people, but they do have a temper.”*
- *“‘Special’ always springs to mind. Different. Other people might say abnormal.”*

Emotions. The emotions aroused by people with Down’s syndrome seemed more difficult to report and evaluate. For example, *“No strong feelings. They are just normal people really... I don’t think about it much.”* The feelings that were reported were mostly ones of sympathy and of protectiveness, for example, *“I feel sorry for the child - people can be cruel and stare.”* and *“Heart-rending. Just really protective towards them.”* One woman expressed ambivalence in her feelings about people with Down’s syndrome.

“Sad – but then not – because they seem so happy with their lives and seem to enjoy life from my experiences. My initial response is ‘what a shame’ but then no because they are quite happy”.

Beliefs about how having a child with Down’s syndrome would affect parental quality of life.

The interviewees all felt that to some degree their lives would be affected by having a child with Down’s syndrome. However, there was a wide range of beliefs expressed, for example,

- *“Someone said it would be really hard to cope with. But it wouldn’t be much of a change – it’s going to be a struggle anyway.”* [18 years, first pregnancy].
- *“Yes I don’t think I could cope – people would point and stare”* [34 years, second pregnancy].

One interviewee (first baby, 24 years) felt that although there would be differences, she could not say what these would be like. All other interviewees managed to evaluate the potential life effects in some way. The positive aspects of having a child with Down’s syndrome were reported to be the rewards associated with the child’s achievement and the loving parent/child relationship, for example *“You still get the love and fun out of them though – take them swimming and go on holiday.”* [28 years, second pregnancy]. Negative aspects were varied, but typical comments

related to the extra care a child with Down's syndrome was perceived to need, the unfavourable attitudes of others, and the impact on existing children. For example,

- *“Other people's attitudes. I imagine it would be quite hard, caring for it 24 hours a day. But then it would be hard anyway!”* [24 years, first pregnancy].
- *“I think it would affect the full family life, y'know of the other children as well, that's a lot of it. I don't think personally I'd be able to give them enough attention all three, like children deserve, and they do need 100% love and attention don't they?”* [21 years, fourth pregnancy].

The link between attitudes and testing intentions

A surprising finding from the interviews was the apparent lack of association between the attitudes women held towards Down's syndrome and whether they intended to have the triple test or not.

The test was viewed positively both by those with favourable and those with unfavourable attitudes. As the interviews progressed the researcher attempted to probe this 'dissociation':

Interviewer: “Does what you know about Down's syndrome influence whether you have the triple test or not?”

- *“No.”* [**Interviewer:** Is it separate?] *“Yes. I think my partner wants the test. I want to think about it”.*
- *“Yes. I've had positive experiences – I don't see it as such a bad thing if I had a handicapped child – not such a bad thing not the end of the world. If I'd had bad experiences it might be different”* [respondent intended to have the screening test].
- *“No. Would have the test anyway – any test offered is a good thing”.*
- *“No, not really”* [**Interviewer:** are you saying that the link isn't so direct?] *“No... I know what you mean. No, I'm not having the test because of that, not specifically just because it's Down's syndrome”.*
- *“No, it's not an influence. I just want to make sure that everything is OK – I just hope they don't find anything. It's a separate issue – Down's syndrome and the test.”*

One woman, who expressed quite unfavourable attitudes towards Down's syndrome did feel that her views influenced her intentions, but more because of a general unfavourable attitude towards having a child with an abnormality:

Interviewer: “Because of what you know about Down's syndrome you think ‘Yes I would like a test for that’?”

“I suppose so, because that's the main thing that everybody hears about when you're pregnant, the triple test for Down's, but any test that they offered me for any abnormality I would take.”

Information about Down's syndrome

Some interviewees had commented that their knowledge of Down's syndrome was limited and so the last two interviewees were asked whether they thought information about Down's syndrome would be beneficial at the screening stage. Neither thought it would be. One interviewee said that information about Down's syndrome would “*put a lot of people off*” having the triple test, but also that she didn't know enough about Down's syndrome to make a decision about amniocentesis and

would want more information then. The final interviewee held “*very strong opinions*” about Down’s syndrome and definitely intended to terminate an affected pregnancy. She felt she did not need any more information at the screening stage but also said that she did not know what the condition “*does or entails during a child’s life*”. At the close of the interview, participants were asked if they had anything they would like to ask the interviewer. During this final interview the following exchange occurred:

Interviewer “I’ve finished what I wanted to ask you. Is there anything you would like to ask me at all?”

“Yes. I wouldn’t mind knowing exactly what it is [Down’s syndrome]? To be honest I don’t. Do you know?” [21 years, fourth pregnancy].

The interviewer then gave a very brief overview of Down’s syndrome, including the range of disability and life expectancy. The interviewee expressed surprise at even the most basic information and commented that perhaps she needed to know more before deciding on the test.

Conclusions from the semi-structured interviews

The interviews demonstrated that women were aware of prenatal testing for Down’s syndrome and were able to generate experiences, beliefs and emotions associated with the condition in an interview situation. They were also generally able to evaluate their response as positive, negative, or neutral. This indicated that the target population should be able to complete the open-ended measures included in the questionnaire. In addition, the apparent dissociation between attitudes towards Down’s syndrome and intentions to have the triple-test was interesting. It was hoped that the results of the questionnaire analysis would throw further light on this issue and identify whether this was a real finding or artefact related to the interview method. For example, a perceived need to report ‘politically correct’ views about the condition to the interviewer might have obscured a more obvious link between attitudes and behavioural intentions. The next stage of the study was the piloting of the questionnaire.

5.5.5 Piloting the questionnaire

Pilot procedure

The pilot stage took place over a three-week period and in two phases. The first phase piloted the first draft of the questionnaire on ten women, and the second phase piloted the revised draft on five women. As for the exploratory interviews, women were invited by their midwives at their booking appointment to fill in the questionnaire at the clinic. All women classified themselves as ‘white’, had a mean age of 27.5 years (range 20 to 36 years) and a mean gestation of 11.7 weeks. Four women were nulliparous and eleven were multiparous (between one and five previous pregnancies). The mean age at leaving education was 16.9 years (range 15 to 21 years). The

questionnaire completion took place in the presence of the researcher in a private room away from the main waiting area. The women were given an information sheet about the purpose of the study and a consent form conforming to the standards of the Trust's ethics committee. They were encouraged to ask questions about the items or to make suggestions about wording and format during the completion process and after they had completed the questionnaire.

Results of pilot phase one

Completion of the full questionnaire took an average of 19 minutes. In most cases the questionnaire was completed as intended although four women did not give evaluations to their responses in the open-ended measures. One woman declined to complete the open-ended measure section as she said she felt uncomfortable doing so in the researcher's presence. As a result of feedback from participants in phase one some changes were made to the questionnaire.

- In the pilot an item related to 'not having' prenatal testing in previous pregnancies was reworded and simplified as some participants found it difficult to answer. Fallowfield (1995) has highlighted the difficulties surrounding negatively worded questionnaire items.
- The open-ended measure response areas were changed from a blank 'box' to a lined grid with a separate column for the evaluations. It was felt that this more structured approach would encourage participants to use short responses and to give an evaluation of each response.
- An evaluation option of 'both positive and negative' was added to the open-ended measure instructions. Although this evaluation was not included in the original measures, a number of participants in this pilot exercise had said that it wasn't always possible to give just one evaluation as they had 'mixed feelings' about a response. Two had spontaneously entered a plus and a minus sign together and so this representation was selected.

Results of pilot phase two

The results from phase two of the pilot were encouraging. All women completed the measures as intended and gave evaluations to most of their responses. Three out of the five respondents used the 'mixed' response at least once supporting the inclusion of this new option. Similar response times to the first pilot phase were achieved and no further amendments were made.

5.5.6 Procedure: administration of the questionnaire

The midwives conducting the booking appointments distributed the questionnaires to potential participants. The women were asked to complete the questionnaire at home and return it by post before their next antenatal visit. This method has been used successfully in a number of studies with pregnant women, including two that investigated attitudinal and informational aspects of

cystic fibrosis screening, which achieved response rates above 65% (Botkin and Alemagno, 1992; Mennie, Liston, and Brock, 1991). As the researcher in this study could not be at the antenatal clinic on a daily basis, this method of administration had the advantage of recruiting the maximum number of participants in the minimum amount of time. In addition, the postal method meant that researcher bias was not an issue and that participants might feel more free to answer sensitive questions honestly. However, it was accepted that the response rate might be lowered as the recruitment was being carried out on the behalf of an 'unknown other', a factor known to reduce motivation to complete questionnaires.

The clinic at the site of the study contained a number of small offices where bookings were carried out during morning clinic sessions. In each of these booking rooms a box containing sets of study materials was provided along with a 'flyer' to remind midwives of the study, the inclusion criteria, and their role in handing out the questionnaires. Each set of materials comprised an information sheet, a consent form, a copy of the questionnaire and a prepaid return envelope placed in a sealed A4 envelope with the University of Leeds logo and an affixed sticker with the words 'TESTING IN PREGNANCY STUDY'. At each booking, the midwife was asked to check if the woman met the inclusion criteria for the study and if so, to ask her if she would take part. If the woman agreed, she was handed an envelope and asked to complete the questionnaire at home. Over the study period one thousand sets of study materials were provided to the clinic in ten boxes of one hundred sets. The envelopes also had the questionnaire identifier ranging from 1 to 1000 marked in the top corner. The clinical midwifery leader (CML) was responsible for keeping the boxes supplied with envelopes and for ensuring that they were distributed in roughly ascending sequence.

Completed and returned consent forms were forwarded in batches to the CML at various points during the study. The clinic staff then used patient records to establish whether the participant had had a serum screening test or not and a list of test uptake by questionnaire identification number was returned to the researcher. Overall uptake figures for the study period, and also the preceding and following six months were obtained from the Immunoassay Laboratory at the hospital.

5.6 STUDY APPROVAL AND ETHICAL CONSIDERATIONS

Prior to the research being carried out, approval was obtained from the hospital's NHS Trust. The Trust's approval process required the submission of a study protocol and relevant forms to the Trust's Local Research Ethics Committee, the Research and Development Quality Group and the

Caldicott Guardian⁵³. Applications were submitted at the beginning of March 2000 and approval for the research to take place was granted at the end of May 2000. In addition, a presentation of the study proposal was given to the Obstetrics and Gynecology Consultant's Committee at the end of May 2000 as a condition of their approval of the study. The research was carried out within the guidelines of the British Psychological Society's 'Code of Conduct, Ethical Principles & Guidelines' for conducting research with human participants (British Psychological Society, 1995). Particular references to key areas of the code are made below.

Consent. Participants were provided with an information sheet that informed them of the purpose of the research and what their participation would entail. Consent was recorded on a form based on the standard suggested by the Ethics Committee of the NHS Trust (see Appendix 7). Separate consent was requested for access to serum screening uptake information. All participants were invited to take part by their midwife and no reward was offered for participation.

Withdrawal from the study. The information sheet told participants that they were not obliged to fill in the questionnaire if they did not want to, and were assured that non-participation would not affect their antenatal care.

Confidentiality. All data provided by participants were kept confidential by separating signed consent forms from questionnaires and returning the consent forms to the antenatal clinic. No data was stored on computer that could be associated with an individual in compliance with the Data Protection Act.

Protection of Participants. In respect of the privacy of individuals, participants were informed that they were not obliged to respond to any questions if they did not wish to. As the topic under inquiry was a sensitive one, every attempt was made to ensure the questions were worded in a way to take account of this. However, it was anticipated that some participants might require further information or support around the issue of testing and termination. The information sheet provided contact details for the Down's syndrome Association and the support organisation ARC (Antenatal Results and Choices).

⁵³ Caldicott Guardians are individuals responsible for the protection and use of patient information within their NHS Trust. Guardians were appointed in accordance with directives from the NHS Executive (1999).

5.7 DATA ANALYSIS

Missing data

Missing data can be treated in a number of ways depending on the extent of the missing data and how these data are distributed across cases; each method has advantages and disadvantages (Tabachnick and Fidell, 1983). In the following analyses (rather than attempt to estimate missing values or replace with mean values) the cases where data were missing were excluded from the relevant individual analyses, but not from the study as a whole. Where cases were excluded for other methodological or statistical reasons, an explanation is provided at the appropriate point.

Analysis

Data were analysed using the Statistical Package for the Social Sciences (SPSS for Windows v. 9.0.0). The chosen analyses examined associative rather than causal relationships between variables, approaches appropriate to the descriptive nature of the research.

- Categorical data were analysed mainly using the chi-square test (χ^2) for independence of variables. Where cell numbers were small, likelihood ratio tests were used, as they are less sensitive to small sample sizes.
- Comparisons of group means were carried out using t-tests and one-way ANOVAs where appropriate. Where assumptions of normality and homogeneity of variance were not met non-parametric Mann-Whitney tests or Kruskal-Wallis tests were used instead. Post-hoc Scheffe tests were used following ANOVAs to identify specific differences between groups.
- Bivariate correlations were carried out using mainly Spearman's rank correlation test due to the non-normal distribution of many of the variable values, however where appropriate Pearson's r was calculated.
- As the data distributions of the main dependent variables were not suitable for linear regression equations, analyses were conducted using binary logistic regression and discriminant analysis. More detail of the decisions surrounding the choice of these procedures is given in the appropriate section.
- The alpha level for all statistical tests was set at $\rho=0.05$.

The results of the data analysis and the discussion of these results have been organised into the following two chapters. **Chapter 6** contains the results from the analysis of the questionnaire data. **Chapter 7** discusses the findings of the study and draws conclusions about the relationships between attitudes towards Down's syndrome, testing and termination intentions and screening uptake.

CHAPTER 6 ATTITUDES TOWARDS DOWN'S SYNDROME IN THE PRENATAL TESTING SITUATION: RESULTS.

This chapter presents the results from the analysis of the questionnaire in two parts: Part 1 presents analyses of the data collected by sections A to D. This includes the findings related to participants' behavioural intentions towards prenatal testing and termination and actual uptake of the serum screening test. Part 2 provides descriptive statistics and the statistical and qualitative analyses of the data collected using the open-ended measures of attitudes towards Down's syndrome.

6.1 RESULTS PART 1

6.1.1 Response rate and sample characteristics

Clinic staff distributed approximately 840⁵⁴ questionnaires over the recruitment period. Of these, 200 were returned giving an overall response rate of 24%. Two of the questionnaires were unusable due to lack of data and one was from a woman younger than 18. The final sample for the study was N=197. Possible reasons for this low response rate are discussed in the next chapter. The women ranged in age from 18 to 43 years (mean 27.3, SD 5.5). Gestations at the time of returning the questionnaires ranged from 8 to 22 weeks (mean 13.2 weeks, SD 2.5). All participants described themselves as 'White' with the exception of one woman who described her ethnic origin as 'Chinese'. Table 6.1 shows some of the known socio-demographic variables of the population for the city where the study was conducted (the 'target population', see 5.5.2) compared with the characteristics of the study sample.

Table 6.1. Comparison of the characteristics of the target population and study sample.

Variable	Target population	Study sample
Age at conception	10% of conceptions in women < 20 years.	9% of study sample < 20 years.
Education	42% complete formal education at 16.	40% completed formal education at 16, 41% by 19, 16% at 20 years or older.
Ethnicity	Largely white European.	All except one participant classed their ethnic group as 'White'.

The majority of participants were married or living as married (86%). Educational level was classified using the age at which participants had completed their formal education; 16 years or younger (40%), 17 to 19 years (41%) and 20 years or older (16%). Three percent did not give this

⁵⁴ The exact number of questionnaires distributed is not known. See section 7.1.1.

information. The majority of the sample (63%) gave their religion as Church of England, 7% were Catholic, 5% gave other or non-specific Christian denominations and the remaining 25% said they had, or gave no, religious affiliation. Eighty-four percent said that religion was not applicable or had no influence over decisions in their life, 14% said it had a little influence, 2% said religion had quite a lot of influence over their decisions. No respondents said that religion influenced important life decisions 'completely'.

6.1.2 Awareness of prenatal testing for Down's syndrome

The first section in the asked whether participants had heard of the tests and the condition before. Test awareness was high with 94% and 91% being aware of serum screening and amniocentesis respectively. Only five women said they had not heard of the tests, and these were all first-time mothers with a mean age of 19.7 years. All respondents said that they had heard of Down's syndrome. However, the questionnaires were distributed after the booking interview had taken place and the questionnaire did not ask whether the woman had been aware of the tests before their current pregnancy. As issues around testing for abnormality may have already been raised at this point, this probably accounts for the high level of awareness.

6.1.3 Parity and previous testing history

Forty-two percent ($n=82$) of women were classified as nulliparous as this was either their first pregnancy or any previous pregnancies had miscarried or been terminated. The remaining 58% ($n=115$) had experienced at least one live birth prior to this pregnancy (range 2 to 11 pregnancies) and they are defined here as multiparous. Of the multiparous women, 113 gave details of their antenatal testing history; 73% had used some form of antenatal testing in at least one of their previous pregnancies (see Table 6.2).

Table 6.2. Testing for Down's syndrome in previous pregnancy: reported frequencies

		Number	Percent
Tested	Serum screen	71	63
	Serum screen and diagnostic test	9	8
	Diagnostic test only	2	2
Not tested	Offered screening but not taken up	15	13
	No tests offered	12	11
Not sure		4	3
Total		113	100

6.1.4 Having a baby with Down's syndrome: anxiety and perceived likelihood

Most women felt only slightly anxious about the possibility that their baby would have Down's syndrome and in general perceived that it was unlikely to happen to them (see Tables 6.3 and 6.4). On a scale of one to four (not at all worried to extremely worried) the mean anxiety score was 1.9 (SD 0.9), and on a scale of one to four (not at all likely to extremely likely) the mean perceived likelihood score was 1.5 (SD 0.6). Nevertheless, 18 women reported that they were extremely worried about having a baby with Down's syndrome, including two women who had previously given birth to a child with abnormal chromosomes (one with Down's syndrome), both of whom had died in infancy. Anxiety about having a baby with Down's syndrome was significantly and positively correlated with perceived likelihood of having an affected child ($r_{ho} = +0.32$, $p < 0.001$). However, while no effect of age on anxiety was found, women 35 years and over rated their likelihood of having an affected child as significantly greater than women under the age of 35 (Mann Whitney U = 1223.5, $N_A = 170$, $N_B = 22$, $p < 0.01$).

Table 6.3. Anxiety about having a baby with Down's syndrome: response frequencies

Anxiety	Not at all worried	Slightly worried	Fairly worried	Extremely worried
	N (%)	N (%)	N (%)	N (%)
N=195	61(31.5)	100(51.5)	16 (8)	18 (9)

Table 6.4. Perceived likelihood of having a baby with Down's syndrome: response frequencies

Perceived likelihood	Not at all likely	Slightly likely	Fairly likely	Extremely likely
	N (%)	N (%)	N (%)	N (%)
N=195	104 (53)	85 (44)	6 (3)	0 (0)

6.1.5 Having a baby with Down's syndrome: evaluation of the event

Women were asked to give an evaluation of having a baby with the condition on a scale ranging from 1 (extremely bad) to 9 (extremely good). The mean evaluation response was 2.8 (SD 1.8); 76% evaluated this event as 'bad' to some degree (a score from 1 to 4), 21% as 'neither good nor bad' (a score of 5) and 3% evaluated having a child with Down's syndrome as 'good' (a score of 6 to 9). Anxiety was correlated with the evaluation variable, with those women who were more anxious about having a baby with Down's syndrome evaluating this event less favourably ($r_{ho} = -0.40$, $p < 0.001$). There was a modest but significant correlation between the age of the participant and their evaluation response ($r_{ho} = -0.18$, $p < 0.05$), thus older women viewed having

a baby with Down's syndrome less favourably in the main. No other socio-demographic variables were significantly associated with the evaluation variable.

6.1.6 Attitudes and intentions towards screening, amniocentesis, and termination

The majority of respondents held favourable attitudes towards the availability of a) screening, b) amniocentesis, and c) termination for Down's syndrome, and agreed they "should be available for every woman that wants one" (93%, 80% and 76% of participants respectively). Table 6.5 gives the responses frequencies for the items measuring behavioural intentions in the current pregnancy. When treated as an ordinal variable (from 1 (definitely have) to 5 (definitely not have), the intention responses all correlated significantly and positively with each other (serum screening with amniocentesis and termination, Spearman's $\rho = +0.55$ and $+0.52$, $p < 0.01$ respectively; amniocentesis and termination $\rho = +0.70$, $p < 0.001$). A number of participants with reported gestations greater than 15 weeks ($n=46$) may have reported screening 'intentions' retrospectively. With the exception of predicting uptake from intention it was decided to include these women in the analyses so that consistent comparisons across different intentions could be made using a larger sample. While it is accepted that some of the relationships between intentions and other variables may be inflated because of this, comparisons of demographic and obstetric variables, intentions, test uptake, and attitudes revealed no effects of gestation significant at $p < 0.05$.

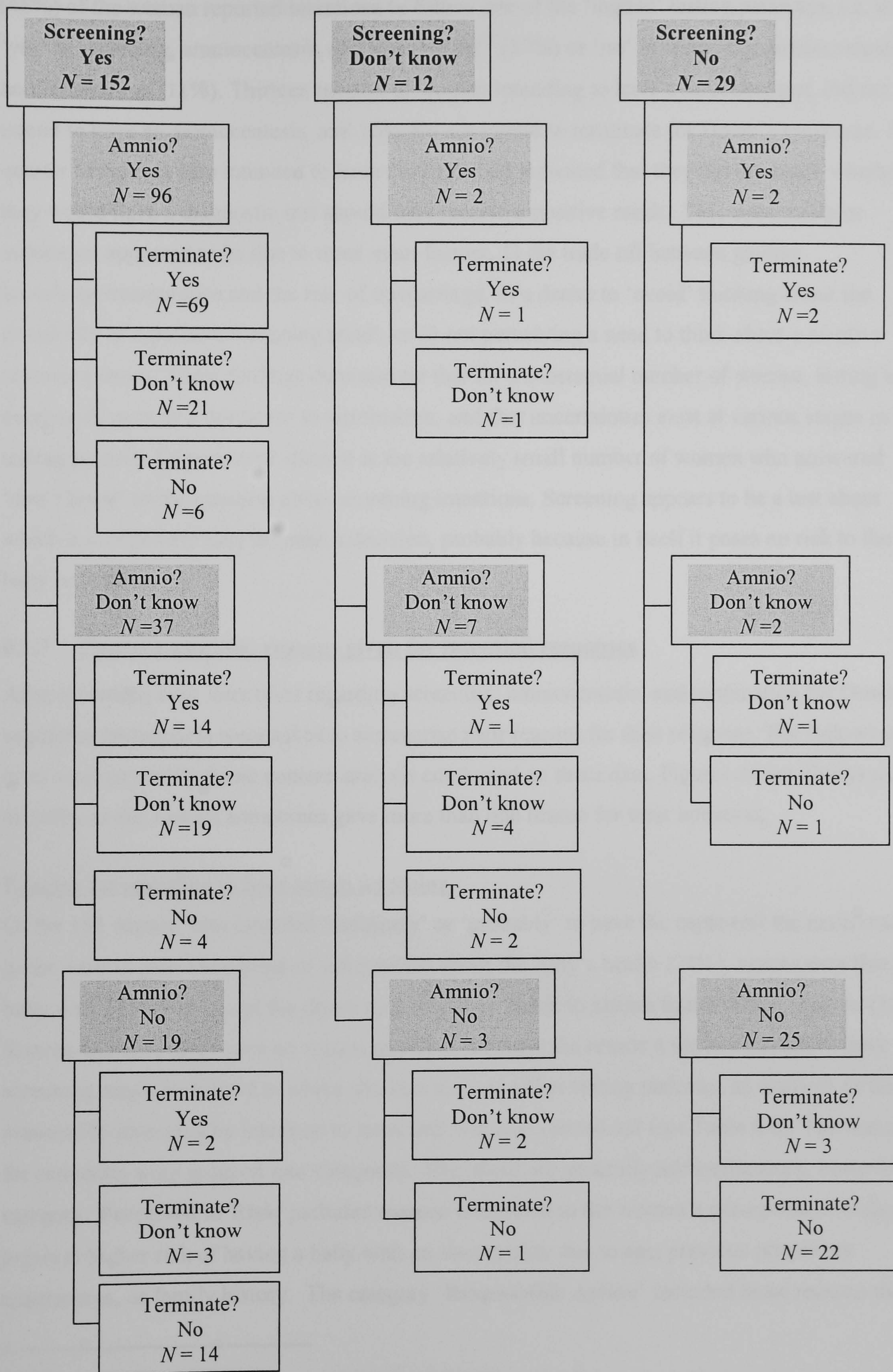
Table 6.5. Intended personal use of screening, amniocentesis and termination: frequencies.

	Behavioural intention				
	Definitely have	Probably have	Don't know	Probably not have	Definitely not have
	N (%)	N (%)	N (%)	N (%)	N (%)
Screening ^{\$}	131 (67)	24 (12)	12 (6)	5 (3)	24 (12)
Amniocentesis*	53 (27)	47 (24)	47 (24)	22 (11)	26 (13)
Termination*	49 (25)	40 (21)	55 (28)	16 (8)	35 (18)

^{\$} N=196, * N=195

Figure 6.1 plots the intended testing pathways for the 193 women who gave responses to all three of the intention items. The five possible intention responses were collapsed into three categories; the responses 'would definitely have' and 'would probably have' formed the 'Yes' category, the responses 'would definitely not have' and 'probably would not have' formed the 'No' category, and the third category was the 'Don't know' response.

Figure 6.1. Intended testing pathways.



Eighty participants (41%) gave at least one ‘don’t know’ intention response. Just under half (48%) of the women reported intentions to follow one of the ‘logical’ testing pathways, i.e. either ‘yes’ to screening, amniocentesis, and termination⁵⁵ (37%) or ‘no’ to screening, amniocentesis, and termination (11%). Thirteen percent of women intending to have a screening test, did not intend to have an amniocentesis, and 16% did not intend to terminate for Down’s syndrome. A quarter of women who intended to have the triple test indicated that they did not know whether they would have a diagnostic test should they receive a positive result. This uncertainty or indecision appeared to be due to three main factors, 1) the trade off between gaining knowledge/reassurance and the risk of miscarriage, 2) a desire to ‘avoid’ thinking about the possibility of a positive screening result, or 3) not perceiving a need to think about a positive screening result. These findings demonstrate that for a substantial number of women, testing is not necessarily seen as a precursor to termination, and that uncertainties exist at various stages in the testing process. Of particular interest is the relatively small number of women who answered ‘don’t know’ to the question about screening intentions. Screening appears to be a test about which it is relatively easy to make a decision, probably because in itself it poses no risk to the baby or mother.

6.1.7 Content analysis: reasons given for intention responses

After recording their intentions regarding screening, amniocentesis, and termination for Down’s syndrome participants were asked to summarise their reasons for their response. The following is a brief summary of the basic content analysis conducted on these data. Figures do not always add up to 100% as the women sometimes gave more than one reason for their intention.

Reasons for intending to have serum screening

Of the 155 women who intended ‘definitely’ or ‘probably’ to have the triple-test the main reasons given were to gain knowledge or information about the baby’s health (24%), reassurance that the baby was ‘OK’ (23%), and the desire to use the risk factor to inform future testing choices (15%). Sixteen women (10%) gave no reason response. Because the reason a woman intends to have screening might be related to where she sees the end of her testing pathway, an analysis of the reasons for screening by intention to terminate was also carried out (see Table 6.6). The reasons for screening were grouped into categories. The labels are generally self explanatory, however the category ‘**Perceived at Risk**’ included reasons connected to the women’s perception of being *a priori* at higher risk of having a baby with an abnormality due to age, previous pregnancy experiences, or family history. The category ‘**Responsible Action**’ included those reasons that

⁵⁵ This figure includes the two women who opted for a diagnostic test without prior screening.

suggested having testing is the responsible response to the offer of antenatal care, for example, testing is “necessary”, “important”, “sensible” and “best for baby”. Three responses were considered unclassifiable, for example, “I would certainly want a test” and these were excluded.

Table 6.6. Reasons for intending to have serum screening by intention to terminate (%)

Reason response	Yes to termination	Don't know	No to termination
Information (health status of the baby)	25	14	36
Reassurance	22	30	16
Inform future choice	21	14	0
Preparation for affected child	1	9	24
Responsible Action	8	14	12
Avoid baby with Down's syndrome	12	0	0
Perceived at risk	5	14	8
Family member with Down's syndrome	2	2	4
Test 'harmless'	4	2	4
No reason given	8	14	8

Although information and reassurance were common responses across all groups, the perceived end of the testing pathway appeared to influence reasons for having a screening test, for example, only those women who intended to terminate saw avoidance of a child with Down's syndrome as a reason for having screening.

“I wouldn't want to go through pregnancy and birth when I knew that there was something wrong with the baby” [Yes to screening, amniocentesis, and termination].

Some women wanted to prepare for the birth of an affected child,

“I would definitely have [screening]...to get me ready for when the baby arrived because I wouldn't have a termination” [Yes to screening, no to amniocentesis and termination].

Others wanted the further information they associated with screening as they perceived this widened their choices, for example;

“Peace of mind, also the choice of rather carrying on with pregnancy or not. Also to discuss the possibility with father and family” [Yes to screening, don't know to amniocentesis and termination].

Women who gave no reason for having the screening test were significantly later on in their pregnancy than those who did give a reason (Mann Whitney U = 636.0, $N_A = 139$, $N_B = 17$, $p < 0.05$). This suggests that some women had already had their test, therefore could not sensibly

give a reason for their 'intention'. No differences were found in the screening, amniocentesis, or termination intention responses of those that did and did not give a reason for having screening.

Reasons for intending not to have serum screening

Of the 29 women who 'definitely' or 'probably' did not intend to have serum screening only one did not give a reason. Thirty-four percent said they did not want to have a termination and 31% believed that they could cope with an affected child. For example,

"It wouldn't matter to me because the baby will be well looked after. It will be given all the help and support it needs to lead a normal life" [No to screening, amniocentesis and termination].

Other reasons included wishing to avoid anxiety raised by testing (21%), not wanting an amniocentesis (13%), and a desire to not know if anything was wrong with the baby (13%). Thirteen percent said they felt the test was not accurate.

Reasons for a 'don't know' response to screening

Twelve women gave a 'don't know' response when asked if they intended to have a screening test. Five did not give a reason for this response. Two women thought that miscarriage might be caused by the screening test, two were unsure they could cope with a positive result, one wanted to discuss it with her partner, and one first wanted to know her age-related risk. One woman said the test gave 'false' information (presumably because of second-hand experiences of false-positive results).

"Because my sister-in-law and cousin were told that their children were Down's after the test but gave birth to healthy children." [Don't know to screening, amniocentesis and termination].

Reasons for intending to have amniocentesis

Of the 100 women who intended 'definitely' or 'probably' to have an amniocentesis 25% said they wanted to gain knowledge or information about their pregnancy, for example,

"Having had the triple test I would want to know more" [Yes to screening, amniocentesis and termination].

Eleven percent said they didn't want or couldn't cope with a child with Down's syndrome, and 10% explicitly said they wanted amniocentesis so they could have a termination if necessary. A further 10% intended to use amniocentesis for reassurance that the baby was OK. Six women said that they wanted amniocentesis to enable them to make a choice about continuing a pregnancy or not. However, all of these six also gave 'definitely' or 'probably' intention to terminate responses to the next item, indicating that they already knew what their choice would be. Other reasons for intending to have an amniocentesis included preparing for a baby with Down's syndrome (3%)

and having an existing family member with a disability (2%). Over one-third of women in this group gave no reason for their intention. These women were more likely to have said they would ‘probably’ have rather than ‘definitely’ have an amniocentesis and termination. They might have found it more difficult to articulate a reason for having the test if there is still some uncertainty about what they would do in the actual situations of a positive screening or diagnostic result.

Reasons for intending not to have amniocentesis

Of the 48 women who ‘definitely’ or ‘probably’ did not intend to have amniocentesis three gave no reason, 75% said because of the risk of miscarriage and 15% said they would not terminate.

“I would refuse the test as there is a possible chance of miscarriage and no test is worth that” [No to screening, amniocentesis and termination].

Some women wished to avoid the anxiety associated with testing (8%),

“Because I just don’t want to be put through unnecessary suffering when it could be nothing” [No to screening, amniocentesis and ‘don’t know’ to termination].

Others said that an affected baby would still be loved and wanted (8%).

“I would love my child whatever” [No to screening, no to amniocentesis and termination].

One woman expecting twins gave that as the reason for not intending to have amniocentesis.

Reasons for a ‘don’t know’ response to amniocentesis

Of 47 women who did not know whether they would have an amniocentesis, 21% gave no reason, 38% said they were concerned about the risk of miscarriage and 34% felt that they could only decide if and when they were given a positive screening result. For example,

“As there is a risk with this sort of test I would have to decide if it was worth it or if it was important to me to know for definite if the baby had Down’s and how I would react if it had, for example, would it make any difference to me? Would I continue with the pregnancy? The decision for this test is a little more complicated” [Yes to screening, don’t know to amniocentesis and termination].

Other ‘don’t know’ reasons given were fear of the procedure (6%), the test being ‘inaccurate’ (one person) and having an existing family member with Down’s syndrome (one person). No differences were found between participants who gave a reason and those that did not.

Reasons for intending to have a termination

Of the 89 women who ‘definitely’ or ‘probably’ intended to terminate an affected child 13% gave no reason. Forty-four percent said they felt that they couldn’t cope with an affected baby.

“I don’t consider myself the type of person who could look after a Down’s child” [Yes to screening, amniocentesis and termination].

A quarter (25%) felt that people with Down's syndrome had a low quality of life.

"As I would not be prepared to bring a child suffering from Down's into the world as I feel I could not subject my child to a less than normal and healthy life" [Yes to screening, amniocentesis and termination].

Other reasons included the belief that continuing the pregnancy would be 'unfair' on other children or family members (17%), concern about long-term care of the affected child (6%) and the prejudice of society (4%). Three women who had previously had a child with a disability (one still living) intended to terminate as they felt they could not cope with having another disabled child. No differences between participants who gave a reason and those that did not were found.

Reasons for intending not to have a termination

Of the 51 women who gave a 'no' response to termination, 35% said that a baby with Down's syndrome was like any other baby and so had a right to life, 33% said that an affected baby would still be 'loved and wanted'. For example,

"I couldn't get rid of a baby just because it had Down's. It would still be my baby and I would love it just as much" [Don't know to screening and amniocentesis, no to termination].

Other reasons included a moral/religious objection to termination (16%) and the belief that they could cope with an affected child (10%). Three women cited miscarriage or previous death of a child, and two felt that termination would be too traumatic as they were already attached to the baby. One person wouldn't terminate because they had a relative with Down's syndrome and one person gave no reason.

Reasons for giving a 'don't know' response to termination

Of the 55 women who gave a 'don't know' response to termination 24% gave no reason; 55% said they would need to be in the situation of having a positive diagnosis before they could decide what to do. For example;

- *"I don't think I can say until I would be in that position as feelings change. I used to think I would definitely have a termination but my feelings are changing"* [Yes to screening, don't know to amniocentesis and termination].
- *"Cannot really say, so many options to consider. It is only when faced with this dilemma then your emotions would dictate the choice you made"* [Yes to screening and amniocentesis, don't know to termination].

The second most common reason given (21%) was that the situation would have to be discussed with their partner before a choice was made. Other reasons included being attached to the unborn child (three women), the need for gaining more information ("*all the facts*"), being unsure about the ability to cope with an affected child and "*not believing in*" abortion. Three women said they

'didn't want to think about' termination until they had to. Those women who did not give a reason for their 'don't know' response were more likely to have left education at a younger age than those who did give a reason (Likelihood ratio = 7.7, $df = 2$, $\rho < 0.05$).

In summary, it was clear that the participants viewed prenatal testing as serving a number of purposes rather than simply being the first step towards termination. Of interest was that women who gave 'no' intention responses were more likely to give a reason for their response than women in the 'yes' and 'don't know' groups. Perhaps women who intended to go against the perceived norm of accepting testing may have felt most need to justify their choice. Alternatively, choosing to 'opt out' might require most thought, making it easier to articulate intention reasons more readily.

6.1.8 Serum screening uptake

Screening test uptake data was collected at a later date than the behavioural intention data and was gathered by midwifery staff from patient notes. Uptake data was not available for 34 of the respondents and the reasons for this are given in Table 6.7. There were no significant differences found by any socio-demographic variable or testing/termination intentions between those women for whom uptake data were available and those for whom it was not.

Table 6.7. Reasons for unavailability of serum screening uptake data: frequencies

Reason	Number
Patient notes missing or unavailable	18
Transfer of antenatal care away from clinic	7
Detail given on consent form insufficient to identify notes	5
Consent not given for access to serum screening result	2
Miscarriage prior to test period	2
<i>Total</i>	34

Of the 163 respondents for whom uptake data were available 77% ($n=125$) took the serum screening test. This compares with a background uptake rate of 64% during the same period ($n=808/1260$)⁵⁶. Screening uptake during the six-month periods prior to, and following the study was 60% and 64% respectively. Possible reasons for the higher uptake in study participants are discussed in a later section.

⁵⁶ The background screening rate was obtained via the Immunoassay Laboratory.

6.1.9 Relationship of intention to screening uptake

Table 6.8 gives the frequencies of serum screening uptake by intention. Although all women who had their booking appointment before 19 weeks gestation were offered the triple test, the test could be conducted as early as 15 weeks. For this reason, participants whose reported gestation was greater than 14 weeks ($n=34$) were not included in this particular analysis to avoid screening ‘intentions’ that may have been reported retrospectively. The figures show that the intention responses were good predictors of test uptake.

Table 6.8. Intention to have serum screening by actual test uptake: frequencies

Serum screening intention	Tested	Not tested	Total
Definitely have test	75	6	81
Probably have test	13	5	18
Don't know	5	4	9
Probably not have test	0	2	2
Definitely not have test	1	17	18
Total	94	34	128

In order to establish whether ‘yes’ and ‘no’ behavioural intentions predicted test uptake and test decline differently, predictive value equations were calculated for the ‘yes’ ($n=99$) and ‘no’ ($n=20$) screening intention categories defined earlier. These equations are commonly used within epidemiology to calculate the predictive values of screening tests i.e. the probability that a person having a positive result has the condition in question, or that one with a negative result does not (Jekel, 1996). The positive predictive value (proportion of women definitely or probably intending to use the test who did so) was 89%. The negative predictive value (the proportion of women definitely or probably intending not to use the test who did not) was 95%. Therefore, while intentions were generally very good predictors of test uptake, an intention **not** to have serum screening was a slightly better predictor of actual test behaviour.

Of the 124 women who had the screening test, six (5%) had a ‘positive’ triple-test result, where the risk of the pregnancy being affected by Down’s syndrome was 1 in 250 or higher. Of these, five women went on to have an amniocentesis. Four had said they definitely or probably intended to have a diagnostic test and one had selected the ‘don’t know’ response. The reason this woman gave for being unsure about having amniocentesis highlights the complexity of testing decisions for some women: she wrote, “*I don’t want to risk miscarriage, but I also don’t want a Down’s baby. I also don’t want to choose to kill my baby if it is Downs.*” The woman who declined to have an amniocentesis had said in the questionnaire that she ‘definitely would not have’ a

diagnostic test because of the risk of miscarriage. One woman who received a negative serum screen result went on to have an amniocentesis following an ultrasound scan. This woman had intended to have the triple test “*for peace of mind*” as she had a cousin with Down’s syndrome, however, while her triple test result was reassuring a subsequent screening process had identified her at risk for an abnormality. The outcomes of these pregnancies are unknown.

6.1.10 Intentions and screening uptake by sociodemographic and obstetric variables

In the following analyses the intention categories of ‘yes’, ‘no’ and ‘don’t know’ defined earlier were used. This was done to reduce complexity within the Results. However, a manual examination of mean scores was conducted and analyses using all five categories for each variable were also carried out, to ensure that a) the probably/definitely and probably not/definitely not pairings were more similar to each other than to other response options, and b) that important differences were not being missed by the category merging. No significant differences were found between the pairs on any variable and the category merging was considered justified.

Nevertheless, it is accepted that in merging these categories some of the more subtle differences between the ‘definitely’, ‘probably’ and ‘don’t know’ groups could have been lost.

Age. Using one-way ANOVAs to compare group means, there was an effect of age by screening intention group ($F(2,190) = 5.52, \rho < 0.01$). Post hoc tests revealed that age did not differentiate between the ‘yes’ and ‘no’ screening intention groups but that women in the ‘don’t know’ category were significantly younger than those who intended to have the test ($\rho = 0.05$). There was an effect of age by termination intention group ($F(2, 189) = 6.48, \rho < 0.01$). Women who intended to terminate for Down’s syndrome were significantly older than the ‘no’ or ‘don’t know’ groups. There was a non-significant trend for older women to be more likely to intend to use amniocentesis ($F(2,189) = 3.08, \rho = 0.052$).

An analysis of the uptake data revealed that those women who did not have the serum screening test were significantly younger than those who did have the test, at 25.4 years and 28.0 years respectively ($t=2.64, df=160, \rho < 0.005$). In addition, the only difference found between the screening ‘don’t knows’ who had actually taken the test ($N=5$) and the screening ‘don’t knows’ who had not taken the test ($N=5$) was their age. Those who took the test had a mean age of 25.8 years compared with 19.8 years in those who did not.

Religion and religiousness. As reported in other studies, the importance of religion rather than religious affiliation was related to behavioural intentions regarding abortion and prenatal testing. There were no significant relationships between religious affiliation and intention, however, the

degree to which religion was reported to affect important life decisions was shown to be significant. The Likelihood ratio test was selected for this analysis, as it is less sensitive than chi-square to small sample sizes. Those participants whose religious upbringing affected their decisions 'quite a lot' ($n=5$) were significantly less likely to intend using the triple test, amniocentesis or to terminate for Down's syndrome than other participants (Likelihood ratio = 11.7, 11.5, and 9.3 respectively, $df = 4$, $\rho < 0.05$). Only one woman, who described her affiliation as Quaker made a specific reference to a religious opposition to abortion. This was combined with a belief that she could successfully parent a child with Down's syndrome despite not living with her partner.

"[I] don't believe in termination from religious perspective. I am quite capable of looking after a Down's syndrome child and view termination as murder."

Most of the women for whom religion was an important life influence spoke of valuing the life of a baby with Down syndrome and of their ability to parent such a child successfully. Uptake data was only available for four of the women who had said their religious upbringing affected their decisions 'quite a lot', but as intended, none had had the screening test.

Education level, parity and marital status. No significant differences were found in intentions by educational level, parity, or marital status.

Previous testing history. For those women who had been offered testing in a prior pregnancy, previous screening behaviour was related to screening intentions. Ninety-six percent of those who said they had used serum screening in a previous pregnancy ($n = 79^{57}$) intended to use it in the current one. Eighty-seven percent of the women who had declined screening in a previous pregnancy ($n=13$) did not intend to use it in their current pregnancy. The remaining two women selected the 'don't know' option. In multiparous women for whom uptake data was available, previous screening behaviour closely matched uptake in this pregnancy. Ninety-one percent of those who said they had used screening in a previous pregnancy used the test in this pregnancy (61/67), and 85% of those who had declined screening in a previous pregnancy did not have the test in this pregnancy (11/13).

⁵⁷ One participant who did not give her screening intention in this pregnancy was excluded from this analysis.

6.1.11 Intentions and screening uptake by psychosocial variables

Anxiety about having a baby with Down's syndrome was greatest in those who intended to have a screening test, an amniocentesis or a termination (Kruskal-Wallis $\chi^2 = 15.6, 26.1$ and 32.3 respectively, $df=2, \rho < 0.001$, see Table 6.9, a higher score reflects greater anxiety). Anxiety scores were also highest in women who had a screening test (Mann Whitney $U = 1637, N_A = 123, N_B = 38, \rho < 0.005$).

Table 6.9. Anxiety about having a baby with Down's syndrome by intention category.

Intention Category	Screening	Amniocentesis	Termination
	M (SD)	M (SD)	M (SD)
Yes	2.1 (0.9)	2.2 (0.9)	2.3 (0.9)
Don't know	1.9 (0.7)	2.0 (1.0)	1.9 (0.8)
No	1.5 (0.9)	1.5 (0.5)	1.5 (0.9)

Perceived likelihood of having an affected child was not significantly associated with screening, intention, termination intention, or screening uptake. However, there was a non-significant tendency for perceived likelihood to relate positively with amniocentesis intention (Kruskal-Wallis $\chi^2 = 5.4, df = 2, \rho = 0.07$) possibly because of the association between age and perceived likelihood and age and amniocentesis intention.

Evaluation of having a baby with Down's syndrome. Those women who intended to use screening, amniocentesis or termination evaluated having a baby with Down's syndrome more unfavorably than those who did not (Kruskal-Wallis $\chi^2 = 25.3, 55.9$ and 105.7 respectively, $df = 2, \rho < 0.001$). Table 6.10 shows the means and standard deviations of these evaluation scores by intention category, a lower score reflects a less favourable evaluation of having a baby with Down's syndrome. As expected from the intention responses, women who had screening generally evaluated having a child with Down's syndrome less favourably than those who did not have the test (Mann Whitney $U = 1131, N_A = 124, N_B = 38, \rho < 0.001$).

Table 6.10. Evaluation of having a baby with Down's syndrome by intention category.

Intention Category	Screening	Amniocentesis	Termination
	M (SD)	M (SD)	M (SD)
Yes	2.5 (1.6)	2.0 (1.4)	1.5 (0.9)
Don't know	3.0 (1.6)	2.7 (1.6)	3.0 (1.4)
No	4.2 (1.8)	4.3 (1.5)	4.6 (1.7)

The **general attitude towards abortion** score was calculated so that the higher the score the more favourable the general attitude to abortion was deemed to be (mean = 1.8, SD = 0.4). This variable was found to discriminate between groups by intentions to have the screening test, amniocentesis and termination for Down's syndrome (Kruskal-Wallis $\chi^2 = 9.8, 17.2$ and 22.8 respectively, $df = 2$, $\rho < 0.01$) with intentions to use testing and termination associated with a more favourable attitude to abortion in general. Neither age, parity, education, nor religion was significantly associated with general attitude to abortion. The relationship between abortion attitude and religiousness was in the expected direction but did not reach significance (Kruskal-Wallis $\chi^2 = 5.3$, $df = 2$, $\rho = 0.07$)

As expected from the intentions data, women who did not have the serum screening test had a significantly less favourable attitude to abortion generally than women who did have the test (Mann Whitney U = 1548, $N_A = 124$, $N_B = 38$, $\rho < 0.01$).

6.1.12 Results Part 1: summary

In summary, the following variables were significantly associated with screening, amniocentesis and termination intentions and with screening test uptake 1) age, 2) religiousness, 3) anxiety about having a child with Down's syndrome, 4) an evaluation of how good or bad such an event would be, and 5) a general attitude to abortion. Screening intentions were good predictors of actual screening behaviour especially in those women who intended not to have a screening test. In multiparous women, testing behaviour in a previous pregnancy was related to intentions to test in the current pregnancy and a good predictor of actual screening behaviour.

6.2 RESULTS PART 2

In this section of the chapter the results of analyses conducted on the data collected using the open-ended measures of attitudes towards Down's syndrome are presented. The first two subsections describe the attitudes data collected. The second section reports the results of the statistical tests and the qualitative analysis used to investigate the relationships between attitudes towards Down's syndrome, testing and termination intentions, and serum screening uptake.

The open-ended measures (Esses *et al.*, 1993) of attitudes towards Down's syndrome constituted the final section of the questionnaire (section E, see Appendix 6). They measured four components of the target attitude; (1) participants' experiences of people with Down's syndrome, (2) stereotypic beliefs about people with Down's syndrome, (3) emotions elicited by meeting or thinking about people with Down's syndrome, and (4) beliefs about how having a child with Down's syndrome would impact on valued aspects of parental life (Parental Quality of life (PQoL))

beliefs). The attitude data were made up of two elements; a) a descriptive response, for example an adjective believed to describe a person with Down's syndrome, and b) an evaluation of that response as either positive, negative, neutral, or mixed (both positive and negative) in valence. The evaluations could range from -2 (very negative/unfavourable) to $+2$ (very positive/favourable). The attitude component scores were computed using the formula $(\sum v)/N$, where v was the evaluation valence and N was the number of responses given. The overall attitude score was the mean value of the sum of the component scores.

6.2.1 Attitudes towards Down's syndrome: descriptive statistics

Sixteen respondents (8% of the total) did not complete any of the open-ended measures correctly. Of these, nine had left the entire section blank and seven had given descriptive responses but had not assigned any valences. A further 41 participants (21%) completed at least one measure incorrectly by leaving it blank or not assigning valences. Table 6.11 gives the response statistics for each of the measures excluding cases where the measure had been incorrectly completed, i.e. responses given but no valence assigned. The number of stereotype and affect responses given are consistent with those found in other studies using the measures (Esses and Beaufoy, 1994; Haddock and Zanna, 1998). However, detailed data for the experience responses have not been reported in other studies, and so it is not known whether the numbers seen here are typical of experience responding generally or specific to experiences of Down's syndrome. As PQoL beliefs were measured in this study for the first time it is not known how representative the response numbers are. However, consistent with findings in others studies, affective responses (emotions) were fewer in number than cognitive responses (stereotypic and PQoL beliefs).

In total, 140 participants completed all measures correctly and in full (71% of the total sample). The women who completed the measures correctly differed significantly from those who did not only in their level of education: 21% of those who completed correctly reported having some form of education beyond the age of 18, compared with 6% of those who not complete correctly ($\chi^2 = 9.5$, $df = 2$, $\rho < 0.01$). There were no significant differences between the two groups' testing and termination intentions or evaluation of having a baby with Down's syndrome.

Table 6.11. Attitudes towards Down's syndrome: descriptive statistics

Attitude component	Mean no. of responses (SD)	Minimum	Maximum
Experiences (N = 160)	1.9 (0.9)	1	5
Positive valences	1.9 (1.4)	0	8
Negative valences	0.9 (1.0)	0	5
Neutral valences	0.2 (0.5)	0	3
Mixed valences	0.4 (0.7)	0	4
Stereotypic beliefs (N = 163)	5.5 (2.7)	1	14
Positive valences	4.0 (2.8)	0	13
Negative valences	2.8 (2.9)	0	16
Neutral valences	0.6 (1.2)	0	6
Mixed valences	0.6 (1.0)	0	6
Emotions (N=163)	3.2 (1.9)	1	10
Positive valences	2.0 (2.2)	0	13
Negative valences	1.9 (1.9)	0	10
Neutral valences	0.4 (0.8)	0	4
Mixed valences	0.4 (0.9)	0	6
PQoL beliefs (N=156)	4.6 (2.6)	1	14
Positive valences	2.0 (2.5)	0	15
Negative valences	3.7 (3.4)	0	20
Neutral valences	0.9 (1.5)	0	6
Mixed valences	1.0 (1.5)	0	9

The attitude scores for the women who correctly completed all the measures ($n = 140$) are given in Table 6.12. Evaluation of experiences of people with Down's syndrome and stereotypic beliefs about the condition were evaluated positively overall. However, the emotions associated with the condition were evaluated negatively overall, and the impact of a child with Down's syndrome on parental quality of life was viewed less favourably still.

Table 6.12. Attitude scores: means and standard deviations

Attitude score	M (SD)
1. Experiences ^b	+0.55 (0.91)
2. Stereotypes ^b	+0.27 (0.80)
3. Emotions ^b	-0.09 (0.94)
4. PQoL beliefs ^b	-0.31 (0.84)
5. Attitude score ^a (mean of 1 to 4)	+0.15 (0.71)

Note. ^a Range is -1.75 to +2, ^b range is -2 to +2, N=140

The coefficients given in Table 6.13 show that each component score correlated positively and significantly with each other. While this indicates a certain intra-attitudinal consistency, the correlations between the four attitude components were not high enough to suggest that they were capturing identical information.

Table 6.13. Inter-correlations among attitude component scores

Attitude component	Experiences score	Stereotypes score	Emotions score	PQoL beliefs Score
Experiences	-	+0.54*	+0.45*	+0.44*
Stereotypes	-	-	0.55*	+0.58*
Emotions	-	-	-	+0.52*

* All Spearman's *rho* significant at the $\rho < 0.01$ level (2-tailed), N=140

6.2.2 Attitudes towards Down's syndrome: qualitative data

The open-ended responses of participants who had completed at least one of the measures correctly were analysed for content (N=181).

Experiences with people with Down's syndrome were reported by 165 participants (84%) and evaluated by 160 participants (81%). The responses were classified into five mutually exclusive categories by common theme (see Table 6.14). Most of the experiences reported were gained through direct contact with people with Down's syndrome; 49% with a specific (but non-related) individual, 24% via working with people with the syndrome, and 14% of participants reported having a family member with Down's syndrome. Indirect experiences via the media (mostly television) were reported by 47% of respondents, and 26% reported indirect experiences such as seeing people with Down's syndrome 'on a bus trip'.

Table 6.14. Experiences of people with Down's syndrome: categories and example responses

	Category and definition	Example response
Direct	Family member with Down's syndrome	"I have a cousin with Down's syndrome".
	Specific (experience referring to a specific person with Down's syndrome, excluding relatives).	"When I was little there was a girl with Down's syndrome in my street."
	Work (experience of dealing with people with Down's syndrome in a voluntary/paid work capacity).	"I have worked with people with Down's syndrome during my career as a nurse".
Indirect	Media (experiences via television, radio, magazines).	"I saw a programme about people with Down's syndrome getting married."
	Non-specific (general experiences of people with Down's syndrome).	"I have occasionally seen Down's syndrome people out and about".

Stereotypic beliefs about people with Down's syndrome were reported by 173 participants (88%) and evaluated by 163 participants (83%). As in the pre-study exercise to obtain example responses, the most frequently expressed beliefs related to the stereotypic personality attributed to people with Down's syndrome. The most frequent response was that a person with Down's syndrome is 'loving' (59% of participants), this was followed by 'friendly' (45%), 'happy' (36%), having 'learning problems' or similar phrase (35%), 'demanding' (29%), vulnerable (23%), and dependent (20%). By contrast, the medical problems associated with Down's syndrome were referred to by only 11% of respondents. It might be that the majority of individuals are unaware of these medical problems, or that these are less salient than other aspects of the condition. Of interest also, was that only one participant expressed the belief that people with Down's syndrome were 'music-loving'. This is a characteristic of people with Down's syndrome frequently included in more traditional closed measures (Wishart and Johnston, 1990).

Emotions associated with people with Down's syndrome were reported by 171 participants (87%) and evaluated by 163 participants (83%). Overall, the most frequent emotions expressed in connection with Down's syndrome were feelings of sympathy or pity for people with Down's syndrome (47% of respondents). Sadness was the second most common feeling (36% of respondents). This was followed by feeling protective towards affected individuals (20%), and feeling lucky or fortunate (19%). These responses were similar to those of the pre-study group in the exercise to obtain example responses,

Parental Quality of Life beliefs were beliefs about how having a child with Down's syndrome would impact on valued aspects (objects) of their parent's lives. PQoL beliefs were reported by 168 participants (85%) and evaluated by 156 participants (79%). The most frequently reported 'valued life object' was 'family' (57% of respondents), followed by 'relationship with partner or husband' (55%), 'physical health' (42%), and 'existing children' (39%). Only three women (2%) made responses related to 'self-actualisation' in contrast to the 13.5% in the pre-study group, perhaps reflecting the academic environment of pre-study group. One woman made a reference to her religious values and perceived that the birth of a child with Down's syndrome would have a neutral impact on this.

Summary

The previous sub-sections describe data collected using the open-ended measures of Down's syndrome. The remainder of the chapter reports results of the statistical tests used to investigate the relationships between attitudes towards Down's syndrome and intentions and screening uptake in the 140 women who correctly completed all measures, and a qualitative analysis of the material captured by the open-ended measures.

6.2.3 Attitudes by sociodemographic and obstetric variables

The age of the participant correlated significantly and positively with the total number of responses given ($rho = +0.21, \rho < 0.05$) and with the number of experiences reported ($rho = +0.34, \rho < 0.01$), symbolic belief and affect responses ($rho = +0.18$ and $+0.17$ respectively, $\rho < 0.05$). This might be expected, in that older women have had a longer period of time in which to collect attitude relevant material. However, age correlated significantly and negatively with the experiences score ($rho = -0.23, \rho < 0.05$), stereotypes score ($rho = -0.31, \rho < 0.001$), emotions score, ($rho = -0.18, \rho < 0.05$), PQoL beliefs score ($rho = -0.27, \rho < 0.01$), and overall attitude score ($rho = -0.31, \rho < 0.01$). Thus, older women tended to hold the least favourable attitudes towards Down's syndrome.

Women who said that their religion had 'quite a lot' of influence over important life decisions reported more experiences of people with Down's syndrome than did women who selected one of the other two categories ('Little influence' and 'No influence' (Kruskal-Wallis $\chi^2 = 15.8, df = 2, \rho < 0.001$). This may be because of contact with people with disabilities through church related functions, or because of the type of situation those with higher levels of religious belief might seek out, such as involvement with disabled groups. However, the overall attitude score and component attitude scores did not show any significant effect by level of religiousness.

No other socio-demographic or obstetric variable, including education level and parity, was significantly related at $\rho < 0.05$ with the number of responses given or the attitude scores calculated

6.2.4 Attitudes by psychosocial variables

Anxiety about having a baby with Down's syndrome was significantly related to all attitude components (Experience, $\rho = -0.30$, Stereotypic beliefs, $\rho = -0.22$, Emotions $\rho = -0.36$, and PQoL beliefs $\rho = -0.32$, all $\rho < 0.01$) and the overall attitude score ($\rho = -0.38$, $\rho < 0.01$). Therefore, as attitudes towards Down's syndrome became less favourable anxiety about having an affected child increased. **Perceived likelihood** of having an affected baby was not significantly associated with attitudes towards Down's syndrome.

Women's **attitudes towards abortion** generally were also related to attitudes towards Down's syndrome with the exception of the experience component (Stereotypic beliefs, $\rho = -0.21$, Emotions, $\rho = -0.17$, overall attitude score, $\rho = -0.20$, all $\rho < 0.05$, and PQoL beliefs $\rho = -0.22$, $\rho < 0.01$). More favourable attitudes towards abortion were associated with less favourable attitudes towards Down's syndrome.

Attitudes and the evaluation of having a child with Down's syndrome

All attitude component scores correlated significantly and positively with the evaluation of having a baby with Down's syndrome at $\rho < 0.01$: Experiences ($\rho = +0.47$), Stereotypic beliefs ($\rho = +0.56$), Emotions ($\rho = +0.56$), and PQoL beliefs ($\rho = +0.68$). The overall attitude score also correlated significantly with this evaluation variable ($\rho = +0.70$, $\rho < 0.01$). A regression analysis was conducted to establish which attitude components best predicted an evaluation of having a baby with Down's syndrome. As the distribution of the dependent variable was strongly skewed linear regression was inappropriate. Instead, the evaluation variable was dichotomised and binary logistic regression was selected. The responses were separated into two categories using the following criterion; if having a baby with Down's syndrome had been evaluated unfavourably (a response of 1 through 4), this was termed the 'Down's syndrome not OK' category (DSNOK), N=106. All other responses, i.e. scores of 5 through 9 (neutral and favourable evaluations), were placed in the 'Down's syndrome OK' category (DSOK), N=34. Despite the unequal group sizes this was considered the most appropriate way to dichotomise the data as a median split would not accurately reflect the distribution of responses.

The attitude component variables were entered simultaneously to establish the unique contribution of each component towards the prediction of the evaluation variable. The goodness-of-fit test produced a non-significant chi-square value ($\chi^2 = 4.95$, $df = 8$, $\rho = 0.76$) indicating that the logistic model fitted the data at an acceptable level (Kinnear and Gray, 2000). The Nagelkerke R^2 value, which gives an approximation of the amount of variance accounted for by the predictor variables, was 47%. **Of the four variables entered, only the Emotion and PQoL beliefs scores contributed significantly towards predicting the evaluation variable** (*Beta* values -0.79 and -1.23, $\rho < 0.05$ and < 0.005 respectively). The analysis was re-run with only Emotion and PQoL beliefs as regressor variables (see Table 6.15). The classification table produced by SPSS showed that 84% of the cases were predicted correctly. However, prediction was more accurate for the DSNOK category (93%) than for the DSOK category (53%). This demonstrates that while unfavourable attitudes towards Down's syndrome indicate an unfavourable evaluation of having a baby with the condition, favourable attitudes towards Down's syndrome do not always indicate a favourable evaluation of having a baby with the condition. As noted in the introductory chapter, most women hope for a 'perfectly normal child' even if they are not willing to actively take steps to avoid having a baby with a disability. It might be expected that past experiences would impact on views about having a baby with Down's syndrome but this might be via emotions and PQoL beliefs associated with the condition. However, as all the attitude components were correlated, an assumption about which variable is causally prior cannot be safely made.

Table 6.15. Regression coefficients for variables predicting evaluation of having a baby with Down's syndrome

Regressor variable	Beta	SE	Sig. ($\rho <$)
Emotion score	-1.01	.303	0.001
PQoL score	-1.44	.398	0.000
Constant	1.37	.284	0.000

6.2.5 Attitudes by testing and termination intentions and screening uptake

Tables 6.16, 6.17, and 6.18 show the descriptive statistics for the attitude component scores by intention category along with the results of one-way ANOVAs used to compare mean attitude scores across groups. For screening intentions significant differences were found only between the 'Yes' and 'No' groups. For the amniocentesis and termination intentions significant differences were found between all groups. The differences were all in the expected direction, that is, participants with the most favourable attitudes towards Down's syndrome were least likely to intend to test and terminate and participants with the least favourable attitudes were most likely to intend to test and terminate. The numbers of positive and negative valences were compared across

intention group for each attitude component. However, the results essentially replicated the main findings and so are not presented here.

Table 6.16. Attitude scores by screening intentions: means, standard deviations, and results of comparisons between groups (F values for one-way ANOVAs)

Screening Intention ^a (n)	Experience (SD)	Stereotypes (SD)	Emotions (SD)	PQoL (SD)	Overall attitude (SD)
Yes (113)	0.5 (1.0)	0.2 (0.8)	0.0 (0.9)	-0.4 (0.8)	0.1 (0.7)
Don't know (6)	0.3 (0.8)	0.3 (0.5)	0.4 (0.8)	-0.3 (0.9)	0.2 (0.5)
No (18)	1.1 (0.7)	0.7 (0.7)	0.8 (0.9)	0.4 (0.7)	0.7 (0.6)
F (2, 134)	3.7*	3.3*	6.9**	9.0**	8.5**

$\rho < 0.05$, ** $\rho < 0.001$

^a Two participants who intended to bypass screening by having an early diagnostic test were excluded from the screening intention analyses.

Table 6.17. Attitude scores by amniocentesis intentions: means, standard deviations, and results of comparisons between groups (F values for one-way ANOVAs)

Amniocentesis Intention (n)	Experience (SD)	Stereotypes (SD)	Emotions (SD)	PQoL (SD)	Overall attitude (SD)
Yes (71)	0.3 (0.9)	0.0 (0.8)	-0.2 (0.9)	-0.7 (0.8)	-0.1 (0.6)
Don't know (31)	0.5 (0.8)	0.2 (0.6)	-0.1 (0.7)	-0.3 (0.7)	0.1 (0.5)
No (36)	1.2 (0.9)	0.7 (0.9)	0.7 (0.9)	0.3 (0.8)	0.7 (0.7)
F (2, 135)	12.4**	9.7**	13.1**	20.5**	23.5**

** $\rho < 0.001$

Table 6.18. Attitude scores by termination intentions: means, standard deviations, and results of comparisons between groups (F values for one-way ANOVAs)

Termination Intention (n)	Experience (SD)	Stereotypes (SD)	Emotions (SD)	PQoL (SD)	Overall attitude (SD)
Yes (62)	0.1 (0.9)	-0.2 (0.6)	-0.5 (0.7)	-0.9 (0.6)	-0.4 (0.5)
Don't know (38)	0.7 (0.8)	0.4 (0.6)	0.2 (0.7)	-0.1 (0.7)	0.3 (0.5)
No (39)	1.2 (0.8)	0.9 (0.8)	0.9 (0.8)	0.3 (0.7)	0.8 (0.6)
F (2, 136)	20.8**	30.8**	39.3**	44.9**	66.9**

** $\rho < 0.001$.

An analysis of test uptake by attitudes towards Down's syndrome revealed significant differences (with the exception of the experiences score) in the expected direction (see Table 6.19).

Table 6.19. Attitudes by screening uptake: means, standard deviations and results of Mann Whitney tests

	Experience (SD)	Stereotypes (SD)	Emotions (SD)	PQoL beliefs (SD)	Overall attitude (SD)
Tested (N=92)	0.5 (1.0)	0.2 (0.8)	0.1 (0.9)	-0.4 (0.8)	0.1 (0.7)
Not tested (N=29)	0.9 (0.7)	0.7 (0.6)	0.6 (0.9)	0.2 (0.7)	0.6 (0.6)
Mann Whitney U=	1016.0	805.5*	879.5*	724.5*	765.5*

* $\rho < 0.01$

The results so far portray a consistent and possibly predictable picture of the relationship between attitudes towards Down's syndrome and testing and termination intentions. However, further analysis of the data revealed an asymmetric pattern not immediately obvious from the comparison of mean attitude scores. Participants were grouped into four categories (quartiles) by their overall attitude score, and an analysis of intention responses of those in the first and fourth quartiles, i.e. those with the least favourable and the most favourable attitudes towards Down's syndrome was conducted (see Table 6.20). Of those with the least favourable attitudes, 97% intended to use screening, 82% intended to use amniocentesis, and 94% intended to terminate. However, even in the group with the most favourable attitudes towards Down's syndrome the majority (65%) intended to have a screening test. By contrast 29% intended to use amniocentesis and only 6% ($n=2$) intended to terminate.

Table 6.20. Participants with 'least favourable' ($n=34$) and 'most favourable' ($n=34$) attitudes towards Down's syndrome: testing and termination intentions

	Attitude towards Down's syndrome	Yes	Don't know	No
Screening	Least favourable attitude	33	1	0
	Most favourable attitude	22	2	10
Amniocentesis	Least favourable attitude	28	5	1
	Most favourable attitude	10	5	19
Termination	Least favourable attitude	32	2	0
	Most favourable attitude	2	8	24

Attitude score ranges: 'Least favourable' = -1.8 to -0.3, 'Most favourable' = +0.6 to +2.0.

Thus, while unfavourable attitudes accurately predict screening, amniocentesis and termination intentions, and favourable attitudes appear to predict amniocentesis and termination intentions reasonably well also, they are not good predictors of intentions to use serum screening. This asymmetric pattern was confirmed by the analysis of screening uptake by attitudes towards Down's syndrome. Positive and negative predictive screening uptake values were calculated for the 'most favourable' and 'least favourable' attitude groups (see Table 6.21). The proportion of women in the 'least favourable' attitude group who used the screening test was 100%. The proportion of women in the 'most favourable' attitude group who did not use the test was 33%. In other words, 67% of those whose attitudes towards Down's syndrome were the most favourable still had a prenatal test to screen for the condition.

Table 6.21. Serum screening uptake by overall attitude score of 'least favourable' ($n=28$) and 'most favourable' ($n=33$) attitude groups

Attitude to Down's syndrome	Tested	Not tested	Predictive value
Least favourable	28	0	100%
Most favourable	22	11	33%

Attitude score ranges: 'Least favourable' = -1.3 to -0.3, 'Most favourable' = +0.6 to +2.0.

In order to explore other factors related to triple-test uptake in the group with the most favourable attitudes towards Down's syndrome, women in this group were categorised as 'tested' ($N=22$) and 'not-tested' ($N=11$). Significant differences were found by age (the tested group were oldest, Mann Whitney $U=43.5$, $\rho < 0.01$), by intention to have a diagnostic test (tested group more likely to intend to have amniocentesis, Likelihood ratio = 15.3, $df = 2$, $\rho < 0.001$), and evaluation of having a baby with Down's syndrome (the tested group gave a less favourable evaluation, Mann Whitney $U=50$, $\rho < 0.05$). However, the tested group were not more likely to intend to have a termination of an affected pregnancy (Likelihood ratio = 4.5, $df = 2$, $\rho = 0.12$). It can be concluded that intending to have (or having) a serum-screening test did not necessarily predict an unfavourable attitude towards Down's syndrome. By contrast, the holding of unfavourable attitudes towards Down's syndrome was a very good predictor of intending to use, and using serum screening. It is proposed that factors associated with an older age group, such as increased saliency of Down's syndrome and a more established lifestyle might take precedence over personal attitude towards the condition when considering screening. For these reasons, reassurance of a healthy child might be a particular motivation in this group.

6.2.6 Content analysis of qualitative data: experiences, stereotypic beliefs, emotions, and PQoL beliefs by intention to terminate for Down's syndrome

Other studies that have used the open-ended measures of attitudes have not reported an in-depth analysis of the descriptive responses as captured by the measures. However, it was felt that in conjunction with the statistical analyses, a qualitative analysis of these responses would contribute to a deeper understanding of the relationship between attitudes towards Down's syndrome and intention responses. It has been said that while quantitative data describe the scope and extent of the topic, qualitative data provide the voices that "*carry through the sense of the phenomena under investigation*" (Parker, 1994, p.15). The relationship between attitudes towards Down's syndrome and termination intentions was identified as the most significant statistically and so it was decided to conduct a content analysis of the descriptive responses by termination intention group in all cases where *that measure* had been correctly completed.

Experiences

Using the categories of direct and indirect experiences of people with Down's syndrome (as defined earlier in this chapter) frequencies for each category were calculated by intention to terminate. Table 6.22 shows relative frequencies by category to be similar across groups, although the 'No to termination' group reported most direct experiences and fewest indirect experiences overall. Intention to terminate for Down's syndrome was analysed by whether someone had a family member with Down's syndrome ($n=21$) or not ($n=176$); 29% with a family member compared with 48% of those without a family member intended to terminate for Down's syndrome. However, while participants across groups reported a similar range of experiences the individual responses were evaluated differently by termination intention. For example, media experiences of Down's syndrome were evaluated negatively overall by the 'yes to termination' group and positively by the 'no' group.

Table 6.22. Experiences of people with Down's syndrome by intention to terminate (proportion of total responses %).

Experience category	Yes to termination ($n=74$)	Don't know ($n=43$)	No to termination ($n=47$)
Work (direct)	15	16	18
Family member (direct)	4	6	13
Specific person (direct)	32	27	28
Media (indirect)	32	31	30
Non-specific (indirect)	17	20	11
<i>Total</i>	100	100	100

Stereotypic beliefs

Using the Stereotypic beliefs categories as defined in the pre-study exercise in Chapter 5 (section 5.4.2) frequencies for each category were calculated by intention to terminate. Table 6.23 gives the frequencies of stereotypic beliefs by intention to terminate for Down's syndrome.

Table 6.23. Stereotypic beliefs about people with Down's syndrome by intention to terminate (proportion of total responses %).

Stereotype category	Yes to termination (n=76)	Don't know (n=48)	No to termination (n=47)
Appearance	9	7	7
Care requirements	13	9	6
Learning difficulty	12	10	9
Medical problems	3	4	2
P-B phenotype	50	58	67
Psychosocial aspects	10	9	5
Differentness	3	3	4
<i>Total</i>	100	100	100

Across all groups, beliefs relating to the personality/behavioural phenotype of Down's syndrome predominated with virtually every participant including at least one of these characteristics in their responses. Beliefs about the care needs of people with Down's syndrome were most frequently expressed by the 'Yes' to termination group, particularly beliefs that a person with Down's syndrome is demanding (40% of the 'Yes' group, 13% of the 'No' to group). Beliefs about the appearance of people with Down's syndrome appeared salient to similar numbers across groups although the 'Yes' group were most likely to view someone with the condition as unattractive. In contrast, three participants in the other two groups commented that people with Down's syndrome are "beautiful". The 'Yes' to termination group most commonly expressed negative beliefs about psychosocial aspects of the condition such as social isolation and prejudice. Twenty-eight percent of the 'Yes' group and 21% of the 'Don't know' group described people with Down's syndrome as 'dependent', compared with 4% of the 'No' group. Responses that fell within the 'Differentness' category represented only a small proportion of the total responses, however, those in the 'Yes' group expressed the belief that people with Down's syndrome are 'different' most frequently ($n=12$) and rated this negatively.

The medical problems associated with Down's syndrome were referred to by few respondents but they were evaluated negatively regardless of intention to terminate. In contrast the characteristic of learning difficulty was reported with similar frequency across groups but was evaluated differently by intention to terminate; 80% of the 'Yes to termination' group evaluated learning difficulty negatively as did 71% of the 'Don't know' group. In contrast, 40% of the 'No to termination' group evaluated learning difficulty negatively with the remainder evaluating it as neutral or mixed.

Emotions

Using the Emotion categories as defined in the pre-study exercise in Chapter 5 (section 5.4.2) frequencies for each category were calculated by intention to terminate. Table 6.24 gives the relative frequencies of emotion responses by intention to terminate for Down's syndrome.

Negative emotions of Anger, Disgust, Fear, Guilt, Sadness and Shame accounted for 60% of the 'Yes to termination' group responses but only 23% of the 'No' group. In contrast, the positive emotions of Acceptance, Joy, and Love accounted for 67% of the emotions elicited in the 'No' group, but 29% in the 'Yes' group. The 'Don't know' group expressed 47% positive emotions and 38% negative emotions. None of the responses of 'Yes to termination' group included feelings of acceptance towards people with Down's syndrome compared with 7% of the responses of the 'No' group.

Table 6.24. Emotions expressed about people with Down's syndrome by intention to terminate (proportion of total responses %).

Emotion category	Yes to termination (n=78)	Don't know (n=47)	No to termination (n=44)
Acceptance	0	3	7
Anger	2	1	0
Disgust	1	0	0
Fear	13	8	6
Feel fortunate	8	9	6
Guilt	5	2	1
Interest/curiosity	3	5	2
Joy	4	5	11
Love/concern	25	40	51
Sadness	29	20	10
Shame/embarrassment	10	7	6
<i>Total</i>	100	100	100

Because the emotions expressed were very different across group it is useful to see the most frequent emotion responses by intention to terminate (Table 6.25). The mixture of positive and negative emotions given by the ‘Don’t know’ group is more apparent here as is the contrast between the type of emotion responses expressed by the ‘Yes’ and ‘No’ to termination groups.

Table 6.25. Most frequently elicited emotions by intention to terminate: proportion of participants giving the response.

Yes to termination (N=78)		Don’t know (N=47)		No to termination (N=44)	
	%		%		%
Sadness	51	Sympathetic towards	36	Admiration	30
Sorry for	27	Sadness	32	Protective	30
Sympathetic towards	26	Protective	26	Caring	25
Lucky	22	Admiration	23	Happiness	23
Awkward	17	Sorry for	23	Loving	23
Nervous	17	Lucky	19	Sympathetic towards	20

Parental Quality of Life beliefs

Parental Quality of Life beliefs were evaluations of how having a child with Down’s syndrome would impact on valued aspects of their parent’s lives (termed valued life objects). The valued life objects (VLOs) were classified into seven categories, six of which were defined in the pre-study exercise (Chapter 5, section 5.4.2). One further category of ‘Social Appraisal’ was added after data input. The need for positive social appraisal (“to be respected and admired by others”) was measured in the study upon which the VLO categories were originally based (Evers-Kiebooms *et al.*, 1993) but no responses of this type were given in the pre-study exercise. However, responses such as “Not standing out abnormally” and “Being accepted socially” were observed in the main study and these were assigned to the Social Appraisal category. Table 6.26 gives the frequencies of the valued life objects by intention to terminate for Down’s syndrome.

Table 6.26. Valued life objects by intention to terminate (proportion of total responses %).

VLO category	Yes to termination (n=75)	Don't know (n=45)	No to termination (n=47)
Health/well-being	14.5	22	15
Job/career	8	4	5
Material aspects	6	9	7
Pleasure/relaxation	16.5	8	14
Relationships	52.5	49	57
Self-actualisation	0.5	1	2
Social appraisal	2	0	0
<i>Total</i>	100	100	100

Relationships with family including partner and existing children were expressed most frequently as valued objects across all groups, but especially by those in the 'No' to termination category. 32% percent of the 'No' group gave 'Love and affection' as a valued life object compared with 19% of the 'Yes' group and 22% of the 'Don't know' group. Those who did not intend to terminate were more likely to believe that having a baby with Down's syndrome would have a neutral to positive impact on their important relationships, whereas those who intended to terminate believed the impact would be negative. This pattern was also seen in the perceived relationship with the new baby which was cited as a 'valued object' by 9% of the 'Yes' group and by 7% of the 'No' and 'Don't know' groups. The 'Yes' group believed that if their baby had Down's syndrome this would impact negatively on the mother-child relationship. For example;

Valued life object = *"Being able to bond with a normal child (not a DS child)"* [negative evaluation].

Those in the 'No to termination' group held less unfavourable beliefs in this respect.

Valued life object = *"My baby. It may be different to my other children but I would still love and care for it as much"* [neutral evaluation].

Across all groups the belief was expressed that having a child with Down's syndrome would have an unfavourable impact on a career, and on pleasure and relaxation. These beliefs were most salient for those in the 'Yes to termination' group. Women in the 'Don't know' group were most likely to report health and well being as a valued life object, but those in the 'Yes' group were most likely to believe that having a child with Down's syndrome would have a detrimental impact on health and well-being. Very few beliefs associated with the importance of positive social appraisal were expressed, and these only arose in the 'yes' to termination group. The responses

reflected a belief that having a child with Down's syndrome would impact negatively on how others perceived them.

Summary of the content analysis

The findings of this analysis suggest six main points in relation to the attitude responses towards Down's syndrome by intention to terminate for the condition.

- Those who reported experiences with a relative with Down's syndrome were less likely to intend to terminate for the condition than those who did not have a relative with the condition.
- The perceived burden of caring for a child with Down's syndrome was a particularly salient concept for those who intended to terminate for the condition. However, burden and dependency does not appear to be a salient issue for many women who would not wish to terminate. This is despite a common belief across groups that a child with Down's syndrome would have a negative effect on leisure time and career.
- Those who did not intend to terminate appeared to view learning difficulty less negatively than did those who did intend to terminate or were uncertain. This might be based on personal experience; for example, other studies have shown that having direct experience of a family member with a condition tends to reduce perception of its severity
- Most women who intended to terminate for Down's syndrome associated unpleasant emotions such as sadness, pity, and fear or discomfort with people with the condition. These feelings would explain why a person would want to avoid a child who was the object of such emotions in self and others. In contrast more positive feelings such as admiration and love were experienced in relation to people with Down's syndrome by those who did not intend to terminate. Protectiveness and caring were experienced more frequently than sadness or pity.
- While respondents valued similar aspects of their lives they differed markedly in the impact they believed that a child with Down's syndrome would have on them - most notably on their family relationships, their feelings towards the new baby, and their physical and mental health. These differences were reflected in intentions to terminate for Down's syndrome.

These aspects of attitudes help explain why some individuals intended to terminate a pregnancy affected by Down's syndrome and why some did not. It is suggested that the concept of 'otherness' was important here. Women who did not intend to terminate for Down's syndrome appeared to view a child with Down's syndrome as essentially similar to a typically developing child – care was not considered burdensome, feelings were similar to those likely to be experienced by parents generally, and although life changes were expected they were not anticipated to be especially negative. In contrast, those who intended to terminate appeared to view a child with Down's syndrome as essentially different to a typically developing child – care as a

burden, a source of sadness, and the cause of negative changes to important aspects of one's life including the relationship with the child.

- Finally, the responses of the 'Don't know' respondents indicated ambivalence in both beliefs and feelings about Down's syndrome, and its potential importance in the uncertainty about terminating a pregnancy for the condition. The findings in relation to attitudinal ambivalence towards Down's syndrome are explored in more detail in the next section.

6.2.7 Attitudinal ambivalence towards Down's syndrome

In order to further understanding of why some women are uncertain about their testing and termination intentions, this study aimed to investigate the role of attitudinal ambivalence in the relationships between such intentions, serum screening uptake and attitudes towards Down's syndrome. For cases where all the measures had been correctly completed (N=140) ambivalence within each attitude component was calculated using the formula specified in the previous chapter⁵⁸. An overall ambivalence score for each participant was calculated by obtaining the mean of the component ambivalence scores. Table 6.27 gives the descriptive statistics for both the component and the overall ambivalence scores. The higher the score the greater the attitudinal ambivalence is taken to be.

Table 6.27. Descriptive statistics for attitudinal ambivalence towards Down's syndrome

Type of ambivalence	M (SD)	Minimum	Maximum
Experiences	27.6 (2.4)	20.0	35.0
Stereotypes	28.7 (5.3)	16.0	44.0
Emotions	27.8 (3.6)	18.0	42.0
PQoL beliefs	27.6 (5.4)	13.0	46.0
Overall ambivalence score	27.9 (2.9)	19.8	36.0

A within-subjects analysis showed that significantly more ambivalence was associated with the stereotypic beliefs than with the other three components ($\chi^2 = 8.2$, $N = 140$, $df = 3$, $\rho < 0.05$, Friedman's rank test for k correlated samples). Inter-component correlations demonstrated significant correlations between all components with the exception of experience ambivalence

⁵⁸ The formula used was $(P + N) - 2 |P - N| + 28$, where P was the number of positive valences, N was the number of negative valences and 28 was the constant added to avoid negative ambivalence scores +2 multiplied by 14 (the maximum number of responses) (Bell *et al.*, 1996). The potential range of ambivalence scores was from zero (maximum number of responses with no conflicting valences) to 56 (maximum number of responses with maximum number of conflicting valences).

with PQoL belief ambivalence (Table 6.28). Overall attitude score did not correlate significantly with overall ambivalence score (Pearson's $r = -0.14$, ns.), supporting the relative independence of ambivalence and attitudes.

Table 6.28. Attitude component ambivalence scores: inter-correlations

	Experience ambivalence	Stereotype ambivalence	Emotion ambivalence	PQoL belief ambivalence
Experience ambivalence	-	0.27**	0.41**	0.14
Stereotype ambivalence	-	-	0.50**	0.17*
Emotion ambivalence	-	-	-	0.30**

* $\rho < 0.05$, ** $\rho < 0.01$ (Pearson's r)

Ambivalence towards Down's syndrome related significantly to only one socio-demographic variable – religiousness. The PQoL ambivalence scores of those for whom religious upbringing influenced life decisions 'quite a lot' were significantly greater than those for whom religion had 'a little influence' or none at all (Kruskal-Wallis $\chi^2 = 7.5$, $df = 2$, $\rho < 0.05$). An analysis of religiousness by overall ambivalence score quartiles found that 34% of the most ambivalent group (N=35, ambivalence score range = 29.8 to 36.0) said that religion influenced their life decisions a little or quite a lot. In comparison, only 9% of the least ambivalent group (N=34, ambivalence score range = 19.8 to 25.8) said that religion influenced their life decisions to any degree at all.

6.2.8 Ambivalence by testing and termination intentions and screening uptake

No effects of ambivalence towards Down's syndrome by intention to screen were found (Table 6.29) although differences in component ambivalence scores by intention to have amniocentesis (Table 6.30) and termination (Table 6.31) were mostly significant.

Table 6.29. Ambivalence scores by intention to screen for Down's syndrome: means and standard deviations, and results of comparisons between groups (Kruskal-Wallis tests).

Screening Intention (<i>n</i>)	Experience (SD)	Stereotypes (SD)	Emotions (SD)	PQoL (SD)	Overall ambivalence (SD)
Yes (113)	27.7 (2.4)	28.9 (5.1)	27.8 (3.6)	27.3 (5.3)	27.9 (2.8)
Don't know (6)	27.8 (1.5)	30.5 (4.0)	27.0 (2.3)	28.7 (6.5)	28.5 (2.8)
No (18)	26.8 (2.4)	27.8 (6.3)	28.0 (3.1)	29.1 (5.5)	27.9 (3.4)
Kruskal Wallis χ^2	2.7	2.3	0.5	1.2	0.4

Table 6.30. Ambivalence scores by intention to use amniocentesis: means and standard deviations, and results of comparisons between groups (Kruskal-Wallis tests).

Amniocentesis Intention (<i>n</i>)	Experience (SD)	Stereotypes (SD)	Emotions (SD)	PQoL (SD)	Overall ambivalence (SD)
Yes (71)	28.0 (2.4)	29.7 (5.2)	27.7 (3.7)	26.3 (4.6)	27.9 (2.6)
Don't know (31)	27.8 (1.8)	29.1 (4.2)	27.8 (2.3)	29.0 (6.8)	28.4 (2.9)
No (36)	26.4 (2.7)	26.9 (5.7)	28.1 (4.3)	28.7 (5.0)	27.5 (3.3)
Kruskal Wallis χ^2	14.0**	6.9*	0.82	6.9*	2.9

* $\rho < 0.05$, ** $\rho < 0.005$ $df = 2$

Table 6.31. Ambivalence scores by intention to terminate for Down's syndrome: means and standard deviations, and results of comparisons between groups (Kruskal-Wallis tests).

Termination Intention (<i>n</i>)	Experience (SD)	Stereotypes (SD)	Emotions (SD)	PQoL (SD)	Overall ambivalence (SD)
Yes (62)	28.1 (2.4)	29.8 (5.2)	27.5 (3.8)	25.2 (4.7)	27.7 (2.9)
Don't know (38)	27.7 (2.2)	29.4 (4.6)	28.8 (3.1)	30.2 (5.3)	29.0 (2.6)
No (39)	26.5 (2.2)	26.3 (5.4)	27.1 (3.5)	28.7 (4.9)	27.2 (2.9)
Kruskal Wallis χ^2	12.3**	14.2**	10.3**	23.3***	11.8**

* $\rho < 0.05$, ** $\rho < 0.005$, *** $\rho < 0.001$, $df = 2$

Participants who did not intend to use amniocentesis or termination had significantly lower experience and stereotypic belief ambivalence scores than the other two groups. In contrast those who intended to use amniocentesis and termination had significantly lower PQoL ambivalence scores. Those in the 'Don't know to termination' group had a significantly higher emotion and PQoL ambivalence scores than those in the 'yes' or 'no' groups. The 'Don't know to termination' group also had significantly higher overall ambivalence scores than the other two groups suggesting that women who were unsure whether or not they would terminate for Down's syndrome held the greatest level of attitudinal ambivalence towards the condition.

Ambivalence by screening uptake showed only one significant difference, women who did have the screening test had a lower PQoL belief ambivalence score than women who did not take the test ($t = -2.80$, $df = 119$, $\rho < 0.01$).

Further analyses of the 'Don't know to termination' respondents

Of the 47 women who gave a 'don't know' response to termination and for whom there was screening uptake data, 35 were tested (75%) and 12 were not (25%). There were no significant differences between these two groups in terms of anxiety about having an affected child, attitude or attitudinal ambivalence towards Down's syndrome. However, the women who were tested held more favourable attitudes towards abortion in general ($t = 2.1$, $df = 45$, $\rho < 0.05$), and there was a trend for the women who were tested to be older than the women who were not tested ($t = 1.95$, $df = 45$, $\rho = 0.06$). These relationships might emerge more strongly in a larger sample.

Ambivalence or neutrality?

In addition to higher overall ambivalence scores, the 'Don't know' to amniocentesis and termination groups also had the greatest ratio of neutral responses to total responses (Kruskal-Wallis $\chi^2 = 7.0$, $df = 2$, $\rho < 0.05$). To test the independence of the ambivalence and neutral constructs, the ratio of mixed responses (allocation of both a positive and a negative evaluation to the same response) to total responses, and the ratio of neutral responses (zero evaluation) to total responses was correlated with the overall ambivalence score. While the mixed response ratio related significantly with the ambivalence score ($\rho = +0.65$, $\rho < 0.001$), the neutral response ratio did not ($\rho = +0.07$, ns.). In addition, the ratios of mixed and neutral responses did not correlate significantly with each other ($\rho = -0.11$, ns.). This supports the existence of attitude ambivalence and attitude neutrality as separate constructs.

6.2.9 Discriminant Analysis

In order to identify those factors that made the greatest contribution to the prediction of the intention and uptake variables some form of regression analysis was required. However, as the distributions of the dependent variables were not normal, this violated one of the primary assumptions of linear regression. Binary logistic was also inappropriate as there were three intention categories ('Yes', 'No', and 'Don't know'). Multinomial logistic regression was considered as SPSS version 9.0.0 supports this analysis, however this option is still relatively new and texts on using the software and understanding the output are not widely available. For this reason the more conventional statistical method of Discriminant Analysis was selected. Discriminant Analysis aims to identify the combination of independent variables that contribute maximally to group separation by the dependent variable (Duarte Silva and Stam, 1995).

Stepwise discriminant analyses⁵⁹ were run for each of the four main dependent variables, 1) intention to have a screening test, 2) actual screening test uptake, 3) intention to have an amniocentesis and, 4) intention to terminate for Down's syndrome. Only cases with a complete attitude data set were included in the analysis. In the screening intentions analysis participants who had given a 'don't know' response were excluded because of their small number ($n=6$) leaving only the 'yes' and 'no' groups. For the screening uptake analysis there were only two possible categories, 'yes' and 'no'. For the amniocentesis and termination analyses three levels of the dependent variables were included, 'Yes', 'No' and 'Don't know'. As group sizes were unequal, the prior probabilities used for group classification were based on observed group sizes (Field, 2000). All variables that had significantly correlated with or differed between the target variables were considered as independent variables in the analysis. These were checked to ensure they met assumptions of independence, multivariate normality, homogeneity of variance, and non-multicollinearity⁶⁰. In addition, as Discriminant Analysis is not robust to outliers, data were examined using the 'explore' option in SPSS and three cases were removed. Table 6.32 shows whether the independent variables were rejected *a priori* of the analysis and the reason for rejection. All variables that had a significant relationship with the dependent variables were considered for input to the analysis with the exception of a) the intention variables themselves, i.e. intention to terminate for Down's syndrome in the model predicting amniocentesis, and b) the overall evaluation of having a baby with Down's syndrome, as it was considered to be more useful to identify the predictive value of individual components rather than the more general attitude.

⁵⁹ The stepwise method of discriminant analysis was selected as it is applicable to most situations (Kinnear and Gray, 2000). However, it is also accepted that debate exists about the best variable entry method, not all of which are supported by SPSS (Duarte Silva and Stam, 1995)

⁶⁰ Multicollinearity exists when variables are highly inter-correlated (Kinnear and Gray, 2000).

Table 6.32. Discriminant analysis: independent variables considered, rejected *a priori*, retained and not retained in the discriminant function model of intentions and screening uptake

Independent variable considered	Screening intention	Screening uptake	Amniocentesis intention	Termination intention
Age	Rejected ¹	Rejected ¹	Rejected ¹	*
Anxiety	✓	*	*	*
Religiousness	Rejected ²	Rejected ²	Rejected ²	Rejected ²
General attitude to abortion	✓	✓	✓	✓
Experiences score	*	Rejected ²	*	*
Stereotypes score	*	*	*	*
Emotions score	*	*	*	✓
PQoL beliefs score	✓	✓	✓	✓
Experience ambivalence	Rejected ¹	Rejected ¹	✓	*
Stereotype ambivalence	Rejected ¹	Rejected ¹	*	*
Emotion ambivalence	Rejected ¹	Rejected ¹	Rejected ¹	*
PQoL beliefs ambivalence	Rejected ¹	*	*	✓

Key: ✓ Included in final model, * not included in final model.

¹ No significant difference in mean scores across groups, ² Unequal variance across groups

Results of the Discriminant Analysis

Intention to screen. Three variables were included within one function that accounted for 22% of the variance in intention to have screening. These were (in order of magnitude of contribution to the model) the PQoL beliefs score, the general attitude to abortion, and anxiety related to having a baby with Down's syndrome. The discriminant function separated the two groups by intention to have the triple test at a significant level (Wilks' Lambda = 0.78, $\chi^2 = 31.1$, $df=3$, $p < 0.001$). Women who intended to have screening held less favourable beliefs about how a child with Down's syndrome would affect their lives, had a more favourable attitude towards abortion generally and a higher level of anxiety about having an affected child than those who did not intend to have screening. Overall, 87% of cases were correctly classified. Participants who intended to have the test were correctly classified in 96% of cases (N=109) compared with 33% of those who did not intend to have the test (N=18).

Screening uptake. Two variables were included within one function that accounted for 17% of the variance in screening uptake. These were the PQoL beliefs score and general attitude to

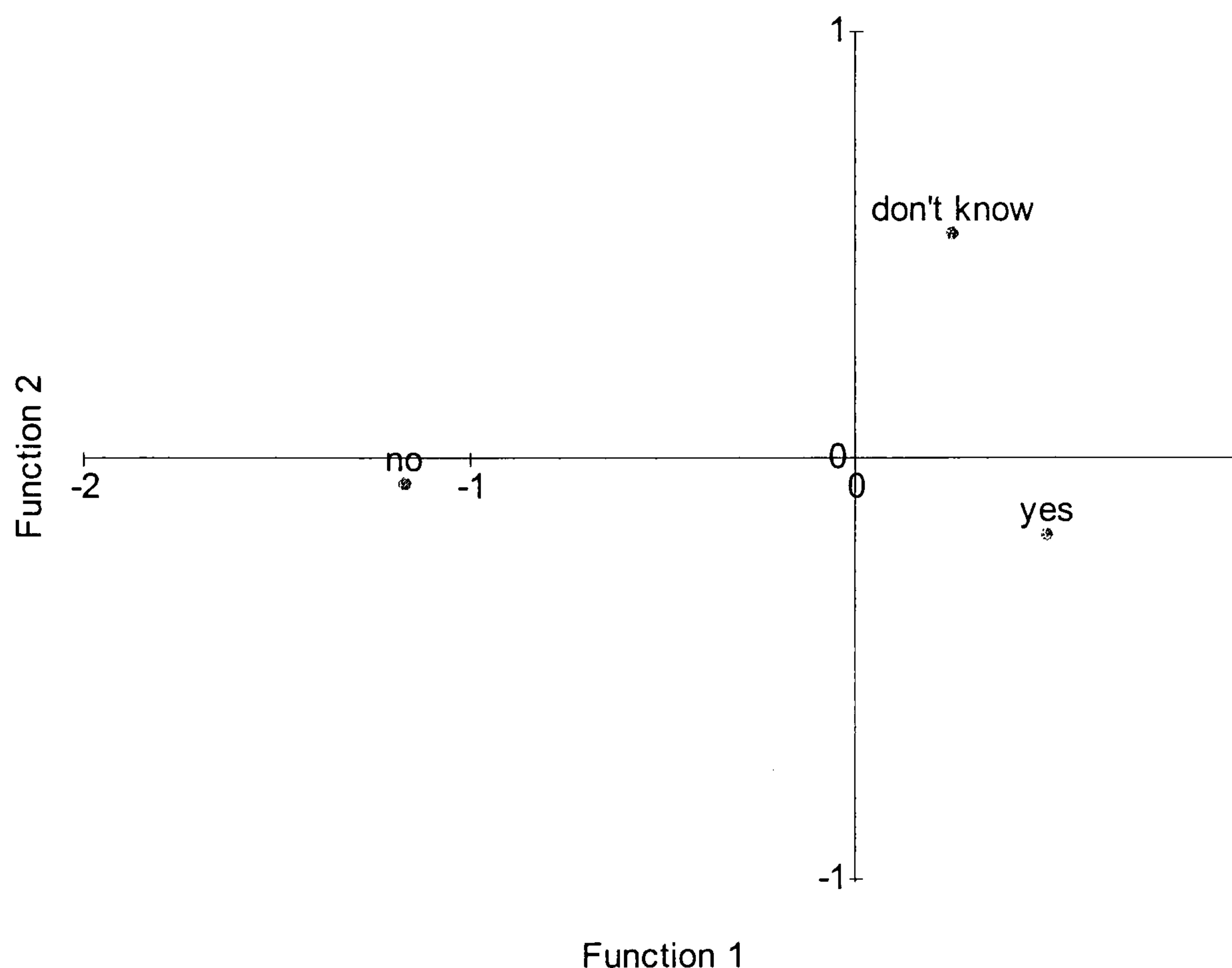
abortion. The function significantly separated those participants who took the screening test from those who did not (Wilks' Lambda = 0.83, $\chi^2 = 21.4$, $df=2$, $\rho < 0.001$). Overall, 74% of cases were correctly classified. Participants who did take the test (N=90) were correctly classified in 92% of cases compared with 17% of those who did not take the test (N=29).

Amniocentesis. Three variables were included within two functions that accounted for 37% of the variance in intention to have an amniocentesis. The *first function*, which accounted for 96% of the variance explained by the two functions, comprised the PQoL beliefs score and the experience ambivalence variable. This function was significant in separating the 'No to amniocentesis' group from the other two groups (Wilks' Lambda = 0.64, $\chi^2 = 57.2$, $df=6$, $\rho < 0.001$). Women who did not intend to use amniocentesis held more favourable PQoL beliefs and had less ambivalence associated with their experiences of people with Down's syndrome than did the other two groups. The *second function* was comprised of the general attitude to termination variable, however it did not explain separation of the groups at a level significantly over and above that explained by function one (Wilks' Lambda = 0.98, $\chi^2 = 2.9$, $df=2$, ns.). Overall, 66% of cases were correctly classified by the model. Participants who did intend to have an amniocentesis (N=69) were correctly classified in 93% of cases, and those who did not intend to have the amniocentesis (N=36) were correctly classified in 67% of cases. None of the 'Don't know' response category (N=28) were classified correctly (0%).

Termination. Four variables were included in two functions that accounted for 66% of the variance in intention to terminate. *Function one*, which accounted for 95% of the variance explained by the two functions comprised the PQoL score, the emotions score, and the general attitude to abortion. Function one discriminated well between all three groups (Wilks' Lambda = 0.39, $\chi^2 = 121.6$, $df=8$, $\rho < 0.001$). Women who intended to terminate held the least favourable attitudes towards Down's syndrome and the most favourable attitude towards abortion generally. *Function two* was comprised of the PQoL ambivalence score and significantly discriminated between those in the 'Don't know to termination' group and the other two groups (Wilks' Lambda = 0.93, $\chi^2 = 9.4$, $df=3$, $\rho < 0.05$). Thus women who were uncertain about whether or not they would terminate expressed significantly more ambivalent beliefs about the effect of a child with Down's syndrome on parental quality of life than did the other two groups. Figure 6.2 shows the within-group means for each intention group plotted by discriminant function. Cases were correctly classified in 70% of cases overall. Participants who intended to have a termination (N=60) were correctly classified in 87% of cases, those who intended not to have a termination

(N=39) were correctly classified in 62% of cases, and those in the 'Don't know' category (N=36) were classified correctly in 53% of cases.

Figure 6.2. Discriminant functions: mean scores for Function One and Function Two by intentions to terminate for Down's syndrome



Interaction between attitudes and ambivalence

In the intention to terminate for Down's syndrome scenario, in the 'yes to termination group' as ambivalence increased attitudes became more favourable, ($r = +0.52$, $p < 0.001$), however, in the 'No' and 'Don't know' groups, as ambivalence increased attitudes become less favourable ($r = -0.69$ and -0.60 respectively, $p < 0.001$). This suggested there was an interaction between attitude and ambivalence dependent on intention to terminate. The significance of the difference between two correlation coefficients for independent samples was calculated (Ferguson and Takane, 1989). The difference between the coefficients for the 'Yes' and 'No' groups and the 'Yes' and 'Don't know' groups was highly significant (z score = +6.69 and +5.86 respectively $p < 0.001$). However, there was no significant difference between the correlations of the 'No' and 'Don't know' group (z score = -0.65, ns.).

6.2.10 Summary of results

Screening. Women who *intended to have screening* were more likely to be anxious about having a child with Down's syndrome and believe that a child with Down's syndrome would have a negative affect on parental quality of life. They were also more likely to find abortion generally acceptable. However, these relationships were shown to be relatively weak, with women holding a wide range of attitudes intending to have screening for a variety of reasons including information, reassurance and to 'be prepared' if the baby had Down's syndrome. Women who *did not intend to have screening* were less likely to be anxious about having a child with Down's syndrome, tended to hold more favourable beliefs about the effects of having a child Down's syndrome and consider abortion unacceptable generally. Due to the small size of the group, factors that discriminated women who were *uncertain about screening* from other groups could not be determined although there was some evidence for the role of poor test related knowledge, relative youth, and not having had previous experience of antenatal care.

Amniocentesis. Women who *intended to use amniocentesis* were more likely to believe that having a child with Down's syndrome would have a negative affect on parental quality of life and to find abortion generally more acceptable. Despite this, nearly one-third of women with favourable attitudes towards Down's syndrome and who did not intend to terminate also intended to have amniocentesis for information and/or reassurance. Women who *did not intend to use amniocentesis* were more likely to find abortion unacceptable and hold more favourable attitudes towards Down's syndrome. They appeared to be more likely to evaluate their experiences with people with Down's syndrome as favourable and with less ambivalence than those who intended to have amniocentesis. The factors that discriminated women who were *uncertain about amniocentesis* could not be determined but are likely to include fear of miscarriage, more neutral/ambivalent attitudes towards Down's syndrome and a less favourable attitude towards abortion.

Termination. Women who *intended to terminate* for Down's syndrome were more likely to believe that having a child with the condition would impact negatively on parental quality of life. They saw very few, if any, positive implications of this situation and were most likely to view a child with Down's syndrome as dependent and demanding. Generally their attitudes towards Down's syndrome were relatively low in ambivalence. The emotions that tended to be elicited by thinking about or meeting people with Down's syndrome were negative ones in the main, such as sadness and pity. In addition they held more favourable attitudes towards abortion generally thus making termination a personally acceptable option for a pregnancy affected by Down's syndrome. For these reasons they intended to use screening tests and amniocentesis if necessary. It is

suggested that this group were more likely to view people with Down's syndrome (and hence a baby with the condition) as more different from than similar to, an unaffected person.

Women who *did not intend to terminate* were more likely to believe that having an affected child would have a neutral to quite positive impact on valued aspects of their life, although the impact on some aspects might be affected negatively. Meeting or thinking about people with Down's syndrome tended to elicit mostly positive emotions, such as feeling 'loving' towards affected individuals. In general the attitudes towards Down's syndrome were held with relatively low to moderate ambivalence. This group held a less favourable attitude towards abortion generally, thus making termination of pregnancy affected by Down's syndrome a less acceptable option. While most of this group viewed having a child with Down's syndrome as a negative life event to some degree, it is suggested that they were more likely to view people with Down's syndrome (and hence a baby with the condition) as more similar to than different from, an unaffected person. They were also less likely to evaluate learning difficulty as a negative aspect of Down's syndrome than the other two groups. These factors might contribute to an unwillingness to actively prevent the birth of a baby with the syndrome.

The measure of religiousness used in this study was not robust enough to include the religiousness variable to be included in the regression analyses. However, for some women religious beliefs or beliefs about the unacceptability of abortion may have influenced their intentions over and above attitudes towards Down's syndrome. Around one-quarter of the women in the 'No to termination' group held quite ambivalent views about how having a baby with Down's syndrome would affect their lives, but also expressed the view that termination was an unacceptable option for them. These women tended to be the ones for whom religious upbringing had at least some influence over their important life decisions. This latter attitude appeared to be a more relevant guide to their behavioural intentions than their attitudes towards Down's syndrome.

Women who were *uncertain about termination* were most likely to hold ambivalent attitudes towards Down's syndrome. In contrast to the other two groups they saw more of a conflict (or a balance) between the favourable and unfavourable aspects of having an affected child. Meeting or thinking about people with Down's syndrome tended to elicit favourable emotions such as loving concern alongside unfavourable emotions such as sadness. For some the ambivalence about the condition may have been related to a more strongly held religious belief. This group was likely to hold a moderately unfavourable attitude towards abortion thus they were unsure whether testing for and terminating a pregnancy affected by Down's syndrome was a personally acceptable option.

The next chapter discusses these findings in relation to the research questions set at the beginning of the study (see Chapter 5).

CHAPTER 7 ATTITUDES TOWARDS DOWN'S SYNDROME IN THE PRENATAL TESTING SITUATION: DISCUSSION

This chapter discusses the results of the study in relation to the objectives as set out in Chapter 5.

The four objectives were:

- To describe attitudes towards Down's syndrome in women in the first trimester of pregnancy.
- To investigate the relationships between testing and termination intentions, serum screening uptake and attitudes towards Down's syndrome.
- To investigate the role of attitudinal ambivalence in the relationships between testing and termination intentions, serum screening uptake and attitudes towards Down's syndrome.
- To identify the variables uniquely contributing to predicting behavioural intentions and screening uptake.

The findings of the study will be discussed in relation to each of these objectives, but first a number of issues associated with the study design and procedure, and their implications for the findings presented, will be considered.

7.1 METHODOLOGICAL ISSUES

7.1.1 Response rate

A questionnaire response rate of 24% (197/840) was achieved and so the number of participants was just less than two-thirds of the target sample size of 300. This low response rate has implications in terms of the power of the study to identify significant relationships, but also for the generalisability of the findings. Reasons for this low response rate will now be discussed.

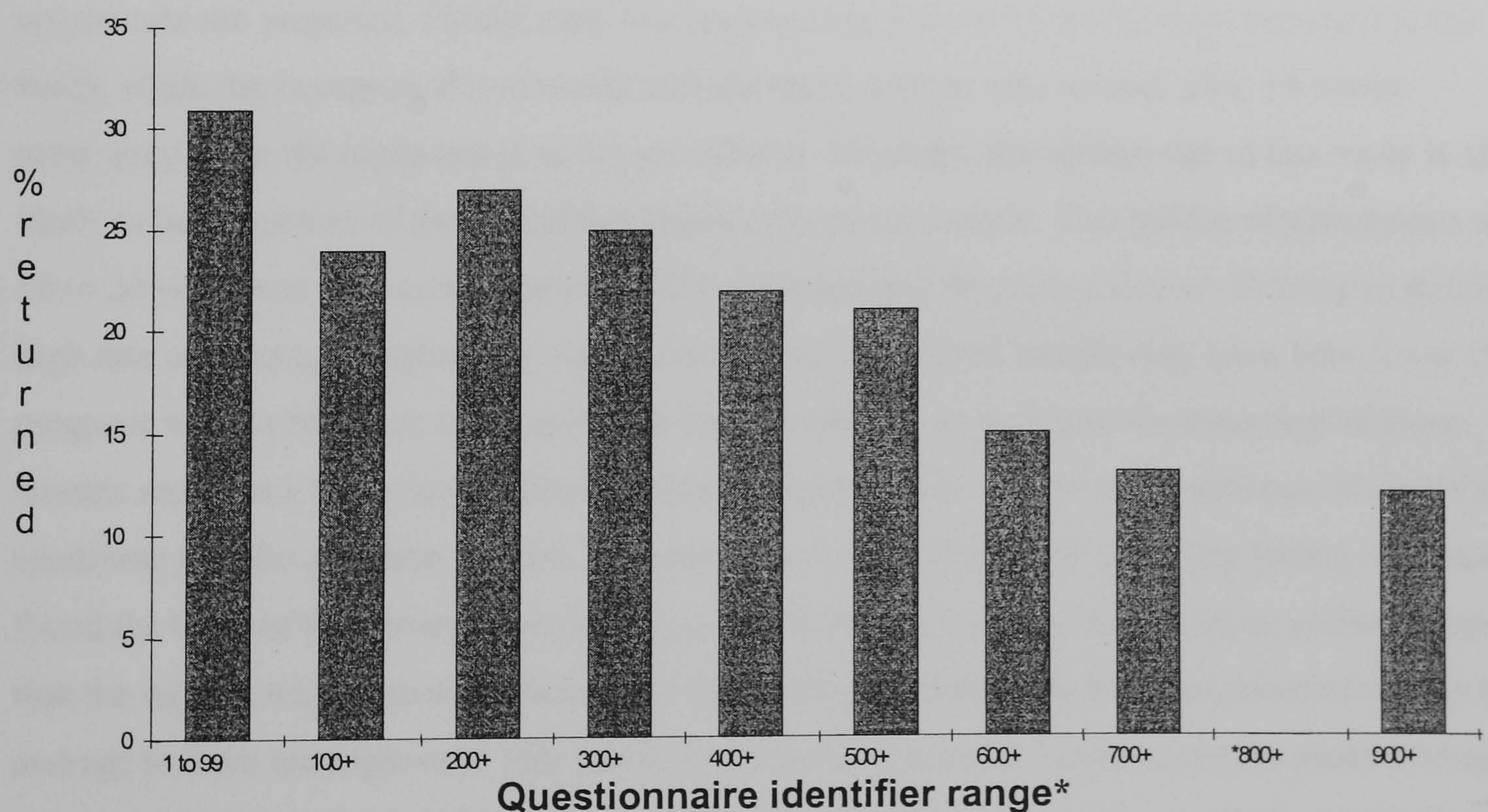
Firstly, surveys of attitudes tend to achieve lower response rates than do surveys of behavioural or factual items (Cartwright, 1989). A number of respondents (8%) returned the questionnaire with only sections A to D completed, suggesting that they found the attitude section (Section E) too difficult or effortful to complete. In a further 21% of questionnaires, the open-ended measures were incomplete or incorrect in some way suggesting that there may have been some difficulty in understanding the instructions or in providing the responses required⁶¹. It might be expected therefore that some other women who started the survey failed to complete the attitude section and then decided not to return the incomplete questionnaire. A sensitive topic is also known to reduce survey response rates and issues around fetal abnormality, prenatal testing and termination of pregnancy are naturally quite sensitive ones in a pregnant population. Higher response rates are

⁶¹ This issue is considered in more depth in the final discussion chapter (Chapter 8) within a general critique of the open-ended attitude measures.

also achieved when the subject is of intrinsic interest to the participant (Shaughnessy and Zechmeister, 1994). The topic may have failed to engage some women sufficiently, or may not have been perceived as salient at such an early stage in their pregnancy.

While questionnaire content is known to impact on response rates, administration procedures are also crucial influences. For example, use of reminders can increase response rates by as much as 50% (Babbie, 1992; Cartwright, 1989) but in this study it was not possible to send reminders out as the details of women who had accepted the materials were not recorded. In addition, the limited window of opportunity between booking and screening appointments would have made a reminder process difficult within the ethical constraints of the study. Knowledge and relevance of the source organisation are also known to influence response rates to questionnaires (Cartwright, 1989). The source of this questionnaire was the University of Leeds rather than the maternity hospital, which might have reduced the perceived relevance of the study to the study population, and/or a commitment to respond. Finally, the way in which the questionnaire was distributed may have been an important factor in the low response rate. Using the questionnaire identifiers, the numbers of returns were plotted by identifier range (see Figure 7.1). The graph shows that as the identifier range increased and the study period progressed there was a general downward trend in the return rate from 31% at the start of the study (Aug 2000) to 12% at the end (Jan 2001). This suggests that the questionnaire distribution rate may have been decreasing as the study continued.

Figure 7.1. Percentage of questionnaires returned by questionnaire identifier range



*One box of study materials with questionnaires in the 800-899 range was 'lost' at the clinic.

At the end of the study, the Clinical Midwifery Leader (CML) reported there were 'about 60' questionnaires remaining at the clinic. Unfortunately, it was not possible to be certain exactly how many questionnaires had been distributed as unused materials had been disposed of at the clinic. Staff in the CML post changed twice during the study period thus the original CML who had been personally involved in setting up the study was no longer able to oversee the questionnaire distribution. It is important to stress that these comments are not criticisms of the midwives at the clinic. The questionnaire distribution was additional to the midwives' workload and was being carried out on the behalf of someone unconnected to the maternity hospital. However, the findings highlight a problem that may occur if the researcher is unable to oversee administration personally. In contrast, a concurrently running 'in house' survey about nausea in early pregnancy achieved a 50% response rate (personal communication). Most items in the nausea survey were factual or behavioural and a consultant based at the maternity hospital oversaw the study. In addition the questionnaires on nausea were posted to the women's homes with the booking appointment information, rather than being dependent on distribution by midwifery staff.

The major concern of a low response rate is response bias. For example, it is accepted that survey respondents tend to be better educated than the average; however, as 40% of participants had finished their formal education at 16 (which is very close to the average figure of 42% reported for the target population) this might not have been an important source of bias in this study. Of more concern was that screening uptake rate of respondents was 77% compared with the background rate of 64%. A number of possible explanations for this higher than average serum screening uptake rate are proposed. Firstly, only women booking prior to 15 weeks were recruited to the study, while the background rate would include those women who booked after 19 weeks pregnancy when the triple-test is no longer offered. Secondly, the uptake rate in this study is also likely to be a function of the ethical age constraints on the sample. Test uptake of participants aged 18 to 20 years was 55% compared with 85% of those aged 30 years and over. Bearing in mind the high rate of 'teenage pregnancies' in the city the test rate of the sample may have been lower if all pregnant women had been made available for recruitment. In addition, the mean age of those women motivated to participate may have been higher due to the increased salience of Down's syndrome to older pregnant women, or those women most interested in having testing may have found the topic of the survey more engaging. Whatever the reasons for the higher uptake it appears that the women who were most inclined to respond to the questionnaire were also more likely than average to have the triple-test. This has to be borne in mind when considering the study findings. In other situations where screening is not routinely offered to all women, or where the 'triple-test' has to be sought privately different results might be obtained. In addition, there might be factors

associated with the economic status, education, ethnicity, and social culture of the study population that makes the findings of this study unrepresentative. It would therefore be necessary to conduct similar research in a variety of clinic settings, using service delivery method as an independent variable. However, a recent review of psychological survey research reported that the main conclusions of a study often remain materially unchanged by obtaining a greater response rate (Krosnick, 1999). It is therefore believed that the findings remain a useful contribution to existing knowledge in their own right, and will hopefully inform further research in this area.

7.1.2 Influence of attitudes towards other conditions

The tests that screen for Down's syndrome are also used to screen for trisomy 18 and neural tube defects (NTDs). It is therefore possible that attitudes towards Down's syndrome might have had a weak relationship with screening intentions and uptake because intentions and uptake were more strongly related to attitudes towards these other two conditions. However, this is unlikely to be the case with attitude towards trisomy 18 as very few people have heard of this condition and the fact that the triple test can be used to screen for it is not widely known. In addition, the clinic's screening information leaflet made no reference to the fact that the triple test can screen for trisomy 18⁶². In contrast, spina bifida is a widely known condition and serum screening tests have been used to identify NTDs for many years. Therefore, it might be that participants' attitudes towards NTDs (or more specifically spina bifida) were less favourable than were their attitudes towards Down's syndrome, explaining why many women with a favourable attitude towards Down's syndrome still had the triple-test. However, there are three lines of evidence to suggest that attitude towards NTDs was not a major factor in serum screening choices. Firstly, the questionnaire items measuring intentions to have screening were specific to Down's syndrome. If attitudes towards NTDs were less favourable than attitudes towards Down's syndrome, and had an influence on test uptake, it would have been expected that uptake of screening would be higher than the intentions data predicted. In fact the opposite was the case, as test uptake was slightly lower than intentions would have predicted. Secondly, the research reviewed in Chapter 2 demonstrated that lay populations (including pregnant women) were likely to view spina bifida as a less serious condition than Down's syndrome. Thirdly, there is now greater emphasis on the relationship between Down's syndrome and serum screening as NTDs are more likely to be identified via ultrasound screening. This argument is supported by both the data collected in the pilot interviews (see Chapter 5), and in the main study. In the questionnaire responses only five women mentioned spina bifida as a reason for having the test, and only two women (both with a

⁶² The leaflet was entitled "Screening for Down's syndrome and spina bifida: Information for parents" and made no reference to trisomy 18.

family member with the condition) mentioned spina bifida without also mentioning Down's syndrome. Ideally, this study should also have measured attitudes towards NTDs although this would have greatly extended the scope of the study and the length of the questionnaire. Nevertheless, the impact of attitudes towards NTDs was not considered during the analysis and this must be taken into account when considering the conclusions drawn in the rest of this chapter.

The findings of the study will now be discussed in relation to each of the four study objectives presented at the beginning of this chapter.

7.2 DISCUSSION OF RESULTS

7.2.1 Description of attitudes towards Down's syndrome

The first study objective was to **describe the attitudes towards Down's syndrome held by pregnant women** prior to them undergoing (or declining) screening for the condition. Was the study successful in capturing these attitudes as intended? The open-ended measures were selected because they allow the respondent to express their attitude in terms of their own feelings, beliefs and experiences, rather than those of the researcher. It could be argued that the measures as used in the study were not truly open-ended, as they included a list of example responses, and it is true that around 80% of responses were ones from the example lists. However, not all the example responses provided were used with equal frequency, in particular, the stereotypic belief examples of 'capable', 'ordinary', and 'healthy' (that were included to balance out some of the more negative characteristics in the list), were used by only four respondents, two respondents, and no respondents respectively. This supports the view that a) the example responses selected were representative of common beliefs, and b) the researcher is not always best placed to decide what aspects of an attitude are important to measure. Support for the claim that open-ended measures facilitate honest responding was also found (Esses *et al.*, 1993). A (small) number of participants with generally unfavourable views of Down's syndrome described people with the condition as "ugly", "nasty", and reported that "they make me feel ill". It is unlikely that a researcher would normally include such items in a measure of attitudes towards disability for fear of causing offence. It is likely therefore, that the full range of views about disabling conditions are not easy to access using more closed measures as they tend to set a ceiling on the expression of extremely unfavourable views.

A key feature of the measures is that the individual always generates the evaluations of the responses themselves, thus the separate evaluative component of the measures allows different participants to give the same descriptive response but to evaluate it in a variety of ways. This is probably the most important quality of the measures and there was good evidence to support that

people did evaluate the same item differently. For example, the evaluations associated with learning difficulty included ones that were neutral, quite negative, very negative, and mixed. The term 'different' was selected by 21 individuals, was rated negative by ten, positive by six, neutral by three, and mixed by two. This range of evaluations might not have emerged from standard measures of attitudes towards disability. Overall, it is argued that the decision to use the open-ended measures was supported, and that the data collected was a valid representation of the attitudes of the women who completed the questionnaires. Nevertheless, there are some concerns about the usability of the measures because of the high rate of missing data and these concerns will be discussed in Chapter 8 within a general critique of the open-ended attitude measures.

The evaluations of the experiences, emotions and beliefs captured using the open-ended measures were positively and significantly associated with an evaluation of 'how bad or good' it would be to have a baby with Down's syndrome. However, the attitude components were more accurate predictors of an unfavourable evaluation of having a baby with Down's syndrome than they were in predicting a favourable evaluation. Thirty-seven percent of those women who held the most favourable attitudes towards Down's syndrome (as measured using the open-ended measures) still evaluated having a child with Down's syndrome negatively to some extent, i.e. below the 'neutral' midpoint on the scale. However, this finding has to be set in context. Other studies have usually created a floor on evaluations of having a child with a disabling condition, for example, by anchoring one end of the scale 'extremely bad', and the other as 'not at all bad' (Figueiras *et al.*, 1999). In this study, there was the option to evaluate having a child with Down's syndrome within the range 'extremely bad' to 'extremely good'. However, it is accepted that most women hope for, and perhaps expect, a baby without any health problems, and so compared to this outcome, any 'abnormality' is going to be disappointing even if not actually distressing. Therefore, an unfavourable evaluation of having a child with Down's syndrome is not incompatible with holding favourable attitudes towards people with the condition or unfavourable attitudes towards terminating an affected pregnancy. These views were made clearer by the responses of some of those who did not intend to terminate for Down's syndrome, for example,

- *As disappointing as it would be that the baby is not healthy I would still love him/her" [No to termination].*
- *"Although I would be upset and worried about the future for us and the child, I do not see a child with Down's syndrome as being less valid or having any less of a right to his or her life than any other child" [No to termination].*

7.2.2 Attitudes towards Down's syndrome and testing and termination choices

The second objective of the study was to **investigate the relationships between testing and termination intentions, serum screening uptake and attitudes towards Down's syndrome.**

During the exploratory interviews at the beginning of this study an apparent dissociation was observed between the attitudes women held towards Down's syndrome and their views and intentions towards the serum screening 'triple test'. The women who expressed unfavourable attitudes towards Down's syndrome intended to have the test, but so did the women who expressed favourable attitudes. It was hoped that the questionnaire would shed more light on this apparent lack of 'consistency'. The results of the study revealed an asymmetry in the relationship between attitudes, intentions and behaviour rather than a complete dissociation: an unfavourable attitude towards Down's syndrome accurately predicted testing and termination intentions and screening uptake, but a favourable attitude did not. A number of explanations for this asymmetry are discussed.

The first explanation is that some women were not expressing their true (unfavourable) attitudes towards Down's syndrome because of a need to demonstrate socially acceptable views of people with a disability. Thus while participants expressed favourable attitudes towards people with Down's syndrome in the open-ended measures, their screening intentions and test choices were more consistent with their actual views. In support of this view a recent American study found that scores on a measure of socially desirable responding contributed significantly towards predicting attitudes towards individuals with Down's syndrome (Hall and Minnes, 1999). However, it is argued that socially desirable responding is an insufficient explanation for the screening intention-attitude asymmetry because participants showed consistent attitude-intention responding with regard to termination for Down's syndrome. Favourable attitudes towards the condition predicted a 'No' intention, unfavourable attitudes predicted a 'Yes' intention, and more neutral or ambivalent attitudes predicted a 'Don't know' intention.

An alternative explanation is that situational factors associated with the antenatal environment are most supportive of attitude-behaviour consistency where the person holds unfavourable attitudes towards Down's syndrome. Screening test appointments are arranged during the booking interview, and in cases where the woman is already between 15 and 18 weeks gestation a test is offered for that same day. In addition, because the test was offered to all women, women with favourable attitudes towards Down's syndrome would have to opt-out of the screening process, and this is a factor known to increase test uptake (Bekker *et al.*, 1993). Such a set-up might therefore be perceived as a cue to an expectation to accept the test. It has previously been noted that women show high compliance with the antenatal care they are offered, and this could act to discourage attitude-consistent behaviour in the case of prenatal testing. Active refusal of the triple-test might require the holding of strong religious or moral beliefs about abortion in addition to

favourable attitudes towards Down's syndrome. For women without these strong beliefs it might be easiest to accept a testing appointment – both psychologically and socially.

These explanations focus on the role of attitudes towards Down's syndrome in predicting testing choices. However, it is also necessary to consider the role of attitudes towards the screening test and attitudes towards *personal use* of a test. In the case of these two constructs it might be concluded that most women are actually displaying high levels of attitude-behaviour consistency. Although attitudes towards the triple-test itself and towards personal use of the test were not measured directly, 93% of participants agreed that screening tests should be available to every pregnant woman who wants one. In addition, the reasons women gave for having serum screening demonstrated that the majority viewed having the test favourably because of its perceived ability to reassure and to inform them. In such a context, the holding of favourable attitudes towards both Down's syndrome and screening tests is not incompatible. This juxtaposition of views is demonstrated by the responses of one participant whose attitude scores and evaluation of having a baby with Down's syndrome placed her into the 'most favourable' attitude category. This woman intended to have screening but did not intend to have amniocentesis or to terminate an affected pregnancy. The reason given for not intending to have an amniocentesis was,

“My first pregnancy was ‘at risk’ with a 1:180 result. I felt I could happily parent a Down's child and therefore decided to proceed no further.”

She commented that she had the triple-test in her previous pregnancy for the purposes of gaining knowledge because if there was a risk, *“I would rather be aware of it”*. This participant could therefore be said to have displayed attitude-behaviour consistency when she went on to have the triple-test a few weeks later.

The results of the study suggested that at different times in the testing process, attitudes towards Down's syndrome and attitudes towards testing might have differing roles to play. The screening information leaflet given to participants at their booking appointment advised them to consider their intentions regarding amniocentesis and termination prior to having the screening test,

“It is better if you decide before having the test what you would do. You may cause yourself a lot of worry if you undergo the triple test, get a high risk result and then do nothing further”.

However, many women did not appear to value this advice and for a significant number a screening test was not viewed as a first step towards either amniocentesis or termination. In addition, the data suggests that some women make clear distinctions between screening and diagnostic testing and do not view them as different stages in the same process. This supports similar findings reported elsewhere (Browner and Press, 1991; Green *et al.*, 1993a). A number of

factors converge to support this perception of separateness. The first is procedural separation; a 'simple' blood sample from the arm versus a needle in the abdomen. The second is the element of risk; a 'no risk' test versus one that might result in miscarriage. A third might be the degree to which the tests are seen to be part of normal care; a routine test versus one offered only where there might be a problem. The fact that detailed information about Down's syndrome was only given to women following a positive screening result might reflect a similar separation in the minds of health professionals as well, with screening being perceived as 'less serious' than diagnostic testing. However, explicitly linking screening with amniocentesis and termination for Down's syndrome is necessary if women are to understand the potential consequences of their screening tests - as definitions of informed choice say they should (Bekker *et al.*, in press).

Explicitly linking the stages of the testing process together might also serve to engage women with their personal values and attitudes associated with termination, which might then impact on attitudes and intentions regarding screening. It has been proposed that for attitudes to affect behaviour they must first be activated, and then be perceived as relevant guides to the behavioural options an individual faces (Snyder, 1982). If the individual does not believe that an attitude towards an object is a relevant guide, the attitude is unlikely to have much influence over that behaviour (Snyder and Kendzierski, 1982). The screening leaflet analysis (Chapter 3), and the preliminary interviews in the antenatal clinic reported in Chapter 5 both demonstrate that the association between screening tests and the condition of Down's syndrome are generally not emphasised in the antenatal setting. Thus for many women, attitudes related to Down's syndrome may not be activated at the screening stage. Even if the attitudes are activated (as presumably they had been for participants in this study) they may not appear relevant to the task of deciding whether to accept the offer of the triple-test or not. The women interviewed prior to the main study said that information about Down's syndrome was unnecessary at the screening stage but if further decisions regarding amniocentesis had to be made, then they would 'need to know more'. Perhaps, when abortion of a 'baby' with Down's syndrome is the behaviour under consideration, attitudes towards the condition might appear to have relevance. This would explain the greater consistency between attitudes towards Down's syndrome and (hypothetical) intentions to terminate. In a report of a study examining the views of Finnish midwives towards prenatal testing and termination for Down's syndrome the authors commented,

"Whereas abortion is clearly a moral question, serum screening may be perceived as a question of more choices, information and self determination. (Jallinoja et al., 1999), p. 1018).

It is suggested that women might also make this distinction between screening behaviour and intending to terminate.

In summary, attitudes towards people with Down's syndrome, and beliefs about parenting an affected child might be more likely to result in attitude consistent screening choices if a person has a) engaged with their experiences, beliefs, and feelings about the condition, and b) perceived these factors as relevant to their decision. However, even attitudes perceived as relevant can be overridden by other factors such as perceived group norms and apparent benefits and costs (Borgida and Campbell, 1982; Snyder and Kendzierski, 1982). Most women, even those who view Down's syndrome favourably, would prefer it if their baby did not have the condition. For some, having the triple-test was viewed as a way of reassuring them of this. Other women were seeking knowledge to prepare them for the possible event of having an affected child. Some women, believing that all antenatal care on offer was in their best interests, might have perceived this belief to be more relevant than any views they might hold about people with Down's syndrome. However, relationships between attitudes and testing and termination intentions were seen and the following sections consider the role that different attitudinal components might play in influencing testing and termination choices for Down's syndrome

7.2.3 The role of experience with people with Down' syndrome

For some women it might be expected that their attitudes towards Down's syndrome would have a very important influence on their behaviour. As discussed previously attitudes towards Down's syndrome should have a more predictable impact on test behaviour in a situation if a person has direct experience with an affected person (Fazio, 1986). However, women who had a family member with Down's syndrome *and* held favourable attitudes towards the condition ($n=12$)⁶³ were not significantly less likely to have serum screening than those who did not have a family member (75% versus 77%). In contrast, 25% of the family group compared with 52% of the rest of the sample intended to use amniocentesis, and none of this family sub-group intended to terminate for Down's syndrome compared with 48% of those without an affected family member. This suggests that even in those with direct positive experience of a family member with Down's syndrome, screening behaviour is not easy to predict from attitudes towards the condition nor can assumptions be safely made about why screening is being used and further supports the distinction that many women make between screening and other stages in the testing/termination process.

⁶³ A positive attitude was defined as one where the overall attitude score was higher than zero.

As noted in previous studies (Haddock *et al.*, 1994; Hall and Minnes, 1999) the evaluations that participants gave to their experiences were significantly related to their attitudes and intentions whereas the *number* of experiences reported was not. This supports the view that the frequency of contact with people with a disability is not the important determining factor in attitude direction. The causal direction of relationships between experiences of people with Down's syndrome and attitudes towards the condition cannot be assumed and the influence is likely to be bi-directional. Evaluations of experiences are probably mediated by the feelings and beliefs associated with Down's syndrome, and this may explain the Experience variable's lack of predictive power. In some circumstances, situational factors were also seen to influence termination intentions, for example, in those women who had already had had one child with a disability. However, if people do not have direct behavioural experience of people with Down's syndrome on which to base their attitudes, their behaviour is more likely to be guided by other factors such as attitudes towards antenatal care in general and beliefs about what you think significant others (midwives, partner, family) would want you to do. It is interesting to note that 15 of the women who did not complete the experiences measure completed at least one other of the measures. Despite being (presumably) unable to report any specific experiences with people with Down's syndrome they were still able to express emotions and beliefs in connection with the condition.

7.2.4 The role of stereotypic beliefs about Down's syndrome

Across all groups the most frequently expressed beliefs combined to portray a consensual stereotype of people with Down's syndrome as 'childlike' – affectionate and happy, but also demanding, dependent, and vulnerable. Wolfensberger defines a number of social perceptions attached to people with disabilities that include being an object of pity, a burden of charity, and an eternal child (Wolfensberger, 1972). For some women the 'eternal child' is quite clearly an unattractive proposition associated with burden and dependency, while for others this stereotype may be irrelevant or even attractive. One participant commenting on a television programme drew on the stereotype of dependency, but gave a not unfavourable evaluation of this characteristic.

"I saw a true story on a Down's syndrome boy and it touched my heart. I wouldn't mind looking after one" [No to termination].

The learning difficulty associated with Down's syndrome was reported by similar numbers across intention to terminate groups but those who intended to terminate evaluated this characteristic as negative most often. Perceptions of learning difficulty might be based on personal experience (or lack of personal experience) although other beliefs and values are also likely to shape views. Hence the 'severity' of Down's syndrome might also be considered to be a characteristic of the individual perceiver rather than a characteristic of the condition itself.

Stereotypic belief ambivalence was found to be significantly higher than ambivalence associated with other attitude components and this is also consistent with the eternal child stereotype of Down's syndrome. The stereotypic combination of people with Down's syndrome as affectionate and happy, yet dependent and disabled paints an inherently ambivalent image of the condition. This common stereotype of people with Down's syndrome may be one reason why stereotypic beliefs did not discriminate significantly between groups by intention to test and terminate or uniquely predict an evaluation of having a baby with Down's syndrome. Esses and colleagues report that agreement with a consensual group stereotype appears only 'minimally related' to a person's overall evaluations of that group (Esses *et al.* 1993, p. 141). In addition, they propose that stereotypes may actually play an indirect role in predicting attitudes by influencing the emotional reactions to a social group.

7.2.5 The role of emotions

The emotions expressed most frequently in connection with people with Down's syndrome were feelings of sadness and sympathy. Similar findings were reported in the MENCAP survey (1982, reviewed in Chapter 1) where 70% of respondents associated feelings of sympathy with people with learning difficulty and 50% associated feeling sadness. Sympathy and sadness were also elicited most frequently in a study that used the open-ended measures to capture attitudes towards a variety of disability groups (Esses and Beaufoy, 1994). A child that engenders pity and sorrow in others is clearly not something that parents would wish for. In the reasons that women gave for intending to terminate for Down's syndrome feelings were seldom included, but where they were, they referred to the suffering of child and parents. For example,

- *"I would feel cruel watching my Down's child struggling from being a child, feeling and being different, though trying to lead a normal life."* [Yes to termination]
- [I would probably have a termination because there would be] *"Less suffering on all parts"* [Yes to termination].

However, those women who did not intend to terminate for Down's syndrome were most likely to report positive emotions connected with Down's syndrome. If someone believes a child with Down's syndrome is to be valued rather than pitied they might not view the birth of such a child as a 'reproductive catastrophe' in the way that has often been portrayed (Firestein, 1989). In the reasons that women gave for not intending to terminate for Down's syndrome feelings about an affected child were included by over one-third of respondents. For example,

"Down's or not, it is still my baby, my flesh and blood and it still has a life and it would be loved and cared for the same as my other three children." [No to termination].

In contrast to the emotion responses of the ‘Yes to termination’ group, the most frequently expressed emotions of the ‘No’ group were very similar to those that might be elicited by any child – pride, concern, happiness, and love. This again, suggests that a perception of ‘differentness’ in relation to Down’s syndrome might be an important factor in the termination intentions reported. This is also supported by the increased frequency with which the ‘Yes’ group reported feeling nervous and awkward around people with Down’s syndrome. The mixture of positive and negative emotions as expressed by the ‘Don’t know’ to termination group, indicated ambivalent feelings towards people with Down’s syndrome. The role of ambivalence in termination intentions is discussed in section 7.2.7.

7.2.6 The role of ‘Parental Quality of Life’ beliefs

Carol Gilligan has previously argued that women make decisions about abortions within a complex network of relationships and responsibilities (Gilligan, 1982). However, while family relationships and responsibilities were clearly of prime importance to most women, there was little support for Gilligan’s view that women often want to continue a pregnancy but terminate because of their responsibilities to others. Instead, the beliefs expressed suggested that most women intended to terminate (or continue) a pregnancy primarily because of personal beliefs and feelings about Down’s syndrome, or more specifically, because of beliefs about how such a child would impact on their lives and their family. Women who intended to terminate believed that a child with Down’s syndrome would have a detrimental impact on their valued relationships, including the anticipated one between themselves and their baby. This was not apparent in those who did not intend to terminate. The PQoL score correlated significantly and positively with all other component scores, which does not support the view that women held favourable personal attitudes towards having a child with Down’s syndrome but negative beliefs about the impact of having such a child on partner and children.

While PQoL beliefs could also be considered stereotypes as they reflect assumptions of how having a child with a disability impacts on personal and family life, they are stereotypes at the individual level rather than the consensual one (Esses *et al.*, 1993). In over 50% of the women who intended to terminate for Down’s syndrome, beliefs about how an affected child would impact on their personal quality of life were cited as the reason for this intention, for example,

“As I already have a healthy two year old I would have to consider his needs and the effects of having a Down’s baby would have on all of us with regards to the care need, our current jobs, financial implications etc.” [Yes to termination].

By contrast, in those who did not intend to terminate, such beliefs were stated much less frequently and usually in a more positive sense.

“I do not think that a Down’s child is not a viable pregnancy. My experience is they can have and give a wonderful quality of life” [No to termination].

The PQoL responses of women who did not intend to terminate for Down’s syndrome reflected the belief that an affected child was more similar to, than different from other children – in terms of rights to life, but also as a potentially loved and valued child. In contrast, the responses of the women who did intend to terminate reflected the belief that an affected child was more different from, than similar to other children. For some women this raised doubts about whether they could love an affected child in the same way as a ‘normal’ child, echoing some of the responses seen in the earlier study using Q methodology. It is also proposed that some women perceived being the parent of a disabled child would also set them apart as ‘different’ in the views of others, which may threaten their own self-concept as normal. It is suggested that these perceptions help explain why the prospect of raising a child with Down’s syndrome is considered ‘burdensome’ for some but not for others.

The perceived burden of caring for a child with a disability has been found to predict attitudes towards prenatal testing and termination for disability (Bryant, 1998; Davies and Doran, 1982; Denayer, Evers-Kiebooms, de Boeck, and van den Berghe, 1992; Marteau *et al.*, 1992a; Priest *et al.*, 1998). Although the word burden is often used in association with physical care and dependency it is also used to describe emotional ‘toll’ – sorrow and suffering, for example. In terms of time, effort, money, and emotional resources the demands of parenting a healthy child are substantial, however, the rewards are usually considered to outweigh the costs and so the concept of burden is not considered applicable (Botkin, 1995). Many women believe a child with Down’s syndrome would be associated with all the usual demands (plus additional ones), but with very few rewards in recompense (Lawson, 2001); such a situation might be considered ‘burdensome’, i.e. difficult to bear. However, it is argued that usage of the term ‘burden’ in the context of measuring attitudes towards disability is problematic because the term is value-laden. Measuring burden assumes that people will consider a child with a disability to be burdensome to a lesser or greater extent. However, if a child with Down’s syndrome is perceived to be more similar to, than different from, any other child, then burden is an inappropriate and possibly irrelevant concept. This is supported by the lack of ‘burden’ type characteristics and negative emotions associated with Down’s syndrome by the ‘No to termination’ group.

7.2.7 Attitudinal ambivalence

The third objective of the study was to **investigate the role of attitudinal ambivalence in the relationships between testing and termination intentions, serum screening uptake and**

attitudes towards Down's syndrome. The psychological antecedents and outcomes associated with '*don't know*' respondents have received very little attention in the prenatal testing literature. In this study, 41% of participants gave at least one 'don't know' response in regard to their testing and termination intentions. It is therefore important to try and understand what factors might be associated with these responses. In a previous study of attitudes towards aspects of pregnancy and antenatal services younger women were more likely to give uncertain or undecided responses (Kafetsios and Green, 1997). In the present study there was also a trend for women who were certain about their intentions to have serum screening ('definitely yes' or 'definitely no') to be older than women who were less certain. In addition, those in the 'Don't know' to screening group were more likely to be nulliparous than women in the 'Yes' or 'No' groups although this did not reach significance. It might be that having previous experience of antenatal testing is a factor in removing uncertainty about screening intentions in the current pregnancy. The inability of the Discriminant Analysis model to accurately predict the 'Don't know' responses to amniocentesis suggests that the variables measured in this study did not fully account for why women were uncertain about having a diagnostic test. It is hypothesised that concern about miscarriage was an important factor as 52% of participants giving a 'don't know' response to the item on amniocentesis cited risk of miscarriage as a reason for their response. In addition, the results suggest that attitudinal ambivalence towards Down's syndrome is another factor in uncertainty in intentions to terminate. Overall, very few women gave a 'don't know' response to screening indicating that the choice to have screening is relatively conflict free. However, attitudes towards Down's syndrome were shown to have greater influence in predicting termination intentions, and ambivalent PQoL beliefs emerged as a discriminating factor between women in the 'Don't know to termination' group and the other two groups. Women who gave a 'don't know' response to the item on termination and had high levels of ambivalence gave reasons that suggested a need for more information, either from health professionals, or 'information' derived from their own or their partner's feelings.

Previous research evidence on the individual factors associated with ambivalence is generally inconclusive (Conner and Sparks, 2002). Certain factors, including personality, knowledge, moral, political, or ideological positions are likely to interact with the situation to produce ambivalent attitudes in some individuals but not others. Religiousness has been shown to be one of the most reliable predictors of unfavourable views towards abortion, however, holding religious beliefs might also result in difficulties for some people in situations where personal desires might conflict with personal morality. This may explain the finding that ambivalence (particularly relating to PQoL beliefs) was higher in those women for whom (Christian) religious belief had some

influence over their life decisions. For these women, important religious values associated with loving acceptance of others might conflict with beliefs about the perceived difficulties of coping with a child with a disability. One participant, whose ambivalence score placed her in the most ambivalent group, and who said her religious beliefs influenced her decisions ‘quite a lot’ commented that,

“I feel that it is wrong to take any life including that of an unborn child. However I would not impose my feelings on anyone else or judge others because of their actions” [No to termination].

It might be that this woman could understand why someone else might choose to terminate for Down’s syndrome because she also held negative beliefs and feelings about the condition. However, it is important to note that only moderate levels of religious influence were observed in this study. Further research might find that very strongly held religious beliefs are associated with low attitudinal ambivalence towards Down’s syndrome. In addition, other moral or ideological beliefs including liberal political views, or views about the inclusion in society of people with disabilities might also be associated with ambivalence and this may be a fruitful avenue of future research (Krishnan, 1991).

Potential consequences of ambivalent attitudes in the prenatal testing context

It has been proposed that health professionals actively steer women in the direction of testing and termination thus undermining their autonomy and the opportunity to make an informed choice (Venn-Treloar, 1998). However, the intention values reported in this study support the alternative view that while situational factors may make certain decisions more likely, most women are not passive recipients of testing who are acting completely against their own values (Dimavicius, 1998a; Green *et al.*, 1993a; Statham and Solomou, 1998). A recent review of the study site’s screening programme reported that 85% of women had a diagnostic test following a positive serum screen result (O’Connell *et al.*, 2000). This is in line with the behavioural intentions reported here, i.e. 13% of those intending to have the triple test intended not to have an amniocentesis. It is likely that *most* women who in advance knew they would definitely not want to terminate a pregnancy for Down’s syndrome would either not have had serum screening⁶⁴, or would not have an amniocentesis after a positive result. While there are almost certainly women who change their mind completely once in the situation or who find themselves acting against their prior ‘moral convictions’ they are probably in the minority. It is hypothesised that the

⁶⁴ This is complicated by the fact that even women who do not have serum screening or amniocentesis generally have an ultrasound scan that may identify markers of Down’s syndrome.

majority of people who actually use testing or termination are those who had either intended to do so in advance, or who hadn't considered/'didn't know' what they would do beforehand.

The research reviewed in Chapter 5 suggested that people with ambivalent attitudes seek additional information when they are required to make a behavioural choice. This suggestion received tentative support from the responses given by those women who were uncertain about their intentions to terminate for Down's syndrome. Because ambivalence may affect how information is processed it is important to consider the potential consequences of holding ambivalent attitudes towards Down's syndrome for someone situated within a prenatal testing scenario. It could be hypothesised that individuals with low ambivalence towards Down's syndrome would be guided more by their pre-existing attitudes towards the condition. For these women, prior attitudes might render further information about the condition irrelevant in terms of informing choice. However, if women high in ambivalence towards Down's syndrome are less able to be guided by their own attitudes they might seek advice from a health professional or attend more closely to material in an information leaflet. In addition, some evidence exists that the information processing of ambivalent individuals may be more susceptible to the influence of mood than that of non-ambivalent individuals (Bell and Esses, 1997). This may be relevant in a situation where the receipt of a positive screening result or diagnosis of abnormality is likely to be associated with unpleasant emotions and anxiety.

These findings raise many questions that cannot be answered in this thesis but merit further investigation. For example, how is ambivalence towards Down's syndrome resolved in the face of choices about diagnostic testing and termination for Down's syndrome? The statistics suggest that most ambivalent women will opt for an amniocentesis or a termination, but what information do women use to help them decide, and how do they process such information under conditions of extreme stress? How might the choice made feed back to inform previously ambivalent attitudes? Would behavioural information help resolve ambivalence or would it exacerbate it? Are ambivalent people more or less satisfied with their choices? They might be more satisfied because of their greater propensity to think systematically about their decision. Alternatively, they might be less satisfied because their favourable views about Down's syndrome have to co-exist with their negative-beliefs-consistent behaviour. In addition to the psychological questions it is also important to understand whether the organisation of maternity care makes the decisions of ambivalent individuals more likely to be in one particular direction, and the implications this has for informed choice.

7.2.8 The relationship between attitudes, intentions, and behaviour

In addition to the relationships between attitudes towards Down's and testing and termination intentions, the study identified a number of other significant relationships that will now be discussed. The social cognition models most associated with the role of behavioural intentions are the Theory of Reasoned Action and the Theory of Planned Behaviour (Ajzen 1985,1988,1991; Azjen and Fishbein, 1980; Fishbein and Ajzen, 1975). As described in Chapter 5, within these two expectancy-value models attitudes exert their influence on behaviour via behavioural intentions, which in turn are considered the proximal determinants of the behaviour. Reviews covering a range of health behaviours report mean correlations in the range of +0.45 to +0.62 between intention and behaviour (Conner and Sparks, 1995). In comparison, intentions to have or not have the triple test appeared to be extremely good predictors of test uptake with predictive values of over 90%. Why might this be? Research suggests that making a plan to implement behaviour increases the likelihood of the intended action being carried out (Gollwitzer, 1999; Gollwitzer and Schaal, 1998). Therefore, an intention to use or decline screening that arose out of involvement in this study might have led to participants' wishing to match their intention with consistent behaviour. Alternatively, women who expressed an intention to have the triple-test via the questionnaire or at the booking appointment would already have been given a date to attend for a blood sample by their midwife, thus putting in place a plan to have the test. A study predicting attendance at health checks found that being given a definite appointment date produced a 70% attendance rate in contrast to a 30% attendance where an open invitation to make an appointment was made (Norman and Conner, 1993). As antenatal care is generally valued highly in the UK, and most women attend for scheduled clinic appointments, this further increases the likelihood that the intention would have been implemented.

Although screening intentions were good predictors of screening behaviour, intentions *not* to have the test were slightly better predictors than intentions to have the test. It may be that those who decline testing invest more cognitive effort in their intention and so are more committed to carrying it out than women who follow the norm of having testing. In addition, some women intending to have the test may have changed their minds after further consideration or discussion with their partner. However, there may be an alternative explanation associated with the logistics of the screening process at the study site. Firstly, the blood sample for the triple-test is taken at a separate appointment from the usual routine check-up. Separate appointments for screening tests have previously been related to lower uptake rates in screening for Down's syndrome (Dormandy *et al.*, 2002b) and cystic fibrosis (Bekker *et al.*, 1993). Secondly, the antenatal outpatients department is situated on the outskirts of the city in a non-residential area, and for public transport

users there is a ten-minute walk from the gates of the hospital to the clinic. For women whose motivation to use testing was not high (perhaps due to non-salience of Down's syndrome or a dislike of having blood taken), a separate appointment may have acted as a barrier to attending. Barriers (the perceived difficulty of engaging in a behaviour) have been shown previously to be reliable predictors of *not* engaging in health behaviours (Armitage and Conner, 1999; Sheeran and Abraham, 1995). In addition, age did not differentiate between those who did and did not intend to have screening, but the mean age of women who actually had the triple-test was significantly greater than the age of women who did not have the test. A reduced salience of prenatal testing for Down's syndrome, plus the fact that younger women are least likely to have access to private transport might have contributed to this finding. In addition, women in the 'don't know' to screening group were significantly younger than the 'yes' group, and the youngest 'don't knows' were least likely to use screening, further increasing the age differential at test uptake.

It has been noted that in many aspects of health care consumers express a preference for things they have already experienced (Porter and Macintyre, 1984; Salkeld, Ryan, and Short, 2000). In this study, past screening behaviour was strongly related to screening intentions and test uptake (although previous test behaviour was obtained via self-report, therefore the relationship might have been inflated due to a desire for behavioural consistency). The ability of prior behaviour to predict current behaviour in health related situations has been discussed by Conner and colleagues (Conner and Sparks, 1995; Norman and Conner, 1996): while they support the existence of a link they suggest that it is a moderator of other variables rather than a useful predictor in its own right. For example, consequences of the past behaviour may impact on beliefs about that behaviour, which then influences intentions to perform that behaviour again (Ajzen, 1988). Around 95% of women who take the triple-test will receive reassuring information about the health status of their baby. It is likely that in many cases this will reinforce their positive beliefs about the test and make them more likely to use the test in a subsequent pregnancy. The reverse may also be true. A study investigating serum screening uptake in multiparous women found that women who had received a 'false-positive' result in their previous pregnancy were significantly less likely to use screening in their next pregnancy than those who received a 'negative' reassuring result (Rausch *et al.*, 2000). Finally, two women in the current study who had used serum screening previously did not intend to use it (and did not use it) in this pregnancy. They both commented that in the previous pregnancy they did not understand the nature or implications of the test. For example one of the women wrote "*I was not sure what the triple test was for until after the test was taken, and then it was explained and I have refused it in other pregnancies.*" Therefore, while prior screening

behaviour is clearly a good indicator of current screening behaviour, it is most likely to be a moderator of other factors associated with intentions to use or decline the test.

7.2.9 The role of socio-demographic variables on intentions and uptake

In this study a number of socio-demographic variables had associations with testing and termination intentions and screening uptake. As reported in other studies (Bell and Stoneman, 2000; Britt *et al.*, 2000; Green *et al.*, 1993a) the importance of religiousness rather than religious affiliation was related to behavioural intentions regarding abortion and prenatal testing. This might have been partly due also to the small numbers of individuals affiliated with religions other than a Protestant Christian one. The finding that older participants were most likely to intend to terminate for Down's syndrome also supports previous research in this area (Kramer *et al.*, 1998; Singer *et al.*, 1999). However, unlike some other studies (Heyman and Henriksen, 2001; Press and Browner, 1998) there was a significant difference in serum screening uptake by age. This difference may partly be explained by the barriers experienced by younger women in attending appointments as discussed previously and possibly because Down's syndrome screening had a lower salience for younger mothers in this sample.

Age also related significantly and negatively to attitudes towards Down's syndrome with older women demonstrating less favourable attitudes towards the condition overall. Although some previous research on attitudes towards people with Down's syndrome has not found age to be a discriminating variable (Bell and Stoneman, 2000; Furnham and Pendred, 1983), a large-scale general public survey of attitudes towards people with learning difficulties did find that younger people held more 'positive impressions' of affected individuals (Mencap, 1982). In addition, the results of the Q-study (Chapter 4) also showed a significant age difference in the valence of beliefs about Down's syndrome with the younger group (18 to 34 years) holding significantly more favourable views than the older group (34 to 80 years). This relationship between attitudes and age could be a cohort effect in that attitudes are generally becoming more favourable towards people with learning difficulties. For example, younger women are more likely to gain direct peer-group experiences of children with Down's syndrome attending mainstream school. Alternatively, as women get older, their attitudes towards the condition could become less favourable due to a wider range of experiences, or because having a child with a disability becomes threatening to a more established lifestyle. Women might also become less idealistic about life generally, and about raising a child with a disability specifically. Finally, there might be a number of differences in pregnant women of different age groups, for example, education and social class, life-stage factors, and reasons for being (or staying) pregnant, which might affect views about having a child

with a disability. A combination of these reasons is likely to be associated with the relationship between age and attitudes towards Down's syndrome.

Despite their associations with intentions and attitudes in this study, neither age nor religiousness met the *a priori* criteria for entry to the Discriminant Analysis model. These two variables may therefore play a more indirect role in predicting the dependent variables measured in this study. For example, age was correlated positively and religiousness was correlated negatively with general attitude to abortion, which itself emerged as a reliable predictor of screening uptake and the testing and termination intentions. However, young women could also hold favourable attitudes towards termination and non-religious women could believe that abortion is immoral. This indirect effect of demographic variables has been noted elsewhere (Statham *et al.*, 1997).

7.3 CONCLUSIONS

The overall objective of the study was to **identify the variables uniquely contributing to predicting behavioural intentions and screening uptake**. The results demonstrate that in addition to previously identified factors such as anxiety and attitudes towards abortion generally, understanding a person's attitudes towards Down's syndrome can help predict their behavioural intentions regarding prenatal testing and termination for the condition. However, while attitudes towards Down's syndrome might play an important role in the testing choices of some women this role is not necessarily consistent across all women, or across all stages of the testing process. Attitudes towards Down's syndrome contributed most significantly to intentions to terminate for the condition – in hypothetical situations at least. In contrast, most women regardless of attitude towards Down's syndrome, intended to use, and did use, prenatal serum screening. This 'inconsistency' between attitude and testing behaviour has relevance to the issue of informed choice. Many women do not have direct behavioural experiences of people with Down's syndrome with which to inform their attitudes and guide their prenatal testing choices. This means their choices are more likely to be informed by other sources of information. Some of these sources might be related to other personal beliefs and values, for example, views about abortion generally, attitudes towards antenatal care, experiences of disability in another context, or their perceived ability to cope with a child with a disability. However, they might also be more likely to be influenced by information coming from external sources that are based on the beliefs and values of others, such as advice from health professionals, information leaflets, situational cues such as 'opt out' screening programmes, and perceived behavioural norms. Even so, women with direct, positive experience of Down's syndrome via a family member with the condition were as likely as the rest of the sample to use serum screening. In addition, many women with favourable attitudes towards Down's syndrome appeared to perceive these attitudes to be irrelevant to their screening

choices but relevant to their termination intentions. This suggests that screening choices might not necessarily be informed either by attitudes about the target condition, or by attitudes towards using termination that has often been assumed. It is suggested that the variable most strongly influencing screening choices might be an attitude towards using the test (not measured directly here) but that this in turn can be influenced by a number of personal and situational factors. Some of these are discussed in more detail in the next (and final) chapter.

CHAPTER 8 DISCUSSION OF THE THESIS

8.1 RESEARCH QUESTIONS REVISITED

The literature reviewed in the first chapter of this thesis demonstrated that research in the area of informed choice and prenatal testing had focused on information and knowledge about the testing process and attitudes towards using the tests. By comparison, little attention had been directed towards information, knowledge, and attitudes relating to the condition(s) being tested for. This is despite consistent evidence that perception of severity of a condition is one of the key predictors of a decision to terminate for abnormality. Specifically, a systematic examination of women's understandings of Down's syndrome and how they inform prenatal testing choices was missing from the literature. The research presented within this thesis has begun to address these gaps. The first study considered the information about Down's syndrome that was provided to women via serum screening leaflets in the early stages of their pregnancy. The second study explored diversity in understandings of Down's syndrome outside the testing context and then considered how different understandings might relate to intentions to test and terminate in hypothetical situations. The third study conducted a systematic assessment of the attitudes of pregnant women towards Down's syndrome and investigated these in relation to actual screening choices. The aim of this final chapter is to discuss the main findings of these three studies with reference to the research questions as set out in Chapter 2. In particular, the discussion will focus on how the findings have contributed to understandings of informed choice in the prenatal testing context and the growing awareness that decisions should reflect the individual's values as well as their knowledge (Marteau *et al.*, 2001).

The first study (reported in Chapter 3) attempted to answer the question **'What information do women receive about Down's syndrome to help inform their prenatal testing choices?'** Recommendations have been that information about the target condition must be provided and that information in general should be delivered in as non-directive a manner as possible (Advisory Committee on Genetic Testing, 2000; Marteau, 1995; Nuffield Council on Bioethics, 1993; Royal College of Physicians, 1989). In order to assess a primary source of information about Down's syndrome, an analysis of the amount and type of information provided in 80 prenatal screening leaflets was conducted. A secondary analysis of the relative balance of positive, negative, and neutral information was also carried out and the findings compared with those of a similar analysis of information about cystic fibrosis (Loeben *et al.*, 1998). Three main findings emerged from this study.

- Firstly, **information about Down's syndrome was frequently absent or insufficient**: in around one-third of the leaflets there was no descriptive information about Down's syndrome, and the median number of descriptive sentences was one. This demonstrated that the basic recommended standards of information provision were not being met by a substantial number of screening leaflets. There is evidence to suggest that in some cases the only information women receive about screening is in leaflet form (Stapleton *et al.*, 2002), and that some women, especially those from certain ethnic groups, have very little knowledge of Down's syndrome. This finding has obvious implications for informed choice at the most basic of levels as many leaflets fail to provide pregnant women with information that is considered essential for informed decision-making.
- The second finding was that **the majority of information** in the leaflets (89%) was related to **medical, clinical, or epidemiological aspects of Down's syndrome**. A minority of leaflets (20%) included some information about education, inclusion in society, and other social issues, but only five leaflets out of 80 (6%) referred to the psychosocial/emotional aspects of the condition for either the affected individual or their family. These data suggest that a rather narrow perspective of Down's syndrome tends to inform the writing of serum screening leaflets. This perspective does not generally appear to be viewed as problematic by those issuing recommendations about informed choice, but this might be because the recommending bodies are also usually allied with the medical model of testing. It is hypothesised that such a limited coverage of the condition may not serve the information needs of women considering testing for Down's syndrome, and might well fail to engage with personal values and understandings. However, this hypothesis remains to be tested through research.
- The third finding was that the **image of Down's syndrome as portrayed by the information in the screening leaflets was generally a negative one**, and that this contrasted with a more neutral portrayal of cystic fibrosis identified in the comparison study (Loeben *et al.*, 1998). Out of 162 sentences of descriptive information about Down's syndrome 63% were classed as negative, 25% as neutral, and 12% as positive. It is argued that the degree of negative emphasis in the information does not accurately represent the complex condition of Down's syndrome or the experiences of many affected individuals and their families, and does not respect the different values and experiences of women considering prenatal testing. Information that is strongly biased in a particular direction cannot be said to support autonomous decision-making.

The second study (reported in Chapter 4) used Q Methodology to help answer the question **'What understandings of Down's syndrome exist independently of the prenatal testing context?'**

The primary aim was to model these understandings and to identify similarities and differences between them. The secondary aim was to explore **how different understandings might relate to intentions to test and terminate for Down's syndrome in hypothetical situations**. It was anticipated that the study would provide information about the types of knowledge that women might bring with them to the prenatal testing situation and generate hypotheses for further study. Previous research attempting to access understandings of disabling conditions had either focused on pregnant women already considering or undergoing amniocentesis, or had taken a 'broad-brush' approach to disability and so had not specifically investigated understandings of Down's syndrome. In this study, a diverse range of participants ($N=76$) were purposively selected for their (anticipated) range of views, and the focus of the investigation was exclusively Down's syndrome. The findings of the Q study are summarised below:

- Using factor analytic techniques, **five statistically separate understandings of Down's syndrome were identified**: (1) People with Down's syndrome were considered to be part of the continuum of normality within which the concept of burden was inappropriate. The problems associated with the condition were seen as having a great deal to do with the attitudes of others. (2) A child with Down's syndrome was not considered equivalent to a 'normal' child and their birth was thought to be a sad misfortune for parents. A person with Down's syndrome was believed to remain childlike and dependent in adulthood. (3) Down's syndrome was seen as an organic disability (as opposed to a social one) associated with a high degree of parental care. While it was felt that the situation could be managed, the view was that it made sense to avoid having an affected child if possible. (4) It was accepted that people with Down's syndrome could be happy but their family would have to sacrifice their own quality of life to achieve this. The view was that a 'handicapped' child meant a handicapped family. (5) The birth of a child with Down's syndrome was viewed as a great disappointment to parents, which was perhaps a fault of the parents rather than the child. Such 'special' children were considered to need special (i.e. different) parents who could accept them as they were.

Despite the diversity of views expressed there was generally a consensus in supporting the right of existing individuals with Down's syndrome to have a good quality of life within an inclusive society. However, no such consensus existed about the right to life for the person with Down's syndrome who is yet to be born. The most obvious differences between the understandings related to how respondents felt that they as parents might cope with, and feel about, having a child with Down's syndrome. This seemed to be associated with whether a child with Down's syndrome was viewed as more similar to than different from a typically developing child. The findings of the Q study emphasised that understandings of Down's

syndrome are informed by a person's beliefs, attitudes, experiences, cultural, and social position. By definition, this also includes the understandings of Down's syndrome as held by health professionals situated within the prenatal testing context, some of whom were participants in the study. The assumption that there is an 'objective viewpoint' about Down's syndrome was challenged.

- **Associations between the different understandings of Down's syndrome and intentions towards using prenatal diagnostic testing and termination in hypothetical situations were identified.** Participants clustered on Factor 1 (Down's syndrome within the continuum of normality) were least in favour of preventing the birth of child with Down's syndrome, while those clustered on Factor 2 (Down's syndrome as a parental misfortune) were especially in favour of prevention. However, these conclusions were cautiously drawn due to the small number of participants and the method of strategic sampling used in the study. Understandings of Down's syndrome appeared to be more closely associated with intentions towards termination than they were with intentions towards using prenatal diagnosis. It was noted that a number of participants whose Q sort loaded significantly on more than one factor displayed relatively **ambivalent beliefs about Down's syndrome**. This ambivalence appeared to be **associated with less polarised views about prenatal testing**.

The final study (Chapters 5, 6 and 7) was designed to answer the question '**What role do pregnant women's understandings of Down's syndrome play in predicting their prenatal testing choices?**' No previously published study had attempted to identify the specific contribution of attitudes toward the condition in predicting testing intention and uptake. Over a six-month period, women in the first trimester of pregnancy attending an antenatal clinic were asked to complete a questionnaire that incorporated open-ended measures of cognitive, emotional and experiential aspects of attitudes towards Down's syndrome. They were also asked about their intentions regarding prenatal testing for Down's syndrome and termination of an affected pregnancy. An objective measure of the participants' serum screening uptake was then collected at a later date from patient records. A variety of statistical tests were employed to explore relationships between variables, and multivariate regression techniques were used to identify those variables that contributed most to predicting screening uptake or test and termination intention. A number of important findings emerged from this third study.

- It was demonstrated that **most women accept screening tests regardless of attitude towards Down's syndrome**. However, intentions towards termination for Down's syndrome were much more reliably predicted by attitudes towards the condition, and in particular by beliefs about parental quality of life and emotions elicited by meeting, seeing, or thinking about

people with the condition. This suggests that the values and attitudes informing termination intentions are not necessarily the same as those informing choices for screening tests, and that acceptance of screening cannot be taken as a proxy for attitudes towards Down's syndrome or the acceptability of termination for the condition.

- **Attitudes towards Down's syndrome and attitudes towards a screening test for the condition appeared to be unrelated constructs for many women.** Previously, this has not been identified as problematic within the context of informed choice. However, such a distinction might impact on the perceived relevance of attitudes and information about the condition and highlights further the complex nature of facilitating informed choice.
- Individuals with a relatively **high degree of ambivalence about Down's syndrome were most likely to be uncertain about termination, but they were not less likely to use serum screening.** In light of evidence suggesting that people high in ambivalence might approach information about an attitude object somewhat differently than do those low in ambivalence, this has implications for understanding informed choice at a somewhat more advanced level than has been examined to date. Further research is necessary to determine how different people use and value information in the prenatal testing context.
- **Beliefs about how having a child with Down's syndrome would impact on parental quality of life emerged as the most important predictor of intentions towards testing and termination in terms of attitudes towards the condition.**

The findings from the three studies will now be discussed more generally, considering their implications for information and choice and the relationship between the two.

8.2 INFORMATION ABOUT DOWN'S SYNDROME

The research presented within this thesis has demonstrated that a person's beliefs about how a child with Down's syndrome might impact on parental quality of life has a significant influence on their intentions to have prenatal testing and termination for the condition. It also appears to influence the actual screening choices that pregnant women make. Despite the importance of these beliefs, good quality information on how others have experienced parenting a child with Down's syndrome is rarely available within the antenatal situation. Participants in both the Q study and the study conducted at the antenatal clinic held a number of beliefs about the impact of a child with Down's syndrome on parents, siblings, and family life in general that are not supported by research evidence. It is argued that women have the right to access to this evidence (in an appropriate form) *even if* it has no discernible impact on the choice they make. Personal belief structures about Down's syndrome that are stable and strong are unlikely to be changed

fundamentally by the provision of information, and attempting to influence a person's beliefs and values in a particular direction goes against the spirit of informed choice. However, the findings suggest that there could be many women who would benefit from better quality information about Down's syndrome, for example, those who are especially ambivalent in their views or those who know very little about the condition. Failing to provide such information serves to maintain outdated stereotypes about Down's syndrome, or excludes those with no knowledge of the condition from making an informed choice. It also allows the message about disability that the offer of testing communicates to remain hidden and unchallenged. This might make testing choices easier for some women, but is it not appropriate in a medical culture that seeks to support informed and autonomous decision-making

Consumer involvement in the planning and delivery of health care services is becoming increasingly recognised as necessary for the provision of patient-centred care within the NHS (Department of Health, 2001a). To continue to see lay expertise in the area of disability as second best - a 'watered down and partially understood version of biomedicine' - is at odds with the overall aims of the modernisation of our health service (Stainton Rogers, 2001). To continue to see clinician's views of Down's syndrome as privileged and objective is at odds with the fact that clinicians too have experiences, beliefs and attitudes about disability that are likely to influence their interactions with patients. Pregnant women may not always share the views of their doctor or midwife but are often unlikely to disagree with them openly; this must be recognised more explicitly if the testing choices of all women are to be supported. It is the responsibility of service providers to make informed choice possible. This includes the basic necessity of providing good quality information about all aspects of testing and alerting pregnant women to its importance. Nevertheless, it has already been argued that informed choice cannot be achieved simply by improving information material (Bekker *et al.*, 1999). A deeper consideration of the psychological and social complexity of informing prenatal testing choices is now required.

8.3 INFORMED CHOICE

Although the importance of respecting an individual's beliefs regarding prenatal testing and termination is now recognised, the role of these beliefs in connection with the target condition has received little attention. This is especially true of research at the screening stage of the prenatal testing pathway. This thesis has argued that the current provision of information about Down's syndrome does not support informed screening choices, and might not sufficiently engage women with their own attitudes towards Down's syndrome and towards disability generally. However, the findings have also suggested that in many cases women themselves may not consider their

attitudes towards Down's syndrome as relevant to their screening choice. What implications might this have for the current conceptualisation of informed choice? Are women going against their values if they hold positive attitudes towards Down's syndrome yet have a prenatal test? Does this mean that they have made an uninformed choice? It is argued that attitudes towards the condition are only one of the many value-laden factors that may influence test choice, and in particular, that attitude towards testing appears to be more of an influence in the screening choices of some women. At different stages in the testing process, different information and different attitudes appear to vary in relevance for different individuals. Is it of concern that attitudes towards Down's syndrome are not always reflected in screening choices? An exploration of why attitudes towards prenatal screening may take precedence over attitudes towards the target condition is now presented.

8.3.1 Routine testing and informed choice

It has been argued previously that when a prenatal test is presented as routine it implies that the test has already been considered of value within a responsible antenatal package of care. Most women would not query the value of a routinely offered blood test for anaemia, for example, and some may see no difference between this and accepting a routinely offered blood test that identifies biochemical markers for Down's syndrome. This perception is supported by information literature aimed at pregnant women. For example, the booklet *Emma's Diary. A Week by Week Guide to Your Pregnancy* (given out at booking appointments and endorsed by the Royal College of General Practitioners) contains the following 'information' about screening tests.

"Week 16, Monday. [My friend says] they'll probably take a blood sample when I go to the hospital clinic tomorrow so they can do a blood test for spina bifida and Down's syndrome. I'm having a scan tomorrow as well. I can't wait to see my baby. Tuesday. At the clinic I had a blood test as well as other routine tests and the results were recorded on my maternity record." (Mackonochie, p. 40).

This is presented with no other information about serum screening tests, Down's syndrome, spina bifida, or choice. In fact there is no further reference to the tests or the test results. Routinisation of testing - by describing it in this way, by presenting the test to all women as part of standard care, or offering it as part of a standard 'check-up' appointment - may have the effect of distancing the consumer from their own values, thus diluting the impact of values on any subsequent behaviour. Evidence for this argument is provided by the study by Dormandy *et al* (2002a, reviewed in Chapter 2). In that study, the numbers of women making an informed choice were compared across two hospitals: one offering serum screening as part of a routine appointment and the other offering the test at a separate appointment. It was reported that attitudes towards having screening were more favourable and uptake was higher at the hospital where tests were offered as part of a

routine antenatal visit. Dormandy and colleagues suggested that conducting serum screening at a routine visit optimises informed choice because physical barriers are removed. While it is desirable that those women who wish to have testing are enabled to do so, removing physical barriers to having the test only facilitates the action, not the informing of that action – these are separate processes. In addition, the instrument used to measure informed choice in the study (Multidimensional Measure of Informed Choice, Marteau *et al.*, 2001) does not measure attitudes towards Down's syndrome or attitudes towards termination for the condition. The authors imply that a person's attitude towards using screening incorporates their values regarding the target condition and termination (see Marteau, et al (2001) pages 102-103). On the basis of the findings from the antenatal study, it is argued that this assumption is unwarranted, and that a measure of a single attitude construct does not provide an adequate assessment of someone's beliefs and values in relation to prenatal testing for Down's syndrome.

Although the evidence suggests that the method of screening delivery is an important influence on test uptake, not all women rely on situational cues to inform their decision-making. There is likely to be a relatively stable proportion of women declining testing regardless of how the service is delivered. This is because, as noted in Chapter 2, there is a relatively consistent proportion of the population who do not find termination acceptable. The findings by Dormandy and colleagues (2002a) also support this argument, as the rate of women declining screening was 23% at both 'routine' and 'non-routine' hospitals. There will also probably be a relatively stable group of women who definitely want to prevent the birth of a child with Down's syndrome (for example those in the Q study who felt that prevention of a child with the condition was particularly important). Many of these women might actively seek out testing if their care provider did not offer it. Little is known about why some women choose to pay privately for prenatal tests not yet available on the NHS, as this population has not been accessed in a research situation. It is suggested therefore, that the majority of the variation in uptake seen across different hospitals occurs in those women whose intentions and attitudes are more ambivalent, or unformed, perhaps because of a lack of personal experience or knowledge of Down's syndrome. For these women, their choices might in effect have already been made for them when their hospital determined how the screening test would be offered.

In most other areas of medicine health professionals can legitimately recommend a course of action. In the case of prenatal testing, however, this recommendation is not generally seen as appropriate. It is not considered acceptable for anyone other than the parents to decide whether a pregnancy affected by Down's syndrome should be continued or terminated. People's values and

attitudes concerning this issue are different, and very well informed individuals are found at each end of the attitude spectrum. It is considered both possible and necessary to provide a more neutral prenatal testing context at an operational and policy level by considering the message that routine test presentation may convey. The presentation of testing as routine clearly suggests that Down's syndrome is a condition that is best avoided (Green *et al.*, 1993b). Within this context individuals with little experience of Down's syndrome are expected to make autonomous choices, despite the accepted knowledge that situational factors exert an important influence on behaviour. In a recent qualitative study of how women make choices in pregnancy it was noted that, "*Women were often alert for clues that indicated approval and a positive attitude regarding their choices and actions*" (Levy, 1999, p. 188).

To facilitate autonomous decision-making a context has to be created within which women do not perceive testing for abnormality as routine even when it is available to all, or offered during a standard antenatal appointment. Prenatal testing for disabling conditions must be seen as an optional service not a recommended course of action. Screening tests should be presented as procedures associated with potential consequences, and as such, requiring a decision to be made. Engaging women's own values and experiences should be of prime importance along with the provision of accurate information about the tests and target conditions. Creating this context might be the most important step towards making informed decisions possible and yet might also be the most difficult to achieve. There is a policy imperative to increase patient choices within NHS services generally⁶⁵ including maternity care, however, research in the area of decision-making suggests that by associating screening with choice, service providers might in fact be guiding decisions in a particular direction.

8.3.2 Informed choice or the lure of choice?

Choice is highly valued in our society, and attempts by the Government to limit health-care options (as witnessed during the recent debate about the MMR vaccination⁶⁶) are seen by some as an attack on personal autonomy. Choice in this context has two meanings; in the active sense of choosing or refusing something, and in the passive sense of having more than one option from which to select. The majority of participants in both the Q study and the antenatal study agreed

⁶⁵ Department of Health press release 11/02/2003. Alan Milburn (Secretary of State for Health) sets out expansion plans for NHS choice. Maternity services are to be put under a national watchdog to 'give women more choice'.

⁶⁶ The 'triple vaccine' for mumps, measles and rubella (MMR) is thought by some researchers to be linked to autism and bowel disease. Some lobby groups have called for the government to increase parental choice by making a course of single vaccines an option available through the NHS.

that pregnant women should have the choice to have prenatal testing even if they did not intend to use testing themselves. However, the very act of presenting prenatal testing within the framework of choice might make the offer difficult to refuse. If a woman opts out of screening she effectively curtails her options from that point, but if she opts in, she can decline or accept further testing after the result of the screening test is known - in theory at least. This desire to keep ones options open is related to the concept of anticipated decision regret: some women foresee that they might regret having declined screening if their baby was later found to have a disabling condition (Tymstra, 1989; Tymstra, 1991).

Research has demonstrated that both animals and people appear to prefer options that offer choice over no choice (Catania, 1980; Suzuki, 1997; Suzuki, 2000). This preference might have general evolutionary advantages, for example, in the selection of habitats where a greater range of food enhances survival. However, further examination of this preference has led some authors to suggest that preference for choice can act to produce sub-optimal decision making (Bown, Read, and Summers, 2002). The phrase 'lure of choice' is used to describe the situation when an individual selects an option that offers choice even when doing so results in an outcome equal to or less good than the one that would have resulted from the 'no choice' option. For example, when doing their weekly shopping someone might prefer to go to a large supermarket rather than a small one because the larger store offers a wider range of products. This is a preference for choice. However, if the person pays more for the same items by shopping at the large supermarket then choice has acted as a lure and produced a sub-optimal decision in financial terms. In the context of prenatal testing, screening offers an option of future choice over no choice, but it is argued that this future choice might be illusory in a psychological sense. *A priori*, women may believe that a positive screening result would leave them unchanged except for having some more information about the health of their baby. However, research shows that receiving a positive result almost always generates high levels of anxiety (Green *et al.*, 2002), and the majority of women do go on to have diagnostic testing, partly in order to relieve this anxiety. Some women choose not to take the test process any further, and for them, the option of screening may not have been in their best interests.

Bown *et al.* (2002) hypothesise that choosing choice is seen as preferable for a number of reasons. First, it is a way to defer commitment to one option for as long as possible and to continue gaining information, even beyond the point at which this information is likely to contribute to the final decision. This might partly explain why women in the antenatal study whose attitudes were highly ambivalent towards Down's syndrome were not less likely to opt for screening or intend to use

amniocentesis than were women whose attitudes were unfavourable with low ambivalence. Over one-third of women said they wanted to have screening to give them information about their baby, including those who said that they did not intend to act on the information if they were found to be in the higher risk group. Secondly, employing a 'choice better than no choice' heuristic may reduce cognitive effort in situations of potentially complex decision-making where information is scarce or difficult to assimilate. It has already been demonstrated that information about testing is often inadequate in antenatal care, and is not always understood. There is no doubt that a decision regarding prenatal testing can be perceived as complex. Bown *et al.* suggest that a preference for choice is favoured as a heuristic in complex situations because in the natural environment choice often leads to the best outcome. They argue that all things being equal, an easy to implement choice will be favoured over a more effortful one. In the testing context, while having a test might require more physical effort than not having one (in terms of attending an appointment) having to decline testing might be seen as more effortful in terms of having to think about and explain to care providers why testing is being declined.

Further evidence for the lure of choice has been demonstrated in studies testing the 'illusion of control' model (Langer, 1975). An illusion of control is defined as, "*An expectancy of a personal success probability inappropriately higher than the objective probability would warrant*" (Langer, 1975, p.313). In games of chance such as a lottery, it has been demonstrated that allowing people to choose their own lottery ticket (as opposed to a no choice option) creates an illusion of control over the outcome (Dixon, 2000; Langer, 1975; Wohl and Enzle, 2002). In one of two studies investigating the effects of offering choice in games of chance, Langer (1975) noted that the offer of choice (i.e. the opportunity to choose ones own ticket) was enough to make people opt for a lottery with an inferior chance of winning over one with superior odds but with no choice of ticket. Langer notes that while people 'pay lip service' to the concept of probability they behave as though chance events can actually be controlled. There is evidence that many women find the concept of probability in relation to serum screening results difficult to understand (Green *et al.*, 2002). Promoting screening as the woman's own choice without considering the context within which this choice is presented might help to foster an illusion of control over a favourable outcome, whereas doing nothing, i.e. declining testing might appear to be leaving the outcome purely to chance. The idea of actively deciding to do nothing about the health of one's fetus runs counter to the psychology of most pregnant women, hence the high value placed on antenatal care.

In summary, it is argued that a number of factors make prenatal screening tests for abnormality especially attractive to pregnant women. These are the natural desire to know that ones' unborn

baby is healthy, an in-built preference for choice and control, and the value that is placed on antenatal care in general. If testing is also presented as routine, then individual and social normative factors combine in a way that might make declining the offer of screening tests psychologically difficult and even counter-intuitive for many women. In addition, it is suggested that many women do not actually hold very strong opinions about prenatal screening tests or people with Down's syndrome, and that they construct their testing preferences based on the context in which it is offered (Bettman, Luce, and Payne, 1998). This might also help explain why attitudes towards the target condition appear to have little influence over many people's screening decisions.

We can now return to the question posed earlier – is it of concern that attitudes towards Down's syndrome are not always reflected in screening choices? It is argued that the answer to this question is 'yes' for a number of reasons. First, the lack of a strong relationship between attitudes towards Down's syndrome and screening uptake can be seen as an indicator of the influence of context over screening decisions. A context that more strongly supports one outcome than another does not facilitate autonomous decision-making. It has already been argued that autonomous decisions are preferable in light of the potential consequences of testing for the individual woman and her family. Secondly, the apparent distancing of women from their values and knowledge (regarding Down's syndrome and termination for example) is of concern within the current conceptualisation of informed choice. Prenatal tests will soon be possible for many rare genetic conditions, and some fear that supporting informed choice will be even more difficult if such blanket genetic testing becomes part of routine antenatal care (Williams, Alderson, and Farsides, 2002b). In the scenario where a woman knows nothing about the condition(s) being tested for, prenatal testing choices may depend almost entirely on beliefs about the value of antenatal care, attitudes towards testing, and perceptions of the social norm. Furthermore, if presenting prenatal testing within a framework of choice does increase the likelihood that a test will be accepted, then widening testing choices is likely to further distance women from their values and attitudes in respect to the condition being tested for. Attitudes toward the specific condition will probably become almost irrelevant unless information about the effects of that condition are specifically presented. In view of the likelihood that blanket prenatal genetic testing will become a reality, a deeper understanding of the role of attitudes towards abnormality and disability is called for.

8.3.3 Recommendations for supporting informed choice in practice

It is the policy of the Department of Health that people who accept screening do so on the 'basis of informed choice' (Department of Health, 2000). The evidence from many sources suggests that

even the basic provisions necessary for this are often missing. Based on the findings and theoretical discussions presented in this thesis the following recommendations for supporting informed choice in the antenatal situation are proposed.

1. More attention must be paid to providing women with information about Down's syndrome prior to serum screening. In particular, such information should be balanced in its construction, with thought given to the needs of the reader and to the tone and the content of the message conveyed. Availability of information that deals with the psychosocial effects of parenting an affected child should be improved and evidence based.
2. Appropriate information must be made available to non-English speaking women. In many cases straightforward translation of existing leaflets will be insufficient, as in some cultures the concept of Down's syndrome as a specific condition does not exist.
3. There should be explicit recognition that midwives and obstetricians are unlikely to be experts on Down's syndrome but are individuals with their own subjectivity. Personal views about Down's syndrome and disability should not be communicated to patients as expert knowledge.
4. Women should be actively encouraged to engage with and discuss their own experiences and attitudes towards Down's syndrome in the prenatal context. However, over-generalisation from a narrow range of personal experience might not give a full picture of the condition, hence the need for accurate information.
5. Prenatal testing for abnormality should not be presented as a routine component of standard antenatal care even when it is offered to all women or within a normal antenatal appointment. The consequences for the women who obtain a screen positive result, for example, are never experienced as routine, and the potential implications of different testing options should be emphasised.
6. Screening should be presented in a context of decision making, rather than in the context of enhanced choice. The relevance of knowledge, attitudes, and information about the target condition to these decisions should be made clear.

The conclusions and recommendations presented in this thesis are based on data collected as part of this Ph.D. in conjunction with the published research and theories of others. In each of the empirical chapters where study findings were discussed, appropriate methodological issues were also raised. While the findings of the studies are considered to be robust, with proper consideration being given to the reliability and validity of all the methods used, a number of limitations apply to the thesis and they will now be discussed.

8.4 LIMITATIONS OF THE RESEARCH PRESENTED WITHIN THIS THESIS

8.4.1 A content analysis of serum screening leaflets

The question that this study was attempting to answer was. **‘What information do women receive about Down’s syndrome to help inform their prenatal testing choices?’** The results of the study provided an in-depth analysis of the information about Down’s syndrome as given to pregnant women via a sample of serum screening leaflets. However, a number of limitations of the study should be considered when assessing the degree to which the findings accurately represented the information about Down’s syndrome that women receive prior to making their screening choices.

Firstly, the analysis was limited to those leaflets collected for a previous study (Murray *et al.*, 2000) and it is possible that leaflets offering a higher quality of information about Down’s syndrome were missed from this initial sampling. However, it is argued that the sample analysed was representative of the total population of screening leaflets offered to women in the UK, and covered maternity units attached to a range of hospitals covering both rural and urban districts. A number of ‘good quality’ leaflets produced by MIDIRS (Midwives Information and Resource Service) and the NCT (National Childbirth Trust) were included in this sample as they were distributed by a number of maternity units. For example, the MIDIRS leaflet is part of the set of ‘*Informed Choice*’ evidence based leaflets produced in association with the NHS Centre for Reviews and Dissemination. However, it contained only three statements of basic medical/clinical information about Down’s syndrome. The NCT leaflet was one of the few to include information on the emotional aspects of parenting a child with Down’s syndrome. Secondly, the leaflets analysed in the study were intended for distribution early in the pregnancy prior to the offer of serum screening. Thus better quality information about Down’s syndrome might be available at later points in the testing process, i.e. prior to decisions about amniocentesis or termination. However, other studies that have considered the information about Down’s syndrome given to pregnant women, including studies of verbal information given by health professionals following a positive screening result, have reported similar findings to those reported in this study (Bekker, 1999; Carroll *et al.*, 2000; Edwins, 2000; Gekas *et al.*, 1999; Helm *et al.*, 1998; Levy, 1999; Marteau *et al.*, 1993; Marteau *et al.*, 1992b; Moyer *et al.*, 1999). In addition, it was noticed during the study conducted in the antenatal clinic (i.e. ‘Attitudes towards Down’s syndrome in the prenatal testing situation’) that the information about Down’s syndrome given to women in a leaflet following a positive screening result was more negative in tone than that provided in the

leaflet distributed prior to screening⁶⁷. Assumptions should not be made on the basis of only one example however, and so a wider analysis of information about Down's syndrome given to women in the prenatal testing context would be valuable.

A third limitation is that the conclusions drawn may no longer stand as written information about Down's syndrome may have increased significantly in quantity and quality since the time when the leaflets were first collected (at the time of writing five years ago⁶⁸). However, while it is likely that some leaflets have been replaced, it is also likely that many are still in use, or that information about Down's syndrome has not been significantly improved in updated versions. For example, the leaflet from the MIDIRS (Midwives Information and Resource Service) '*Informed Choice*' series was updated in 2001⁶⁹, however, the information about Down's syndrome remains the same as in the leaflet analysed as part of this thesis. Two other recent publications have also made reference to a lack of good quality information about Down's syndrome (Marteau and Dormandy, 2001; Williams *et al.*, 2002c). In sum, there is no reason to believe that there has been a great improvement in provision of written information about Down's syndrome in the prenatal screening context. It would, however, be desirable to revisit the leaflet analysis at some point in the future (for example, in 2008 ten years from the original data collection) to assess how the information about Down's syndrome has changed. By this time the government's policy of universal screening availability should have been in operation for four years, and a standardised information package disseminated.

A fourth limitation of this study was that it only considered information written in English. While it was noted that few leaflets included information in other languages, this was not identified as a variable of interest at the time of the analysis. Therefore, while it is argued that conclusions about the lack of information about Down's syndrome in languages other than English are probably valid, the full nature and extent of this lack of information remains to be verified. It is also important to note that the comparison study by Loeben *et al.* (1998) showed that significantly more positive statements about CF were found in the US leaflets than in the UK leaflets. The

⁶⁷ The amniocentesis leaflet referred to Down's syndrome "commonly being called Mongolism", noted that "from a medical point of view many people underestimate the difficulties involved in looking after a child with Down's syndrome" and that "as a matter of record some relationships break down under the strain". None of these items of information were provided in the screening leaflet.

⁶⁸ The leaflets were originally collected in 1998, see Murray *et al.* (2001).

⁶⁹ The MIDIRS leaflets are now available electronically at <http://www.midirs.org/nelh.nsf/>.

findings of this study might, therefore, not be broadly applicable to other countries that provide serum screening for Down's syndrome.

In summary, the content analysis of screening leaflets has considered only one source of information about Down's syndrome that women might use to inform their prenatal testing choices. The limitations discussed above must be taken into consideration when assessing the degree to which the study addressed the initial research question. In his chapter on possible research designs of studies using content analysis Holsti (1969) notes, "*The investigator may analyze messages to make inferences about the characteristics of the text, the causes or antecedents of messages, or the effects of communication*" (p. 24). This study addressed only the characteristics of the text using two fairly basic approaches. The possible reasons for the nature of the message were explored in Chapter 3 but these require independent verification. Most urgently however, the possible effect of the lack of information about Down's syndrome (and its negative bias) on prenatal testing choices has yet to be objectively and systematically investigated.

8.4.2 Understandings of Down's syndrome: a Q methodological investigation

The primary research question addressed by this study was, '**What understandings of Down's syndrome exist independently of the prenatal testing context?**' and the main aim was to characterise these understandings and to identify similarities and differences between them. A critique of Q methodology was provided in the introduction to Chapter 4. This section will not revisit this critique, but instead will look at how issues arising from the way that Q methodology was operationalised could impact on the conclusions drawn about the findings of the study.

The first issue relates to the 'forced choice' sorting technique that was selected. This technique was selected to help reduce socially desirable responding by forcing the sorter to discriminate between variables. Sorters cannot allocate the same position on the grid to more than a few items in contrast to the way they could allocate the same score to any number of items when completing a standard questionnaire. However, using a forced distribution also has the potential to distort responses in a different way. For example, the neutral point on the scale at the bottom of the grid is not an anchor in the usual sense, as all items are being ranked as 'a piece'. Most participants found they had to place some items they had originally agreed or disagreed with into the neutral column on the grid. In some cases the neutral column became the 'slightly agree' or 'slightly disagree' column, and the neutral items were shifted along. This made it unsafe to assume that the score of the items placed in the zero, +1, and -1 columns were exactly as had been originally intended. However, the aim of using Q methodology is to give structure to subjectivity, and the

interpretation of the overall pattern is of more interest than the score allocated to an individual item. Statements placed in the zero column represent the physical centre of the ranked items rather than an absolutely neutral view⁷⁰. Interpretation of the factors should therefore focus on the items falling at the end points of the scale as their meaning can be more reliably perceived, although the relative position of the more central items can be seen to provide additional insights. This issue is likely to relate to many Q studies and should be considered when assessing the researchers' interpretations.

The second issue is connected with the selection of factors prior to the rotation procedure. Despite the use of techniques such as the scree test, the selection of five factors was a subjective one. Another researcher may decide to rotate fewer or more factors and therefore report slightly different results with the same data. Although proponents of Q method exhort researchers not to make claims as to the concreteness of the factors, it is easy for those unfamiliar with the method to interpret factors as categories of people and make conclusions about their prevalence in the population. Care must be taken when reading reports of Q studies to view the findings as an interpretation of a phenomena rather than a definitive mapping. One way of validating the selection of factors would have been to ask participants whether the researcher's interpretation of their viewpoint agreed with their own. The decision was made not to approach participants directly for their view, mainly due to time constraints and lack of contact details for all participants. A summary of the study findings was sent to participants for whom contact details were known, and while feedback was invited, none was received. In retrospect, a more explicit request for feedback might have provided useful information about the decisions that were made regarding factor selection and interpretation.

Thirdly, a limitation of the study is that people with Down's syndrome were not included in the expert group. At the time the study was developed it was not felt that the methodological and ethical considerations necessary to ensure the safe inclusion of people with Down's syndrome were warranted. However, it is accepted that by not including the views of people with the condition an important gap in the subjective understandings of Down's syndrome might exist. If the views of people with Down's syndrome were to be assessed using Q methodology, the materials, design, and procedure would certainly need to be revisited, and the benefits and costs to participants should be considered in detail. Another approach would have been to purposefully seek out text that included the views of people with Down's syndrome during the development of

⁷⁰ The neutral position is called 'distensive zero' in Q terminology because it is the point from which "all the information, so to speak, bulges out or distends" (Stephenson, 1953, pp. 195-196).

the Q sample. This was not done, and in retrospect this omission highlights how easily the voices of those with learning difficulty are excluded from the debate about the quality of their lives.

The secondary aim of the Q study was to explore **‘the relationship between understandings of Down’s syndrome and intentions towards undergoing prenatal testing for the condition’**. While the findings of the study gave some pointers as to how understandings might relate to attitudes towards prenatal testing, they also highlighted the difficulties of trying to identify patterns of association between factor membership and other variables when only a few individuals represent some factors. Associations in terms of ability to predict testing and termination intentions from factor membership were not provided by this study, and this might be considered a limitation. However, it is argued that this is not a limitation of Q methodology but rather a problem with attempting to draw ‘quantitative’ conclusions from qualitative data. The aim of the study was to explore potential links between variables not make predictive statements. Q methodology is not suitable for identifying predictive links because of the nature of the participant recruitment and because the data is not normative in nature. Running the study with a larger group of participants would not have resolved this issue; instead a different type of approach would be required such as a survey using a normative measure of attitudes administered to a representative sample of individuals.

Q methodology is still a fairly novel approach to studying attitudes within psychology, although as noted in the introduction to Chapter 4, other areas of social science, including health studies, have used the method more widely. It is believed that despite its limitations Q held certain advantages over more traditional techniques in a study of understandings of Down’s syndrome. The views of seventy-six people from a diverse range of backgrounds were collected, analysed and interpreted in a systematic way within a relatively short space of time. Had in-depth interviewing been the method of choice (and for example, discourse or narrative analysis) this diversity would have been very much more difficult to access and model, and the number of participants would have had to be greatly reduced. The factor analytic techniques employed made the identification of themes, similarities and differences in understandings relatively straightforward to identify. The interpretations are accessible to others via the presentation of full factor arrays in a way not possible with narrative data. However, the merging of views that is part of the Q methodological approach might have meant some subtleties of individual views were lost. This makes the method, as operationalised in this study, unsuitable where it is essential to hear the ‘voice’ of each

individual⁷¹, or where the researcher wants to explore a topic in a more dynamic, data driven way. As described in Chapter 1, studies measuring attitudes towards Down's syndrome have most frequently used questionnaires. It might be considered that the items selected from the concourse could have been developed into a questionnaire using Likert-type scaling and administered in the usual way. Some researchers have taken this approach and Stephenson's original book on Q methodology contains a chapter on developing a Q set into a survey⁷². However, presenting the items as a standard questionnaire removes the key advantages associated with 'forcing' people to make comparisons and choices including the reduction of socially desirable responding. Q sorting is generally considered more effortful than questionnaire completion and arguably might require the participant to engage and think through the issues of the topic more thoroughly. The Q sorting technique also enables both participant and researcher to see the value/belief patterns more clearly and can prompt the participant to comment on the items and their placement giving further insights into aspects of people's understandings of the topic. For these reasons, the Q sorting technique has been the approach generally favoured by researchers using the methodology and was considered the most appropriate technique to answer the main research question addressed by this study.

8.4.3 Attitudes towards Down's syndrome in the prenatal testing situation

The questions that this study aimed to answer were, **“what is the relationship between understandings of Down's syndrome and behavioural intentions towards undergoing prenatal testing for the condition?”** and **“what role do understandings of Down's syndrome play in pregnant women's prenatal actual testing choices?”**. The tool used to access understandings of Down's syndrome was an open-ended measure of attitudes (experiences, beliefs, and emotions) adapted from those developed by Esses and colleagues (Esses *et al.*, 1993). It has been claimed that open-ended measures are easy to use, as *“respondents have the relatively simple task of reporting the beliefs and affects that ‘come to mind’”* (Eagly *et al.*, 1994, p.118). However, in this study 8% of respondents did not complete any of the open-ended measures, and failure to provide at least one response per attitude component ranged from 12% (stereotypic beliefs) to 16% (experiences). Overall, data from 29% of participants were excluded from the attitude analysis because one or more of the measures were not 'correctly' completed. There are a number of reasons why this high rate of missing data may have occurred.

⁷¹ However, Q sorting is used in single-case studies, often in clinical research. In this situation, the individual is often required to Q sort a set of materials from a number of different perspectives (Goldman, 1991).

⁷² Stephenson, 1953, Chapter 9, *“The prior analysis of questionnaires”*.

Firstly, it is possible that the completion instructions were unclear or difficult to understand. However, it is unlikely that this is a major contributing reason for the high data loss as the instructions were designed to be as close to the original ones as possible and the measures were checked for readability and clarity with members of the target population. Secondly, a lack of interest in the topic matter may have reduced motivation to complete the measures or to complete them correctly. However, if salience in issues related to testing for Down's syndrome is connected to maternal age, motivation about the topic may have not been a significant factor as no difference in age was found between those who correctly completed the measures and those who did not. Thirdly, some participants may have had little knowledge of people with Down's syndrome and therefore believed that they had no attitudes to record. Four women who had left the experiences measure blank used the response box to say that they had no experiences of people with Down's syndrome. In further support of this, a study measuring attitudes towards people with disabilities rejected 14% of questionnaires due to 'incorrect completion' (Esses and Beaufoy, 1994), which is higher than previously reported loss rates of between 3% and 5% (Haddock and Zanna, 1998). Esses and Beaufoy (1994) suggested that the failure of some respondents to complete the measures might have been due to a lack of first-hand experience of people with disabilities. If lack of knowledge of Down's syndrome contributed towards non-completion of the measures this is of interest in itself as the 'non-completing group' was still able to express their intentions related to testing and termination for the condition. Neither screening intentions nor test uptake differed significantly from the rest of the sample.

While the reasons outlined above probably contributed to the high rate of incorrectly completed returns, it is proposed that the most important contributory factor was level of education. Those women who did not complete the measures, or had completed them incorrectly, had left education at a significantly younger age than those who correctly completed the measures. It is therefore suggested that the level of education required to complete these measures correctly might have been underestimated. The developers of the measures have argued that use of the open-ended measures is not limited to populations with high verbal abilities (Esses and Maio, 2002; Haddock and Zanna, 1998), and cite the view of Geer (1988) that "*only 5% of the general public are not articulate enough to answer open-ended questions*" (Geer, 1988, p.367). This assumption does not appear to have been tested, however, and there are few studies using the open-ended measures with non-student populations. The possibility of low completion rates should therefore be taken into consideration by researchers wishing to use these measures and research that evaluates the use of the measures on the wider population is required.

A further limitation of the measures as employed in this study was that they produce a uni-dimensional attitude score (favourable to unfavourable) while attitudes towards people with disabilities are known to be multi-dimensional (Antonak and Livneh, 1988; Furnham and Pendred, 1983). Multiple dimensions of attitudes towards Down's syndrome were also apparent in the data collected in this study, for example, the dimension 'normality - abnormality' in terms of perceptions of people with the condition, as well as 'favourable - unfavourable'. It is accepted that treating the data as a uni-directional construct undoubtedly hides a much richer and more complex pattern of associations, and that further analysis of the attitude data could be conducted at a future date. In addition, the attitude construct measured in this study was attitude towards Down's syndrome, rather than attitude towards *existing people* with Down's syndrome. This construct was considered to incorporate aspects of attitudes towards having a baby with the condition as well as attitudes towards existing individuals. It was believed that measuring attitudes only towards people with Down's syndrome would not have been particularly useful in light of the findings from the Q study that attitudes towards the 'yet-to-be-born' person with the condition were most important in relation to testing and termination intentions. Thus, while the experiences, stereotypic beliefs, and emotions components measured aspects of attitudes towards *people* with Down's syndrome, the PQoL beliefs tapped a component of an attitude towards *having a baby* with Down's syndrome. This might explain why the PQoL emerged as the attitude component with the greatest predictive power in the Discriminant Analysis, and it could be argued that to amalgamate the separate scores into an overall attitude score was a confounding of the two constructs. In retrospect, an overall evaluation of people with Down's syndrome as favourable or unfavourable might have been a useful addition to the questionnaire as this would have enabled separate analyses of the role of the attitude components in predicting attitudes towards people with Down's syndrome and of having a baby with the condition oneself. It is recommended that any replication of this study incorporates such a measure.

A general limitation of this study is that it only considered the viewpoint of the women, and therefore, the influence that partners may have had on screening choices and testing and termination intentions was not considered. While the main focus of the study was pregnant women it is known that beliefs about what significant others think a person should do in a situation can have an impact on their behaviours. For example, the Theory of Planned Behaviour (TPB) includes the concept of 'subjective norms' to account for this influence or 'social pressure' (Conner and Sparks, 1995). In relation to the concept of ambivalence, it was noted that those women who were uncertain about their intentions to terminate for Down's syndrome were more likely to mention the need for discussion with their partner in the reason they gave for a 'don't

know' response. For these reasons, it would have been useful to measure the degree to which women felt their partner's views on testing and termination were important to them, and what they believed these preferences might be.

In summary, the study of attitudes towards Down's syndrome in the prenatal testing situation had a number of limitations and it is recommended that a study seeking to replicate these findings should bear these in mind. In particular, the conclusions about the weak relationship between attitudes towards Down's syndrome and screening choices, and the relationship between ambivalence and uncertainty towards termination resulted from a single study at one clinic. This clinic offered screening as part of a routine package of antenatal care to all pregnant women, and as already discussed, situational factors might have a very important moderating role on how attitudes towards the target condition influence testing choices. Data relating to the potential impact of situational factors could not be measured using a single-site study and this was a limitation of the study design.

On a more general point, the focus of this thesis was Down's syndrome, which necessarily precluded consideration of other tested-for conditions. However, screening for neural tube defects (NTDs) has also been central to the development and dissemination of prenatal testing. While a number of studies have considered other psychosocial aspects of screening tests for NTDs such as anxiety there is an absence of research considering people's understandings of NTDs and spina bifida. It could be argued that most women are unlikely to have much experience of the wide-ranging nature and effects of NTDs, and this is a topic for further research. While there might well be some commonality between the findings of research on understandings of NTDs and the research presented in this thesis, assumptions should not be made, and to do so further encourages the homogenisation of disability and the disabled in our society.

8.5 FUTURE RESEARCH DIRECTIONS

A number of issues have emerged from this thesis that are considered worthy of future research. These issues fall within two main topics, (1) further research on information and informed choice, and (2) research aimed at a theoretical consolidation within the area of prenatal testing.

8.5.1 Information and informed choice

- It has been shown that despite many recommendations, information about Down's syndrome is frequently sparse, missing, or overly negative in nature. While some women would like more information, some do not perceive information about the condition as a necessary

component of an informed screening choice. There is currently no way of knowing what impact missing or biased information has upon screening test uptake or prenatal testing choice in general. It is unlikely that this will be resolved unless a **reliable measure of knowledge of Down's syndrome is developed** that validly addresses the complexity of the condition.

- Where information about screened for conditions is missing or not perceived as useful, other situational cues may have a greater influence on testing choices. Attitudes towards using testing might well be influenced by service delivery. Further research is needed to assess **the role of service delivery and other social variables on attitudes towards testing and test uptake**. There are a number of useful places to start in this research. For example, the 'tentative model of attitudes towards prenatal testing' presented by Jørgensen (1995, p. 428) includes societal factors although these are mainly high level or conceptual variables, for example the "norms and ethics of society". However, method of service delivery, the independent variable used in the informed choice studies by Dormandy and colleagues (Dormandy *et al.*, 2002a; Dormandy *et al.*, 2002b) is a variable that can be captured easily. The degree of routine with which tests are presented is a more abstract concept but one for which a measure might usefully be devised.
- Different women may use and value information about Down's syndrome in different ways. Information might be most useful to those women who have ambivalent attitudes towards Down's syndrome. Research **investigating the relationship between attitudinal ambivalence and aspects of the testing environment** would therefore be worthwhile, for example;
 - (1) A comparison of the effect of the provision or absence of information about Down's syndrome on screening intentions in those with high or low levels of ambivalence.
 - (2) An assessment of satisfaction with information provision and decisions made in women with high and low levels of ambivalence towards Down's syndrome. Women who have highly ambivalent attitudes might be more likely to follow the normative direction (whatever that might be). This might result in them being less satisfied with their decisions than women whose views were less ambivalent. Alternatively, women who are highly ambivalent might be more satisfied with their choices because they have thought through the issue from a number of angles.
 - (3) An investigation into the effect of ambivalence on the processing of attitude relevant information and of the decision-making processes of different groups regarding prenatal testing would also be informative.

Such research would usefully contribute to a better understanding of how information is used and valued by different women, as well as inform more theoretical aspects of research into ambivalence and attitudes. Further exploration of the effects of attitude ambivalence in the prenatal testing context is required to understand the information needs of individual women. Information might help resolve ambivalence in some women for whom ambivalence is an uncomfortable state. Alternatively, ambivalence might not necessarily be something that must be resolved, and so supporting women to work through conflicting feelings and beliefs might be required in some cases.

- The role of men in ‘informing’ the testing choices of their partner has generally received little research attention. As many men will receive their information about prenatal testing ‘second-hand’ from their partner, it might be anticipated that their knowledge levels would be even lower than those seen in pregnant women. How this then applies to knowledge and attitudes towards Down’s syndrome is unknown. The findings from the Q study did not suggest that the views of men and women about the condition were very different, although the limitations of using Q methodology to address these questions make assumptions unsafe. It is recommended that **future research investigating the influences on prenatal testing also considers the role of the father’s attitudes towards testing and the target condition.**

8.5.2 Theoretical consolidation

Over the last decade or so, research has identified variables that consistently predict the acceptance or rejection of prenatal tests and termination for Down’s syndrome. These include individual factors such as attitude to abortion generally, anxiety, religiosity, education, and maternal age. It is argued that attitudes towards the target condition should also be included in the list, particularly in relation to termination intentions. Situational variables, such as the way a test is presented, i.e. in a routine context or as something to enhance individual choice, also appear to be important but have been less extensively researched in a systematic fashion. It is argued that a lack of appropriate theory in the area of prenatal testing hampers a more rigorous and necessary examination of the influence of situational constraints on testing choices. There is now a need to consolidate existing research from a theoretical perspective, in order to complement the recent synthesis of research on psychosocial aspects of screening by Green and colleagues (2002), and the movement towards theoretical interests in prenatal decision-making.

One approach to this would be to test existing models of health behaviours using the factors as identified in the literature. Previous research has demonstrated some limited value in using models and theories from social and health psychology to predict prenatal testing attitudes and behaviour,

for example, Ley's Cognitive Hypothesis, Subjective Expected Utility Theory, and the Theory of Reasoned Action (Marteau *et al.*, 1992a) and the Health Locus of Control (Furr and Seger, 1998). However, in general these models were not implemented rigorously, and therefore further research might demonstrate them to be more useful than previously supposed. However, existing models might not be best suited to explaining the complexities surrounding prenatal screening choices. As discussed in an earlier chapter, the Theory of Planned Behaviour (TPB) (Ajzen, 1985, 1988, 1991) does not give attitudes towards the target a formal explanatory role in explaining behavior. Although the TPB includes normative beliefs, environmental factors such as the method of service delivery are not included. The Health Belief Model (HBM) (Becker, Haefner, and Maiman, 1977) and the Protection Motivation Model (PMM) (Rogers, 1983) account for 'perceived severity' of a health-threatening condition but do not allow explicit investigation of the influencing roles of attitudes towards related behaviours (i.e. abortion) and do not include a role for social and environmental influences.

As noted in Chapter 5, Eagly and Chaiken (1993) have proposed a composite model of the attitude-behaviour relation that brings together attitudes towards behaviour (for example, screening) and attitudes towards the target (for example, Down's syndrome). The model sets out a causal sequence whereby attitudes towards targets affect behaviour via their impact on the attitudes towards the behaviour itself. It is argued however, that the findings from this thesis demonstrate that (at least) three attitudes are important when considering the influences on testing and termination choices; (1) attitudes towards Down's syndrome, (2) attitudes towards the procedures (screening, amniocentesis and termination for Down's syndrome), and (3) attitudes towards undergoing these procedures. While the attitude towards the procedure may precede the attitude towards undergoing it, the attitude towards Down's syndrome itself is a separate construct that may act independently on behavioural intentions and choices. Internal processes, including the perceived relevance of the attitudes under consideration, may play a crucial role in deciding which of the attitudes goes on to exert the greatest influence over the final choice. Further research is required to investigate and define the moderating role of these different influences on the relationship between attitudes towards Down's syndrome and attitudes towards testing and termination for the condition. Such research could also usefully take into consideration other known influences on behavioural intentions in this context such as past behaviour, subjective norms and environmental factors.

The development of a theory-driven model to consider all the variables known to influence testing choices would be a useful contribution to the prenatal testing literature. Any model would need to

explain the varying influences of important variables at different stages of the process, from screening through diagnostic testing and termination. For example, anxiety is likely to operate differently before screening and after receiving a positive screening result. Such a model would help specify the roles of test context and attitudes toward the target condition more clearly and so have applications beyond testing for Down's syndrome.

8.6 FINAL POINTS

The research presented in this thesis was driven by perceived gaps in the literature in the area of informed choice for prenatal testing for Down's syndrome. In particular the issue of interest was information, knowledge, and understandings of Down's syndrome. It is believed that this thesis has significantly contributed to this area in a number of ways. The findings of the leaflet analysis demonstrate that in terms of informed choice regarding prenatal testing for Down's syndrome, information regarding the condition itself rarely meets adequate standards of quantity, quality, and non-directiveness. However, the second two studies consider issues beyond the simple provision of the 'right' information. They demonstrate the subjectivity of understandings of Down's syndrome and how they permeate the individual and collective values attached to prenatal testing. They highlight the complexity and range of knowledge about the condition and dispute the ownership of objective information by health professionals. They point to the need for developing a better understanding of how individuals use (or don't use) such information when making choices about prenatal testing. They suggest health service providers look again at the routine context within which testing and its associated information is presented and how this might distance the consumer from their own values. It has been proposed here that information has varying degrees of relevance for different individuals at different stages of the testing process. It is also argued that these findings point to a need to understand informed choice from the perspective of the consumer within the prenatal testing context, and what choice means to them.

As new technologies come along, prospective parents will be given more choices (or face more decisions) about their unborn children. Although some form of gene therapy for Down's syndrome remains a (very) long-term possibility, the complex genetic nature of the condition would make this a major challenge (Wishart, 1995). In the foreseeable future there will be no medical treatment for Down's syndrome other than the improved detection and termination of affected fetuses. Despite the concerns of some, new prenatal testing technologies are developing apace. In a few years at most, early diagnostic testing of chromosomal abnormalities will be possible via the isolation of fetal cells in maternal blood. Non-invasive diagnostic testing is seen as the 'holy-grail' of prenatal test development as it will remove the threat of miscarriage, which is a major barrier to

acceptance of amniocentesis or CVS (Zamerowksi *et al.*, 2001). However, the risk of miscarriage is also used as a justification for rationing an expensive procedure. Attitudes toward Down's syndrome will also continue to evolve and change although this will undoubtedly occur at a slower pace (Rees, Spreen, and Hamadek, 1991). It is difficult to predict how such changes in attitude will manifest, and whether technological advances will influence them, or vice versa (Pessione, 2001). At the same time, it is likely that social and educational opportunities for people with Down's syndrome will continue to develop with further steps being taken towards their inclusion in society. This could make the apparent paradox of the offer of prenatal testing and termination for Down's syndrome even more acute for some. Without a determined attempt on behalf of service providers and policy makers the outlook for facilitating informed prenatal testing choices for Down's syndrome does not look promising.

It is argued that there is a need to move the informed choice debate forward. A focus on the purely psychological factors situates responsibility for testing decisions within the context of the individual woman and the communication of the right information. This ignores the major role of a clinical context that embraces biomedical developments unproblematically with relatively little thought about the impact of these developments on society or the individual. Equally, to conclude that the dissemination of testing is purely an example of technological determinism deprecates the potential for autonomy in the individual, and the complex and emotionally difficult decisions that some people make. A psycho-sociological explanation of the factors that predict and inform prenatal testing choices is missing from the literature. Without this, attempts to facilitate informed choice will continue to take a piecemeal approach and significant advances in understanding and supporting informed choice could fail to develop in time for many parents whose interests testing aims to serve.

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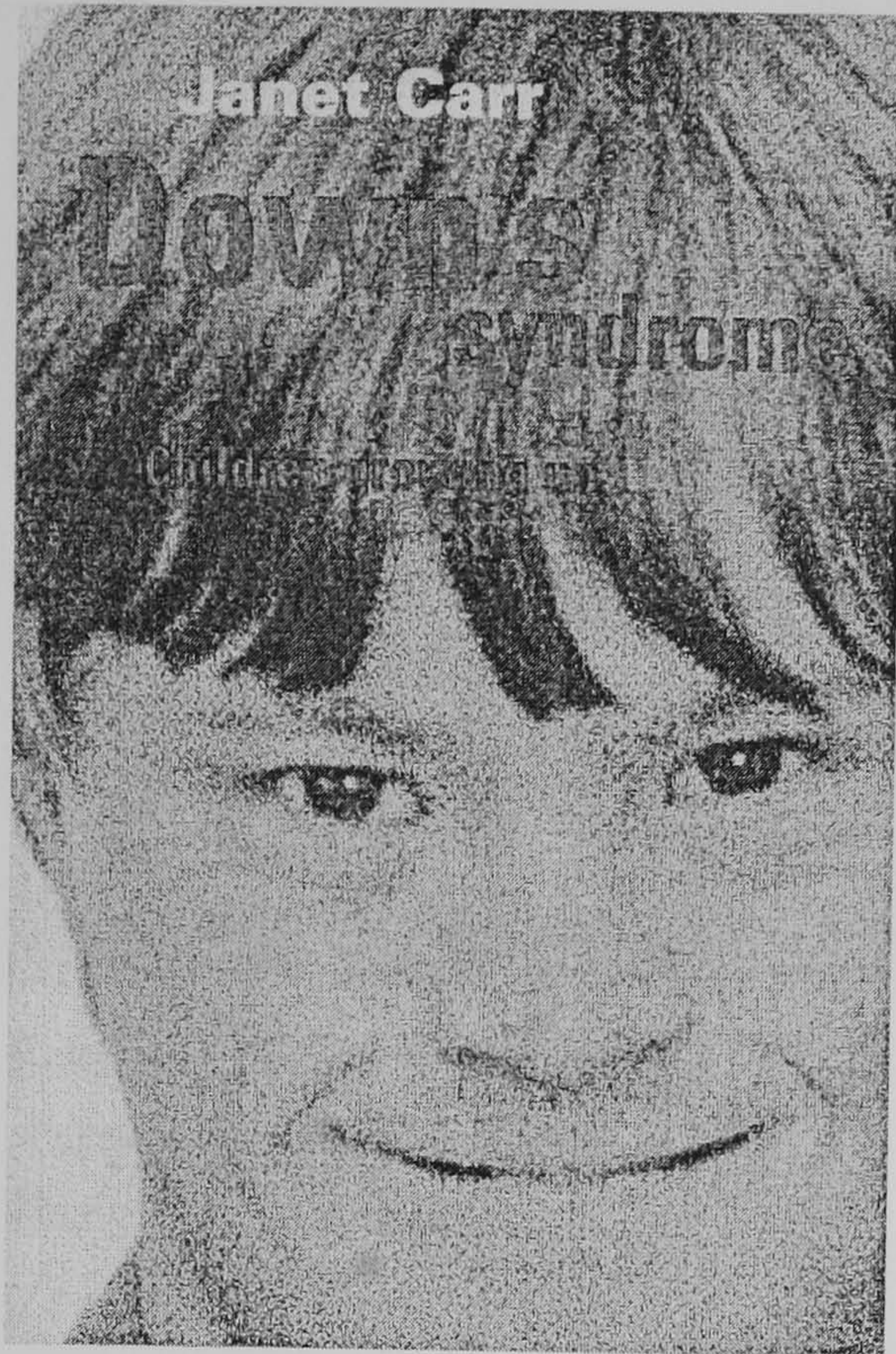
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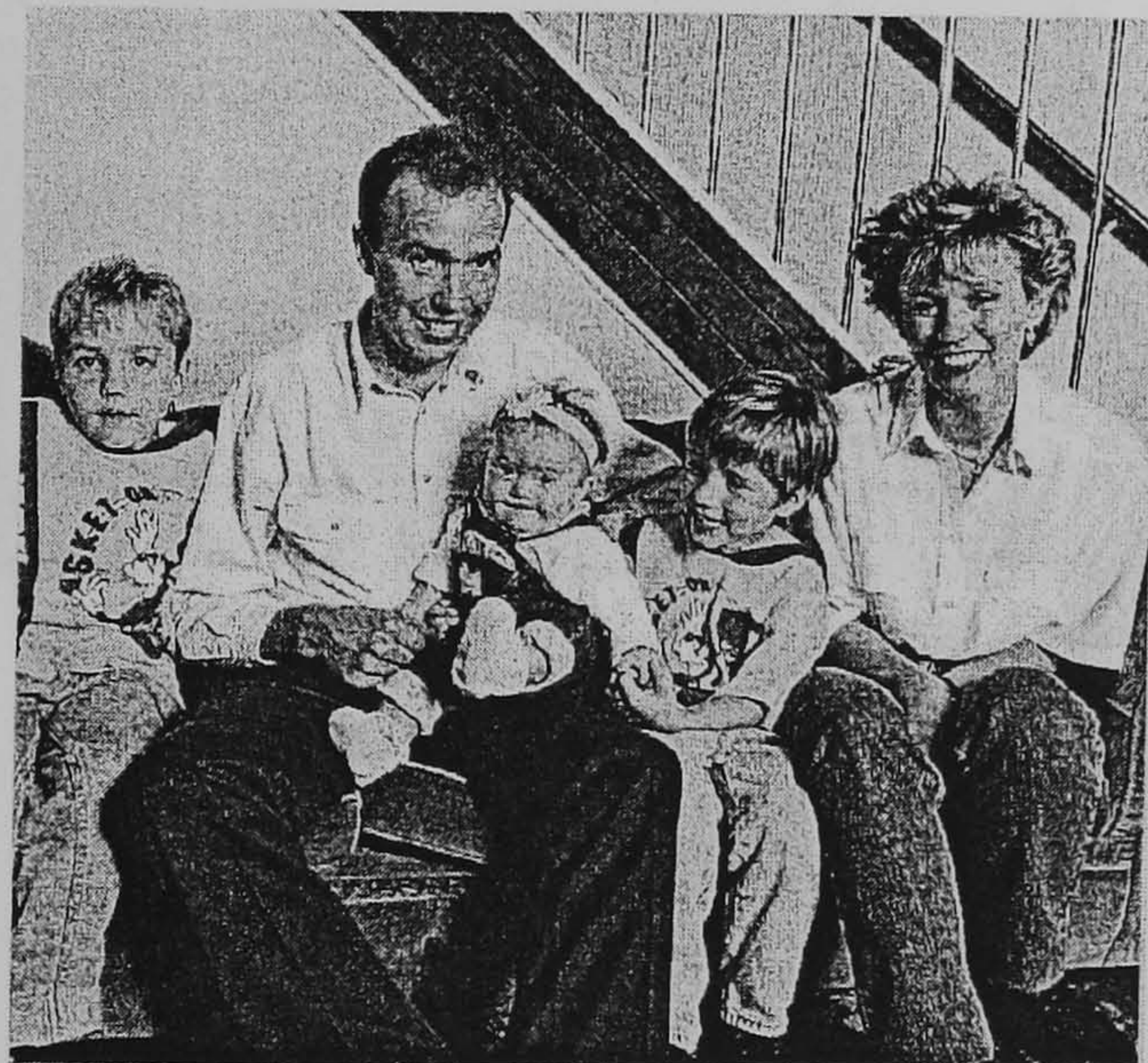
Appendix 1: Q study: photographs used in the focus group



“Freddie”. Source: Carr (1995)



Source: Clarke, N (1998). Should this girl have been given plastic surgery to disguise her Down's syndrome? *Daily Mail*, November 17, pg. 40-41

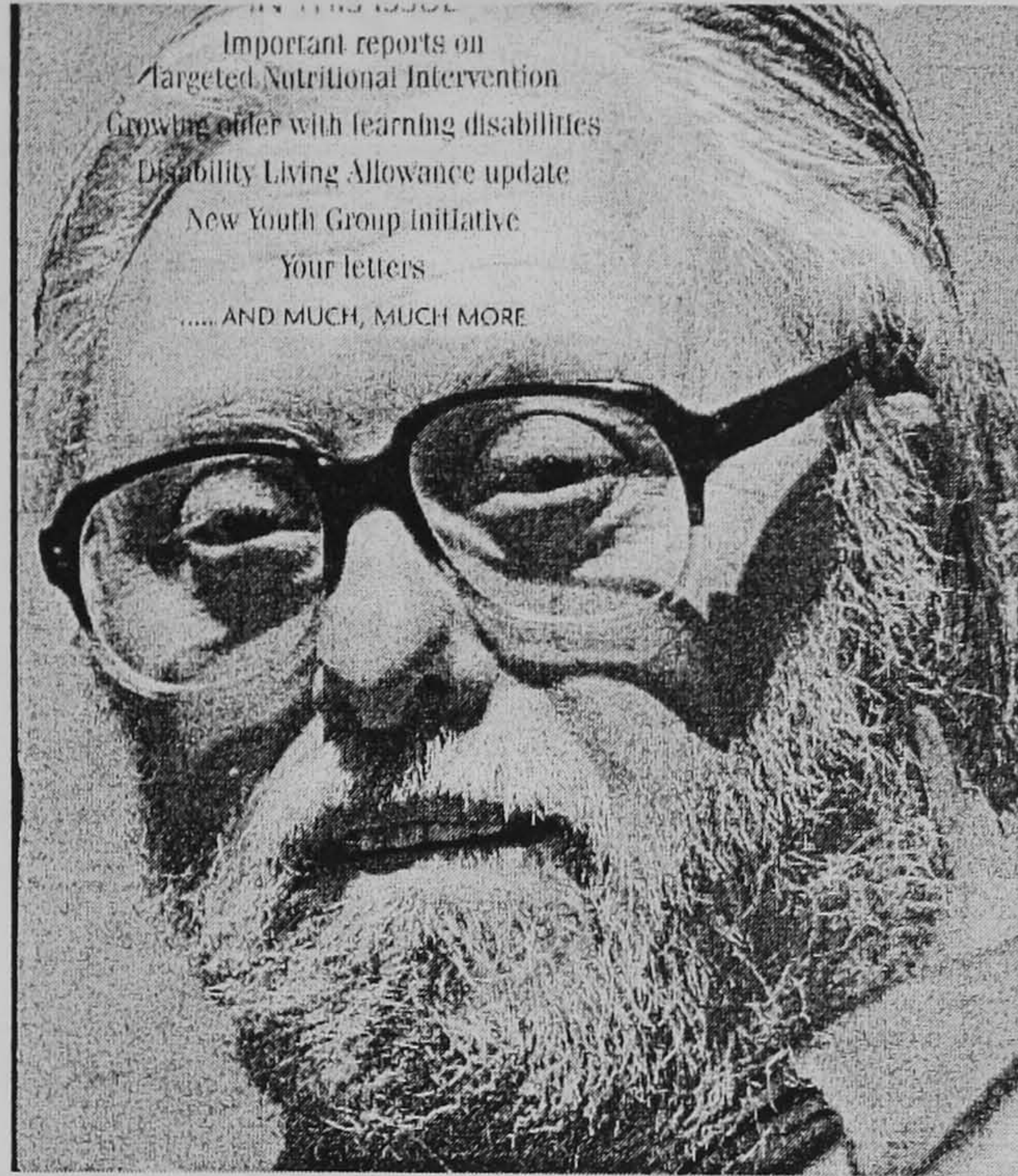


Source: Down's Syndrome Association (1997). *Your Baby Has Down's Syndrome*. London, DSA



Source: *Down's Syndrome Association Newsletter*, (1998) Issue 89, Winter

Appendix 1 continued.



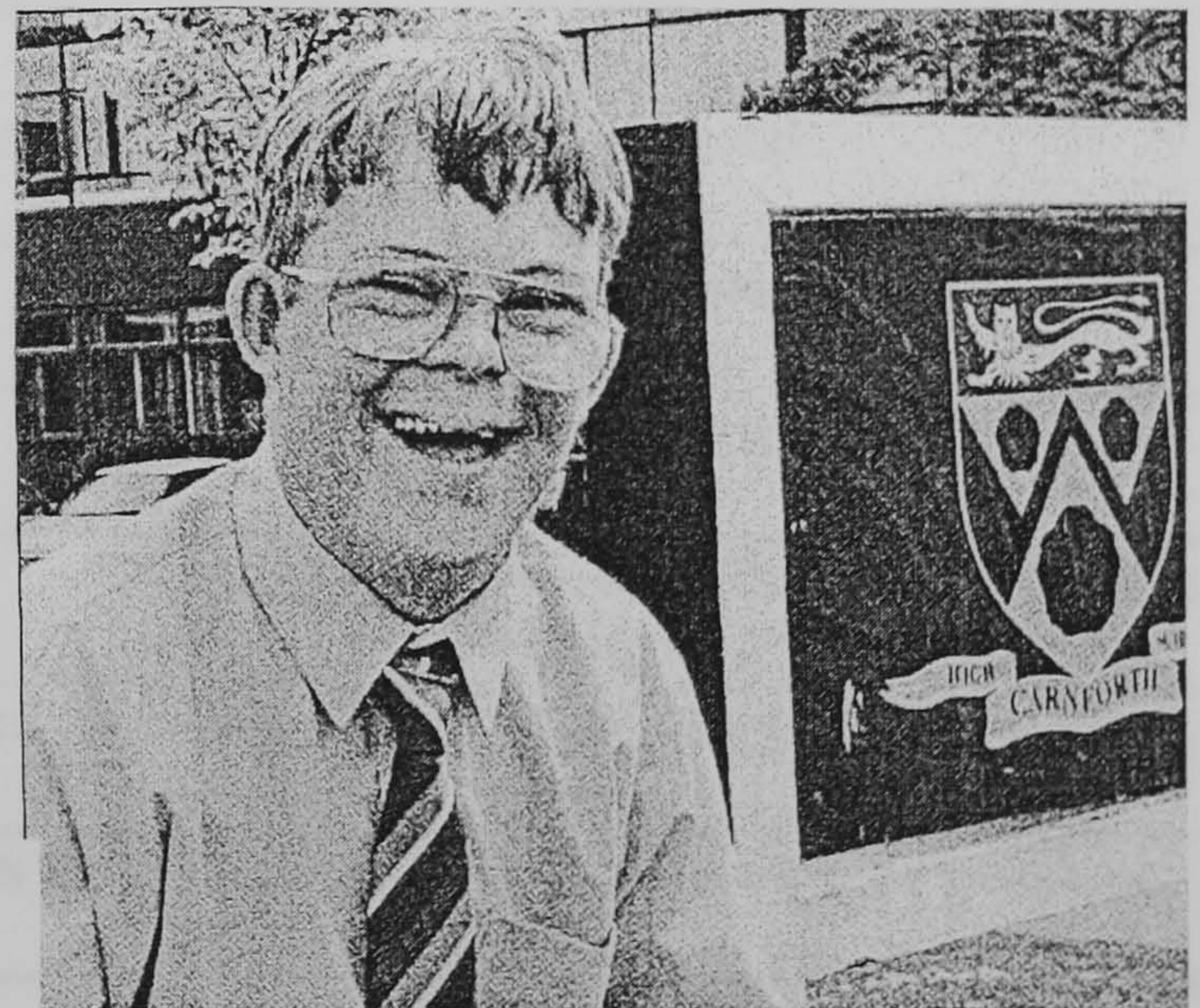
Source: *Down's Syndrome Association Newsletter* (1998)
 Issue 88, Summer.



Source: *Down's Syndrome Association Newsletter* (1998),
 Issue 88, Summer.



Source: Down's Syndrome Association
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Source: *Down's Syndrome Association Newsletter*, (1997)
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Appendix 2. Q study: the Q set, fifty statements about Down's syndrome

#	Statement	Category	Source
1.	Children with Down's syndrome can achieve a great deal.	Achievement	Discussion group
2.	You can be as proud of a child with Down's syndrome as you can be of any child.	Achievement	Information booklet for parents (Brinkworth and Collins, 1973)
3.	If you have a baby with Down's syndrome it may be better to have it adopted and try again.	Adoption	Research report (Rutter and Seyman, 1999)
4.	A child with Down's syndrome is a family tragedy.	Affect on family	Research article (Williams, 1995)
5.	The normal siblings of children with Down's syndrome suffer as well.	Affect on family	Booklet on sibling experiences (Fairbrother, 1988)
6.	A problem with children with Down's syndrome is that they will probably outlive their parents.	Aging	Research report (Rutter and Seyman, 1999)
7.	It's not right to submit a child with Down's syndrome to cosmetic surgery, they should be accepted the way they are.	Appearance	Focus group
8.	I find people with Down's syndrome rather unattractive.	Appearance	Research article (Kerr, Cunningham-Burley, and Amos, 1998)
9.	If you have a child with Down's syndrome it is because God chose you.	Attribution	Book on parental experience (Merriman, 1999)
10.	If a child with Down's syndrome died, it might be a blessing.	Better if died	Research report (Rutter and Seyman, 1999)
11.	Children with Down's syndrome are a burden throughout their lives.	Burden/Coping	Focus group
12.	Normal children are just as demanding as children with Down's syndrome.	Burden/Coping	Focus group
13.	Nobody would choose to have a child with Down's syndrome.	Choice	Discussion group
14.	Choosing to bring a child with Down's syndrome into the world is just selfish.	Choice	Book on parental experience (Boston, 1994)
15.	People with Down's syndrome are a financial burden on the state.	Cost to society	Focus group
16.	A person with Down's syndrome will always be totally dependent on others.	Dependence	Research article (Williams, 1995)
17.	People with Down's syndrome remain like children all their lives.	Dependence	Focus group
18.	For people with Down's syndrome, the biggest obstacle is not their learning disability but the attitudes of others.	Discrimination	Information leaflet (Scottish Down's Syndrome Association, 1998)
19.	If I had a child with Down's syndrome I would be worried about people staring at us.	Embarrassment	Research article (Sjögren and Uddenberg, 1987)
20.	Knowing someone with Down's syndrome enriches our understanding of what it is to be human.	Enrich society	Book on parental experience (Boston, 1994)

Appendix 2 continued.

#	Statement	Category	Source
21.	Down's syndrome is an abnormality and an error of nature. It makes sense to try and prevent it.	Eugenics	Research article (Williams, 1995)
22.	I think that euthanasia of babies with Down's syndrome is acceptable if that is what the parents want.	Euthanasia	Research article (Shepperdson, 1983)
23.	People with Down's syndrome make me feel uncomfortable.	Fear	Research report (Sinson, 1985)
24.	People with Down's syndrome have the same feelings as anybody else.	Feelings	Research report (Sinson, 1985)
25.	The world would be a worse place if no more babies with Down's syndrome were born.	Future world	Interview statement (reversed)
26.	People with Down's syndrome give as well as receive love.	Giving to others	Dissertation (Bryant, 1998)
27.	It is wrong to treat people with Down's syndrome as a group. They are all individuals.	Individual	Research report (Rutter and Seyman, 1999)
28.	I would find it as easy to love a child with Down's syndrome as to love any other child.	Loving a child	Research article (Sjögren and Uddenberg, 1987)
29.	I think you are lucky if you have a person with Down's syndrome in your family.	Lucky	Dissertation (Bryant, 1998)
30.	People with Down's syndrome should have the same health care as any other person even if that means an expensive operation.	Health	Research report (Rutter and Seyman, 1999)
31.	I wouldn't call Down's syndrome a major health problem.	Health	Research report (Rutter and Seyman, 1999)
32.	The medical profession paints an overly gloomy picture of what it is like to have a child with Down's syndrome.	Medics	Dissertation (Bryant, 1998)
33.	Having to say 'Down's syndrome' instead of Mongol, is just another example of political correctness.	Mongol	Research report (Rutter and Seyman, 1999)
34.	Saying that having a child with Down's syndrome is as good as a normal child is just denying reality.	Normal vs abnormal	Interview
35.	For me, having a child with Down's syndrome wouldn't be the end of the world.	Parental distress	Research article (Marteau <i>et al.</i> , 1993)
36.	A child with Down's syndrome must bring continual sorrow to its parents.	Parental distress	Booklet on sibling experiences (Fairbrother, 1988)
37.	People with Down's syndrome shouldn't be called 'sufferers'.	Suffering	Research report (Rutter and Seyman, 1999)
38.	I feel so sorry for people who have a baby with Down's syndrome.	Pity for parents	Research report (Rutter and Seyman, 1999)
39.	It must be awful to have Down's syndrome.	Pity for person	Interview
40.	You would get a lot of joy from having a child with Down's syndrome.	Joy	Focus group

Appendix 2 continued.

#	Statement	Category	Source
41.	People with Down's syndrome can live very happy lives.	Quality of life	Information leaflet (Scottish Down's Syndrome Association, 1998)
42.	People with Down's syndrome can have as good a quality of life as everyone else.	Quality of life	Information leaflet (Scottish Down's Syndrome Association, 1998)
43.	People with Down's syndrome have a right to be heard within society, especially when it comes to decisions that affect them.	Rights of people with DS	Information leaflet (Scottish Down's Syndrome Association, 1998)
44.	A family with a child with Down's syndrome is just like any other family.	Special parents	Booklet on sibling experiences (Fairbrother, 1988)
45.	Looking after a child with Down's syndrome needs certain qualities I don't think I've got.	Special parents	Magazine article (Allott, 1997)
46.	I think mixing children with Down's syndrome into ordinary schools is a good thing.	Education	Interview
47.	People with Down's syndrome are just a bit different from other people.	Seriousness	Interview statement (reversed)
48.	People with Down's syndrome are severely mentally disabled.	Seriousness	Information leaflet (Down's Syndrome Association, 1998)
49.	People with Down's syndrome should be allowed to have a normal sex life like everyone else.	Sex	Focus group
50.	People with Down's syndrome should mix together with other people as much as possible.	Social integration	Information booklet for parents (Brinkworth and Collins, 1973)

Appendix 3. Q study: participant details

Q#	Age	Biographical details	Factor
1	24	Female postgraduate student, no children	Mixed
2	64	Mother of an adult son with Down's syndrome, three children	1
3	45	Female computer programmer, no children	2
4	50	Female researcher in psychology, two children	3
5	23	Female postgraduate student, no children	Mixed
6	32	Male genetic counsellor, three children	1
7	32	Female postgraduate student, no children	1
8	34	Female postgraduate student, no children, sibling of man with DS	1 ^s
9	41	Female clerk, one child (pregnancy via IVF treatment)	Mixed
10	30	Female postgraduate student, one child	1
11	31	Female postgraduate student, no children	Mixed
12	31	Female postgraduate student, three children	1
13	42	Male manager of housing scheme for adults with LD, no children	4
14	48	Female parent counsellor at a nursery for children with LD, no children	1
15	33	Male postgraduate student, no children	Mixed
16	28	Male computing manager, no children	1
17	80	Father of a woman with severe learning difficulties, one child	Mixed
18	33	Female computer professional, two children	1
19	33	Male, sociology researcher with special interest in disability issues	1
20	40	Female secretary, no children	2
21	29	Male postgraduate student, no children	2
22	22	Female clinical psychologist working with adults with LD, no children	1
23	37	Female archaeologist, one child, sibling of person with DS	1
24	52	Male medical researcher/specialist in prenatal screening, two children	2
25	55	Teacher, mother of adult daughter with DS, two children	1
26	54	Female manager of care home for adults with LD, two children	1
27	41	Female manager of support organisation for adults with LD no children	1
28	51	Male manager of support organisation for adults with LD, four children	1
29	24	Female postgraduate student, no children	1
30	47	Female researcher in area of fetal abnormalities, two children	4
31	24	Female clinical psychology trainee, no children	1

32	36	Male senior lecturer and consultant obstetrician, two children	1
33	29	Male postgraduate student, no children	Mixed
34	44	Male consultant obstetrician, two children	1*
35	42	Female general practitioner, four children	2
36	36	Male researcher and lecturer in disability studies, children	1
37	45	Male health care researcher, no children	4
38	53	Mother of son with Down's syndrome, four children	1
39	25	Female researcher and biologist, no children	Mixed
40	26	Female postgraduate student, no children	1
41	27	Male postgraduate student, no children	2
42	26	Female postgraduate student, no children	1
43	27	Male researcher and psychologist, no children	1
44	26	Male postgraduate student, no children	Mixed
45	32	Female postgraduate student, no children	1
46	27	Female midwifery student and nurse (pregnant with first baby)	Mixed
47	37	Female computer analyst programmer, two children	Mixed
48	41	Male computing manager, two children	Mixed
49	40	Female midwife, children	1
50	43	Male computer systems analyst, no children	Mixed
51	43	Male computer programmer, no children	Mixed
52	37	Female student, two children	2
53	33	Female teacher at school for children with LD, no children	Mixed
54	43	Male computer analyst programmer, two children	2
55	53	Female computer programmer, aunt of woman with DS, two children	1
56	41	Female medical laboratory assistant, children	1
57	38	Male cytogeneticist, children	Mixed
58	32	Male medical scientist	5
59	45	Male clinical cytogeneticist	1
60	35	Female cardiac nursing specialist, one child	5
61	32	Female medical doctor and postgraduate student, no children	1
62	34	Male warehouse manager, one child	Mixed
63	50	Male computer programmer, no children	1

Appendix 3 continued.

64	38	Female probation officer, two children	1
65	35	Housewife, two children	2
66	36	Female pharmacist, three children	1
67	38	Female teacher, four children	1
68	43	Female genetic counsellor, no children	2
69	30	Male hospital haematologist, one child	Mixed
70	23	Female youth support worker, no children	1
71	33	Male engineer, two children	3
72	40	Teacher and mother of adopted son with DS, three children	1
73	39	Female clinical cytogeneticist, children	3
74	26	Female clinical cytogeneticist, no children	1
75	23	Female clinical cytogeneticist, no children	Mixed
76	34	Male scientific officer, children	Mixed

§ = the researcher, * = negative loading, LD = Learning difficulty

Appendix 4. Q study: questionnaire ‘Testing in Pregnancy for Down’s syndrome’

Your views on prenatal diagnosis

1. Many women now have to make important decisions about whether or not to be tested in pregnancy for Down’s syndrome, and different people hold different views. Can you please indicate how strongly you agree or disagree with the statement that “*prenatal diagnosis for Down’s syndrome should be freely available for everybody*”

Strongly disagree 1 2 3 4 5 6 7 Strongly agree

2. This question asks what you think you would do if you (or your partner) were pregnant and were told that there was a **higher than normal risk** that your pregnancy was affected by Down’s syndrome. Even if you think that this situation is very unlikely to affect you, could you please answer the question as best you can.

I would definitely **not** have prenatal diagnosis 1 2 3 4 5 6 7 I would definitely have prenatal diagnosis

Your views on selective termination for Down’ syndrome

3. Abortion is a subject that many people feel strongly about. I am interested to know what your attitudes towards terminating a pregnancy affected by Down’s syndrome might be, allowing for the fact that in real-life situations, such decisions are often not straightforward. Can you please indicate how strongly you agree or disagree with the statement that “*termination for Down’s syndrome should be freely available for everybody*”

Strongly disagree 1 2 3 4 5 6 7 Strongly agree

4. This question asks what you think **you** would do if you (or your partner) were pregnant and were told that your pregnancy was affected by Down’s syndrome. Even if this situation is very unlikely to affect you, could you please answer the question as best you can.

I would definitely **not** have a termination 1 2 3 4 5 6 7 I would definitely have a termination

Question 3: What things are important to you in your life?

What are the things that you value in your life (for example, your career, your family), or what aspirations do you consider very important to you (for example becoming wealthy)? In the left-hand column of the box below, please list these valued things or aspirations. For each one then indicate whether you think these things would be positively affected, negatively affected or not altered by having a child with Down's syndrome, by placing a tick in one of the columns. You can also list values or aspirations that you think other people may hold.

Valued object or aspiration	Positively affected	Not altered	Negatively affected
Example: <i>my career</i>		✓	

If you have any comments about this exercise please write them here. Thank you for your time.

Appendix 6. Attitudes towards Down's syndrome in the prenatal testing situation: Main study questionnaire

(see over)

PLEASE READ THESE NOTES FIRST!

Thank you for taking the time to help me with my research. I am interested in anything that you would like to say about this topic so feel free to write on the questionnaire at any point. There is also a space at the end that you can use for this if you want.

Questionnaire

Testing in Pregnancy for Down's syndrome

Some questions will ask you to tick the box or boxes that match up best with your situation; some questions will ask you to circle a number that best matches your point of view. Some questions will ask you to write a short answer in the space provided.

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I know that this is a sensitive subject and that some of the questions may seem noseey, but please try and answer all the questions. However, if you do not want to answer anything remember that you do not have to. You do not have to fill in the questionnaire all in one go. **Be as honest as you can as your answers will be kept private.**

Please fill in and return the questionnaire to me as soon as you can but before your next antenatal clinic appointment.

Louise Bryant
University of Leeds

SECTION A: Tests for Down's syndrome in pregnancy

Before you start, I would like to make sure that you understand some of the terms used, as this will make the questions more clear.

There are two main types of tests in pregnancy that are used for checking if a baby has problems. These are **Screening tests** and **Diagnostic tests**.

Screening tests can only estimate the chance that the baby has Down's syndrome and do not say for certain whether the baby has it or not. The most common screening tests are called serum screening tests. These include the Triple Test, which is offered at Hull Maternity Hospital. These tests use chemicals in the mother's blood to work out the chances of the baby having Down's syndrome. The tests are done at around 15-18 weeks of pregnancy. Having this test does not carry any risk to the baby or the mother.

1. Have you heard of these blood tests before? **Please circle one number.**

Yes 1 No 2

Diagnostic tests can say almost for certain if a baby has Down's syndrome or not. The most usual test is called amniocentesis. In this test a small amount of the liquid around the baby is taken from the mother. This test is done at about 16-18 weeks of pregnancy, sometimes as a result of a screening test, sometimes for other reasons. There is a chance that a miscarriage will happen in around 1 in every 100 amniocenteses carried out.

2. Have you heard of amniocentesis before? **Please circle one number.**

Yes 1 No 2

Tests can be for a number of conditions and problems but I am just interested in tests for **Down's syndrome** in this questionnaire.

3. Have you heard of Down's syndrome before? Please circle one number.

Yes 1 No 2

SECTION B: Your experiences of pregnancy

4. How many weeks pregnant are you at the moment?
5. How many pregnancies have you had, including this one?

If this is your first pregnancy go to question 9. If this is not your first pregnancy please answer question 6.

6. In any of your previous pregnancies, did you have tests for Down's syndrome? **Please circle one number.**

Yes 1 No 2 Not sure 3

If you answered 'Yes' please go to question 7. If you answered 'No' go to question 8. If you answered 'Not sure' go to question 9.

7. If you **did** have tests for Down's syndrome in a previous pregnancy can you say which test(s)?

Serum screening (e.g. the Triple Test) Tick

Amniocentesis

Other tests

Please say which.....

Can you tell me about why you had the tests for Down's syndrome?

8. If you **did not** have tests for Down's syndrome in any of your pregnancies, can you tell me about why you did not have them?

9. In this pregnancy how worried are you about the baby having Down's syndrome? **Please circle one number.**

- Not at all worried 1
- Slightly worried 2
- Fairly worried 3
- Extremely worried 4

10. How likely do you think you are to have a baby with Down's syndrome? **Please circle one number.**

- Not at all likely 1
- Slightly likely 2
- Quite likely 3
- Very likely 4

11. On a scale of 1 to 9, could you say how good or bad it would be for you if you were to have a baby with Down's syndrome?
Please circle one number.

- | | | | | | | | | | |
|------------------|---|---|---|---------------------|---|---|---|------------|------------------|
| extremely | | | | | | | | | extremely |
| good | | | | neither good | | | | bad | |
| 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | |

SECTION C: Your views on testing in pregnancy for Down's syndrome

Your views on screening tests (e.g. the Triple Test)

12. Many women now have to make a choice about whether or not to have a screening test in pregnancy. Please say how strongly you agree or disagree with the statement that;

"Screening tests for Down's syndrome should be available for every pregnant woman who wants one". Please circle one number.

- Agree 1
- Slightly agree 2
- Don't know 3
- Slightly disagree 4
- Disagree 5

13. What do you think you would do at the moment if you were offered a screening test for Down's syndrome (e.g. a Triple Test)?
Please circle one number.

- I would definitely have a screening test 1
- I would probably have a screening test 2
- Don't know at the moment 3
- I would probably not have a screening test 4
- I would definitely not have a screening test 5

Could you briefly give the reason for your answer to question 13?

Your views on diagnostic tests (e.g. amniocentesis)

14. Some women have to make important decisions about whether or not to have a diagnostic test when they are pregnant, sometimes because of the result of their screening test. Can you please say how strongly you agree or disagree with the statement that;

“Diagnostic tests for Down's syndrome should be available for every pregnant woman who wants one.” Please circle one number.

- Agree 1
- Slightly agree 2
- Don't know 3
- Slightly disagree 4
- Disagree 5

15. What do you think you would do at the moment if you were offered a diagnostic test (e.g. an amniocentesis)? This could be because of the result of a screening test or for some other reason. Even if you think that this is very unlikely to happen to you, could you please answer the question as best you can. **Please circle one number.**

- I would definitely have a diagnostic test 1
- I would probably have a diagnostic test 2
- Don't know at the moment 3
- I would probably not have a diagnostic test 4
- I would definitely not have a diagnostic test 5

Could you briefly give the reason for your answer to question 15?

Your views on termination

16. Abortion is a subject that many people feel strongly about. Please say how strongly you agree or disagree with the statement that;

“A termination for Down’s syndrome should be available for everybody who wants one”. *Please circle one number.*

Agree 1

Slightly agree 2

Don’t know 3

Slightly disagree 4

Disagree 5

17. What do you think you would do if after having a diagnostic test you were told that your baby had Down’s syndrome? Even if this is very unlikely to happen to you, please try and answer the question as best you can. **Please circle one number.**

I would definitely have a termination 1

I would probably have a termination 2

Don’t know at the moment 3

I would probably not have a termination 4

I would definitely not have a termination 5

Could you briefly give the reason for your answer to question 17?

18. Could you say whether or not you think you would terminate a pregnancy for the following reasons? **Please circle one number for each statement**

Would you terminate if.....? Yes No Don’t know

You became pregnant through rape 1 2 3

Your health was in severe danger 1 2 3

You were on a low income, and could not afford a child 1 2 3

You were **not** in a stable relationship and didn’t want the baby. 1 2 3

You were in a stable relationship and didn’t want the baby 1 2 3

SECTION D: A few questions about you

It is helpful to know that I have the views of women in different situations. I hope that you do not mind giving me the following information.

19. What is your date of birth?

20. Are you still at school, college or university? **Please circle one number.**

Yes 1 No 2

21. If you answered 'No' to question 20, how old were you when you finished your education?

22. Are you? (Please circle one number that best matches your situation)

- Single with no partner 1
- In a relationship but not living together 2
- Married or living with your partner 3
- Divorced or separated 4
- Widowed 5

23. How would you describe your ethnic group or origin? (For example, Chinese, Black, Pakistani, White etc.)

24. What is your religion? (If you have no religion please write 'none')

25. Does your religion or religious upbringing affect the sorts of decisions that you make about your life?

- Not at all 1
- A little 2
- Quite a lot 3
- Completely 4
- Not applicable 5

Section E. Your thoughts and feelings about Down's syndrome

The questions in this last section have a different style from those in the rest of the questionnaire. Try and answer all the questions as best you can.

Please be as honest as you can because your answers will be kept completely private.

26. Your experiences of people with Down's syndrome

Have you ever had any experiences of, or with, people with Down's syndrome? Try to remember these for a moment.

These might have happened a while ago or recently. You might have seen a programme on TV about it, or have read about Down's syndrome in a magazine. You might have seen somebody with Down's syndrome in the street when you were a child. You may know someone with Down's syndrome. You may have worked with someone with Down's syndrome.

In Box 1, in the column marked 'A', write a bit about any experience you have had (see the example below). Write down as many or as few experiences as you want to, but especially those that stand out in your mind most strongly.

Box 1. My experiences of people with Down's syndrome

	B
<i>Example:</i>	
<i>I saw a programme on TV about Down's syndrome.</i>	

NOW...

Think about each of the experiences you have written in Box 1.

- If it was a positive one (e.g. good or happy) put a plus sign (+) in the last column of Box 1 marked 'B'. If it was very positive put two pluses (+ +) e.g.

<i>I saw a programme on TV about Down's syndrome</i>	++
--	----

- If it was a negative one (e.g. bad or unhappy) put a minus sign (-) in column B. If it was very negative put two minuses (- -) e.g.

<i>I saw a programme on TV about Down's syndrome</i>	-
--	---

- If it was neither negative nor positive put a zero (0) in column B, e.g.

<i>I saw a programme on TV about Down's syndrome</i>	0
--	---

- If it was both a positive and a negative experience put a plus and a minus sign (+ -) in column B, e.g.

<i>I saw a programme on TV about Down's syndrome</i>	+ -
--	-----

27. How would you describe a person with Down's syndrome?

What words or phrases come into your head when you think about a person with Down's syndrome? Try to picture a person with Down's syndrome in your mind for a moment if you can.

In Box 2, column A write down the words or phrases that you think best describe your picture of a person with Down's syndrome. Write as many or as few as you want. If you like you could use some of the examples given at the bottom of the next page.

A	B

NOW...

Think about each of the words or phrases you have written in Box 2. For each one, do you think it is a positive or a good thing to be described as, or is it a negative or a bad thing to be described as?

- If you think the word or phrase is positive put a plus sign (+) in column B. If it is very positive put two pluses (++).
- If you think it is negative put a minus sign (-) in column B. If it is very negative put two minuses (--).
- If you think it is neither negative nor positive put a zero (0) in column B.
- If you think it is both positive and negative put a plus and a minus sign (+ -) in column B.

Some examples:

friendly	difficult	innocent	aggressive
capable	attractive	huggy	short
different	slow	independent	healthy
demanding	emotional	loving	stubborn
childish	unhealthy	vulnerable	kind
clumsy	happy	ordinary	trusting
dependent	fun	noisy	unattractive
Plump	learning problems	uninhibited	good-natured

You have now finished - thank you!

Please check that you have answered each question as best you can. Then send the questionnaire back to me with the signed consent form using the prepaid envelope. Please return this as soon as you can but **BEFORE YOUR NEXT ANTENATAL CLINIC APPOINTMENT.**

If you are at all worried by anything raised by this study, please tell your doctor or midwife. Your midwife can be reached at Hull Maternity Hospital on 01482-675468 during the day.

If you would like to make any comments about this questionnaire or about testing in pregnancy please use this space.

**Appendix 7. Attitudes towards Down's syndrome in the prenatal testing situation:
information sheet and consent form**

INFORMATION SHEET. Testing in Pregnancy for Down's syndrome

Most pregnant women are now offered testing for Down's syndrome as part of their antenatal care. This study is looking at how women make choices about testing and their views about Down's syndrome. This should help us know more about the needs of pregnant women. I would like to invite you to take part in this research. I have tried to answer some of the questions you might have about the study, but if you have any others please ask your midwife or ring me on the number at the bottom of the page.

"What will I have to do?"

You are asked to fill in a questionnaire. This has some questions about you, your pregnancy and your views on testing for Down's syndrome. It also asks about any experiences of people with Down's syndrome you may have had. The questionnaire is for you to take home to fill in. You will be given a pre-paid envelope to send the questionnaire back to me (not the clinic).

"How long will it take?"

The questionnaire will take about 15-20 minutes to fill in. You do not have to fill it in all in one go.

"Will my answers remain private?"

Yes. Your name will not be written on the questionnaire only on the consent form, which will be kept separate. Only I will be able to read your questionnaire. Your midwife or doctor will not be able to read your answers.

"What if I do not want to take part?"

If you think that taking part in the study will upset you for any reason, you do not have to complete the questionnaire. If you decide later on that you no longer want to take part, you can tell your midwife and your details will be removed from the study. **Your antenatal care will not be affected in any way.**

"What do I do now?"

If you would like to help me with this research, please fill in the 'Consent Form', and then fill in the questionnaire. Then send both of them back to me in the prepaid envelope. If you do not want to take part in the study, you have nothing else to do.

Thank you very much for your time.

Louise Bryant, University of Leeds, Telephone: 0113-2336697

The following information may be helpful to you:

The **Down's Syndrome Association** has up to date information on testing for Down's syndrome and about the condition itself.

Down's Syndrome Association, 55 Mitcham Road, London SW17 9PG.

Tel: 0181 682 4001. Web page: www.downs-syndrome.org.uk

ARC (Antenatal Results and Choices) offers help to women thinking about, or undergoing tests or termination for conditions such as Down's syndrome.

ARC, 73-75 Charlotte Street, London W1P 1LB

Tel. 0171 631 0280, email: arcsatfa@aol.com

Appendix 7 continued.

CONSENT FORMTesting in Pregnancy for Down's syndrome

Filling in and signing this form shows that you are happy to take part in the study and understand what you are asked to do. It does not mean that you have to do anything that you do not want to.

Please write your initials in the box

1. I have read and understood the information sheet for the above study.

2. I understand that my helping with this study is voluntary and that I am free to pull out **at any time** and without having to give a reason for doing so.

As part of this study you are asked whether you *think* you would have testing for Down's syndrome in pregnancy or not. It would be very helpful for the researcher to be able to match up your early thoughts with the actual choices that you make in the future. **However, the Trust must have your permission to pass this information on.** This information would be kept completely private and *only* information about whether you had tests or not would be passed on. Your name and address details would not be given to the researcher.

3. I give permission for the researcher to be given information about whether or not I choose to have tests for Down's syndrome in the future.

4. I agree to take part in the study (you can still take part even if you do not want your test choice information passed on).

Please fill in the following details:

Your name in capital letters _____ Your date of birth _____

Your consultant (his name will be on your notes, e.g. Mr Hay) _____

Your signature _____ Today's date _____

****Please fill in this form and send it in with your completed questionnaire, thank you.****