

LONDON  
SCHOOL of  
HYGIENE  
& TROPICAL  
MEDICINE



LSHTM Research Online

Shakespeare, Tom; Hull, Richard; (2018) Termination of Pregnancy After Non-Invasive Prenatal Testing (NIPT): Ethical Considerations. *JOURNAL OF PRACTICAL ETHICS*, 6 (2). pp. 32-54. ISSN 2051-655X <https://researchonline.lshtm.ac.uk/id/eprint/4651219>

Downloaded from: <http://researchonline.lshtm.ac.uk/4651219/>

DOI:

**Usage Guidelines:**

Please refer to usage guidelines at <https://researchonline.lshtm.ac.uk/policies.html> or alternatively contact [researchonline@lshtm.ac.uk](mailto:researchonline@lshtm.ac.uk).

Available under license: <http://creativecommons.org/licenses/by-nc-nd/2.5/>

<https://researchonline.lshtm.ac.uk>

# Termination of Pregnancy After Non-Invasive Prenatal Testing (NIPT): Ethical Considerations

Tom Shakespeare

*International Centre for Evidence in Disability, London School of Hygiene and Tropical Medicine*

Richard Hull

*Philosophy & COBRA, NUI, Galway*

## ABSTRACT

This article explores the Nuffield Council on Bioethics' recent report about non-invasive prenatal testing. Given that such testing is likely to become the norm, it is important to question whether there should be some ethical parameters regarding its use. The article engages with the viewpoints of Jeff McMahan, Julian Savulescu, Stephen Wilkinson and other commentators on prenatal ethics. The authors argue that there are a variety of moral considerations that legitimately play a significant role with regard to (prospective) parental decision-making in the context of NIPT, for example, views on the morality of abortion and understandings of the impact of disability on quality of life. The variable nature of such considerations, both singularly and combined, suggests that any approach to NIPT should be sensitive to and understanding of similarly variable parental assessments and decisions. The implications of the approach developed for current and future policies in this area are explored, along with the impact of such arguments on ideas about procreative beneficence.

## INTRODUCTION

While the ethical and social dimensions of prenatal testing and selective termination of pregnancy have been debated for many decades (Shakespeare 2006), the advent of non-invasive prenatal testing (NIPT) and its rapid and widespread adoption has reawakened these debates, together with associated anxieties about eugenics and equality. Because NIPT removes the risk of iatrogenic miscarriage associated with invasive procedures such as chorionic villus sampling (CVS) and amniocentesis, there seems to be less reason for pregnant women and their partners to be cautious about consenting to screening for Down syndrome and other conditions (Chitty et al 2016). Moreover, NIPT is more accurate in identifying Down syndrome than previous screening tests (Taylor-Phillips et al 2016), and appears simpler to understand, at least in the case of Down syndrome screening. This suggests that more pregnant women are likely to opt for screening without fully considering the implications. This in turn means more identification of affected pregnancies, possibly at an earlier stage of pregnancy. Given that upwards of 90% of pregnant women who are identified carrying a pregnancy affected by Down syndrome currently chose to terminate, this suggests that there may be more selective abortions, and potentially fewer babies born with Down syndrome or other trisomies.<sup>1</sup> However, the trend towards women becoming pregnant later in life means a somewhat higher incidence of Down syndrome and other trisomies, so this has to be set against the increase in diagnosis and selective abortions.

Further, NIPT may increase the number of other conditions that are detectable prenatally. Until now, mainstream screening has been largely limited to trisomy screening and detection of anomalies on ultrasound. NIPT is already being used to identify microdeletions in the genome, where the test has a much lower accuracy. The universal acceptance of NIPT raises the option of whole exome or genome sequencing (WES/WGS). The cost of sequencing has been reducing year on year

1. Down syndrome results when a fetus carries 3, rather than 2 copies of chromosome 21, hence 'trisomy'. Other common trisomies affect chromosome 18 (Edwards syndrome) and 13 (Patau syndrome). These trisomies affect infants more significantly than Down syndrome: half of children with Edward syndrome die within a week of birth, with less than 10% of surviving to age 10; more than 80% of children with Patau syndrome die within the first year of life, and around 13% survive to age 10.

for decades: WES will soon be cheaper than \$500. Sequencing has the potential to identify both significant and insignificant genetic conditions, as well as variants of unknown significance (VUS). For example, in some clinics in the USA, prospective parents are offered WES if any structural anomalies—such as unusual nuchal translucency findings—are detected on ultrasound or through other screening tests, with three-quarters accepting the offer. These developments may have a number of implications. Information about microdeletions and single gene conditions will be more uncertain, more complex, harder to understand, and demand more genetic counselling support. Where the risk of a genetic condition is high, more invasive tests may be required for accurate diagnosis, and these tests may cause more iatrogenic miscarriage. More conditions may be identified prenatally, at an earlier stage of gestation. This may result in earlier terminations of pregnancy.

It should be noted that NIPT is not diagnostic for trisomies or many other changes (Taylor-Phillips et al 2016). In a high chance population (women who have been identified by serum screening as being at a higher than 1:150 chance of carrying an affected fetus) the positive predictive value for Down syndrome may be as high as 91%; whereas in the general population the positive predictive value falls below 82%, meaning that 18 out of 100 pregnancies identified are not in fact affected by Down syndrome. The test performs even worse for the other trisomies, where positive predictive value in the general population is less than 50%. This explains why NIPT is better offered as a second tier screen after serum screening, as the NHS National Screening Committee proposes, and why a diagnostic test is still required to confirm that the pregnancy is affected (NSC 2016).

The Nuffield Council on Bioethics is an independent advisory body in the UK, funded by the Medical Research Council, Nuffield Foundation and the Wellcome Trust. It is often considered to be the closest that the UK comes to having a national bioethics committee. In 2016-17, a working group of the Nuffield Council on Bioethics spent approximately one year considering the ethics of NIPT resulting in a report that was published in March 2017. The working group, comprising expertise in law, philosophy, psychology, public health, social science, and genetics, deliberated the issue with input from both experts in the field and lay people, including people with lived experience of genetic conditions. The deliberations of the working group thus drew on empirical data—about scientific and clinical practices, and about public opinion—and on normative arguments, as well as on responses to an open consultation.

The working group's inquiry into NIPT considered all kinds of uses of NIPT—for screening for trisomies (sometimes called Non-Invasive Prenatal Screening, NIPS); diagnosing single-gene conditions (sometimes called Non-Invasive Prenatal Diagnosis, NIPD); and for determining fetal sex. The working group also looked to the future, when whole genome sequencing of fetuses using NIPT might become clinically available. The inquiry considered both public sector provision of NIPT in the NHS, but also explored how NIPT was marketed and provided in the commercial sector.

The working group developed an ethical approach to the formation of policy in this area based on the three sets of values:

1. The value of reproductive choice;
2. The importance of reducing or avoiding harm to different individuals and groups;
3. The importance of ensuring equality and social inclusion.

This led the working group to support the offer, whether in the NHS or the private sector, of NIPT for significant medical conditions and impairments, but to argue that certain preconditions would have to be met. These preconditions included: that the test gave an accurate prediction of the condition being tested for; that women/couples were provided with high quality information and support; that disabled people receive high quality health and social care; and that the discrimination and exclusion experienced by disabled people was tackled. The working group had particular concerns about availability of information and genetic counselling in the private sector.

The working group had more concerns about extensions to NIPT, particularly in the private sector. The report argued that NIPT should not normally be used to test whether a fetus has a less significant medical condition or an adult onset condition, or to find out if the fetus is the carrier of a gene for a medical condition. The report argued that NIPT should not be used to disclose the sex of the fetus at the 10-week stage, or for non-medical traits at any stage. In particular, the report argued that there should be a moratorium on the use of whole genome sequencing with NIPT. The argument was that WGS information had limited clinical utility, that information

would create anxiety, that it would lead to more confirmatory invasive testing, and that lay people generally lacked the information and support necessary to make an informed and ethical decision. The working group made an exception for situations where there is a family history of genetic conditions, or where NIPT is used diagnostically after anomaly has been detected on ultrasound.

The authors will end up agreeing with the Nuffield Council on Bioethics recommendations on these points, perhaps unsurprisingly given that one author chaired the working group. However, the current paper offers a chance to go into the question of the ethics of NIPT in more depth, and to consider the recommendations more philosophically, particularly regarding what might appear to be an inconsistency in the Council's position. We will show that, given the considerations we explore, the Council's position need not be regarded as inconsistent.

#### NUFFIELD COUNCIL ON BIOETHICS RECOMMENDATIONS

As can be seen, in this report, the Nuffield Council on Bioethics adopted a compromise position on NIPT and selective termination. Contrary to the advocacy of the Down Syndrome community—represented by highly vocal campaigns such as 'Don't Screen Us Out'—the Council supported, with qualifications, the provision of NIPT services as a second tier screening test for trisomies in the NHS. In other words, it would be offered to women who were found to be at a higher chance (greater than 1:150) of having a pregnancy affected by Down syndrome as a result of first tier serum screening. But contrary to the advocacy of pro-choice advocates such as the British Pregnancy Advisory Service, the Council recommended prohibition of NIPT for sex selection, and a moratorium on whole genome sequencing/exome sequencing.

The report justified this compromise position in terms of a balance between the ethical value of choice, and the countervailing values of avoiding harm, and promoting equality and inclusion. For example, on trisomy, the Council argued that choice should be balanced with more extensive information provision, balanced and non-directive prenatal counselling, and welcoming of Down syndrome babies.

To take another example, the Council argued that NIPT for sex selection would be intrinsically sexist, and could lead to undesirable social consequences. Moreover, in a global context where some countries—China, India and some Middle Eastern countries—have used prenatal testing to favour male rather than female births with consequences for their national sex ratios, the Council argued that the UK should

not undermine the international efforts to counter this deleterious social practice by permitting NIPT for sex selection.

Yet in recommending restrictions on the use of testing and implicitly the availability of information to pregnant women and their partners, the Council could be accused of paternalism. Indeed, this was exactly the charge levelled by the British Pregnancy Advisory Service.

While the Report explored ethical issues, placing NIPT in the context of values such as choice, avoidance of harm, and equality and inclusion, it did not give a closely argued justification of the position adopted. In recommending that prospective parents be able to use NIPT to discover the trisomy status of the fetus, the Report followed the ethos of reproductive autonomy and informed choice. However, when it came to NIPT for sex selection or NIPT with whole genome/exome sequencing, the Report took the position that this information should essentially be denied to the parents, unless there was a compelling reason arising from family history or ultrasound indication of anomaly.

The resulting recommendations are thus vulnerable to the accusation of inconsistency, as well as paternalism. A straightforward and consistent position to adopt could be that parents are able to find out all available information about their fetus, and that women should have control over their own bodies, and thus the right to terminate on any grounds. This is what the principle of reproductive autonomy might demand. But in the era of whole genome sequencing and perhaps particularly given that NIPT can convey results at week 10-12, rather than week 18-20, the implications are troubling to those who want to see an acceptance of diversity and who worry about the dangers of “laissez faire eugenics” (Kitcher 1996). If prospective parents can discover every difference or anomaly at an early stage of pregnancy, then the scenario envisaged by Barbara Katz Rothman (1993) in *The Tentative Pregnancy* can come to pass. Particularly for those—middle class and empowered people—who are accustomed to choice and control in their lives, it might be tempting to reject any fetus which carries an obviously deleterious allele—not just chromosomal anomalies like Down syndrome, Edward syndrome, Turner syndrome, Klinefelter’s syndrome, Patau, not just serious genetic conditions such as haemophilia and cystic fibrosis, but also anything untoward, however trivial, including unwanted social or behavioural traits, such as being female or having a higher chance of being gay or on the autistic spectrum. The implications are also troubling for anyone who worries about increasing numbers of terminations of pregnancy, and who might consider that such a morally weighty act

can be best justified on the grounds of characteristics of the fetus which are significant and non-trivial. While all this seems worrying enough, some theorists go further and argue that it is now morally wrong to have disabled children when the severity of a genetically transmitted condition is “great enough that particular parents are morally obligated to prevent it, given the specific means necessary for them to do so” (Buchanan, Brock, Daniels and Wikler 2000, 243).

### PROCREATIVE BENEFICENCE AND NIPT

Several philosophers would place on pregnant women and their partners a particular duty, when it comes to reproductive choices. In the context of pre-implantation genetic diagnosis, rather than prenatal screening, Julian Savulescu writes: “couples (or single reproducers) should select the child, of the possible children they could have, who is expected to have the best life, or at least as good a life as the others, based on the relevant, available information.” (Savulescu 2001, 415).

In a slightly different formulation, Jeff McMahan presents his ‘Impersonal Comparative Principle’: “If in either of two possible outcomes the same number of people would ever live, it would be worse if those who live are worse off, or have a lower quality of life, than those who would have lived” (McMahan 2005, 145). Neither McMahan nor Savulescu makes the mistake of holding that a person is harmed by being brought into existence in these cases, even if the person’s quality of life is low. Their position acknowledges Parfit’s famous non-identity problem, which is brought in to play given that these cases involve identity-affecting choices (Parfit 1984). The problem is expertly articulated by Buchanan, Brock, Daniels and Wikler where they point out that, in the case of disability, for example, “a person’s disability uncontroversially leaves him or her with a worthwhile life”, so “it would not be better for the person with the disability to have had it prevented, since that could only be done by preventing him or her from ever having existed at all” (Buchanan, Brock, Daniels and Wikler 2000, 245).

Nonetheless, the implication of McMahan’s and Savulescu’s claims, particularly Savulescu’s, is that, where the same number of people will exist, there is a positive duty on prospective parents to exercise procreative beneficence and thus to produce or select the best fetus and hence best child they can.

The practical implications of the duty of procreative beneficence are hard to understand. For those people who are at known risk of passing on genetic conditions



to their child, then procreative beneficence presumably demands and must surely recommend that they either access PGD services, or use prenatal diagnosis and selective termination if such conditions can be avoided. One might think that it would also recommend forgoing reproduction in favour of adoption or childlessness where they cannot be avoided, yet the principle is consistent with the view that if the only possibility is to have a child with a given condition, then it is acceptable to go ahead and do so (Savulescu 2001, 415).

The very many couples affected by skeletal dysplasia, or deafness or other avoidable conditions who knowingly reproduce and have children with their own conditions might be guilty of violating the Reproductive Beneficence Principle, and potentially the Impersonal Comparative Principle. However, this depends on what is meant by “the best life, or at least as good a life as the others” and understandings of the comparative “worse off, or lower quality of life”. Many disabled people report that they have as good a quality of life as non-disabled people, and indeed quality of life evidence suggests that measures of well-being and quality of life are indeed comparable among people with and without disability (Amundsen 2010).

For those who are not at any known chance of carrying a baby with a genetic condition, then these two Principles demand caution when it comes to reproduction, perhaps specifically imposing a duty that people should avail themselves of all available screening services, and terminate selectively. With the advent of NIPT, this would suggest using this new screening service, in association with whole genome/exome sequencing, to avoid possibly deleterious alleles (ignoring the point that it would not be a trivial problem to ascertain what might be deleterious, neutral or even positive in the context of a particular genome and socio-historical environment).

As Rothman argued in *The Tentative Pregnancy*, these Principles place a potentially draconian obligation on women who wish to reproduce, and their partners. Rather than leaving the fate of their prospective offspring to karma, God or the random assortment of genes, prospective parents would have to go through the stress of waiting to get pregnant, then a screening test—with possible need for confirmation via diagnosis invasive test—then termination if any of the alleles turn out to be problematic.

Given that every genome contains around 100 mutations, it might be thought that almost every genome has something that could raise alarm. This being so, the prospective mother and father, as Rayna Rapp (1999) has written, are turned into bio-ethicists, weighing up whether this life or that life would have “at least as good a life as the others”, often in the absence of good empirical evidence about the natural history

of genetic conditions, let alone the lived experience of people who themselves have the condition.

Prospective parents are turned not only into bioethicists but also, arguably, into consumers, considering whether this or that possible offspring would be of sufficient quality, or should be rejected. Data on the sequelae of abortion is scant and contested, but the implications of serial quality control terminations of pregnancy must be daunting to at least some prospective parents. Therefore, we would argue that, if they were applied in the context of NIPT, the implications of the purported duty of Reproductive Beneficence or the Impersonal Comparative Principle are repellent.

Of course, one could restrict the application of a principle of Procreative Beneficence to selection cases involving pre-implantation genetic diagnosis during IVF, when decisions regarding which embryo to implant must often be made regardless, and thereby avoid some of these worries. Moreover, termination of pregnancy might be considered to be an issue that would override the demands of Procreative Beneficence if applied in the context of NIPT. However, it is not at all obvious that this need be or is the case. For example, Savulescu asserts that his argument “extends in principle to selection of fetuses using prenatal testing and termination of affected pregnancy” (Savulescu 2001, 421) and it is hard to see why prospective parents would not avail of that option if they considered Procreative Beneficence to be *the* principle, or the most significant one, to follow. On the other hand, Savulescu acknowledges the psychological harms associated with termination of pregnancy (Savulescu 2001, 421) and states that Procreative Beneficence is a principle that must be balanced against others (Savulescu 2001, 425). We acknowledge here too, that the application of a principle of Procreative Beneficence in the context of NIPT would involve some controversial, even if reasonably common, assumptions about the nature and moral status of human development (that might impact in turn on beliefs about termination of pregnancy explored below). That is, at the particular stage of pregnancy in a given case, the potential life in question would have to be considered as not yet being a morally relevant entity that can be harmed or benefitted. Altogether then, it would seem that either the principle can be read as quite a radical thesis whereupon the significant concerns outlined above remain or, more generously, it is not as controversial as it can first appear. It can be limited in application to cases of pre-implantation genetic diagnosis during IVF, leaving open the contentious issue of whether or not advocates of Procreative Beneficence believe that PGD during IVF should become

the procreative norm. Moreover, both with IVF and in a more general sense, it is a principle that is one of many contextual considerations that we explore below.

Some notion of beneficence is likely to play a significant role in (prospective) parental decision-making. Michael Parker argues, for example, that

*insofar as we have reason to believe that it is possible to say something meaningful about the conditions under which it is possible to live a good life, the concept of beneficence will have content and will require of us, where we have choice and insofar as it is possible to discern, that we choose to reproduce in ways that make it possible for our children to grow up under such conditions (Parker 2007, 283).*

In addition, however, there are a variety of other considerations that, we suggest, legitimately factor in to such decisions. They will likely combine as well, to add content to any notion of beneficence.

One such consideration in the context of NIPT is a (prospective) parent's beliefs about termination of pregnancy. It goes without saying that most, if not all, of the considerations discussed here are intensely personal, and views about termination of pregnancy are no exception. Indeed, they tend to be held very deeply and so, in turn, can have a critical role in deliberations. Some views will also have different implications for decision-making, depending on the stage of the pregnancy in question.

The Nuffield Council report, for example, cites research exploring the factors that influence women's decisions to continue or terminate a pregnancy following a diagnosis of fetal anomaly. Reasons for continuing a pregnancy after a diagnosis include, amongst other things, "religious beliefs" and "not wanting to experience a termination" (Nuffield Council 2017, 11). Karpin and Savell similarly suggest that what women are prepared to do to prevent the birth of a child who has an abnormality or genetic condition or disease is a crucial contextual matter (Karpin and Savell 2012, 283). They also observe that, as a pregnancy proceeds, the willingness to terminate that pregnancy can decrease, citing evidence that the stage of gestation correlates with clinicians' willingness too, to facilitate termination for certain disabilities (Karpin and Savell 2012, 283).<sup>2</sup>

This is reflected in a concern expressed in the Nuffield Council report, that of-

2. Karpin and Savell (2012, 283) further postulate that the interaction between severity and gestational age might work in reverse, "that is, that the threshold for seriousness might be lower early in pregnancy or before pregnancy".

fering NIPT only to women after they have had an initial combined test may lead to a delay in diagnosis for some women. The report states that such a delay (of a week or longer) “will be significant to some women, particularly those considering a termination” (Nuffield Council 2017, 42). The significance is said to lie in the fact that some research suggests that later terminations are associated with higher levels of stress and therefore increased harm to women, at least in the short term, which is clearly likely to be bound up with women’s considered beliefs about the issue and procedure. The report thus recommends that “women are able to go straight to diagnostic testing after a high chance combined test result if they wish” (Nuffield Council 2017, 42).

It is obvious, then, that beliefs about the issue of termination of pregnancy can have a significant impact on decision-making with regard to NIPT. Indeed, the significance of the issue and its impact is highlighted further in the Nuffield report, where it is said that the decision to terminate a pregnancy following diagnosis of fetal anomaly “is frequently described by pregnant women and couples as shocking, painful and distressing, with some reporting feeling unprepared for making such a decision” (Nuffield Council 2017, 11). The report cites research from The Netherlands, which found that “a significant number of women experienced post-traumatic stress symptoms and depression in the 16 months following the termination, particularly among those who felt high levels of doubt during the decision-making period, lacked partner support, were religious, and were at more advanced stages of pregnancy” (Nuffield Council 2017, 11-12). This was the case even though most women did not report regretting their decision to have a termination (Nuffield Council 2017, 12), which both illustrates the gravity of the issue and implores a deep sensitivity to women’s considered decisions in such cases (in either direction).

Decisions to terminate pregnancy on the basis of a diagnosis of fetal anomaly are made on a number of grounds, according to the Nuffield Council report (Nuffield Council 2017, 11). They correspond to three loosely defined and interrelated sets of considerations that, along with beliefs about the issue of termination, legitimately factor in to (prospective) parental decision-making. The first involve an evaluation of the impact of the diagnosed condition on the life of the prospective child. The second involve an evaluation of the impact of having a child with the diagnosed condition on the parent(s) and other family members, along with consideration of (prospective) parental hopes and aspirations. The third involve the social, economic and personal context in which a decision is being considered. While balancing such con-

siderations is no easy task, we want to suggest that any serious and sensitive evaluation will attempt to do so.

A diagnosed condition is likely to concern us because of the impact that it might have on a prospective child's life or, as Jonathan Glover puts it, their capacity to flourish. In the context of disability, for example, Glover argues that it

*requires failure of functioning. But failure of functioning creates disability only if (on its own or via social discrimination) it impairs capacities for human flourishing. It would not be a disability if there were a failure of a system whose only function was to keep toenails growing. With arrested toenail growth, we flourish no less (Glover 2006, 9).*

Similarly, having one gender or another does not impair capacity for flourishing.

Here, then, the concern of (prospective) parents speaks “not to imperfection, but to human suffering” (Karpin and Savell 2012, 155). Moreover, as Wilkinson rightly points out in his analysis of Glover, the relationship between a diagnosed condition and human suffering is indirect because “the a priori connection is not between being disabled and *not flourishing* but rather between being disabled and having a reduced *capacity to flourish*” (Wilkinson 2010, 63). Clearly, as has already been described, individuals with disabilities can and do live flourishing lives, which leads Wilkinson to argue that it is inadvisable to generalize about the relationship between disability and quality of life (Wilkinson 2010, 68). Having said that, the impact that a diagnosed condition is likely to have on a child's capacity to flourish is an obvious concern of (prospective) parents, however difficult it is to assess. We note here the danger that prospective parents may be ignorant about, or even biased against disability, given the prevailing negative valuation of disability in modern societies (Kaposy 2018).

Judging the impact of a given condition on a potential child's capacity to flourish is very problematic and controversial. It is perhaps least controversial in what tend to be described as ‘wrongful life’ or ‘sub-zero’ cases.<sup>3</sup> These are cases where it is said that a prospective child's life will not be worth living or, put another way, that the life created will contain negative net utility. Wilkinson argues that a child with a negative quality of life

3. This is not to deny that some people's beliefs about termination of pregnancy may preclude that course of action, even in these types of case.

*does have a valid complaint and can legitimately claim that it has been harmed by being created. For if the child could compare the state of affairs in which it exists (one with sub-zero quality of life) with another in which it does not (one with no life, and hence no quality of life) it would (and rationally should), other things being equal, prefer the latter (Wilkinson 2010, 70-71).*

Wilkinson goes on to contend that wrongful life cases are extremely rare. “Most people with disabilities have a positive quality of life, even if their disabilities, or society’s responses to them, cause them to have a lower quality of life than that of the average non-disabled person” (Wilkinson 2010, 71). Thus, unless we wanted to raise the threshold of wrongful life to include the types of conditions tested for via NIPT, which would be very contentious, the concept of wrongful life has little to offer (prospective) parents when trying to assess the impact of a diagnosed condition in the context of NIPT.

Above the wrongful life threshold, it is very hard to know what we can legitimately say about decisions made on the basis of an assessment of the likely impact of a diagnosed condition. Given the non-identity problem described earlier, we cannot claim that it is wrong to continue with a pregnancy after most diagnoses of fetal anomaly, since it will likely result in a child with a worthwhile life who prefers to exist than to not exist.<sup>4</sup> However, for reasons already given (and more to follow), we do not want to claim that it is wrong to continue with a pregnancy after a positive NIPT result.

What discussion of the wrongful life threshold and the non-identity problem encourages, perhaps unexpectedly, is a more nuanced discussion of what we consider might add to, detract from, or have a questionable impact in either direction on, our capacity to flourish. For it is fair to assume that no decent (prospective) parent would desire to have a child whose quality of life was a morsel above zero. Thus, while we do not wish to claim that it is wrong to have a child whose quality of life hovers just above the zero threshold, there are a myriad of serious moral considerations that legitimately apply above that threshold, and which help us to form a view about whether or not to continue with a pregnancy in any specific circumstances.

4. Of course, a child may not consider their life to be worthwhile, but this may be for reasons entirely independent of a diagnosed condition or for reasons that cannot be predicted or assumed. By implication, they cannot form the basis of a defensible reproductive decision.

Some of those considerations about the impact on a prospective child's capacity to flourish that apply above the zero threshold will be explored below.

While it is hard to claim that a child is harmed when it is brought into existence above the zero line, there are a number of ways of articulating worries about the impact of certain conditions. One is to question whether the level of suffering or restriction likely to be bound up with a given condition is reason enough to prevent it where that choice is available. These are cases where, as Herrisone-Kelly puts it, "the possible child's life would be worth living, but would contain what the prospective parent considers a greater load of suffering than she is prepared to allow her child to endure" (Herrisone-Kelly 2006, 169). While this sounds like a sensible and humane approach, Wilkinson questions the logical implications of harm prevention arguments more generally. Given that, as he puts it, *all* people will suffer harmful experiences of some sort during their lives, the reasoning behind harm prevention arguments suggests the "banning of *all* conceptions, because *all* children will suffer harm at some point" (Wilkinson 2010, 84). Wilkinson concludes that this style of argument must therefore be rejected.

While the point is appealing, we wonder if it has merit. For example, we could qualify the claim that "all people will suffer harmful experiences of some sort during their lives" with 'within reason'; and where the severity and likelihood of suffering exceeds what we consider to be ordinarily acceptable, we could argue that we are justified in wishing to prevent it. It is one thing to suggest that a life will have a fair chance of containing, for example, a range of injuries, exposure to crime, toothache, a broken heart, a road traffic collision, periods of deep sadness, illnesses, job loss and the like. It is quite another to suggest that, as a matter of near certainty, it will contain, for example, deep psychological trauma, protracted chronic pain, subjection to chemical warfare, prolonged imprisonment, or absolute poverty. And while the contrast is admittedly extreme, it illustrates that a contrast can indeed be drawn between harmful experiences of different severities and likelihoods and that, although it is true that all people will suffer harmful experiences of some sort during their lives, we may justifiably think seriously about the latter types of projections while being far less perturbed by the former.

There may be, then, defensible reasons to prevent suffering when we can, where it is both severe and highly likely to be experienced. Moreover, this need not entail raising the threshold of wrongful life. We are not claiming that such lives are wrongful: we are recognizing legitimate moral concerns about quality of life (way) above that

threshold, which help to determine what prospective parents are prepared to bring about. The level of suffering or restriction that a child is likely to experience is clearly a legitimate concern, which reinforces the point that the wrongful life threshold is far from the end of the story when it comes to (prospective) parental considerations, responsibilities and permissible choices. Rather obviously, it also explains why prenatal testing, including NIPT, exists.

Another way of moving beyond the wrongful life threshold is by comparison with other possible lives. As already observed, the decisions being considered are identity affecting, so one strategy is to compare a prospective child's life (with a diagnosed condition) with an alternative future child's life (without that condition). Wilkinson cites Parfit's 'same number quality claim' as exemplifying such a strategy, which states that "If in either of two possible outcomes the same number of people would ever live, it would be worse if those who live are worse off, or have a lower quality of life, than those who would have lived" (Parfit 1984, 360, Wilkinson 2010, 91).<sup>5</sup> He argues further, that same number quality claims seem plausible: "For in the absence of strong countervailing reasons (an important qualification) it is patently irrational, one might even say perverse, knowingly to select the worse of two states of affairs. Indeed, part of what it means to say that a state of affairs is 'better' is that we have reason to prefer it" (Wilkinson 2010, 91). This reflects Jonathan Glover's observation about the non-identity problem, that it "shows that we can make the world a worse place without harming particular people, and that this matters" (Glover 2006, 49).

Along the above lines, then, one might consider termination of pregnancy after a diagnosis of fetal anomaly, on the grounds that a future child without the diagnosed condition is likely to have a better quality of life than the one with it. Otherwise, as Wilkinson puts it, "a worse-off future person is created when a better-off future person could have been created instead" (Wilkinson 2010, 92). However, while many find the above sort of approach appealing, there are significant conceptual problems with it as well as some insensitive implications, which have been well articulated elsewhere (Bennett 2014, Herissone-Kelly 2006, Hull 2009, Wilkinson 2010, 91-96). These difficulties suggest that a defensible analysis will not hinge solely on a same number quality claim. Moreover, we do not want to restrict our evaluation to same number quality claims. That is, we want to argue that there are compelling reasons to not continue with pregnancy in some cases (reasons concerning harm preven-

5. A similar type of claim can be found in Buchanan, Brock, Daniels & Wikler (2000, 249).



tion, for example,) *irrespective* of who is or is not subsequently born. Having said that, the above approach does introduce comparative considerations that might, even if largely hypothetical to most (prospective) parents, make a useful contribution to an assessment of the likely impact of a given condition.<sup>6</sup>

When thinking about the likely impact of a diagnosed condition in terms of what we may wish to prevent, irrespective of any alternatives, concerns about autonomy can add further substance to the analysis. During an instructive discussion of the topic, Wilkinson argues that frequently cited ‘open future’ arguments are, at root, attempts to articulate more plausible ethical principles relating to autonomy (Wilkinson 2010, 47). He describes one such principle as “the view that parents ought to do what they can to ensure that their children develop into autonomous adults who are capable of making independent rational choices, based on autonomously held beliefs and desires”. In turn, autonomy is taken to be “a psychological property of persons and ‘respect for autonomy’ is a term for the moral constraints that a person’s having this psychological property places on the way in which we should treat her” (Wilkinson 2010, 48).

Concerns about autonomy take two forms, according to Wilkinson, which he terms “the Failure to Respect Autonomy Worry and the Failure to Promote Autonomy Worry”. The Failure to Promote Autonomy Worry is a worry about choosing to create future people whose autonomy will be limited (Wilkinson 2010, 51) (presumably, as a psychological property). The Failure to Respect Autonomy Worry is a worry about choosing to create future people with limitations that will, in adulthood, lead to the frustration of their autonomous desires and the failure of their autonomously formulated aims and projects (Wilkinson 2010, 53). While he argues that there are problems with the Failure to Respect Autonomy Worry, he concludes that, in as much as we value autonomy, “both ‘worries’ seem ultimately to count *in favour* of some forms of selection” (those that will increase autonomy) (Wilkinson 2010, 51, 54). Although this view clearly has weight among persons and in cultures where individual autonomy is particularly favoured, others do take contrary positions (Kittay 1999, Kaposy 2018).

Wilkinson’s discussion takes place in the context of embryo selection prior to implantation but, even so, the ethical concerns clearly carry over to cases of diagno-

6. Herissone-Kelly (2006, 168) seems to argue against this, where he says the sort of transpersonal “better for” judgment of what he calls the external perspective will have no force: “it cannot be regarded as any sort of reason, let alone a moral one”. We are suggesting that such a perspective does provide reason, even if it is inappropriate or irrelevant in many contexts and for many (prospective) parents.

sis during pregnancy. In tandem with the desire to prevent suffering, they comprise legitimate and weighty considerations about whether or not to continue with a particular pregnancy. Moreover, they are reflected in the practical concerns of healthcare providers, as evidenced by national and international guidelines about the seriousness of a given condition. The Nuffield Council report, for example, cites the Royal College of Obstetricians and Gynaecologists' 2010 guidance on termination for fetal anomaly, which states that doctors should weigh up the following factors when reaching a decision:

1. The potential for effective treatment, either *in utero* or after birth;
2. On the part of the child, the probable degree of self-awareness and of ability to communicate with others;
3. The suffering that would be experienced;
4. The probability of being able to live alone and to be self-supporting as an adult;
5. On the part of society, the extent to which actions performed by individuals without disability that are essential for health would have to be provided by others (Nuffield Council 2017, 24-25).<sup>7</sup>

Here, concerns about suffering and lack of autonomy are abundantly clear; and the Nuffield Council largely endorses the views of the RCOG, including their conclusion that an assessment of the seriousness of a fetal anomaly should be considered on a case-by-case appraisal (Nuffield Council 2017, 25). The reasons for that conclusion, as quoted in the Nuffield report, are that “the consequences of an abnormality are difficult to predict, not only for the fetus in terms of viability or residual disability but also in relation to the impact in childhood as well as on the family into which the child would be born” and that “sufficiently advanced diagnostic techniques capable of accurately defining abnormalities or of predicting the seriousness of outcomes are not currently available” (RCOG 2010, Nuffield Council 2017, 25). As we explore

7. Karpin and Savell observe further, that the RCOG guidance draws on the World Health Organization's definitions of Assisted and Dependent performance (Karpin and Savell 2012, 260).

below, this acknowledges the point that a condition like Down syndrome might not be thought of as serious at all, by affected individuals and families (Slotko et al 2011; Slotko et al, 2016). Indeed, it is interesting to note here that, as well as concerns about suffering and lack of autonomy on the part of the potential child, the impact of having a child with a diagnosed condition on the parent(s) and other family members is also recognized as a significant factor. This brings us to the second cluster of considerations that, along with beliefs about the issue of termination, legitimately contribute to (prospective) parental decision-making.

Assessment of the seriousness of fetal anomaly is further complicated by the fact that (prospective) parental perceptions about what is serious legitimately weigh in to the assessment. It is possible, for example, that one (prospective) parent could consider a particular condition to be serious, while another could consider it to be a little less, far less or not at all serious. In turn, this is highly likely to be bound up with distinct yet often related perceptions about the impact of a given condition on (prospective) parents and other family members, as well as with personal experience of different conditions. Karpin and Savell, drawing on the work of Wertz and Knoppers, suggest that there is little consensus about where to draw the line between serious and non-serious conditions; that there is a broad spectrum of opinions with greater agreement at the extremes (Karpin and Savell 2012, 273). They then follow Rosamund Scott's analysis, arguing that, between the extremes, disagreement is most prevalent with regard to what Scott describes as 'mid-spectrum' conditions (Karpin and Savell 2012, 274).<sup>8</sup> In these types of cases (as well as others, in our view), perceptions of the impact on the parent(s) and other family members may be decisive. As Scott puts it, "the point of recognizing that parents will be the most important judges of the impact on them of a given fetal condition is to suggest that, given there is *room for doubt* about seriousness in the mid-spectrum area, parents' perceptions may legitimately *tip the balance*" (Scott 2003, 212, Karpin and Savell, 2012, 275). For Karpin and Savell, this reinforces their point that "the question of what is serious may not be a purely clinical determination, as parents will have views about whether or not the disability being described to them is one with which they feel they can cope" (Karpin and Savell 2012, 275).<sup>9</sup> Indeed, given their interaction and engagement with the other considerations discussed throughout, we believe that (prospective) parental views will

8. These are conditions that are harder to classify. They may entail a good or reasonable quality of life, yet have a significant impact on parents and other family members.

9. Karpin and Savell (2012, 275) suggest that there will be further complexities that inform the interpretive matrix. We explore some of these in the section on context.

'tip the balance' quite a lot. While we believe that any ignorance and bias should be challenged by provision of evidence-based information and counselling (e.g. Slotko et al 2011, Slotko et al 2016), we consider that for many people, there will remain valid reasons for concern about the impact of certain conditions.

So far, the analysis has considered (prospective) parents' beliefs about termination of pregnancy, considerations about the impact of a given condition on a (potential) child's capacity to flourish and perceptions of the impact of having a child with a diagnosed condition on (prospective) parents and other family members. All of these things will be clearly bound up with (prospective) parental hopes and aspirations, both about their own lives and the lives of their potential children. As already stated with respect to quality of life, such hopes and aspirations are likely to extend way beyond any zero threshold and, we argue, legitimately so. Add to the analysis the moral optionality of parenting in general and, given the moral concerns described above, we should be sympathetic to decisions to terminate pregnancy when they involve such concerns. As Wilkinson argues, "if we believe in the moral optionality of parenting in cases... where disability and disease are not an issue, then surely we should believe that the moral optionality of parenting applies a fortiori to cases... where the decision is whether or not to have a child with a serious genetic disorder" (Wilkinson 2010, 182). Moreover, given the personal and variable nature of such assessments, exclusionary messages can be avoided. Karpin and Savell argue for example that, given the assessments described are made by the individuals closely concerned and carry the meaning of the woman/couple affected, they need not be assessments upon which we must necessarily agree and, by implication, the negative social effects of the choice to avoid a particular outcome are lessened (Karpin and Savell 2012, 347-348). They add that "it then becomes open to the community to challenge the individual's imaginary account of that outcome, since it is not enshrined in any kind of legal or policy doctrine" (Karpin and Savell 2012, 348).

Another reason for the likely variability of (prospective) parental assessments and decisions is that they will inevitably take place in different contexts. Different individuals bring varying personal and material resources to bear. As Rapp has argued, people's thinking about risks and chances will be very different, depending on the risks they already face in their daily life, which will be very different for people who are poor or socially excluded than they are for people who are economically privileged. Equally, the consequences of having a disabled child depend greatly on the material resources an individual or family can draw on. For example, where a couple

can afford to have one parent who does not do paid work, or works part time, it may be easier to support a child who has additional needs in their life; similarly if they can afford to pay privately for therapy, assistance or transport. The resources may not be about individual households, they might pertain to better and more supportive local networks or wider social environments being inclusive, via national policies on healthcare, education, and accessibility. As barriers are removed and provision is improved, it may become easier to parent a child with additional needs, or to contemplate a good life for an adult with disability.

As well as these individual and structural resources—which include availability of services and networks, as well as funds—there are also individual resources at the psychological level. Some individuals and families are more resilient than others. They can cope with difficulties, and maybe even thrive and grow as a result (Traustadottir 1991, Slotko et al 2016). Some individuals are more open to different experiences, and indeed different forms of embodiment, and others are more prejudiced against disability. People's values differ, and they may consequently place different weight on individual accomplishment, as opposed to other strengths such as sociability or inclusiveness. Individuals are all different in their responses to disability, and so are disabled people themselves.

## CONCLUSIONS

In line with the conclusions in the Nuffield Council on Bioethics report, in stressing the moral nature of individual choices, we do not wish to imply that prenatal diagnosis and selective abortion is all down to individual choice. We have written elsewhere of the ways that near-patient and wider social processes can undermine choice and push women and men into making particular decisions (Shakespeare 1996). For example, the availability of balanced information, the extent to which counselling is directive or non-directive, and the 'conveyor belt' of routinisation of testing and onward march of decision making all render the notion of choice problematic. In wider society, there remain pressures on people to avoid disability, and strong social messages that disabled lives are inferior to non-disabled lives. For these reasons, it is very significant that the Nuffield Council on Bioethics report states very powerfully that social and cultural pressures, and unbalanced information, need to be amended, and also that society should welcome disabled babies into the world,

even as it also offers prospective parents better technological means to avoid that eventuality, should they so wish.

NIPT is one of a panoply of techniques available, alongside diagnostic ultrasound, gamete donation, pre-implantation genetic diagnosis and other measures. But it is currently far-fetched to suggest that pre-implantation genetic diagnosis, for example, might or should become the procreative norm; and we would argue that this is likely to be the case for the foreseeable future. However, NIPT is becoming the norm, which is why it is important to question whether there should be some ethical parameters regarding its use.

There are numerous ethical considerations in the context of NIPT, many of which are intensely personal and reasonably variable, especially in combination. As such it would be both insensitive and misguided to prescribe responses to test results when such weighty and contextual considerations are involved. By implication, there should be no blanket policy with regard to termination of pregnancy as a result of NIPT.

Our analysis further suggests that there can be no single metric upon which a principle of procreative beneficence could or should operate, given legitimate variations in beliefs, perceived outcomes and impacts, parental hopes and aspirations; and context (both singularly and combined). Moreover, many of those variations will serve at least to partly comprise a view of what beneficence actually entails. That conclusion is reinforced by the non-identity problem and, in the current context of NIPT, the morally objectionable idea of encouraging more widespread selective termination of pregnancy.

The use of NIPT for sex selection (except where to avoid certain genetic conditions) is not justified on the basis of the above discussion either, since that use does not involve any of the moral concerns we have outlined and, as such, could be said to be a superficial reason to terminate pregnancy in most contexts, arising from bias rather than genuine concerns about the impact of gender on the life chances of the potential child. The same applies to any principle of beneficence, where it does not draw upon the types of moral considerations discussed that are weighty enough to justify termination of pregnancy on the grounds of a test result (that is, where there is little or no perceived suffering, loss of autonomy, impact on (prospective) parent(s) or family members; and further contextual issues do not apply).

In addition, the option of whole genome/ exome sequencing can be considered to have the potential to offend the principle of autonomy and should be resisted in

the absence of significant countervailing benefits. It could also be used to bolster the unjustified application of a principle of beneficence (where it does not draw on weighty moral considerations, as noted above). With that in mind it is worth remembering that, presumably, the reason that this technology has been developed is to detect issues or problems, not to create them.

*Acknowledgements: With thanks for the support and interest of Jeff McMahan, Julian Savulescu and Dominic Wilkinson, and in particular the contribution of the anonymous referees. We would like to dedicate this paper with love and respect to the memory of Rev. John Hull (14/8/1936—1/8/2017), father of the second author and family friend of the first author.*

#### REFERENCES

- Amundsen, R. 2010. "Quality of life, disability, and hedonic psychology." *Journal for the Study of Social Behaviour* 40 (4): 374-392.
- Bennett, R. 2014. "There can be no obligation to eradicate all disability." *Cambridge Quarterly of Healthcare Ethics* 23: 30-40.
- Buchanan, A., Brock, D., Daniels, N., and Wikler, D. 2000. *From Chance to Choice*. Cambridge: Cambridge University Press.
- Herissone-Kelly, P. 2006. "Procreative beneficence and the prospective parent." *Journal of Medical Ethics* 32: 166-169.
- Chitty, L.S., Wright, D., Hill, M., Verhoef, T.I., Daley, R., Lewis, C., Mason, S., McKay, F., Jenkins, L., Howard, A., Cameron, L., McEwan, A., Fisher, J., Kroese, M., Morris, S. 2016. "Uptake, outcomes and costs of implementing non-invasive prenatal testing for Down's syndrome into NHS maternity care: prospective cohort study in eight diverse maternity units." *British Medical Journal* 354: i3426.
- Hull, R. 2009. "Projected disability and parental responsibilities." In K. Brownlee and A. Cureton (eds) *Disability and Disadvantage*, 369-384. Oxford: Oxford University Press.
- Glover, J. 2006. *Choosing Children*. Oxford: Clarendon Press.
- Kaposy, C. 2018. *Choosing Down Syndrome: ethics and new prenatal testing technologies*. Cambridge MA: MIT Press.
- Karpin, I., and Savell, K. 2012. *Perfecting Pregnancy*. Cambridge: Cambridge University Press.
- Kitcher, P. 1996. *The Lives to Come: The Genetic Revolution and Human Possibilities*. London: Allen Lane/Penguin Press.
- Kittay, E.K. 1999. *Love's Labor: Essays on Women, Equality and Dependency*. New York: Routledge.

McMahan, J. 2005. Preventing the existence of people with disabilities, in D. Wasserman, J. Bickenbach, R. Wachbroit (eds) *Quality of Life and Human Difference: genetic testing, health care and disability*. Cambridge: Cambridge University Press: 142-171.

UK National Screening Committee. 2016. UK NSC non-invasive prenatal testing (NIPT) recommendation, available from: [https://legacyscreening.phe.org.uk/policydb\\_download.php?doc=57095](https://legacyscreening.phe.org.uk/policydb_download.php?doc=57095) [accessed on 29 November 2018]

Nuffield Council on Bioethics. 2017. *Non-invasive prenatal testing: ethical issues*. London: Nuffield Council on Bioethics.

Parfit, D. 1984. *Reasons and Persons*. Oxford: Oxford University Press.

Parker, M. 2007. "The best possible child." *Journal of Medical Ethics* 33: 279-283.

Rapp, R. 1999. *Testing Women, Testing the Fetus: The Social Impact of Amniocentesis in America*. New York, NY: Routledge.

Rothman, B.K. 1993. *The Tentative Pregnancy: how amniocentesis changes the experience of motherhood*. New York: W.W.Norton Company.

Royal College of Obstetricians and Gynaecologists. 2010. *Termination of pregnancy for fetal abnormality in England, Scotland and Wales*. London: Royal College of Obstetricians and Gynaecologists.

Savulescu, J. 2001. "Procreative beneficence: why we should select the best children." *Bioethics* 15 (5-6): 413-426.

Scott, R. 2003. "Prenatal Screening, Autonomy and Reasons: The Relationship between the Law of Abortion and Wrongful Birth." *Medical Law Review* 11: 265-325.

Shakespeare, T. 2006. *Disability Rights and Wrongs*. Abingdon: Routledge.

Skotko, B.G., Levin, S.P., Goldstein, R. 2011. "Self-perceptions from people with Down syndrome." *American Journal of Medical Genetics Part A* 155: 2360-2369.

Skotko, B.G., Levin, S.P., Macklin, E.A., Goldstein, R. D. 2016. "Family perspectives about Down Syndrome." *American Journal of Medical Genetics Part A* 170: 930-941.

Taylor-Phillips, S., Freeman, K., Geppert, J, et al. 2016. "Accuracy of non-invasive prenatal testing using cellfree DNA for detection of Down, Edwards and Patau syndromes: a systematic review and meta-analysis." *BMJ Open* 6:e010002. doi:10.1136/bmjopen-2015010002

Traustadottir, R. 1991. Mothers who care: gender, disability, and family life. *Journal of Family Issues* 12 (2): 211-228.

Wilkinson, S. 2010. *Choosing Tomorrow's Children*. Oxford: Oxford University Press.