

**A COMPARISON OF ATTITUDES TOWARDS PRENATAL DIAGNOSIS AND PRE-
IMPLANTATION GENETIC DIAGNOSIS**

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ABSTRACT

Technological advances in prenatal screening and diagnosis mean that it is now possible to test for a wide range of congenital conditions (Hewison et al., 2007). Traditionally testing has been carried out during pregnancy (prenatal diagnosis, PND). However, advances in technology have made it possible for diagnosis of an embryo created through in vitro fertilisation, prior to implantation into the womb (pre-implantation genetic diagnosis, PGD). This means that women can avoid the birth of a child with a genetic condition without the stress of terminating a pregnancy. This raises questions about what women want from reproductive technologies, as it means they are making decisions based not only on the condition diagnosed but also on the technology used to test.

Two studies were carried out to examine this further. In the first study, 216 participants completed a questionnaire either based on PND or PGD. Participants were asked whether they would terminate a pregnancy (PND condition) or avoid implantation (PGD condition) following diagnosis of five different genetic conditions, ranging in severity.

The results suggest an interaction between the technology (PND or PGD) and the severity of the genetic condition diagnosed, such that for the most and least severe conditions, the number of people choosing to terminate/avoid implantation was similar for the PND and PGD groups. However for conditions in the middle range of severity significantly more people said they would avoid implantation. A within subjects interview study was carried out to explore this further and thematic analysis identified a number of themes that influenced participants' responses.

Overall, the results suggest that PGD may be more acceptable for women in some cases. Women considering diagnoses are likely to benefit from detailed information about both PND and PGD in order to make a fully informed decision as to which is best for them.

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ABBREVIATIONS

CVS:	Chorionic villus sampling
DMD:	Duchenne muscular dystrophy
HFEA:	Human Fertilisation and Embryology Authority
IVF:	In vitro fertilisation
PGD	Pre-implantation genetic diagnosis
PKU:	Phenylketonuria
PND:	Prenatal diagnosis
NHS:	National Health Service
NICE:	National Institute for Health and Clinical Excellence
TPB:	Theory of Planned Behaviour

CHAPTER 1: INTRODUCTION

1.1 Introduction

Technological advances in prenatal screening and diagnosis mean that it is now possible to test for a wide range of congenital conditions (Hawkey, 2005; Hewison et al., 2007). Traditionally testing has been carried out during pregnancy (prenatal diagnosis, PND). Therefore until recently the only way to avoid the birth of a child with a genetic condition was to terminate the pregnancy following PND or avoid having children altogether. More recent advances in technology have made it possible for diagnosis of an embryo created through in vitro fertilisation (IVF), prior to implantation into the womb (pre-implantation genetic diagnosis, PGD). This raises important psychological questions about what women want from reproductive technologies as it is now possible for them to make decisions based on the technology used to test as well as the genetic condition diagnosed. This chapter describes the current literature on attitudes towards prenatal diagnosis (PND) and pre-implantation genetic diagnosis (PGD).

1.2 What are Prenatal diagnosis and Pre-implantation genetic diagnosis?

Prenatal diagnosis (PND) is the identification of congenital conditions during pregnancy. PND usually occurs following prenatal screening (PNS), where non-invasive methods such as ultrasonography and maternal serum screening are used in the first instance to indicate risk of the foetus having a congenital condition. If risk is indicated through PNS, other methods can be used in order to carry out a definitive prenatal diagnosis. Ultrasonography can be used to diagnose some conditions, involving structural abnormalities. However, two more commonly used methods of PND are amniocentesis and chorionic villus sampling (CVS). Amniocentesis involves a needle being inserted through the abdominal wall and into the amniotic sac of fluid surrounding the foetus. A syringe is then used to remove a small sample of amniotic fluid from the amniotic sac for analysis in a laboratory (NHS Direct, 2009a). Chorionic

villus sampling involves obtaining a small sample of the chorionic villi (placental tissues), either by passing a thin needle through the wall of the abdomen, or by passing a small tube through the vagina and the cervix (NHS Direct 2009b). Both procedures are carried out under the guidance of ultrasound scanning. CVS has the advantage that it is generally carried out earlier than amniocentesis (11-13 weeks rather than 15-18 weeks). However, it also carries higher risk of miscarriage (1-2% versus 1% loss rates) (National Screening Committee, 2010). Although earlier amniocentesis is possible, the risk of foetal loss is even greater than CVS (3-5%) (Cederholm & Axelsson, 1997). One of the advantages of having an earlier test (CVS) is that women have often felt foetal movements by the stage of amniocentesis, making termination much more difficult psychologically. Due to the reduced emotional and physical strain on couples, particularly those in high risk groups, CVS is increasing in popularity (Papp & Papp, 2003). If given a positive diagnosis through either procedure, women have the option of terminating the pregnancy.

For women at high risk of having a child with a genetic condition, pre-implantation genetic diagnosis (PGD) is an alternative to prenatal diagnosis. It involves screening an embryo created through in vitro fertilisation (IVF), prior to implantation into the womb. The IVF process initially involves taking fertility drugs to stimulate egg production. Eggs are then retrieved under the guidance of ultrasound. A needle is inserted through the vaginal wall into the ovaries and eggs are then collected through a special catheter. Following egg retrieval women are given hormones to prepare the uterus for pregnancy, while the eggs are fertilised with sperm in a laboratory (Human Fertilisation and Embryology Authority (HFEA, 2007a). Once the eggs have been fertilised PGD takes place and implantation of embryos with a positive diagnosis can be avoided. Until 2001 in the United Kingdom (UK), criteria for using PGD were broadly in line with those for abortion, which state, "that there is a substantial risk that if the child were born it would suffer from such physical or mental abnormalities as to be

seriously handicapped' (HFEA, 1990, chapter 37, p.3). However, in 2006 the HFEA agreed to consider the use of PGD for some adult onset conditions where there is clear family history of the condition (HFEA, 2006). More recently in 2007, the HFEA issued a licence to screen for the homozygous (more serious) form of familial hypercholesterolemia (HFEA, 2007b). In the UK currently over 100 conditions have been licensed by the HFEA (see HFEA, 2009a).

1.3 Current practice

It is Department of Health policy that all pregnant women should be offered routine prenatal screening. These tests do not give definite diagnoses but indicate risk and prenatal diagnosis is offered as standard to women if they have positive antenatal screening results. Some women may be offered PND regardless of antenatal screening for the following reasons:

- Family history of an inherited condition
- Previous pregnancy with foetal abnormality
- Exposure to viral illness during the pregnancy
- Exposure to teratogens during the pregnancy
- If the woman has type 1 diabetes, epilepsy or myotonic dystrophy.

Unlike PND, PGD is not widely available on the NHS and is considered on a case by case basis. The acceptable reasons for requesting PGD include people with chromosomal disorders and couples at risk of transmitting serious genetic disorders to their offspring. However, the clinical indications for PGD are widening and will continue to do so in the future. For example, some centres offering PGD suggest that it should be offered as a screening procedure to couples undergoing IVF for infertility (Department of Health, 2002).

1.4 Attitudes towards PND and selective abortion

Previous research has shown that women's decision to terminate following PND is linked to a number of factors. Mansfield, Hopfer, and Marteau (1999) carried out a

systematic review of termination rates after prenatal diagnosis of Down Syndrome, Spina Bifida, Anencephaly, and Turner and Klinefelter Syndromes. They found that termination rates remained stable from 1980 to 1998. They also found termination rates to vary according to condition. The largest proportions of terminations occurred following diagnosis of Down Syndrome and the least for Klinefelter Syndrome. The authors suggest that perceived severity of condition, timing of the diagnosis and information given to parents about the diagnosis affect decisions about termination.

Bell and Stoneman (2000) surveyed women to find out what they would do following PND for three conditions - Down Syndrome, Spina Bifida and Haemophilia. They found that women were most certain that they would continue with the pregnancy if the child had Haemophilia and least certain for Down Syndrome. The authors found modest support that negative attitude towards people with disabilities was associated with increased likelihood of abortion. They also found church attendance to be related to decisions concerning abortion for all three conditions.

Hewison et al. (2007) asked Pakistani and white women in the UK whether they would want a prenatal test and whether they would consider termination for 30 conditions. They found that overall, Pakistani women were less likely to consider termination than white women. However, Pakistani women were more in favour of prenatal testing than white women. Hewison et al. (2007) found that for more serious conditions such as Anencephaly and Duchenne Muscular Dystrophy, women considered termination even when they rejected the idea of termination on the whole. Multi Dimensional Scaling (MDS) was used to determine whether the conditions could be grouped in a meaningful way in order to inform the development of consent procedures. The MDS analysis revealed a 2-dimensional configuration. One dimension represented 'seriousness of the conditions'. However, the authors were unable to identify the second dimension. They concluded that there is too much individual

difference to be able to group conditions and that women should be given information about testing on an individual basis.

Hawkey (2005) carried out a study aimed at seeing if genetic conditions could be grouped. Overall, Hawkey (2005) found that women grouped conditions according to 'manageability' and 'lifespan'. However, as with the Hewison et al. (2007) study, there were individual differences in how women grouped the conditions again suggesting that they need to be given this information on an individual basis. In this study participants were not asked to consider termination. However, it is possible to hypothesise that women would be more likely to consider PND and termination for conditions they consider to shorten lifespan and/or be less manageable.

Other factors alongside women's attitudes towards conditions may influence their attitudes towards PND and termination. Research suggests that religion in particular may play an important role in attitude formation. For example, the Catholic Church states that life begins at conception. Use of PND is supported as a means of identifying risks to the unborn child and providing appropriate intervention to safeguard the mother and foetus as long as the risks to mother and child are not disproportionate to the benefits. However the use of PND is not condoned if it is carried out with the aim of possibly terminating the pregnancy depending on the results (Congregation for the Doctrine of the Faith, 1987). In Orthodox Jewish law abortion is allowed up to 40 days after conception as this is when the foetus gains human status. While this makes testing more feasible, results would have to be available early in the pregnancy, for termination to be an option if a condition is diagnosed (Papp & Papp, 2003). Although this is possible as CVS has been performed as early as week 6, loss rates are high (9%) (Wapner et al., 2002). Islam forbids the termination of a pregnancy after soul or 'Ruh' is given to the foetus. There is disagreement within Islam as to when this happens but the most commonly held belief is that it is at 120 days. It is rare that termination would be permitted any later than this unless the life of the mother was at

risk. Like Catholicism, PND is permitted if the aim is to offer intervention or prepare for the birth of a child with specific needs. Different schools of Muslim law have different views on acceptable reasons for termination. However if a foetus has a condition that will cause substantial suffering to the child then it is generally considered to be acceptable to abort before 120 days (Omran, 1992).

Unsurprisingly, the decision to terminate following diagnosis is often a difficult one. Grief after termination of pregnancy following diagnosis of foetal abnormality has been described as similar to grief following the death of a newborn (Kenyon, Hackett, & Campbell, 1988). Chamayou et al. (1998) observed that couples often decide against having another child after repeated terminations following β -thalassaemia diagnosis for this reason.

1.5 Attitudes towards PGD

As a relatively new technological advancement, there has been limited research into women's attitudes towards PGD. However, it has been argued that PGD is a preferable alternative to PND as it avoids the difficult decision of whether to terminate the pregnancy and the risk of miscarriage due to procedures used in PND such as CVS and amniocentesis (Vergeer, van Balen, & Ketting, 1998). Indeed, women who have had a child with a serious genetic condition and women who have terminated a pregnancy following diagnosis are particularly supportive of PGD (Chamayou et al., 1998; Hui et al., 2002; Palomba et al., 1994). Kalfoglou, Scott, and Hudson (2005) summarised the findings from studies assessing participants' attitudes towards PGD. Advantages included avoiding termination following PND, avoiding the birth of a child with a genetic disorder, avoiding the stress of waiting for prenatal testing to find out results and avoiding risk of miscarriage due to a genetic condition (Chamayou et al., 1998; Fernandez, De Vincentiis, Chillik, & Brugo-Olmedo, 2004; Lavery et al., 2002; Pergament, 1991; Snowdon & Green, 1997). Disadvantages included difficulty conceiving through IVF (approximately 21.8% success rate, HFEA, 2002), risks to both

mother and child, the physical risks associated with IVF, cost, and the ethical issue of what to do with unused embryos (Chamayou et al., 1998; Fernandez et al., 2004; Hui et al., 2002; Katz, Fitzgerald, Bankier, Savulescu, & Cram, 2002; Lavery et al., 2002; Pergament, 1991; Snowdon & Green, 1997). Kalfoglou et al. (2005) also identified two studies in which unreliability of testing was a perceived disadvantage (Hui et al., 2002; Pergament, 1991).

Kalfoglou et al. (2005) carried out a study on PGD patients' and providers' attitudes towards the use and regulation of PGD. They found that while participants advocated the use of PGD as a means to avoid the birth of a child where there is high risk of a serious genetic condition, attitudes varied for less severe conditions and non-medical characteristics. Again, one of the advantages identified by people who had used PGD was that it avoids the stress of prenatal testing and the possibility of having to decide whether or not to terminate. Kalfoglou et al. (2005) also found that while some women would not consider abortion on moral grounds, they felt more comfortable using PGD and avoiding implantation of embryos. The authors also looked at the views of patients and providers of PGD to avoid having a child with less serious conditions, for example adult onset diseases. They found that 11 out of the 32 participants were either unsure or felt that this was inappropriate use of PGD. Reasons for this included the fact that the disease is not immediately life threatening, that there may eventually be effective treatments and cures, and that affected individuals can still lead productive lives.

Religious viewpoints on PGD and avoiding implantation vary according to the belief as to when a foetus is given human status. For Judaism and Islam, where the foetus is not considered to have human status at the 4-8 cell stage (when PGD would take place), avoiding implantation of an embryo following PGD is considered to be preferable to termination following PND (El-Hashemite, 1995). However the Catholic Church states that life begins at conception and for this reason avoiding implantation of

an embryo following PGD is not considered to be acceptable (Congregation for the Doctrine of the Faith, 2008).

Alsulaiman and Hewison (in press) compared four groups of Saudi parents' attitudes towards the process and outcome of PGD. In Saudi Arabia, PGD is permissible under Islamic law. Two of the groups in this study were at genetic risk for either haemoglobin disorders or non-syndromic deafness but had no experience of PGD procedures. The third group had used PGD and the final group (control group) had used IVF for infertility. The authors found that parents in both of the genetic condition groups viewed PGD as a positive way of avoiding the birth of a child with a disability and this group were less concerned about the technical limitations of PGD. The largest concerns for the PGD group were the moral status of the embryo and the technical limitations of PGD. The IVF group were the most concerned about the need for confirmatory PND once a PGD pregnancy has been established, due to the risks this entails. They were also the most concerned about the views of family and friends. Overall the results suggest that PGD is a viable option for parents in Saudi Arabia, although as in any country, there is a need for support and counselling for couples making that decision.

Vergeer et al. (1998) discussed PGD from a medical-biological and a social-ethical perspective using panels made up of different professionals. The medical-biological panel consisted of biologists and physicians and the social ethical panel consisted of psychologists, social scientists and ethicists, all of whom had expertise and interest in the area of reproductive technology. A number of issues were discussed. Both panels agreed that IVF would be a barrier for PGD but the degree to which it would be a barrier varied according to previous experience of prenatal diagnosis (PND) and seriousness of the condition. The second issue discussed was selective abortion or destruction of embryos. The study found that both practices were considered acceptable. However, destruction of embryos after PGD was considered

more ethically acceptable than selective abortion. The panels were also asked about their expectations of use of PGD according to severity of the condition diagnosed. Both panels said that PGD would be used more frequently to diagnose more serious conditions. Less serious conditions and gender were not considered acceptable reasons for PGD from an ethical point of view according to both panels. The social-ethical panel were more critical about ethical acceptability than the medical-biological panel. Both panels rated anticipated societal acceptance as lower than their own acceptance. Finally, the panels were asked about their opinions on the acceptability of Down Syndrome in mothers over 36 years of age and children born with a fatal condition that the family are aware of. Both panels agreed that social acceptability of these scenarios would decrease significantly in the future. In actual fact, 12 years later, these scenarios do not appear to have become less acceptable. Attitudes towards people with disabilities are improving and society is increasingly well set up to support individuals with disabilities. In many ways, this conflicts with the fact that new technologies have the potential to reduce the number of people being born with disabilities/genetic conditions.

Lavery, Aurell, Turner, Taylor, and Winston (2000) found that only 48% of couples who had been through PGD would consider it again for future pregnancies and 41% of participants considered PGD to be severely stressful.

Finck, Meister, Stobel-Richter, Borkenhagen, and Brahler (2006) reviewed attitudes towards PGD in Germany, where it is not legal. They found the main perceived benefits to be relief of pain and suffering for those concerned and the development of medical and scientific progress. The main disadvantages were the destruction of embryos and the effect on acceptance of people with genetic conditions. They also looked at the effect of religious attitudes on attitudes towards PGD. They found that self-report on how religious people thought they were had an effect on attitudes towards PGD; however, affiliation to a particular religious group did not. As

PGD is relatively new and much less common, it is possible that people are less aware of the religious guidance about it compared to PND.

Katz et al. (2002) draw attention to the issue of whether embryos identified as having genetic abnormalities, should ever be transferred. Approximately a third of participants in the Katz et al. (2002) study said that if a couple request it, and if there are no unaffected embryos then implantation should be allowed. Katz et al. (2002) suggest that desire to have children may outweigh the possibility of having a child with a genetic condition. Draper and Chadwick (1999) note that as clinicians have a statutory obligation to consider the future interests of the embryo (HFEA, 1990), power is somewhat shifted from the mother to clinician, due to the clinicians integral role in the process.

1.6 Comparison Studies

Chamayou et al. (1998) asked participants to rank some of the advantages and disadvantages for PGD compared to PND. Advantages included the elimination of anxiety and stress while waiting for the PND result, the elimination of the psychophysical trauma during selective pregnancy termination in the case of β -thalassaemic diagnosis and loss (death) in the case of positive PND. Disadvantages of PGD included IVF for fertile couples, ovarian stimulation, oocyte retrieval and the failure to become pregnant after transfer of non β -thalassaemic embryos. Chamayou et al. (1998) compared the views on PGD of couples at high risk of having a child with β -thalassaemia. The groups compared were couples coming for their first PND (group A), high risk couples coming for a second or third PND with previous experience of selective abortion (group B), high risk couples coming for a second or third PND without previous experience of selective abortion (group C) and infertile couples from the reproductive medicine centre undergoing IVF or intracytoplasmic sperm injection treatment (group D). All groups agreed that the biggest advantage was avoidance of selective pregnancy termination in the case of positive PND. However, group A placed

similar importance on reducing psychological trauma of couples during abortion.

Possible failure to become pregnant was considered the greatest disadvantage of PGD by all four groups.

Katz et al. (2002) compared the views of people attending a clinic for PGD (for either single gene or aneuploidy screening) with those attending for their first cycle of IVF (control group). Table 1 illustrates the questions and the responses they received.

Table 1. Responses in Katz et al.'s (2002) study

Statement	PGD Single gene disorders	PGD Aneuploidy screening	Control group (1 st cycle IVF)
IVF pregnancy rates range between 15 to 40%, indicating that more than one IVF cycle may be needed to become pregnant.	No concern 51%	No concern 19%	No concern 53%
Would you terminate the pregnancy if the secondary confirmation in the form of prenatal testing showed that the foetus was genetically affected?	Yes 51%	Yes 29%	Yes 25%
Is the destruction of an embryo prior to implantation less wrong than the destruction of a foetus in pregnancy?	No 4%	No 4%	No 25%
If given the choice, would you accept the transfer of an embryo identified as being a healthy carrier?	Yes 63%	Yes 8%	Yes 22%
Should there be restrictions as to who should be offered PGD or limitations on the types of disorders that should be analysed by PGD?	Yes 2%	Yes 10%	Yes 25%
Is there a concern regarding the availability of IVF and PGD technology for couples who are fertile, including the Australian federal government rebate?	No concern 80%	No concern 38%	No concern 53%

The results from the Katz et al. (2002) study show that people's attitudes differ somewhat depending on their experience of PGD and the type of screening/diagnosis they are having. It is particularly interesting that participants actually going through PGD had much more favourable attitudes towards it. This adds emphasis to the PND studies which have found that information about testing needs to be given to people on an individual basis. This paper also highlights the ethical dilemmas of PGD for example whether embryos identified as healthy carriers should be transferred. These debates are considered in more detail below.

Research suggests that there may be some cultural differences in women's attitudes towards PGD. Alsulaiman and Hewison (2006) interviewed Saudi couples about their attitudes towards PND and PGD. They found that 27% of participants found PGD to be acceptable, 13% found PND to be acceptable and 10% found either technology acceptable. The authors also found that for thalassaemia, 86% of couples were interested in PGD. However, this figure was lower for other conditions such as cystic fibrosis (27%) and haemophilia (25%). The majority (77%) of participants said that they would not terminate a pregnancy for religious reasons. However, overall 38% said they would consider PGD and avoiding implantation. Although this figure is lower than previously found in Western countries, as found in Alsulaiman and Hewison's (in press) research described above, it further indicates that PGD may be more acceptable to people with particular religious beliefs.

1.7 Ethical debates

The development of reproductive technologies and the option of terminating pregnancies/avoiding implantation of diagnosed embryos raise important ethical questions in terms of eugenics. The issue of eugenics in terms of people being born with disabilities is not a new one and Shakespeare (2003) illustrates how it has changed over the years; from stopping certain people with disabilities from becoming

parents through eugenic sterilisation, to giving people the opportunity to choose to avoid the birth of babies with disabilities through PND and PGD.

The disability movement has argued that the use of PND and PGD to prevent the birth of a child with disabilities devalues the lives of people who have these disabilities and sends the message that these people should not have been born (Parens & Asch, 2003). Indeed, a study by Helm (1998) found that some genetic counsellors reacted negatively to parents who wished to knowingly have a child prenatally diagnosed with Down syndrome. However, the HFEA (1999) suggests that an alternative view is that it is acceptable for people to choose not to have a child with a particular condition, whilst still acknowledging that people living with such conditions have the same rights as anyone else.

The HFEA (1999) highlight some ethical questions raised specifically by PGD, such as whether an embryo diagnosed with a condition should be implanted. The HFEA give the example of a congenital deafness where parents who are deaf may argue that having a child who is also deaf would be preferable in order to integrate them into their home environment. Another possibility is that when diagnosing one condition/chromosomal abnormality another could be identified. Parents would then have to make a decision on whether to implant based on the new diagnosis.

Concerns have also been raised about the future of PGD and the possibility that diagnosis might extend to physical and social characteristics. The HFEA currently state that it would not be acceptable to test for any 'social or psychological characteristics, normal physical variations, or any other conditions which are not associated with disability or a serious medical condition' (HFEA, 1999, p. 7). However, it is difficult to ignore the fact that the potential for this exists and that it may be reconsidered in the UK in the future or that it may be available in other countries. For example, in the UK, sex selection for social reasons is currently banned (HFEA, 2010). However this is not the case in other countries such as the USA and Russia.

1.8 Attitudes and behaviour

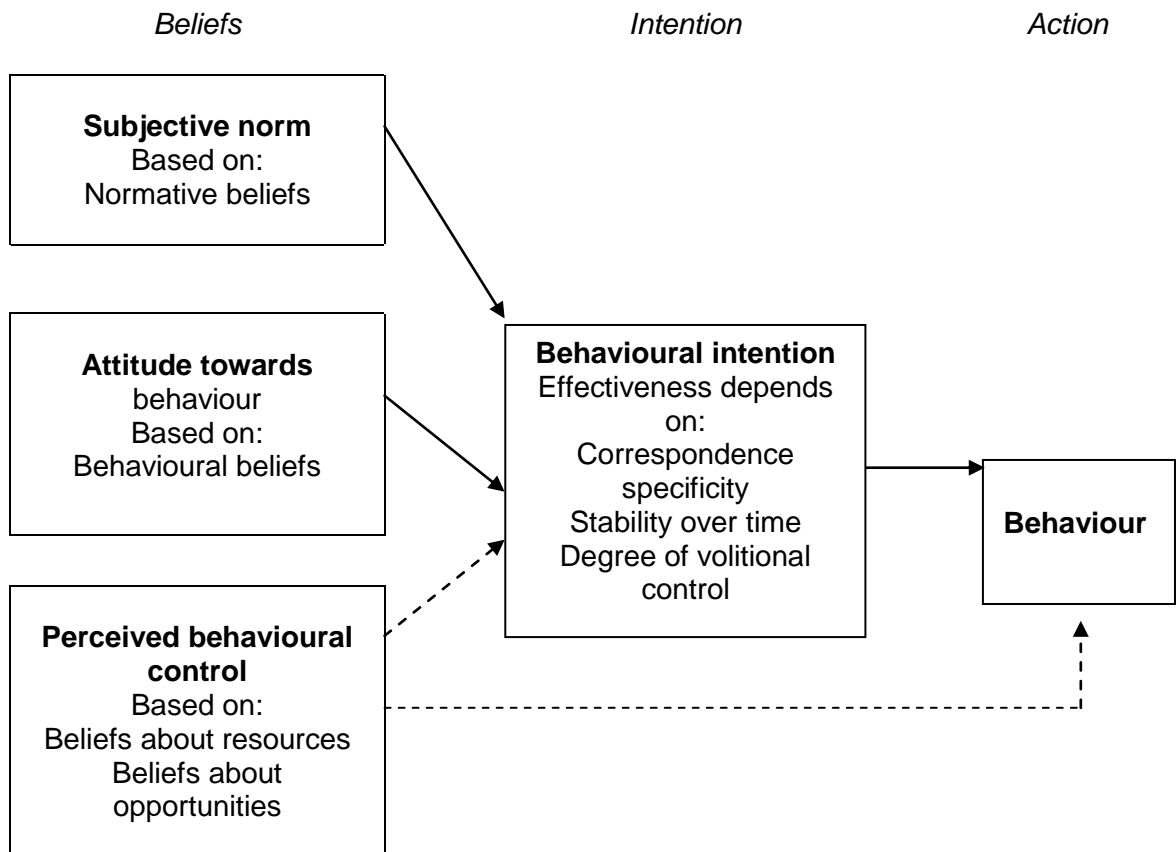
There are a number of theories that help to inform understanding of the relationship between attitudes and behaviour. It is widely accepted that attitudes do not predict behaviour in isolation but that there are a number of factors that can affect the relationship between attitudes and behaviour. Fishbein (1971) argues that the basic ingredient of an attitude is affect. However, this alone cannot accurately how people will behave and better prediction relies on understanding the interaction between attitudes, beliefs and behavioural intentions and the relationship between these and subsequent actions.

Ajzen and Fishbein (1980, Fishbein and Ajzen, 1984) have found that in order to look at the relationship between attitudes and behaviour, specific rather than general attitudes must be considered. For example a study by Davidson and Jacard (1979) looked at attitude specificity in relation to women's use of the contraceptive pill. More general attitudes such as 'attitude towards birth control' were significantly less predictive of behaviour than specific attitudes such as 'attitude towards using birth control in the next two years'. In the case of PND, this would mean that looking at women's attitudes towards termination as a means of predicting this behaviour is not likely to be accurate. Instead one would have to look at women's attitudes towards termination, in specific circumstances, following diagnosis for a specific condition.

Ajzen and Fishbein (1980) and Fishbein and Ajzen (1974) combined the processes of beliefs, intention and action to develop the theory of reasoned action. Specifically, this model states that behaviour is determined by behavioural intention to carry out the behaviour. Intention is determined by attitudes and subjective norms. Each of these predictors have their own influences, including beliefs about the consequences of the behaviour (attitudes) and the subjective likelihood that significant others think the person should perform the behaviour (subjective norms).

Ajzen (1989) extended the theory of reasoned action and developed the theory of planned behaviour (TPB). This includes the role of perceived behavioural control - beliefs about how easy it is to perform an act, based on consideration of past experiences and present obstacles envisaged by the individual. The theory of planned behaviour is the dominant model used to understand health behaviours. The theory of reasoned action and theory of planned behaviour are illustrated in Figure 1. In terms of PGD a woman might be more likely to go ahead with implantation of an embryo diagnosed through PGD if she believes she will value the outcome, believes people whose views she values think she should go ahead with implantation and believes she has the resources/opportunity to go ahead (Conner & Norman, 1995).

Figure 1. Theory of Planned Behaviour and theory of reasoned action



The solid lines show the theory of reasoned action and the dotted lines the addition of the theory of planned behaviour. From Hogg and Vaughan (2008)

In this model the attitudes component comprises a combination of the perceived likelihood that performance of the behaviour will lead to a particular outcome and the evaluation of that outcome. A person may possess many beliefs about a behaviour but only some of these beliefs will become salient at any one time and it is these salient beliefs that determine attitude (Conner & Sparks, 1995).

The TPB has been used in the area of prenatal testing to develop the multi-dimensional measure of informed choice (MMIC) (Marteau, Dormandy & Michie, 2002). The MMIC comprises three dimensions – knowledge, attitude and uptake on the basis that a woman has made an informed choice to undergo prenatal testing if she has a positive attitude towards the test, relevant knowledge and undergoes it. However, it was found that attitude but not knowledge predicted outcome, suggesting that the TPB cannot fully predict informed choice for prenatal testing. To date the TPB has not been used to look at abortion following PND or behaviours relating to PGD.

The TPB has been criticised due to the fact that it states that people's attitudes are formed through a trade off between the expected good or bad outcomes. This does not allow for people to be ambivalent in their attitudes (March, 1978; Conner & Sparks, 1995). People may want to go ahead with termination/avoid implant on the one hand but feel it is immoral or unethical on the other. The potential usefulness of the TPB for PND and PGD will be reconsidered in the discussion, in relation to the findings from the current studies.

1.9 Summary of literature and research questions

As this review has shown, the use of PND and PGD has been debated from a number of perspectives. It is apparent from the literature that there are both similarities and differences between PGD and PND. Both raise issues relating to the severity of the condition diagnosed and issues relating to the development of the embryo. Issues specific to either PND or PGD are also apparent, such as that of whether to terminate in the case of PND and whether diagnosed embryos should be implanted in the case of

PGD. Other factors such as religion also play a clear role in both PND and PGD. The issue of eugenics is identified as important for both PGD and PND, and there is some suggestion in the Katz et al. (2002) study, that PGD and avoiding implantation may be more acceptable to women in this respect. Overall, PGD enables people to avoid bringing up a child with a genetic condition without the stress and guilt of terminating a pregnancy. Prior to PGD this was impossible, other than by avoiding childbearing altogether. Therefore, the availability of PGD raises profound psychological questions about what women want from reproductive technologies. A number of previous studies have found the severity of the genetic condition diagnosed to have an impact on women's attitudes towards PND/PGD and decisions around termination/avoiding implantation. However, to date there has not been any research that looks directly at the interaction between the genetic condition diagnosed and the technology used to test. It was hypothesised that attitudes towards termination following PND and avoiding implantation of embryos following PGD may differ according to the severity of the condition diagnosed. The aim of this thesis was to investigate this further.

Research questions

1. Do women's attitudes towards termination following prenatal diagnosis differ from their attitudes towards non-implantation of embryos following pre-implantation genetic diagnosis?
2. If there is a difference, does this difference vary according to the condition diagnosed?
3. If there are differences, what are the explanations for these differences?

Two studies are described; an online quantitative study aimed at answering questions 1 and 2 (Study 1) and a brief interview study aimed at answering question 3 (Study 2).

CHAPTER 2: PILOT STUDY

2.1 Introduction

It was conjectured that the proportion of women saying that they would terminate (PND) or avoid implant (PGD) would vary systematically by the apparent severity of the condition. Moreover it was conjectured that relative to PND, when offered PGD women would be more likely to end the potential pregnancy (avoid implant) for each of the conditions. A pilot study was carried out to examine:

1. Whether these conjectures were broadly correct
2. The feasibility of a vignette simulation methodology
3. The advantages and disadvantages of two response formats.

2.2 Method

2.2.1 *Sample*

An opportunistic sample of eight female friends of the researcher was used. Participants were aged between 23 and 31.

2.2.2 *Design*

Participants considered both PND and PGD options and responded using both continuum and forced choice response options. This was chosen so that discussion about differences in responding according to technology and/or genetic condition could be facilitated.

2.2.3 *Procedure*

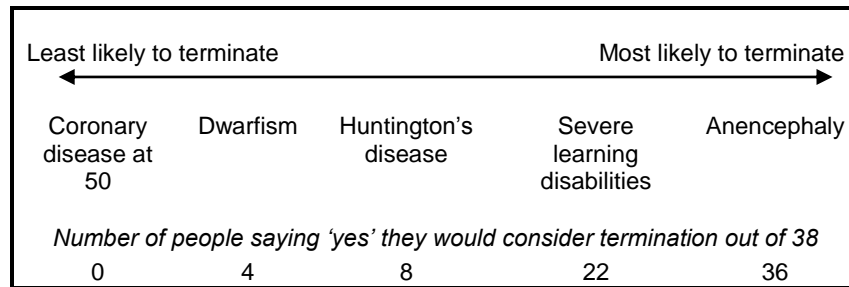
Participants were asked to fill in two questionnaires, one based on PND and the other on PGD (see appendices 1 and 2). Questionnaires were counter balanced to prevent order effects. After completing the questionnaires participants were asked for informal feedback on their experience of filling them in.

2.2.4 *Questionnaires*

Both questionnaires listed descriptions of five genetic conditions and participants were asked whether they would consider terminating the pregnancy (PND

questionnaire, appendix 1) or avoiding implantation (PGD questionnaire, appendix 2) following diagnosis of each condition. The actual names of the conditions were not available to participants as it was felt that any pre-conceived beliefs about conditions may cloud judgement. The choice of conditions included in the questionnaires was based on a doctoral thesis which asked women whether they would terminate a pregnancy following PND for 30 different conditions (Deeks, 2003). Five conditions that varied from least to most 'likely to terminate' were identified for use in the questionnaires. These are shown in Figure 2.

Figure 2. Conditions varying in 'likelihood to terminate' based on Deeks (2003)



Two different response formats were tested in the questionnaires - a continuum representing likelihood of termination/avoiding implantation and forced choice (yes, no, not sure). The aim of piloting two types was to see whether the different response styles had any effect on participants' responses and/or experience of filling in the questionnaires.

Continuum design: For the PND condition participants were asked, 'Imagine you are pregnant and a prenatal test shows that the baby has this condition. On a scale of 0-100 where 0 = extremely unlikely and 100 = extremely likely, how likely is it that you would terminate the pregnancy?' For the PGD condition, participants were asked, 'Imagine you are having a baby through in vitro fertilisation (IVF) and there is one embryo ready to be implanted. A test shows that the embryo has this condition. On a scale of 0-100, where 0 = extremely unlikely and 100 = extremely likely, how likely is it

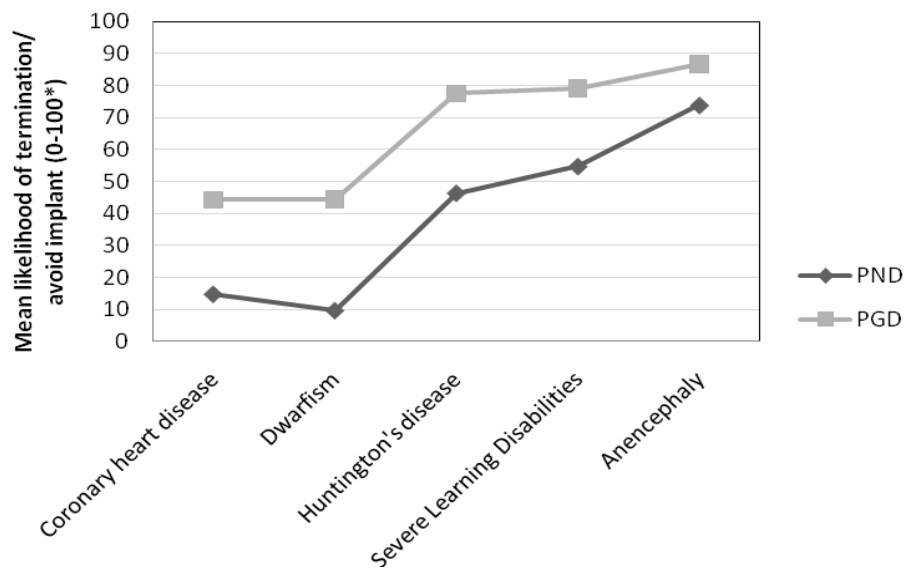
that you would avoid the implantation?' Participants were then asked to indicate their responses on a scale.

Forced choice design: For the prenatal testing condition, participants were asked 'imagine you are pregnant and a prenatal test shows that the baby has this condition. Would you consider terminating the pregnancy?' For the pre-implantation genetic diagnosis condition, participants were asked 'imagine you are having a baby through in vitro fertilisation (IVF) and there is one embryo ready to be implanted. A test shows that the embryo has this condition. Would you go ahead with the implantation?' Participants were then asked to respond yes, no or not sure.

2.3 Results

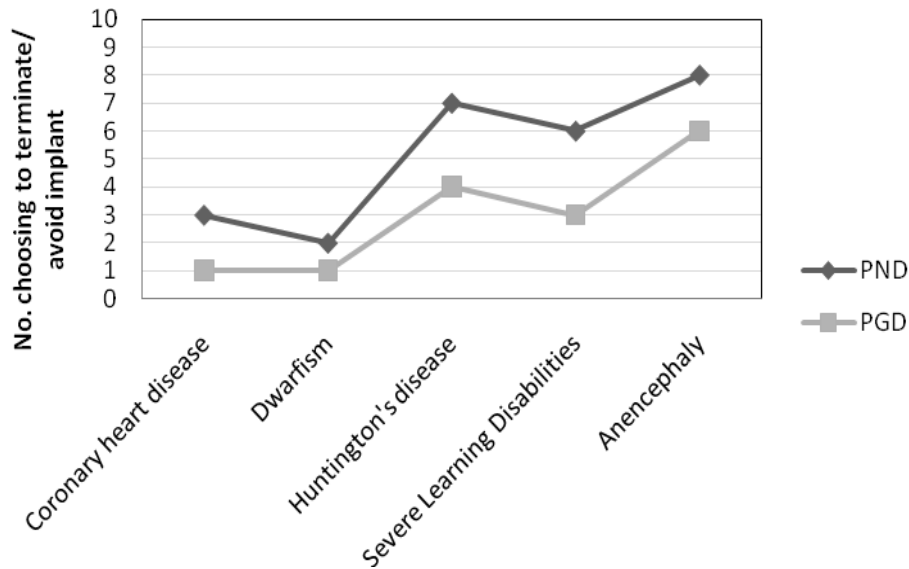
The data for each condition and response option were plotted (see Figures 3, 4 and 5). These plots suggest a clear difference in responding between the PND and PGD groups. The difference appears more pronounced for some conditions than others. Both response formats appear to result in a similar pattern of responding.

Figure 3. Average rating for each condition for termination and avoiding implantation, continuum format (N=8)*



*0=extremely unlikely to terminate/avoid implantation, 100=extremely likely to terminate/avoid implantation

Figure 4. Number of participants who responded ‘yes’ to terminating pregnancy/avoiding implantation, forced choice format (N=8)



2.4 Informal feedback from pilot study

Participants were asked how they had found the experience of filling in the questionnaires and whether they had any comments. All participants were keen to talk about the subject and discuss factors that influenced their decisions. Many commented that this wasn't a topic that they had thought about before and were interested by the difference in their own opinions on termination (PND) versus avoiding implantation (PGD).

One participant commented that she would be more likely to avoid implantation than to terminate, as a non-implanted embryo was one step removed in that it 'wasn't yet inside' her. Similarly, several participants felt that their decision to terminate would be affected by how far into the pregnancy they were. One participant felt that her religious views had a large effect on attitude to termination. However, she felt that these views were not clearly formed in relation to non-implantation of an embryo following PGD.

Two participants felt that it was easier to imagine being pregnant than it was to imagine that they were trying for a baby through IVF. One participant felt that this was

due to the stage she was at in her life - at present she did not want children, so it was hard to imagine going to great lengths to have a child through IVF. However, an unplanned pregnancy was a scenario that was easier to imagine. Several participants commented that it was easier to give a definitive response in the 'yes', 'no', 'not sure' condition than it was to indicate the likeliness they would terminate or avoid implantation on a scale. Participants felt that this was not as difficult if they were very clear that their decision was a 'yes' or a 'no'. However, if they were unsure, they found it hard to indicate exactly how unsure they were on the scale.

When asked whether they had found the study distressing, participants said that they had not found it too distressing although it had generated some feelings of guilt and discomfort. None of the participants currently had children and it was felt that it might be more difficult and/or distressing for people who have children, are pregnant, those who are unable to have children or people with personal experience of the conditions.

2.5 Conclusions

The pilot study suggests a clear difference between women's decision making and attitudes towards termination following PND versus non-implantation of embryos following PGD. In line with previous studies (Deeks, 2003; Hewison et al. 2007), it also seems that attitudes vary according to the genetic condition that has been diagnosed.

The study is limited by the small sample size and opportunistic sample. The following studies aim to consolidate the quantitative findings from the pilot study (Study 1) and develop more detailed qualitative understanding of why such patterns arise (Study 2). Participant feedback from the pilot study is considered in more detail in the method sections for both studies.

CHAPTER 3: STUDY 1 METHOD

3.1 Design

This was an internet based questionnaire study with a between subjects design. A within subjects design with a questionnaire, followed by semi-structured interview was considered instead of carrying out two separate studies. However, a much smaller number of participants were needed for the interview part of the study. It would therefore have been both un-economical and unethical to interview everyone who filled in the questionnaires. It was also felt that for the quantitative analysis, a between subjects design would be more rigorous and carry-over effects from the PND to PGD condition or vice versa would be prevented. This was followed by an interview study where participants were interviewed about their responses to both the PND and PGD questionnaires (described in Chapter 5).

3.1.1 Internet studies

Over the past decade there has been an increase in the number of researchers carrying out web-based studies (Bewick, Gill, Mulhern, Barkham, & Hill, 2008). Research suggests that university populations are generally willing and able to respond to internet-based studies (Richards & Tangney, 2007). A Eurostat report (2005) found that 94% of students and 51% of all employees use the internet in the UK. One may hypothesise that the percentage of university employees using the internet is likely to be higher than this average figure. There are of course issues of representativeness such as the characteristics of people who fill in online studies. For example, a study by Bewick et al. (2008) found that 74% of respondents in their study were female. Although issues of representativeness, for example students' ability to use the internet and possible differences between staff and students must be considered it was decided that this would not impact the results enough to warrant using paper-based questionnaires. It was hoped that using an online study would maximise recruitment for the quantitative part of the study as people could access the questionnaires when and

where they chose to and submit them more easily than returning them by post. Similar internet studies used previously on students at Leeds University have been successful at recruiting large numbers of participants (e.g. Sherlock, 2009).

3.2 Sample

3.2.1 Sample size

Two-hundred and sixteen participants were recruited for the study. This is discussed in more detail in the power analysis section below.

3.2.2 Inclusion criteria

Women aged between 18 and 50 were included in this study. This age limit was chosen based on the office for national statistics data which shows that the fewest births per 1000 women are from women aged under 20 and over 40 (Office for National Statistics, 2009a). In 2008, the mean age for giving birth in the UK was 29.3 in 2008 (Office for National Statistics, 2009a). The average age for women having IVF was 36 in 2007 (HFEA, 2009b). As the average age of women giving birth is increasing (Office for National Statistics, 2009b), it was hoped that an inclusion age range of 18-50 would encompass the majority of women who would be considering PND/PGD now and in the near future.

Participants were recruited from Leeds University. Both staff and students were invited to take part. It has been argued that university students are not representative of the general population. However, decisions of this kind have the potential to affect any woman considering child bearing now or in the future. Therefore, it was felt that this sample was appropriate for a baseline study with the aim that future research could be carried out on a more representative sample.

3.2.3 Exclusion criteria

Following feedback from the pilot study, it was decided that women who were pregnant should be excluded from the study due to the potentially distressing nature of the questionnaires.

3.2.4 Sample Characteristics

A total of 216 participants took part in the study, 110 in the PND condition and 106 in the PGD condition. Participants were recruited via email lists and posters displayed around the university (appendix 3). Participants were randomised to fill in either the PND or PGD questionnaire. Table 2 presents demographic data for the two groups.

Table 2. Demographic characteristics of Study 1 participants

		Condition		
		PGD	PND	
		N=106	N=110	
		N (%)	N (%)	
Age in years	Mean (SD)	32.02 (8.42)	29.93 (6.92)	
Occupation	Undergraduate student	9 (8.5)	11 (10)	
	Postgraduate student	27 (25.5)	30 (27.3)	
	Academic staff	26 (24.5)	35 (31.8)	
	Admin/Clerical staff	24 (22.6)	16 (14.5)	
	Other	20 (18.9)	18 (16.4)	
Ethnicity	White	British	79 (74.5)	86 (78.2)
		Other	13 (12.3)	12 (10.9)
		Mixed		
	Mixed	White and Black Caribbean	1 (0.9)	-
		White and Black African	1 (0.9)	-
		White and Asian	-	1 (0.9)
	Asian or Asian British	Indian	5 (4.7)	2 (1.8)
		Pakistani	-	-
		Bangladeshi	-	-
	Black or Black British	Other	4 (3.8)	3 (2.7)
		Caribbean	-	-
		African	1 (0.9)	1 (0.9)
	Other	1 (0.9)	-	

Table 2 continued

	Chinese or other ethnic group	Chinese	-	2 (1.8)
		Other	-	-
	Missing		1 (0.9)	3 (2.7)
Religion		Christian	39 (36.8)	39 (35.5)
		Atheist	26 (24.5)	20 (18.2)
		None	22 (20.8)	29 (26.4)
		Other	8 (7.5)	5 (4.5)
		Agnostic	6 (5.7)	13 (11.8)
		Muslim	2 (1.9)	-
		Hindu	1 (0.9)	1 (0.9)
		Buddhist	1 (0.9)	2 (1.8)
		Jewish	1 (0.9)	1 (0.9)
Number of participants with children		Yes	29 (27.4)	17 (15.5)
		No	77 (72.6)	93 (84.5)
Plan to have children in future?		Yes	59 (55.7)	69 (62.7)
		No	24 (22.6)	21 (19.1)
		Don't know	23 (21.7)	20 (18.2)

3.3 Sample comparisons

Comparisons between participants in the PND and PGD conditions were calculated for age, occupation and ethnicity using the independent *t*-test. Results of these comparisons showed that participants in the PGD group ($M = 32.02$, $SE = 0.82$) were significantly older than the PND group ($M = 29.93$, $SE = 0.66$, $t(214) = 1.20$, $p < 0.5$). However the effect size was small $r = 0.27$. There were no significant differences for ethnicity (PGD, $M = 1.91$, $SE = 0.22$; PND = 1.78, $SE = 0.23$, $t(210) = 0.40$, $p > 0.5$) occupation (PGD, $M = 3.18$, $SE = 0.12$; PND, $M = 3.00$, $SE = 0.12$, $t(214) = 1.07$, $p > 0.5$) or parity (PGD, $M = 1.73$, $SE = 0.04$; PND = 1.85, $SE = 0.04$, $t(214) =$

2.15, $p > 0.5$). Effect sizes were trivial ($r = 0.14$ for occupation and parity and $r = 0.06$ for ethnicity).

3.4 Questionnaires

The questionnaires were created using Bristol Online Survey system (BOS, 2007). As BOS is unable to randomise participants to different questionnaires a web page with a link that randomised participants to either the PND or PGD questionnaire was created. Participants were provided with the web page address in the posters (see appendix 3) and then clicked on the link on the web page to be randomised. The questionnaires were live from 23rd July 2009 for two months. Participants were given a participant information sheet, consent form, a demographic questionnaire, the PGD or PND information sheets and then the corresponding PGD or PND questionnaire. The PGD questionnaire asked participants to *'Imagine you are having a baby through in vitro fertilisation (IVF) and there is one embryo ready to be implanted. A test shows that the embryo has this condition. Would you go ahead with the implantation?'* The PND questionnaire asked participants to *'Imagine you are pregnant and a prenatal test shows that the baby has this condition. Would you consider terminating the pregnancy?'* Both listed descriptions of five different genetic conditions. As in the pilot study, names of conditions were not given to participants so that preconceptions about conditions did not affect responses. For examples of the full PND and PGD questionnaires, please refer to appendices 4 and 5. A forced choice 'yes' 'no' response format was chosen as it was felt that this design reflected real life more closely where women would have to make a yes/no decision if faced with positive diagnosis.

Conditions were chosen from a study by Hewison et al. (2007) as this study used a larger sample size than the Deeks (2003) thesis that was used to choose conditions for the pilot study. It was therefore considered to offer a more robust continuum of severity. The descriptions of conditions used in Hewison et al.'s study were generated by a team of social scientists and medical doctors including a clinical

geneticist, and an obstetrician specialising in prenatal testing. The conditions for this study were chosen by establishing the percentage of participants who said 'yes' to termination for a number of different conditions in the Hewison et al. (2007) study. Hewison et al. (2007) used four groups of women: Pakistani women with either high levels of education or low education levels and white women with high or low levels of education. Frequencies were taken from the white high education group from the Hewison et al. (2007) study as it was felt that this was most representative of the student sample used in the current study. The spread of percentage of participants responding 'yes' to termination for genetic conditions was 0.96-84.1%, such that for some conditions 0.96% participants said yes to termination and for others 84% said yes. Conditions were chosen to represent even midpoints (0.96, 21.75, 42.53, 63.32, 84.1). Where this was not possible (for example there were no conditions to which 21.75% of participants said they would terminate), the condition with the nearest number of responses was used (in this case Huntington's disease with 19.8% of participants saying they would terminate).

Table 3. Conditions varying in severity (data from Hewison et al. 2007)

Least likely to terminate		Most likely to terminate		
←		→		
Phenylketonuria	Huntington's disease	Duchenne muscular dystrophy	Trisomy 13	Anencephaly
<i>Percentage of people saying 'yes' they would consider termination</i>				
0.96	19.8	40.9	66.6	84.1

Full names and descriptions of conditions are listed below.

Phenylketonuria (PKU): Child would have a blood condition that could cause mental problems if left untreated, have a normal lifespan and have strict diet restrictions throughout life.

Huntington's disease: Child would develop an incurable condition by age 40, which has both severe mental and physical deterioration, require constant looking after and medical help and have a shortened lifespan.

Duchenne muscular dystrophy (DMD): A male child would have a progressive muscle-wasting disease, be wheelchair-bound by 11 or 12 years and have a much shortened lifespan (death probably before 20 years of age).

Trisomy 13: Child would have severe learning disabilities/mental handicap, requires a lot of looking after and die within first few months of life.

Anencephaly: Child would be born without a brain and die before or soon after birth.

In addition to the main questionnaire there was space for participants to write comments about their personal experience of the conditions and any other comments, if they chose to. Participants were also asked how easy they found it to imagine themselves in the scenarios described. This was included due to feedback from the pilot study, where participants said they found it difficult to imagine themselves in the scenarios described. Feedback from the pilot study also suggested it might be more difficult for participants to imagine themselves in the PGD condition as some participants said this wasn't something they had considered before. If participants were to find one condition easier to imagine than the other then this may affect results, hence the importance of measuring this.

3.5 Ethical Considerations

Ethical approval was obtained from LIHS/LIGHT Research Ethics Subcommittee. As this was a potentially difficult and/or distressing topic there was a recognised need for care and sensitivity in all aspects of the study. However, it was also felt that there would be potential benefit to women making decisions about testing in the future. Informed consent was obtained and all participants were given the contact details of appropriate services to contact if they needed further support (appendix 6). These services were contacted to let them know that their details had been listed.

3.6 Power Analysis

A power analysis was used to calculate the minimum sample size required to accept the outcome of chi-square test at the $p < 0.01$ level. This significance level was chosen in order to allow for multiple comparisons. The power analysis was calculated for chi-square as this is less sensitive than Cochran's Q test and therefore requires more participants. It was calculated using the proportions of participants saying yes to termination/avoiding implantation for anencephaly in the pilot study. Anencephaly was chosen as in the pilot study this was the condition with the smallest difference between PGD and PND conditions with proportions of 1.0 (avoid implant) and 0.75 (terminate). Anencephaly was also used in the pilot study and therefore unlike other conditions used it is consistent across the pilot and current study. The following equation was used:

$$\text{Standardized difference} = \frac{p_1 - p_2}{\sqrt{\bar{p}(1 - \bar{p})}}$$

$$\text{where } \bar{p} = \frac{(p_1 + p_2)}{2}$$

$$\text{So } \bar{p} = \frac{(1 + 0.75)}{2} = 0.875$$

$$\frac{1 - 0.75}{\sqrt{0.875(0.125)}} = 0.755$$

A graphical calculating device (nomogram) was used to determine the actual number of participants needed in each condition which was found to be 45. As this is a sensitive topic and questions will not be mandatory, the aim was to recruit approximately 200 participants in total to ensure that enough data was collected.

3.7 Main analyses: Attitudes towards PND and PGD

Ideally, the data would have been analysed in a way that looks at the interaction between PGD/PND and condition. However, there does not seem to be a clear method

of analysing dichotomous data in this way. Consultation with a statistician did not provide any further ideas and it was felt that chi-square test for within subjects comparisons, and Cochran's Q test for between subjects comparisons, were sufficient in order to answer the research questions.

Within subjects comparisons. Cochran's Q analysis was used to look at the variation in responding within the PND and PGD groups. Cochran's Q is an extension to the McNemar test for related samples, which tests more than two sets of proportions, in this case the proportion of participants saying yes for each condition (Siegel & Castellan, 1988).

Between subjects comparisons. Chi-square test was used to compare responding between subjects, for each genetic condition. Chi-square test calculates the difference between proportions, in this case, the proportion of people saying yes to termination/avoid implantation for the PGD and PND conditions.

Data management and simple statistical comparisons were conducted using SPSS Version 10.0 (SPSS Inc, 1999).

3.8 Response Patterns and Scaling

Guttman scaling was used to look at the patterns of responding shown by participants (McIver & Carmines, 1981). Guttman scaling was developed as a way of measuring the degree to which data is scalable. In this study this would mean the degree to which it could be assumed that if a person said yes to termination/avoiding implant for a particular condition then they would have the same response for all conditions that are considered to be more severe. Error (the number of participants who deviate from the scale) was calculated using the Goodenough-Edwards coefficient of reproducibility method (CRge). Guttman suggests that data is considered scalable if error is 10% or less so the scalability criterion is $CRge > .90$. This is compared to the Minimal Marginal Reproducibility (MMR) which reflects the reproducibility of a series of items based upon knowledge of the item marginal

distribution. The MMR values need to be greater than the CRge values to be considered scalable.

3.9 Additional Questions

Participants were asked how easy they found it to imagine themselves in the scenarios described: very difficult, difficult, neutral, easy, very easy. This data was analysed using chi-square test. Answers to the qualitative questions were not analysed in a structured way but they were used to help understand participants' individual responses to the questionnaires and to help explain patterns in the data.

CHAPTER 4: STUDY 1 RESULTS

4.1 Introduction

This chapter reports the results of Study 1. Main findings are reported first followed by further analyses of response patterns. Responses to additional questions in the questionnaire are then reported.

4.2 Main analyses: Attitudes towards PND and PGD

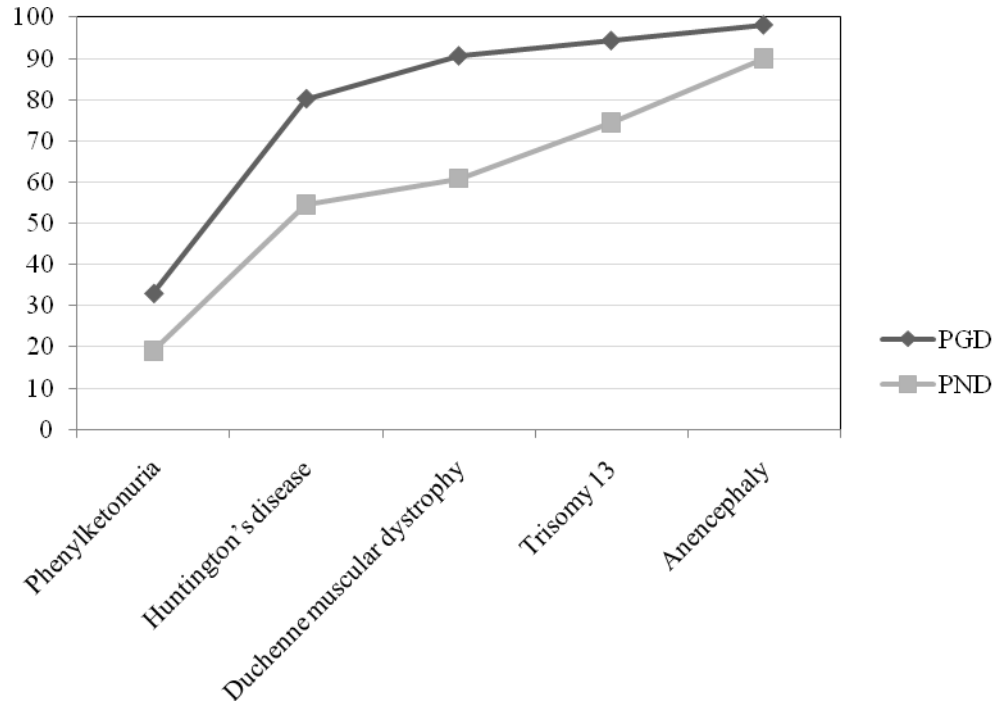
Table 4 shows the number and percentage of participants who said that they would avoid implantation in the PGD condition and terminate the pregnancy in the PND condition.

Table 4. Number and Percentage of participants who would avoid implantation (PGD)/ terminate pregnancy (PND) in Study 1

	Phenylketonuria	Huntington's disease	Duchenne muscular dystrophy	Trisomy 13	Anencephaly
PGD N=106	33% (n=35)	80.2% (n=85)	90.6% (n=96)	94.3% (n=100)	98.1% (n=104)
PND N=110	19.1% (n=21)	54.5% (n=60)	60.9% (n=67)	74.5% (n=82)	90% (n=97)

Figure 6 illustrates the percentages graphically. Observation suggests that attitudes towards PND and PGD do indeed vary according to the condition diagnosed and that there is convergence around the most and least severe conditions (PKU and anencephaly). However, for other conditions there is more variation in attitudes depending on which technology (PND or PGD) is used.

Figure 5. Percentage of participants avoiding implantation/terminating pregnancy in Study 1



Within subjects comparisons. Cochran's Q analysis was used to look at the variation in responding within each condition (PND and PGD). Cochran's Q analysis for within subjects comparisons showed significant variation in responding according to the genetic condition diagnosed (PND: Cochran's $Q=200.125$, $p<0.001$), (PGD: Cochran's $Q=164.339$, $p<0.001$). This supports the previous findings by Hewison et al. (2007) that women's attitudes towards termination following PND vary according to the genetic condition diagnosed. These results suggest that this variation in attitudes also exists for PGD.

Between subjects comparisons. Chi-square test was used to compare responding between subjects, for each genetic condition. Table 5 shows the results of each comparison. In order to account for multiple comparisons α is set at 0.01.

Table 5. Chi-square analysis for between subjects comparisons adjusted for multiple comparisons ($p/5$)

	Phenylketonuria N=211	Huntington's disease N=211	Duchenne muscular dystrophy N=211	Trisomy 13 N=211	Anencephaly N=211
χ^2	5.48	16.09	23.98	15.10	6.28
P	$p=0.19(\text{ns})$	$p=0.00$	$p=0.00$	$p=0.00$	$p=0.12(\text{ns})$

As illustrated in the table, a significant difference between the PND and PGD groups was found for Huntington's disease $\chi^2(1, N=211) = 16.09, p<0.001$, Duchenne muscular dystrophy $\chi^2(1, N=211) = 23.98, p<0.001$ and trisomy 13 $\chi^2(1, N=211) = 15.10, p<0.001$. PND/PGD comparisons were not significantly different for PKU or anencephaly.

Summary of main analyses. To summarise, within subjects comparisons show significant differences in likelihood to terminate/avoid implant, according to the condition diagnosed. Between subjects analysis shows that for some genetic conditions decision to terminate/avoid implant varies significantly depending on technology (PND/PGD). However, for the most and least severe conditions (anencephaly and PKU), decision to terminate/avoid implant does not differ significantly according to the technology when a conservative $\alpha \leq 0.01$ level was used.

4.3 Response Patterns and Scaling

Further analysis was carried out looking at the patterns of responding displayed by participants. Tables 6 and 7 illustrate these patterns of responding. The first 6 rows are ordered to represent 'consistent' patterns of responding. Thus in row one a participant chooses to continue with the pregnancy/implantation for all conditions. In row 6 the participant would choose to terminate the pregnancy/avoid implantation for all conditions. The rows in between represent increasing likelihood that a person would

choose to terminate/avoid implantation. Therefore the first 6 rows suggest that there is a hierarchy in the judged severity of the condition which is shared by many participants, especially for PGD where 97% of participants followed this pattern. The 7-9 rows in Table 6 and 7-12 rows in Table 7 represent the participants who are regarded as having made inconsistent choices if the conditions are ordered in accord with the majority. In the PND condition 9 participants (8.5%) followed an alternative pattern of responding. For all but one of the alternative patterns only 1 participant followed the pattern. However in the PND condition 5 participants (4.72%) followed an alternative pattern whereby they said they would not terminate for DMD but would for Huntington's disease (see table 7, row 9).

The overall pattern of results suggests a Guttman scale in which both participants and conditions can be scaled (McIver & Carmines, 1981).

Key for Table 6

- ✓ go ahead with pregnancy/implantation
- × terminate pregnancy/avoid implantation

Table 6. Response patterns for PGD in Study 1

					PGD
PKU	Huntington's disease	DMD	Trisomy 13	Anencephaly	N (%)
✓	✓	✓	✓	✓	0 (0%)
✓	✓	✓	✓	×	3 (2.86%)
✓	✓	✓	×	×	5 (4.76%)
✓	✓	×	×	×	10 (9.52%)
✓	×	×	×	×	50 (47.62%)
×	×	×	×	×	34 (32.38%)
×	✓	✓	×	✓	1 (0.95%)
✓	✓	×	✓	×	1 (0.95%)
✓	×	×	✓	×	1 (0.95%)

Key for Table 7

- ✓ go ahead with pregnancy/implantation
- × terminate pregnancy/avoid implantation

Table 7. Response patterns for PND in Study 1

PKU	Huntington's disease	DMD	Trisomy 13	Anencephaly	PND N (%)
✓	✓	✓	✓	✓	8 (7.55%)
✓	✓	✓	✓	×	15 (14.15%)
✓	✓	✓	×	×	10 (9.43%)
✓	✓	×	×	×	13 (12.26%)
✓	×	×	×	×	33 (31.13%)
×	×	×	×	×	18 (16.98%)
×	✓	✓	✓	✓	1 (0.94%)
✓	✓	×	✓	✓	1 (0.94%)
✓	×	✓	×	×	5 (4.72%)
×	×	×	✓	✓	1 (0.94%)
×	×	✓	×	×	1 (0.94%)

Guttman scaling was developed as a way of measuring the degree to which data is scalable. The Goodenough coefficient of reproducibility (CRge) values are PGD = 0.987 and PND = 0.962. These figures were compared with the Minimal Marginal Reproducibility (MMR). The MMR values are PGD = 0.866 and PND = 0.724. So in both the PGD and PND conditions CRge > 0.0 and > MMR showing that both the PND and PGD data sets are scalable. However, it is important to remember that the conditions chosen for this study were intentionally spread out over the range of severity. Had more conditions been included, or more that were considered to be of similar severity, results may not have been scalable and it is likely that there would have been more alternative patterns of responding.

4.4 Additional questions

Participants were asked how easy they found it to imagine themselves in the scenarios described. Responses are illustrated in Table 8 and show that participants' ability to imagine themselves in the conditions varied substantially.

Table 8. Ease of imagining self in scenario in Study 1

	Condition	
	PGD (N=106)	PND (N=110)
Very difficult	23.6% (N=25)	21.8% (N=24)
Difficult	28.3% (N=30)	31.8% (N=35)
Neutral	18.9% (N=20)	17.3% (N=19)
Easy	24.5% (N=26)	17.3% (N=19)
Very easy	4.7% (N=5)	11.8% (N=13)

Chi-square analysis showed no significant differences between the PND and PGD groups $\chi^2(4, N=216) = 5.0, p > 0.05$.

Participants were also asked whether they had any personal experience of the conditions described. A number of participants described having some kind of personal experience (38.7% in PGD group and 30.9% in PND group). The type of experience varied substantially e.g. relative/friend with a genetic condition, work, personal experience of diagnosis through PND/PGD. Responses to the question '*if you would like to elaborate or comment on any of your answers please do so*' varied enormously. Twenty two participants left comments in each condition. Comments totalled 1165 words in the PND condition and 1501 words in the PGD condition. Examples of comments are illustrated below. These illustrative comments have been chosen to reflect the variety in the comments that were left and to emphasise the complexity of influences on women's attitudes towards PND and PGD for different conditions. A more detailed analysis of women's attitudes to PND and PGD is given in Study 2.

The thought of designer babies, a la Gattaca, is quite horrifying, but I also feel that knowingly birthing a child which is pre-destined to suffer seems cruel. Condition d (PKU) could go either way with me, as I know plenty of

people with diet restrictions who are quite happy and healthy. (PGD condition)

I found this very difficult to imagine considering that my baby is sitting in front of me playing happily. (PGD condition)

I would not now (if I were planning more children) have any antenatal testing. For our eldest son we thought that we would consider termination if a serious condition were picked up, but after having our younger son, we know now that although it is so very hard for him, us and his brother, we would not have wanted a termination. Therefore if having IVF I would choose NOT to have any pre-implantation testing. That is a personal choice - not one I would impose on others. (PGD condition)

I also think that to not continue with the IVF also depends on the situation I would be in. If unstable with little money, no partner and little family help my answers would probably be different. (PGD condition)

Condition a (Huntington's disease) would have been a maybe if there'd been an option. From a selfish point of view the person not getting ill until they are 40 wouldn't affect my role in their upbringing but a prior knowledge of what life would be like for them later on would be enough to not put them through it. (PND condition)

Have personal experience of a positive screening test, declined prenatal diagnosis. Child is alive and well. (PND condition)

If I was to get pregnant I would like to have as much prenatal testing as possible and I would consider terminating the pregnancy for any major health problem. That is not to say I would definitely terminate, but I would consider it. (PND condition)

CHAPTER 5: STUDY 2 METHOD

5.1 Design

Women's attitudes to avoiding implant following PGD /terminating following PND have not previously been compared according to the severity of conditions diagnosed. The aim of this study was to answer research question 3, which was to look into explanations for the pattern of responding shown by participants. A brief baseline interview study exploring these attitudes was carried out. Participants filled in both the PND and PGD questionnaires and then took part in the interview about their responses.

5.2 Sample

5.2.1 Sample size

Sandelowski (1995) states that having an appropriate sample size in qualitative research is important. Too small a sample size can fail to support claims and too large a sample size can prevent deep, case-oriented analysis. Qualitative studies have been published with as few as one or as many as 15 participants and Smith (2008) states that there is no ideal sample size. It was decided that this study would aim to recruit 10-12 participants. This number was chosen as although this was intended to be a small scale study, it was hypothesised that there would be substantial variation between women in their responses. A smaller number of participants may not have allowed for accurate identification and consolidation of themes. Additionally as interviews were brief, this sample size still allowed for detailed enough analysis of the data.

5.2.2 Inclusion criteria

The same criteria were used as for Study 1: female students and staff from Leeds University aged between 18 and 50 who were not pregnant.

5.2.3 Exclusion criteria

People who had already completed Study 1 were excluded from taking part in Study 2 so that they were not 'primed' by already having filled in one of the questionnaires.

5.2.4 Sample Characteristics

Eleven participants were interviewed. Participants responded to study advertisements sent out via email or as posters displayed around the university (appendix 7). Twenty people responded to the advertisement showing interest in taking part in the study and all were sent a participant information sheet. Of these, four did not get back in touch, a further five were unable to attend the interview at a mutually agreeable time with the researcher and the remaining 11 took part in the study. Demographic characteristics of the participants are presented in Table 9.

Table 9. Demographic characteristics of Study 2 participants

		N
Age in years	Mean	26.00
Occupation	Undergraduate student	4
	Postgraduate student	4
	Academic staff	2
	Other	1
Ethnicity	White	
	British	6
	Other	2
	Black or Black British	2
	Chinese or other ethnic group	1
Religion	Atheist	3
	Christian	3
	Agnostic	2
	None	2
	Buddhist	1
Number of participants with children	Yes	2
	No	9
Plan to have children in future?	Yes	8
	No	0
	Don't know	3

5.3 Procedure

After informed consent was obtained (see appendices 8 and 9 for participant information sheet and consent form), demographic information was collected (appendix 10). Participants were then asked to read the PGD/PND information sheets and

corresponding questionnaires (appendices 11 and 12). The order in which participants read and filled in the PGD/PND information sheets and questionnaires was counterbalanced to prevent order effects. After questionnaires had been completed, participants took part in a brief semi structured interview about their experience of filling in the questionnaire and factors that had influenced their responses (appendix 13).

5.4 Questionnaires

It was decided that the same questionnaires and conditions would be used as for Study 1 so that patterns of responding from the two studies could be compared.

5.5 Interviews

A semi-structured interview was developed which consisted of specific questions and prompts. A semi-structured approach was felt to be most appropriate as it allowed exploration of the main areas of interest. However, unlike a structured interview it also offered the flexibility to explore some areas in more detail (Smith 2008). Smith (2008) suggests that this allows the interviewer to enter into the psychological world of the respondent more fully and also allows respondents to introduce ideas that the interviewer may not have thought of. As there are no previous studies directly comparing PND and PGD in this way, flexibility and potential for the introduction of new ideas was important.

A pilot interview was conducted in order to ensure that the questions were worded clearly and that participants could relate their questionnaire responses to the questions in the interview.

In the interview, participants were initially asked a general question of 'Was there anything that particularly influenced your responses in the questionnaire?' It was hoped that this would, for example, determine whether people have strong religious views that affected responding. Following this question, participants were asked more specific questions directly linked to their responses. For example, if a participant had

responded 'no' to termination but 'yes' to avoiding implantation, they were asked why this was (for full interview protocol see appendix 13).

At the end of the interviews participants were asked whether they had anything else they would like to add or whether they had any questions about the interview process. Interviews lasted approximately ten minutes and were recorded on a digital voice recorder.

5.5.1 Interviewer effects

Some research suggests that interviewer effects such as race, ethnicity and gender can affect responses (Davis et al, 2010). Davis et al. (2010) also report that respondents often have a desire to portray a positive self image. The authors suggest that interviewer effects can be controlled for by using a large sample. Using a larger sample would be beyond the scope of this small scale study. However, as mentioned above, quantitative results from this study can be compared to those from Study 1. If the pattern of responding were to differ substantially then it may be that interviewer effects were present. If this was found to be the case, further studies looking at the effect of methodology on responding would be warranted.

5.6 Ethical Considerations

As in Study 1, ethical approval was obtained from LIHS/LIGHT Research Ethics Subcommittee. As this was a potentially difficult and/or distressing topic there was a recognised need for care and sensitivity in all aspects of the study. However, it was also felt that there would be potential benefit to women making decisions about testing in the future. Informed consent was obtained and all participants were given the contact details of appropriate services to contact if they needed further support. These services were contacted to let them know that their details had been listed.

5.7 Analysis

5.7.1 Transcription

As recommended by Reismann (1993), all interview recordings were transcribed by the researcher in order that initial familiarisation with the data could be established.

5.7.2 Thematic analysis

As this was a preliminary study thematic analysis was chosen to analyse the data. Thematic analysis can be used as a simple method for identifying, analysing and reporting patterns (themes) within data (Braun and Clarke, 2006). Other methods of qualitative analysis such as interpretative phenomenological analysis and grounded theory were considered. However, as this research was time limited, with half of the focus on carrying out the quantitative study, it was decided that thematic analysis was the most practical and economical method.

Themes can be identified in one of two ways – inductive or deductive (e.g. Boyatzis, 1998; Frith & Gleeson, 2004; Hayes, 1997). For this project themes were identified through inductive analysis, as this is data rather than theory driven, and helps to reduce the impact of the researcher's preconceptions (Braun & Clarke, 2006). Braun and Clarke (2006) describe six phases of thematic analysis, which were followed closely.

1. Familiarising yourself with your data: The transcription process was valuable in familiarization with the data. The interview recordings were transcribed and then re-listened to in order to check that transcription was accurate. Initial ideas were noted.
2. Generating initial codes: Interesting features of the data were coded.
3. Searching for themes: Initial codes were collated into potential themes and data re-examined to see whether it fitted potential themes.

4. Reviewing themes: Themes were checked to see whether they worked in relation to the coded extracts and the entire set of interviews. A thematic analysis map was developed.
5. Defining and naming themes: Once data had been reviewed in relation to themes, names and definitions of themes were generated.
6. Producing the report: Extract samples were chosen for the report, based on the reliability check. A report of the analysis was produced.

5.8 Reliability

Inter-rater reliability was established by asking two other raters to sort quotes into the themes and sub-themes identified by the main researcher. Results of the reliability check are reported in the next chapter.

CHAPTER 6: STUDY 2 RESULTS

6.1 Introduction

As reported in chapter five, the results of Study 1 show that for some genetic conditions, there is a difference in women's attitudes towards non-implantation of embryos following PGD and termination of pregnancy following PND. This difference varies according to the genetic condition diagnosed. The aim of Study 2 was to develop understanding of why such differences occur. This chapter describes the results of Study 2 firstly reporting descriptive statistics, followed by thematic analysis findings.

6.2 Attitudes towards PND and PGD

6.2.1 Percentage of participants who would terminate pregnancy/avoid implantation

Table 10 shows the number and percentage of participants who said that they would avoid implantation in the PGD condition and terminate the pregnancy in the PND condition.

Table 10. Number and Percentage of participants who would avoid implantation/terminate pregnancy in Study 1

	Phenylketonuria	Huntington's disease	Duchenne muscular dystrophy	Trisomy 13	Anencephaly
PGD	27.3 (n=3)	81.8 (n=9)	90.9 (n=10)	100 (n=11)	90.9 (n=10)
PND	0 (n=0)	27.3 (n=3)	36.4 (n=4)	81.8 (n=9)	90.9 (n=10)

Figure 7 illustrates the percentages graphically. As found in Study 1, observation suggests that attitudes towards PND and PGD vary according to the condition diagnosed and that there is convergence around the most and least severe conditions (PKU and anencephaly). However, for other conditions there is more variation in attitudes depending on which technology (PND or PGD) is used.

Figure 6. Percentage of participants terminating pregnancy/avoiding implantation in Study 2

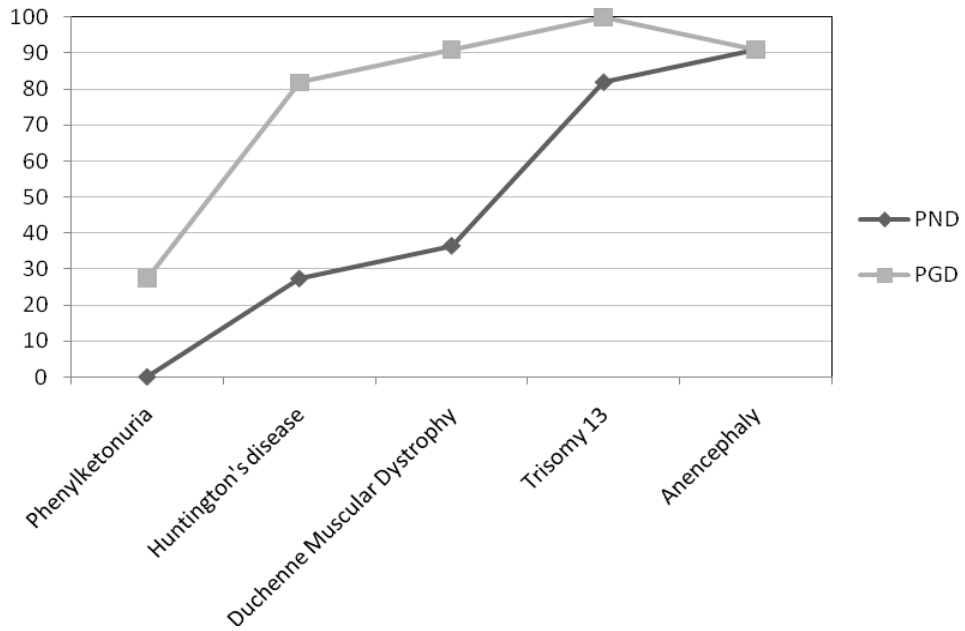
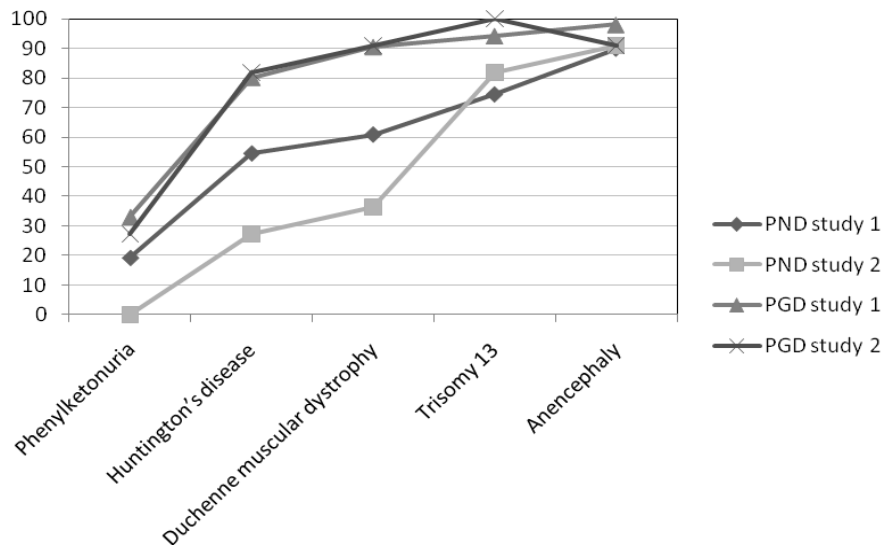


Figure 8 shows the results for Studies 1 and 2. This figure shows that for PGD, responses across the two studies were similar. There appears to be more of a difference for PND. However, in both studies there was the same increase in the number of people avoiding implant/terminating with the increase in severity of condition.

Figure 7. Percentage of participants terminating pregnancy/avoiding implantation in Studies 1 and 2



6.2.2 Response patterns

Table 11 illustrates the patterns of responding followed by participants. As found in Study 1 the majority of participants follow a pattern of responding whereby the likelihood of avoiding implantation/terminating pregnancy increased with severity of condition. However, there are exceptions to this.

Key for Table 11

- ✓ go ahead with pregnancy/implantation
- × terminate pregnancy/avoid implantation

Table 11. Response patterns in Study 2

					PND	PGD
PKU	Huntington's	DMD	Trisomy 13	Anencephaly	N	N
✓	✓	✓	✓	✓	0	0
✓	✓	✓	✓	×	2	0
✓	✓	✓	×	×	5	1
✓	✓	×	×	×	0	1
✓	×	×	×	×	3	6
×	×	×	×	×	0	2
✓	Missing	×	×	✓	1	-
×	×	×	×	✓	-	1

6.2.3 Individual participants' responses

Participants' individual responses are shown in Table 12. These have been reported in order to aid understanding of the themes and quotes illustrated in the thematic analysis section of this chapter. They are displayed in order of most to least likely to avoid implant/terminate pregnancy calculated by adding the total number of instances in which they would avoid implantation/terminate pregnancy with a maximum score of 10.

Key for table 12

- ✓ go ahead with pregnancy/implantation
 x terminate pregnancy/avoid implantation

Table 12. Individual participants' responses in Study 2

Participant number	Condition	PKU	Huntington's	DMD	Trisomy 13	Anencephaly
1	PND	✓	x	x	x	x
	PGD	✓	x	x	x	x
2	PND	✓	x	x	x	x
	PGD	✓	x	x	x	x
3	PND	✓	x	x	x	x
	PGD	✓	x	x	x	x
4	PND	✓	<i>Missing</i>	x	x	✓
	PGD	✓	x	x	x	x
5	PND	✓	✓	✓	x	x
	PGD	x	x	x	x	x
6	PND	✓	✓	✓	✓	x
	PGD	x	x	x	x	x
7	PND	✓	✓	✓	x	x
	PGD	x	x	x	x	✓
8	PND	✓	✓	✓	x	x
	PGD	✓	x	x	x	x
9	PND	✓	✓	✓	x	x
	PGD	✓	✓	x	x	x
10	PND	✓	✓	✓	✓	x
	PGD	✓	x	x	x	x
11	PND	✓	✓	✓	x	x
	PGD	✓	✓	✓	x	x

6.3 Thematic analysis

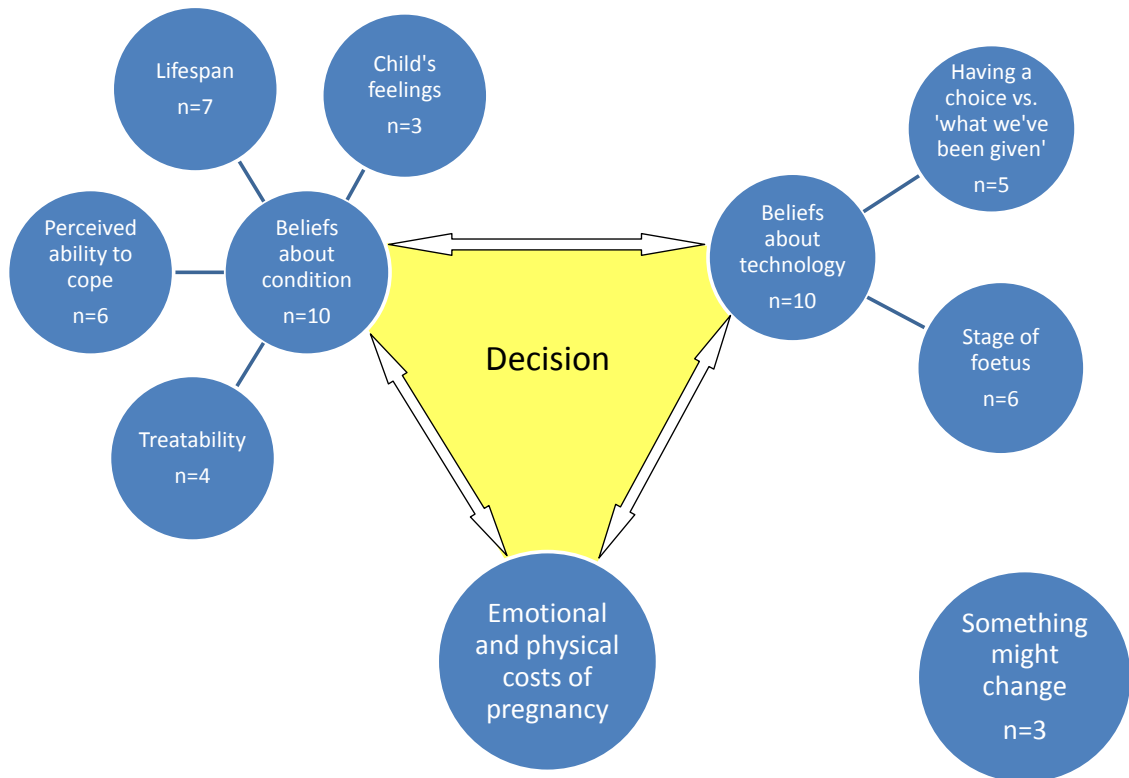
Transcripts were analysed using inductive thematic analysis (Braun and Clarke, 2006). Transcripts were read several times and themes were identified based on the data, without imposing the structure of any previous findings/theories.

6.3.1 *Inter-rater reliability*

Inter-rater reliability was established by asking two other raters to sort quotes into the themes and sub-themes identified by the main researcher. Extra quotes that did not fit any theme were added and an 'other' theme was available for the raters to sort quotes into. For 67% of quotes at least two out of the three raters placed the quotes within the same sub-theme. The highest consistency between raters was within the 'beliefs about condition' theme and subthemes. A number of quotes spanned more than one theme and were placed in one of the two themes by raters, reducing the overall consistency between raters. However, this emphasises the importance of the interaction between themes. A total of 15 quotes were sorted into the 'other theme' by the two raters. Eight of these were quotes intentionally added in by the researcher and had not originally been allocated to a theme in the analysis. The remaining seven quotes sorted into the 'other' theme belonged to the 'beliefs about technology' theme, six out of the seven in the 'stage of cells' sub-theme.

Quotes illustrated below to represent themes or subthemes are those that showed the greatest consistency between raters. Quotes in the 'interaction between themes' section are those that were placed in more than one theme by raters. Four main themes and six sub-themes were identified as illustrated in Figure 9. The 'n' in Figure 9 refers to the number of participants who made comments within a theme. This does not reflect the number of quotes that fell into a theme as some participants talked about a theme more than once over the course of the interview. Themes and subthemes are described below.

Figure 8. Themes identified in Study 2



6.3.2 Theme 1: Beliefs about the condition

Within the theme 'beliefs about condition' four sub-themes were identified:

Sub-theme 1: Treatability of condition

Four participants identified the treatability of the condition in question as important in making their decisions. All four of these participants mentioned PKU and the fact that the description had stated that the condition 'could cause mental problems if left untreated'.

Erm, on the one that I ticked no for (PKU)... it's only 'could cause mental problems if left untreated' and they've just got, would have, a normal lifespan and they've just got to have diet restrictions which... a lot of people have diet restrictions. It's not a big thing. But especially 4 and 5 (Trisomy 13 and anencephaly) they're very much more serious and it was just a definite yes. (Participant 3)

One participant talked about possible changes to medicine in the future for later onset conditions and the possibility that currently untreatable conditions may become treatable in the future (e.g. Huntington's disease).

I thought as well, for these two, I thought well it's quite a long time and there could be changes in medicine by then. Especially this one (Huntington's). It says it's an incurable condition now but in 40 years there might be changes so I thought about that as well. (Participant 11)

Sub-theme 2: Perceived ability to cope with a child with condition

Six participants reported their own ability to cope with the child as affecting their decision. Participants talked about feeling they would be unable to cope practically and emotionally. Two participants mentioned guilt at knowingly bringing a child who has a genetic condition into the world.

I think probably people who have disabled children, even though it's not their fault, I think they probably feel quite a lot of guilt and I think if you already knew that they were going to have problems (pause) I don't think I could live with myself knowing that I'd caused a child to be born into the world with problems. (Participant 3)

Yeah. I would want to continue with my pregnancy. I wouldn't want to terminate the pregnancy. But, yeah, I also wonder, if my child is born with this kind of condition I can take care of my child because I am a nurse and I work at the hospital for disabled children. (Participant 6)

Sub-theme 3: Lifespan of condition

Lifespan was one of the strongest themes with the highest inter-rater reliability. There appeared to be a strong link between lifespan and likelihood of terminating pregnancy/avoiding implantation such that for more severe conditions, likelihood of terminating pregnancy/avoiding implantation was greater.

Yeah. I think I have more of a sort of, idea about that one, wheelchair bound by 11 or 12 and dead by 20 seems awful, mainly because I'm older than 20 but I'm not yet 40... being dead by 20 seems awful but being dead by 40 still seems like a relatively long time. (Participant 8)

... the bottom two (anencephaly and Trisomy 13) I struggle to find a reason why you would have a child if they're definitely going to die. I'm definitely sure of that but erm, I think the other one, number two (Huntingdon's) live to age 40, well lots of things could happen by 40 so I think that's still a very worthwhile 40 years of life and the first one (DMD) is kind of between ages. (Participant 9)

Sub-theme 4: Child's feelings

The child's feelings, how they would cope with their condition and the idea of knowingly 'subjecting' a child to a genetic condition were talked about by 3 participants.

...so I can take care of my child but I will wonder how my child feels about his or her condition. So if I consider the child's feelings, this kind of life is very hard for her or him so that's why it's difficult to answer. (Participant 6)

I'm thinking about the fact that I knew that if I went ahead with this pregnancy this would be the outcome. I don't think it would be fair to the child because I had prior information. (Participant 4)

To summarise, in line with previous research, participants' beliefs about the conditions help to explain the difference in responding according to the condition in question. In particular, participants considered the treatability of the condition, perceptions of their ability to cope with caring for the child, the lifespan of the condition and how the child might feel growing up with the condition. However, this theme alone can not explain differences in responding according to the technology (PND or PGD).

6.3.3 Theme 2: Physical/emotional cost of pregnancy

The physical and emotional costs of pregnancy were relevant to participants for both the PGD and PND conditions. Risks of pregnancy, labour and the emotional costs were illustrated.

I think in the last two where it says the child is going to die very quickly, like within months or very soon then I would say, as well, it depends on whether you already have kids, but it's probably not worth putting your own body through the risk of pregnancy when that's definitely going to be the outcome. You'd probably be better having a go at getting pregnant again. But that's probably not quite the case with the other ones (conditions).

(Participant 9)

Ok, a child would be born without a brain. Like I said, it would be more traumatic. I would already know that I found it difficult to get pregnant naturally then I will not accept to carry a baby that will die after birth. I think that would be more traumatic. (Participant 4)

I just, I don't, I can't imagine putting myself through having a child and then them dying almost instantly or having lots of problems. (Participant 3)

In both the PND and PGD conditions participants considered the physical and emotional costs of carrying a child. It appeared that in some cases participants were weighing up the costs of pregnancy against the beliefs about the condition, whereby for the most severe conditions it may not be worth the physical and emotional costs of the pregnancy if the child is going to die quickly or have significant difficulties. This may explain the finding that the number of participants saying they would terminate/avoid implant is similar for the more severe conditions. It may be that the costs of having a child with a severe condition combined with the costs of pregnancy outweigh beliefs about the technology, discussed below. So, for these more severe conditions, participants

might be more likely to terminate/avoid implantation even if they reject the idea of terminating/avoiding implant on the whole.

6.3.4 Theme 3: Beliefs about the technology

1. Having a choice vs. 'what we've been given'

Five participants contrasted the idea of IVF and PGD being artificial with the idea that pregnancy was more natural. They believed that PGD gave them more choice about whether to go ahead than PND.

I wouldn't really put the same emotional attachment to it as a foetus at that stage. And if it's a naturally conceived foetus then there's a certain amount of 'well this it's what's meant to happen.' It was complete chance and this is the foetus that we've been given and so let's, you know, do what we can. But if it's sort of at implantation stage then it's that much more artificial anyway and I feel you can, sort of, have more choice about it. (Participant 8)

It would be hard to continue with this treatment (IVF). After that if you got an embryo with this kind of condition I think hmmm, I think I would make a choice to continue this treatment to get, how can I say, an alternative embryo. But it's difficult. To select a child, it's difficult to make a choice. (Participant 6)

2. Stage of cells/foetus

The idea of the foetus either being 'a group of cells' or a 'life' and the stage at which testing occurred was mentioned by six participants.

Erm. I felt there was a difference between terminating (pause) is it 10-18 weeks into the pregnancy? And avoiding implantation, erm, because at the point of implantation I think it's not really (pause). It's still in my mind just a bunch of cells at that point. (Participant 8)

This, the pre-implantation one, there's obviously less emotional attachment to an embryo than to something that's already growing inside you.

(Participant 3)

Religion also had an impact on this theme for some participants. An example, where a participant talks about Catholicism and beliefs about the beginning of life, is shown in the following extract:

I'm catholic and I think that has quite a big impact on what I believe about life and er I'm not a strict catholic in some ways but the way I was brought up and taught was that life begins at fertilisation pretty much and also that a disabled life is better than no life at all.

Interviewer: Do you think that would have affected your responses in the questionnaires?

Yeah it definitely did actually because with the first one (PND) I was thinking more along the lines of an actual life. Whereas the second one (PGD), with the embryo, because it's outside of the body and it's not yet implanted that makes me think that it's not (pause) it could become a life but it's not at the same stage. Although I did feel like I was contradicting myself a little bit because an embryo could become a life if it was fertilised. It's difficult.

It is likely that beliefs about the technology explain much of the difference in responding between the PND and PGD conditions in the middle range of severity. As discussed above, for the most severe conditions beliefs about the condition and the costs of pregnancy appear to override beliefs about the technology. Additionally for the least severe, beliefs about condition override the other themes and a large number of participants would go ahead with the pregnancy, regardless of the technology. However, for conditions in the middle range of severity, beliefs about the technology have more of an influence. With

these conditions beliefs about the technology and the stage and moral status of the foetus meant that avoiding implantation was more acceptable than termination.

6.3.5 Theme 4: Something might change

Three participants talked about the possibility that the baby would be ok. Interestingly this theme only emerged for the more severe conditions.

I just think that out of all of them that's (anencephaly) the most likely one where I probably would (go ahead with implantation). Yeah. Something might change. Something might happen that would make it different....my reasoning is not (pause) it's sound in my head but on paper it doesn't really make much sense. (Participant 7)

If you're already pregnant and then they were like 'oh, it might die in the first few months', I don't know whether I'd think in my head 'oh well I'll do it anyway, they might not die'. So I guess you don't know until you're in that situation. (Participant 11)

6.3.6 Interaction between themes

Although four distinct themes were identified there was clear interaction between themes, as illustrated in Figure 9. For example, a number of participants talked about weighing up the physical cost and emotional costs of pregnancy against beliefs about the condition, as illustrated in the following quotes:

Well I thought if it was born without a brain and would die straight away, if it was guaranteed that it was going to be born without a brain then I wouldn't see the point of going through with the whole pregnancy but like the one where it would develop the condition at 40, well that person would still live 40 years and that's, I'm presuming, 40 years of a normal life, so I'd keep it. (Participant 11)

... having a child and then, 9 months and then going through labour and then having the child to die. No. No. Wheelchair bound for life, incurable disease, mental disturbance, I can't. Other people can but I can't.

(Participant 2)

The following quote illustrates the interaction between beliefs about the condition and beliefs about the technology.

Yeah. Erm, it was really difficult. I really struggled with number 2 (Huntington's) with the IVF one. Because by the time you've...if you're healthy until age 40 it seems sort of wrong to prevent that person from being born, but then again, if it's at implantation stage I don't think it is a person that you're preventing from being born yet. But it's very difficult and it's all the sort of, the implantation stuff, it's all a bit like, you feel like you're playing God and trying to create these perfect people and it's slightly uncomfortable. (Participant 8)

Although the majority of participants followed a similar pattern of responding, there were clear exceptions to this. The interaction between the themes identified in this study can help to explain these less common patterns of responding. Two examples will be discussed to illustrate this.

Key for examples 1 and 2	
✓	go ahead with pregnancy/implantation
×	terminate pregnancy/avoid implantation

Example 1 (Study 2):

	PKU	Huntingdon's	DMD	Trisomy 13	Anencephaly
PND	✓	×	×	×	✓
PGD	✓	×	×	×	×

In this example the participant has followed the 'common' pattern of responding for PGD in that they have said they would only go ahead with implantation for the least

severe condition. In this transcript the participant is considering their beliefs about the conditions. For example, in the following extract the participant talks about their own ability to cope with a child who had PKU.

To have a strict diet (pause) a lot of people have strict diets especially children who have diabetes have very strict diets. It seems like something that I could manage.

However, for PND a different pattern is followed whereby the participant says they would go ahead with the pregnancy for PKU, would terminate for Huntington's, DMD and trisomy 13. However, they then indicate that they would continue with the pregnancy for anencephaly, the most severe condition. The following extract illustrates the participant's reasons for this choice which fall into the theme 'There's a chance the baby won't have the condition'.

For here (PND) why I said no (to termination) was that I was hoping. I would pray. I would be hoping for the best that the child would live – that a miracle would happen and it would have some chance. I would want to give this baby a chance. If it died before or soon after birth at least I gave it a chance.

Example 2 (Study 1):

	PKU	Huntingdon's	DMD	Trisomy 13	Anencephaly
PND	✓	✓	x	✓	✓

As Study 1 was online and detailed qualitative information was not collected, it is not possible to know the exact reasons for this participant's response pattern. However, the themes identified in Study 2 allow for hypotheses to be made that could explain this pattern. One hypothesis is that this participant was combining beliefs about lifespan with beliefs about their own ability to cope with the child. So, PKU and Huntington's (due to its late onset) may be perceived as more manageable and

therefore the participant would continue with the pregnancy. Although trisomy 13 and anencephaly are considered to be more severe than DMD, the lifespan is short and the participant may have thought that they would be able to cope with caring for the child for this short time but not for longer. Another explanation may be that the participant was considering how the child might feel growing up with DMD.

The way in which themes interacted varied for each participant. However, it appeared that for most, their decisions reflected a combination of themes one to three. Unlike the themes one to three, theme four 'something might change' appeared to override themes one to three for three participants, as illustrated in the following quote:

For here (PND) why I said no (participant said no to termination for anencephaly) was that I was hoping. I would pray. I would be hoping for the best, that the child would live, that a miracle would happen and it would have some chance. I would want to give this baby a chance. If it died before or soon after birth at least I gave it a chance. (Participant 4)

To summarise, Study 2 identified a number of themes that answer research question 3 – 'if there are differences (in responding according to technology and genetic condition), what are the explanations for these differences?' The themes 'beliefs about the condition', 'physical and emotional costs of pregnancy' and 'beliefs about technology' are the most prominent. Each of these carries different weight according to the severity of condition in question. For example, with PKU, beliefs about the condition and costs of pregnancy have the most influence. Participants tended to believe that having a nearly healthy baby was worth the costs of pregnancy and generally participants would not consider terminating or avoiding implantation in this case. For more severe conditions such as anencephaly, the physical and emotional costs of pregnancy were too great when the child would die before or soon after birth. For conditions considered to be in the middle range of severity beliefs about the condition and costs of

pregnancy also had an influence. However, beliefs about the technology and the status of the foetus in PND versus PGD conditions had much more influence on women's decisions. As discussed above, theme 4 'something might change' seemed to override the other themes for some women.

CHAPTER 7: DISCUSSION

7.1 Summary of main findings

The aims of this research were to answer the following questions:

1. Do women's attitudes towards termination following prenatal diagnosis differ from their attitudes towards non-implantation of embryos following pre-implantation genetic diagnosis?
2. If there is a difference, does this difference vary according to the condition diagnosed?
3. If there are differences, what are the explanations for these differences?

Consistent with previous research (Hewison et al. 2007), the results of Study 1 found that the likelihood of women saying they would consider terminating a pregnancy following PND varied significantly according to the severity of the genetic condition diagnosed. Findings from Study 1 show that similar variation is observed when PGD and the likelihood of women avoiding implantation of an embryo is considered. In addition to this within subjects variation in responding, there were also differences between the PND and PGD groups, such that for the most and least severe conditions (anencephaly and PKU) the difference between likelihood of terminating/avoiding implant was non-significant. However, for conditions placed in the middle range of severity for this study (Huntington's disease, DMD and trisomy 13) the difference between the PND and PGD conditions was significant. This suggests that women's responses were the result of an interaction between the technology (PND and PGD) and the genetic condition diagnosed.

The aim of Study 2 was to develop an understanding of why such differences occur. Four themes and six sub-themes were identified that help to explain the differences between the PND and PGD groups, according to genetic condition.

Theme 1: Beliefs about the condition

Sub-theme 1: Treatability of condition

Sub-theme 2: Perceived ability to cope with a child with condition

Sub-theme 3: Lifespan of condition

Sub-theme 4: Child's feelings

Theme 2: Physical/emotional cost of pregnancy

Theme 3: Beliefs about the technology

Sub-theme 1: Having a choice versus what we've been given

Sub-theme 2: Stage of foetus

Theme 4: Something might change

Beliefs about the condition, physical and emotional costs of pregnancy and beliefs about technology were the most prominent themes and interacted differently according to the severity of condition in question. For PKU, beliefs about the condition appear to have the most influence. As PKU is treatable, generally participants would not consider terminating or avoiding implantation. For more severe conditions such as anencephaly, the emotional and physical costs were too significant if the child would die before or soon after birth. For conditions considered to be in the middle range of severity beliefs about the technology, including the artificiality of PGD versus PND being natural and beliefs about the stage of the foetus, had more of an influence whereby for most people PGD was more acceptable. The 'something might change' theme did not appear to interact directly with the other themes. Instead it seemed to override the other themes, for some women.

In addition to the main findings, both studies found that the majority of participants showed a distinct pattern of responding whereby the likelihood of them terminating/avoiding implant increased as the severity of the genetic condition increases. However, there were a small number of people who did not follow this pattern of responding.

7.2 Findings in the context of existing literature

7.2.1 Attitudes towards PND

The results from Study 1 support the previous findings that the perceived severity of the condition predicts the likelihood of termination for most participants (Mansfield, Hopfer and Marteau (1999). In this study severity has been defined by the percentage of participants who said they would terminate a pregnancy following PND for a number of genetic conditions in a study by Hewison et al. (2007). The implications of measuring severity in this way are discussed in more detail in the methodological considerations section below.

Mansfield, Hopfer, and Marteau (1999) also suggest that timing of the diagnosis and information given to parents about the diagnosis affect decisions about termination. The idea that timing of diagnosis affects decision making may be explained by the 'stage of foetus' theme identified in Study 2 in which a number of participants felt that in PGD the foetus was at an earlier stage and for this reason PGD was more acceptable to them.

Hewison et al. (2007) used Multi Dimensional Scaling (MDS) to determine whether 30 genetic conditions could be grouped in a meaningful way. The MDS analysis revealed a 2-dimensional configuration. The authors were unable to identify one of the dimensions. However the other represented 'seriousness of the conditions' and within this there was a cluster of conditions that stood out from the rest because women would consider termination for these even when they rejected the idea on the whole. These conditions were anencephaly, trisomy 13 or 18, quadriplegia, Duchenne muscular dystrophy, and severe learning difficulties. A similar pattern was found in the current studies in that the majority of participants were more likely to consider termination for the more severe conditions. It is important to note that three of the conditions used in the current studies (DMD, trisomy 13 and anencephaly) were in the 'severe' cluster identified in Hewison et al's study. Whilst the conditions chosen were

spread over the continuum of percentage of people saying they would terminate in Hewison et al's study, future studies may like to include a larger number and variety of conditions in order to see whether similar patterns of responding emerge.

Hawkey's (2005) study investigating whether genetic conditions could be grouped, found that overall women grouped conditions according to 'manageability' and 'lifespan'. Lifespan was one of the subthemes identified for Study 2 and manageability could incorporate a number of the themes and subthemes identified in this study including the 'beliefs about the condition' theme and sub-themes and the 'physical and emotional costs of pregnancy' theme. The other themes identified in this study were 'beliefs about the technology' and 'something might change'. Beliefs about the technology would not have come up in Hawkey's study as it did not ask people to consider testing procedures. Similarly, it did not allow for participants to think that there was a chance the baby would not have the condition as they were grouping descriptions of the conditions. As found in Studies 1 and 2, Hewison et al's (2007) research found significant individual differences in how women grouped the conditions, suggesting that they need to be given information about testing on an individual basis, depending on the person and the condition in question.

Unlike the findings by Bell and Stoneman (2000) and Papp and Papp (2003), religion was not identified as a theme that influenced attitudes towards termination. Although religion was not identified as a theme in its own right, religion appeared to affect a number of different themes for some participants. For example, within the theme 'there's a chance the baby won't have the condition' some participants talked about praying for a miracle, particularly for more severe conditions. Religion also had some affect on the 'stage of foetus' theme, for example, one participant talked about Catholicism and the belief that life begins at conception.

It is important to note that religion was not investigated as part of the quantitative analysis. It may be of benefit for further research to be carried out on the impact of

religion and its influence on attitudes and decision making relating to PND and PGD. Research by Alsulaiman and Hewison (2006, in press) suggests that religion can have an impact on these decisions for some women. For example in their (in press) study they found that PGD was more acceptable than PND for Saudi Arabian parents due to Islamic law which states that PGD is acceptable as long as sperms and oocytes are from the husband and wife.

Previous research has suggested that termination of pregnancy following diagnosis of foetal abnormality is similar to grief following the death of a newborn (Kenyon, Hackett, & Campbell, 1988). Chamayou et al. (1998) observed that couples often decide against having another child after repeated terminations following β -thalassaemia diagnosis. The stress of termination was not identified explicitly as a theme in Study 2. However, this does not mean that it didn't affect people's responses and it may have influenced responses that led to the development of the themes 'stage of cells' and/or 'emotional and physical costs of pregnancy'.

7.2.2 Attitudes towards PGD

Research by Vergeer, van Balen, and Ketting (1998) found PGD to be preferable to PND as it avoids the decision of whether to terminate the pregnancy. The results of Study 1 suggest that for some conditions, avoiding implant following PGD is more likely than terminating following PND. Themes identified in Study 2 help to explain these results and suggest that beliefs about the stage of the foetus affect responding. However, the current studies suggest that the stage of the foetus has an influence for some genetic conditions more than others. Importantly, other factors such as beliefs about the condition in question and the physical and emotional costs of pregnancy also have an influence. For example, for anencephaly, a similar proportion of people would avoid the birth of that child regardless of whether PND or PGD was used. However, with Huntington's disease, significantly more people would avoid implant than would terminate.

Much of the previous research on PGD focuses on the testing process and ethical issues to do with discarding embryos (Chamayou et al., 1998; Fernandez, De Vincentiis, Chillik, & Brugo-Olmedo, 2004; Lavery et al., 2002; Pergament, 1991; Snowdon & Green, 1997). The current studies asked participants to imagine scenarios where testing had already taken place. However, in the additional questions section of Study 1 where participants were asked *'if you would like to elaborate or comment on any of your answers please do so'* several commented that they would not use PGD in the first place. For example, *'I personally would never go through IVF because the borders of playing God are quite shaded'* and *'just to clarify that I don't particularly believe in the need or the right for PGD. However, I have answered the questions on the basis that PGD has already taken place.'* Comments such as these support the previous findings that PGD raises ethical concerns for some people.

Kalfaglou et al. (2005) found that while participants advocated the use of PGD where there was high risk of a serious genetic condition, attitudes varied for less severe conditions and non-medical characteristics. The results of the current studies support Kalfaglou's findings, in that people are generally more likely to avoid implant following PGD for more severe conditions. Reasons for not advocating PGD for less severe conditions included the fact that the disease is not immediately life threatening, that there may eventually be effective treatments and cures, and that affected individuals can still lead productive lives. Similar reasons for going ahead with an implant following PGD for less severe conditions were identified in Study 2, particularly within the 'beliefs about condition' theme. It is important to note that Kalfaglou's research differs from the current studies as it was looking at the use of PGD in the first place, rather than attitudes and decision making following diagnosis.

Vergeer et al. (1998) found that destruction of an embryo after PGD was considered more ethically acceptable than selective abortion. This was reflected in the theme 'beliefs about technology' where participants talked about the impact of the

stage of the foetus on their responses and also the idea that in PND, there is sometimes a fatalistic belief that 'this is what we've been given'. Both panels in the Vergeer et al. (1998) study said that they thought PGD would be used more frequently according to the seriousness of the disorder. The results from Study 1 support the findings that severity of condition affects people's attitudes and that decision to avoid implant following PGD increases in likelihood according to severity of condition, for most participants.

7.2.3 Comparison studies

Chamayou et al.'s (1998) research identified advantages and disadvantages of PGD over PND. Advantages included the elimination of anxiety and stress while waiting for the PND result, the elimination of the psychophysical trauma during selective pregnancy termination in the case of diagnosis and loss (death) in the case of positive PND. Disadvantages of PGD included IVF for fertile couples, ovarian stimulation, oocyte retrieval and the failure to become pregnant after transfer of non β -thalassaemic embryos. Chamayou et al. (1998) found that the greatest advantage was avoidance of selective pregnancy termination in the case of positive PND. Possible failure to become pregnant was considered the greatest disadvantage of PGD by all four groups. Again, this research differs from the current studies as it asked participants to consider the process of PGD/PND rather than to imagine a scenario where it had already taken place. However, it emphasizes the findings from the current studies in that although the process of IVF is stressful, once it has taken place PGD may be preferable in that it avoids termination, as illustrated in the 'beliefs about technology' and 'physical and emotional costs of pregnancy' themes.

Katz et al. (2002) compared the views of people attending a clinic for PGD (for either single gene or aneuploidy screening) with those attending for their first cycle of IVF (control group). They found that overall participants believed that destruction of an embryo prior to implantation was less wrong than the destruction of a foetus in

pregnancy and that there should be restrictions as to who should be offered PGD or limitations on the types of disorders that should be analysed by PGD. Again, this supports the findings that attitudes differ according to technology and the condition diagnosed. However, the current research adds to the literature in that it emphasises that the point at which people say they will avoid implant/terminate occurs at different severity thresholds for the PND and PGD groups.

7.3 Attitudes and behaviour

In the introduction, theories on attitude and behaviour were discussed. These are reconsidered here in relation to the research findings.

Ajzen and Fishbein's (1980) findings that specific rather than general attitudes affect behaviour fit well with the findings in the current studies. Rather than considering whether to give birth to a child with a genetic condition or not, participants were considering the specific procedures involved in avoiding the birth of a child with a genetic condition (PND or PGD) and the specificities of the condition itself.

The theory of planned behaviour (Ajzen, 1985) states that behaviour is determined by intention to carry out the behaviour and perceived behavioural control (see Figure 1 in the introduction). Intention is determined by attitudes, subjective norms and perceived behavioural control. Each of these predictors have their own influences including beliefs about the consequences of the behaviour (attitudes), the subjective likelihood that significant others think the person should perform the behaviour (subjective norms) and beliefs about the ability to perform the behaviour in terms of resources/opportunities (perceived behavioural control). Some components of the theory of planned behaviour do map onto the themes identified in Study 2. The 'beliefs about condition' theme suggests that participants are considering the consequences of termination/avoiding implant and that these factors have a strong influence on their attitudes. The beliefs about technology theme may relate closely to subjective norms in that on a societal level avoiding implant following PGD may be viewed as more

acceptable than termination. However, the influence of significant others was not identified as a theme in its own right in this study. In terms of perceived behavioural control, participants were asked to imagine a scenario where testing had already taken place where they had a significant level of perceived behavioural control. Attitudinal ambivalence (March, 1978; Conner & Sparks, 1995), described in the introduction would not have been identified in this study in relation to the technology used as again, participants were asked to imagine a scenario where testing had taken place. However, it may be more apparent in a clinical situation.

To summarise, it is possible that the TPB could account for some of the themes identified in Study 2. The exception to this is the 'something might change' theme which seemed to override the other themes. The effect of perceived behavioural control and the specifics of the way in which TPB components might interact in this area warrant further investigation.

7.4 Methodological and sampling considerations

7.4.1 Sample

Previous research has been criticised for using a student sample. In these studies both staff and students from the university were invited to take part. This resulted in a reasonably varied sample in terms of occupation and ethnicity. It also meant that the average age of participants in Study 1 (29.3) was older than the average student age and was in fact very close to the average age of mothers at childbirth in the UK (29.1 years in 2000, Office for National Statistics, 2000). However, the sample is likely to be biased in other ways. For example, the socio economic status of staff and students at a university is unlikely to be representative of the whole population. In Study 2 the average age of participants was slightly younger (26) and a smaller percentage of participants currently had children than in Study 1. However, 8 participants said they planned to have children in the future suggesting that the

scenarios were not unimaginable for them. However, it would be beneficial for future research to be conducted on a sample more representative of the general population.

Sample comparisons for age in Study 1 showed that participants in the PGD group were significantly older than those in the PND group, although the effect size was small. However, the same pattern of interaction between genetic condition and method of diagnosis (PND and PGD) was observed in both the pilot study and in Study 2. It is therefore unlikely that this age difference will have affected the pattern of results significantly.

As a self selecting sample was used for both studies it may be that it attracted certain people, for example, those interested in reproductive technology or people with personal experiences of testing. This was illustrated in some of the comments participants left in response to the question *'Do you have any personal experience or knowledge of the conditions described?'* such as *'My younger son has a chromosome mosaic condition. This was not picked up on CVS (it would possibly have been on amniocentesis)'* and *'through work in paediatrics, have seen quite a lot of the above situations.'* In total 65.2% of participants did not have any experience or knowledge of the conditions. However, this re-emphasises the possible benefits of carrying out the study on a wider sample.

It is also important to note that it is rare that women would be making decisions such as these in isolation. In Study 1 a few participants emphasised the influence of partners/family members on their decisions. Although this was not identified as a theme in Study 2, it is possible that participants' perceptions of significant others attitudes had an influence on their responses. It would be beneficial for future research to look at the role of significant others on attitudes/decisions relating to PND and PGD.

7.4.2 Design

A between subjects design was chosen for Study 1 in order that participants were not primed by having already filled in either the PND or PGD questionnaires. The

aim of this was so that their judgment on one technology was not clouded by beliefs about/attitudes towards the other. On reflection it is possible that a within subjects design would have more closely resembled a real life situation where people are often very aware of the options available to them regarding testing.

7.4.3 Analysis

As a time limited initial study it was felt that thematic analysis was sufficient and appropriate for the analysis of interviews in Study 2. This allowed for the identification of simple themes within the data. Future research may like to use a larger sample size, more in-depth interviews and detailed analysis such as grounded theory. This may allow for the development of a model or theory to explain women's attitudes towards the technologies.

7.4.4 Interviewer effects

Overall, the same pattern of interaction between genetic condition and technology was observed in both studies. However, when compared graphically (see Chapter 6), it seems that although the PGD responses were very similar in both studies, the PND responses differed with fewer participants saying they would terminate for the less severe conditions in Study 2. It is possible that interviewer effects contributed to this difference and that participants wanted to portray a positive self image as suggested by Davis et al (2010). If it is the case that interviewer effects affected responses for PND, then this may add support for the previous research suggesting that PGD is more ethically acceptable to people (Kalfoglou et al., 2005).

7.4.5 Validity

A non-clinical sample was used for this study. Some participants reported that they found it difficult to imagine themselves in the scenarios described and some felt their responses may be different if they were actually faced with such decisions. Participants were asked how easy they found it to imagine themselves in the scenarios described and responses varied substantially ranging from 'very easy' to 'very difficult'.

It is important to consider the possibility that results would have been different if people who were currently undergoing PND and PGD were used. Research by Alsulaiman and Hewison (in press) suggests that PGD may be more demanding, psychologically and ethically, than people anticipate, emphasising the difficulties people may have imagining the scenarios. Additionally, the Katz et al. (2002) study identified that people going through PGD had more favourable attitudes towards it than an IVF control group.

In these studies women were asked to make a decision for a number of different conditions. In a real life clinical situation it is likely that they would be making a decision based on one condition. Linked to this, it is also possible that by presenting a number of different conditions, participants automatically compare them and patterns may emerge that would not be present if only one condition was presented. This could be explored further by carrying out a study where participants are only presented with one of the conditions, to see whether similar patterns emerge.

Participants were also presented with a brief summary of PND/PGD procedures and in a clinical situation would be provided with more detailed information. It is therefore possible that participants did not have a thorough understanding of the procedures. For example, for PGD, one participant stated that '*an embryo could become a life if it was fertilised*' suggesting that she did not fully understand the details of PGD. A future study could include questions to check that participants have fully understood the procedures.

In the samples used a number of women reported experiences of PND but only one reported experience of PGD. However, these types of decisions do have the potential to affect any woman and the results of the current studies still offer valuable insight into factors that influence attitudes towards the two technologies.

As mentioned above, severity was defined by the percentage of participants who said they would terminate a pregnancy following PND for a number of genetic conditions in a study by Hewison et al. (2007). It is important to note the limitations of

this definition of severity in that it is based on the results of one study and reflects the majority opinion of a continuum that the authors defined as severity. People who did not follow the response pattern of the majority are not necessarily considering severity less, they may just be measuring severity differently. It is also likely that the conditions are not on one simple continuum of severity but that each has specific factors that contribute to participant perceptions of each condition. For example, age of onset is likely to have influenced decisions for Huntington's and DMD but not the other conditions.

7.4.6 Generalisability

As discussed above, a similar study looking at the interaction between technology and condition using a clinical sample may be of benefit in order to ensure that these results are generaliseable to people actually undergoing PND/PGD.

Previous research suggests that factors such as religion and ethnicity have an influence on attitudes towards PGD and PND (Hewison et al., 2007, Alsulaiman & Hewison, 2006). The present study was UK based and although an ethnically diverse sample was used, it is important to consider the possible impact of cultural and societal beliefs about PND/PGD. For example, results may have been different in a country where PGD is currently illegal as societal rules and regulations are likely to impact on the beliefs of some individuals.

7.5 Clinical implications

The results of these studies show that women's attitudes and decision making about PND and PGD are complex and although the majority of participants followed a similar pattern of responding, in both studies there were exceptions to this. The themes generated in Study 2 can help to explain these less common patterns of responding.

The different patterns and explanations emphasise the findings by Hewison et al. (2007) and Hawkey (2005), which showed that information about screening/testing should be tailored to meet the needs and preferences of individual women. The results

also suggest that for some women and for some genetic conditions, avoiding implant following PGD is more acceptable than termination following PND. This suggests that where possible (e.g. where PGD is available for a particular condition and the condition runs in the family so the risk is known to be increased), women considering diagnoses are likely to benefit from detailed information about both PND and PGD in order to make a fully informed decision as to which is best for them.

7.6 Conclusions

To conclude, these studies add to the current literature in that they suggest that the interaction between the method of diagnosis (PND or PGD) and the genetic condition diagnosed influences women's attitudes towards termination/avoiding implantation. Study 2 identified a number of themes to help explain why such differences occur. It found that the technology used to test had more influence on women's attitudes depending on the severity of the genetic condition in question. It would be of benefit for future research to look in more detail at theoretical explanations for these results, to conduct the study on a wider, more representative sample and also to conduct a similar study on women currently undergoing PND or PGD.

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APPENDIX 1: Pilot study questionnaire PND

Main features of the condition	Imagine you are pregnant and a prenatal test shows that the baby has this condition. Would you consider terminating the pregnancy? Please tick ✓		
	Yes	Not Sure	No
1. Child would develop an incurable condition by age 40, which has both severe mental and physical deterioration, require constant looking after and medical help and have a shortened lifespan.			
2. Child would be at high risk of heart attack (before age 50).			
3. Child would be born without a brain and die before or soon after birth.			
4. Child would have severe learning disabilities/mental handicap, unable to speak or understand, require a lot of looking after and have a nearly normal lifespan.			
5. Child would have a normal lifespan and be extremely short.			

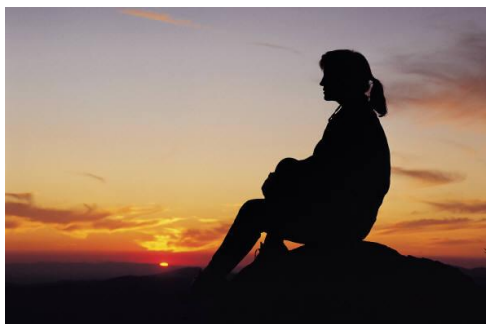
Main features of the condition	<p>Imagine you are pregnant and a prenatal test shows that the baby has this condition. On a scale of 0-100 where 0 = extremely unlikely and 100 = extremely likely, how likely is it that you would terminate the pregnancy?</p> <p>Please place an x on the line e.g.</p> <p>0 _____ 100</p> <p>Extremely unlikely _____ Extremely likely</p>
1. Child would develop an incurable condition by age 40, which has both severe mental and physical deterioration, require constant looking after and medical help and have a shortened lifespan.	<p>0 _____ 100</p> <p>Extremely unlikely _____ Extremely likely</p>
2. Child would be at high risk of heart attack (before age 50).	<p>0 _____ 100</p> <p>Extremely unlikely _____ Extremely likely</p>
3. Child would be born without a brain and die before or soon after birth.	<p>0 _____ 100</p> <p>Extremely unlikely _____ Extremely likely</p>
4. Child would have severe learning disabilities/mental handicap, unable to speak or understand, require a lot of looking after and have a nearly normal lifespan.	<p>0 _____ 100</p> <p>Extremely unlikely _____ Extremely likely</p>
5. Child would have a normal lifespan and be extremely short.	<p>0 _____ 100</p> <p>Extremely unlikely _____ Extremely likely</p>

APPENDIX 2: Pilot study questionnaire PGD

Main features of the condition	Imagine you are having a baby through in vitro fertilisation (IVF) and there is one embryo ready to be implanted. A test shows that the embryo has this condition. Would you <u>avoid</u> going ahead with the implantation? Please tick ✓		
	Yes	Not Sure	No
1. Child would develop an incurable condition by age 40, which has both severe mental and physical deterioration, require constant looking after and medical help and have a shortened lifespan.			
2. Child would be at high risk of heart attack (before age 50).			
3. Child would be born without a brain and die before or soon after birth.			
4. Child would have severe learning disabilities/mental handicap, unable to speak or understand, require a lot of looking after and have a nearly normal lifespan.			
5. Child would have a normal lifespan and be extremely short.			

APPENDIX 3: Study 1 recruitment poster

Are you female and aged between 18 and 50?



I am looking for women aged between 18 and 50 who are not pregnant to take part in an online study looking at women's attitudes towards different types of reproductive technology.

For more information please contact
Chloe Miller

email: reproductivetechnology@googlemail.com

or go to

www.reproductivetechnology-survey.nonblinkingeye.com

100 printer credits will be given to the first 100 people to complete the questionnaire.

APPENDIX 4: Study 1 questionnaire - PND

APPENDIX 5: Study 1 questionnaire - PGD

APPENDIX 6: Signposting information for participants Study 1

Thank you for taking part in this study.

If you feel distressed after taking part in this study, you can contact me at:
reproductivetechnology@googlemail.com.

Alternatively you may find it helpful to contact one of the following:

Leeds Student Counselling Centre: 0113 3434107

Student Medical Practice: 0113 2954488

Nightline: 0113 3801290

Chaplaincy: 0113 3435071

Welfare Office: 0113 3801300

Samaritans: 08457 909090

APPENDIX 7: Study 2 recruitment poster

Are you female and aged between 18 and 50?



I am looking for women aged between 18 and 50 who are not pregnant to take part in an interview study looking at women's attitudes towards different types of reproductive technology.

This will involve filling in two questionnaires and then answering questions about your responses in a short interview.

For more information please contact
Chloe Miller

email: reproductivetechnology@googlemail.com

500 printer credits will be given to all participants.

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APPENDIX 8: Study 2 participant information sheet

Women's Attitudes Towards Reproductive Technology Participant Information Sheet

Developing an understanding of women's attitudes towards reproductive technology

We would like to invite you to take part in a research study. Before you decide you need to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully. Talk to others about the study if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

What is the purpose of the study?

Advances in reproductive technology mean that it is now possible to test for a wide range of genetic conditions using a variety of different tests. The aim of this study is to develop an understanding of women's attitudes towards different types of testing for a number of genetic conditions.

Why have I been invited to take part?

You have been invited to take part because you are female, aged between 18 and 50 and you are not currently pregnant.

What will I be asked to do if I take part?

You will be asked to fill in two questionnaires which ask how you would respond to a positive test for a number of different conditions, for two different types of technology. You will then be asked to take part in a short interview about your responses to the questionnaires. This interview will be tape recorded. The questionnaires and interview should take approximately 45 minutes to complete.

Will my data be confidential?

All information collected is confidential. I will need your email address in order to give you free printer credits. However, this will be kept separately to your questionnaire and interview responses. Tape recordings will be transcribed and will then be deleted.

Do I have to take part?

You do not have to take part in the study. If you decide to take part and then later change your mind, either before you start the study or during it, you can withdraw without giving your reasons.

Where can I obtain further information if I need it?

If you have any questions or would like any further information please contact:

Chloe Miller

email: reproductivetechnology@googlemail.com

Supervised by:

Professor Stephen Morley and Professor Jenny Hewison

This project has been approved by the LIHS/LIGHT Research Ethics Subcommittee.

APPENDIX 9: Study 2 consent form

Consent Form

Women's attitudes towards reproductive technology

Please delete as applicable

I have read and understood the participant information sheet. YES/NO

I have had the opportunity to ask questions and discuss the research. YES/NO

I am satisfied with the answers to my questions. YES/NO

I have received enough information about this research. YES/NO

I understand that my participation is voluntary and I am free to withdraw from the research at any stage without giving a reason. YES/NO

I agree to take part in this research. YES/NO

Signature.....

Name.....

Date.....

APPENDIX 10: Study 2 demographic questionnaire

Demographic information

1. Are you currently pregnant?

Yes*

No

**If you answered yes to this question, please do not continue with this study*

2. How old are you? _____

3. What is your current occupation?

Undergraduate student

Postgraduate student

Academic staff

Admin/clerical staff

Other (please specify) _____

4. What is your ethnic group?

Choose ONE section from A to E, then tick the appropriate box to indicate your ethnic group.

A. White

British

Other (please specify) _____

B. Mixed

White and Black Caribbean

White and Black African

White and Asian

Other (please specify) _____

C. Asian or Asian British

Indian

Pakistani

Bangladeshi

Other (please specify) _____

PTO

D. Black or Black British

 Caribbean African Other (*please specify*) _____

E. Chinese or other ethnic

 Chinese Other (*please specify*) _____**5. How would you describe your religious beliefs?**

6. Do you have any children? Yes No

If yes, how many? _____

7. Do you plan to have children in the future? Yes No Don't know

If you would like to receive printer credits for taking part in this study I will need your university email address. This will be kept separately to the rest of your questionnaire responses.

Email address _____

APPENDIX 11: Study 2 PND information sheet and questionnaire**Prenatal diagnosis (PND)**

Prenatal diagnosis (PND) is offered to women when they are considered to be at risk of carrying a foetus with a genetic condition. Two commonly used methods of PND are amniocentesis and chorionic villus sampling (CVS). Amniocentesis involves having a needle inserted into the abdomen. The abdomen can be numbed with anaesthetic which involves a small injection which may sting slightly. A needle is then inserted through the abdominal wall and into the amniotic sac of fluid surrounding the foetus. A syringe is then used to remove a small sample of amniotic fluid from the amniotic sac for analysis in a laboratory. Chorionic villus sampling obtains a small sample of the chorionic villi (placental tissues), either by passing a thin needle through the wall of the abdomen, or by passing a small tube through the vagina and the neck of the womb (cervix). Both procedures are carried out under the guidance of ultrasound scanning.

These procedures are carried out between 10 to 18 weeks into the pregnancy. Both carry some risk of miscarriage (between 1.5-2.5%). If the foetus is diagnosed with a condition through either procedure, women have the option of terminating the pregnancy.

Main features of the condition	Imagine you are pregnant and prenatal diagnosis shows that the baby has this condition. Would you consider terminating the pregnancy? Please tick ✓	
	Yes	No
1. A male child would have a progressive muscle wasting disease, be wheelchair-bound by 11 or 12 years and have a much shortened lifespan (death probably before 20 years of age).		
2. Child would develop an incurable condition by age 40, which has both severe mental and physical deterioration, require constant looking after and medical help and have a shortened lifespan.		
3. Child would have a blood condition that could cause mental problems if left untreated, have a normal lifespan and have strict diet restrictions throughout life.		
4. Child would be born without a brain and die before or soon after birth.		
5. Child would have severe learning disabilities/mental handicap, require a lot of looking after and die within first few months of life.		

APPENDIX 12: Study 2 PGD information sheet and questionnaire**Pre-implantation genetic diagnosis (PGD)**

Pre-implantation genetic diagnosis (PGD) is offered to women when they are considered to be at risk of carrying a foetus with a genetic condition. Pre-implantation genetic diagnosis involves screening an embryo created through in vitro fertilisation (IVF). The IVF process initially involves taking fertility drugs to stimulate egg production. Eggs are then retrieved under the guidance of ultrasound. A needle is inserted through the vaginal wall into the ovaries and eggs are then collected through a special catheter. This procedure takes around 20-30 minutes. Some patients may experience some discomfort during and after this procedure. Following egg retrieval women are given hormones to prepare the uterus for pregnancy, while the eggs are fertilised with the sperm in a laboratory. Once the eggs have been fertilised PGD takes place. If the embryo is diagnosed with a condition through PGD women then have the option of avoiding implantation.

Main features of the condition	Imagine you are having a baby through in vitro fertilisation (IVF) and there is one embryo ready to be implanted. Pre-implantation genetic diagnosis shows that the embryo has this condition. Would you consider <u>avoiding</u> going ahead with the implantation? Please tick ✓	
	Yes	No
1. A male child would have a progressive muscle wasting disease, be wheelchair-bound by 11 or 12 years and have a much shortened lifespan (death probably before 20 years of age).		
2. Child would develop an incurable condition by age 40, which has both severe mental and physical deterioration, require constant looking after and medical help and have a shortened lifespan.		
3. Child would have a blood condition that could cause mental problems if left untreated, have a normal lifespan and have strict diet restrictions throughout life.		
4. Child would be born without a brain and die before or soon after birth.		
5. Child would have severe learning disabilities/mental handicap, require a lot of looking after and die within first few months of life.		

APPENDIX 13: Study 2 semi structured interview protocol**Interview protocol****1. General question**

- Thank you for filling in the questionnaire. Is there anything that particularly influenced your responses when filling in the questionnaire?
- In what way did x affect your responses?
- Do you think that x affected different conditions / technologies in different ways?

2. Examples of questions about specific responses

- I noticed that you responded 'yes' to avoiding implantation but 'no' to termination. Could you tell me a bit more about why that is?
- I noticed that you responded 'yes' for everything. Could you tell me a bit more about why that is?
- I noticed that for some conditions you responded yes to avoiding implantation and no for termination and for others you responded yes for both. Could you tell me a bit more about why that is?

3. Is there anything else you would like to say about your experience of filling in the questionnaires.**4. Do you have any questions?**