

Title

Parents' decision-making following diagnosis of a severe congenital anomaly in pregnancy: practical, theoretical and ethical tensions

Highlights

- *Insight into parental decision-making following diagnosis or suspicion of a congenital anomaly in pregnancy*
- *Highlights ethical and theoretical tensions alongside practical difficulties*
- *Enactment of and tensions arising from parental decision-making are explored*

Keywords

Decision-making, congenital anomaly, qualitative, empirical ethics, sociological bioethics, patient autonomy, bioethics

Abstract

Patient involvement, in the form of shared decision-making, is advocated within healthcare. This is informed by the principlist account of patient autonomy that prioritises informed understanding, and decision-making free from coercion. This arguably over-simplifies the role of the social, whilst overlooking the role of culture

and context in medical decision-making. Clinicians encourage patients to demonstrably make decisions in the principlist 'style' that fit with their understandings of ethically 'correct' ways to support patient decision-making. However, this expected 'style' is often not achieved in practice. In this article, we use empirical data from a qualitative study exploring parental decision-making following diagnosis or suspicion of a severe congenital anomaly in pregnancy. Our study was based in four fetal medicine clinics in England, comprising semi-structured interviews with 38 parents whose pregnancy was affected by a severe congenital anomaly, 18 interviews with fetal medicine clinicians, and audio-recordings of 48 consultations. Examination of the dynamics at play within different approaches to decision-making highlights how the idealised concepts proposed in theory fail to capture real-life experiences of medical decision-making. The influence of the patient-clinician relationship on decisions is brought to the fore, highlighting the influence of power dynamics in implicitly and explicitly influencing patient decisions, and the need to better address this in policy and practice.

Background

Patient involvement in decision-making is advocated within healthcare.¹ In the United Kingdom (UK), a shared decision-making (SDM) model dominates policy, practice and rhetoric, informed by the principlist account of autonomy and reflecting the National Health Service's (NHS) commitment to person-centred care.² The principlist account of autonomy prioritises informed understanding, with non-directive counselling a basic requirement. Individuals are supported to make informed decisions without interference or coercion from internal or external constraints.^{3, 4} The principlist account is currently the dominant form of autonomy taught to medical students and practicing clinicians.⁵ As a result, SDM that prioritises the patient making a distinct and demonstrable decision about their care is widely considered the morally correct way to practice medicine.²

To ensure patients make decisions in the 'correct' way, clinicians are required to counsel non-directively, providing information about the options available, and helping patients identify their preferences in the context of their values.⁶ Whilst there is a logic to the underlying rationale, practical implementation is more complex, with patient-clinician interactions requiring "conversation, not just information, and care, not just choice".⁷ Work in the NHS has shown clinicians expect demonstrable decision-making according to the principlist 'style' in line with their understanding of ethically 'correct' ways to support patients.⁶ However, this expected 'style' of decision-making can be challenging to achieve.⁸⁻¹¹ In particular, clinicians' expectation about how information should be requested and understood that does not always match patients' experiences of how they themselves access and

use information¹². In the UK context, existing General Medical Council (GMC) and NHS guidelines align with this expectation by emphasising the role of information provision (usually provided by clinicians) as a fundamental part of SDM.

The expectation that 'good' decisions are made following a 'correct' process is problematic in an environment where the clinician-patient relationship continues to be fraught with inequalities.⁹ Yet the relationship is often the context within which patients' decisions are made, and clinicians remain a primary source of information.¹³ Indeed, GMC and NHS guidance suggests that access to treatment information is at the heart of the patient-clinician relationship,^{2, 14} suggesting "the information that the doctor has given the patient is enough"⁷, where 'enough' can be interpreted as both content and quantity of the information provided. This reinforces a power dynamic in which the doctor is "the more powerful and knowledgeable of the two, linking power with information, and the communication of that information."⁹ As a result, power, information, and the clinician-patient relationship are "closely woven together, impacting on how patients make decisions"⁹ and thus this relationship impacts on how 'well' patients are able to engage in the expected decision-making process.

We argue the principalist account over-simplifies the role of the social and overlooks the role of culture and context in medical decision-making.^{4, 9-11} Instead we look to relational autonomy. Relational autonomy is a feminist conceptualisation developed from the premise that people are socially embedded. Identities are formed through the complex social structures, relationships and social determinants that form the context within which people live.¹⁵ Relational

autonomy, with its focus on social and cultural influences (both positive and negative) on decision-making, provides a different theoretical approach that acknowledges the power dynamics that can restrict informed understanding, while simultaneously giving weight to those sources outside of formalised medical knowledge that patients draw on to make autonomous decisions.^{9, 15} Relational autonomy recognises the role cultural and socio-economic background have in constructing the values and beliefs that inform decisions.^{9,16} It also acknowledges that these are not all positive forces, arguing it is necessary to recognise “the effects of oppressive socialization that bring about power dynamics that traditional theories of autonomy often ignore.”^{9, 15} The principlist account often minimises these important sources down to confounding factors hindering the idealised decision-making process.^{5,10}

Decision-making in the context of severe congenital anomaly in pregnancy

One of the most pervasive themes in the sociology of medical knowledge is uncertainty, the enduring relevance of which was underscored by Fox’s observation that ‘Scientific, technological, and clinical advances change the content of medical uncertainty, and alter its contours, but they do not drive it away’.¹⁷ The difficulties in facilitating patient autonomy are particularly pronounced in situations with high levels of uncertainty. Parental decision-making following diagnosis or suspicion of a severe congenital anomaly in pregnancy is such a situation, and is the clinical context for our work.

Congenital anomalies affect 2-3% of pregnancies in high income countries.¹⁸ Screening programmes, such as the UK’s Fetal Anomaly Screening Programme

(FASP), identify congenital anomalies antenatally, enabling parents and clinicians to plan for the pregnancy and beyond, or to consider termination. In the UK, eleven anomalies are screened for: serious cardiac, anencephaly, spina bifida, exomphalos, renal agenesis, lethal skeletal dysplasia, congenital diaphragmatic hernia, trisomies 13 and 18, cleft lip and gastroschisis. The first nine (FASP9) pose significant morbidity or mortality risk, while cleft lip and gastroschisis are generally identified for early postnatal intervention. In around 70% of FASP9 anomalies, the affected pregnancy is terminated, representing around 3,300 terminations annually in the UK. Nonetheless, congenital anomalies remain a leading cause of neonatal and infant mortality, accounting for over 30% of these deaths. Evidence examining parental decision-making has focused on linking factors such as anomaly type, gestational age at diagnosis, severity and demographics to the decision to continue or terminate the pregnancy.²³ However, contradictory findings on the influence of these factors make interpretation difficult.²⁴ Other literature has focused on the difficulties encountered by parents, exploring aspects of parental experience,^{25, 26} care provision,²⁷ and the impact of a diagnosis.

Much of the social science literature relates to antenatal screening and the decision to accept or reject the offer, rather than the decision to continue or end an affected pregnancy. While both decisions are made antenatally, important differences exist. Although, in theory, an informed decision on screening would include consideration of whether to continue or end an affected pregnancy, any decision made at this point would essentially be hypothetical and therefore likely to differ from a decision based on a 'real-life' scenario.³² Furthermore, attitudes to screening are not necessarily a good indicator of attitudes towards termination.^{33, 34} Whilst the

screening literature provides some insights into the broader issues of non-directiveness, informed choice and risk, application of the evidence to post-diagnosis decisions is not without difficulty.

Drawing on a qualitative study of decision-making after the suspicion or diagnosis of a severe congenital anomaly, this paper contributes to debates around how best to support patient decision-making, autonomy, and informed consent. Our analysis offers empirical insights into the nuances of the decision-making process. As we will show, different styles of decision-making result in different tensions between parents and clinicians, as the latter seek enactment of an 'ideal' decision-making process that cleaves to the principlist approach. Examination of the dynamics at play within different decision-making styles highlights how the idealised concepts proposed in theory fail to capture real-life experiences of medical decision-making in practice. The influence of the patient-clinician relationship on decisions is brought to the fore, highlighting the influence of power dynamics in implicitly and explicitly influencing patient decisions, and the need to better address this in policy and practice.

Methods

This study examined parents' decision-making following antenatal suspicion or diagnosis of a FASP9 anomaly. A qualitative approach comprising interviews with clinicians and parents and audio-recordings of consultations between them was employed, enabling comparisons of real-time descriptions whilst providing contextual understanding of how participants made sense of the situations in which they found themselves.

Patient and public involvement was integral to planning the research. In particular, guidance on how best to approach parents at an emotionally charged time was sought. Ethical permission was granted by the [blinded]Research Ethics Committee(REC reference [blinded]).

We recruited from four tertiary referral centres, within two hospital trusts. Twenty women and 18 partners whose pregnancy was suspected or had been diagnosed as affected by a FASP9 anomaly were approached and recruited. Consent was a two-stage process. Parents were approached at their initial fetal medicine consultation, and with consent, the consultation recorded. Parents were formally enrolled into the study later, when they were approached for consent to use the consultation recording and be interviewed. Nineteen of the women had a partner at the time of diagnosis, although one couple separated before enrollment. Of the 18 couples, 15 were interviewed together, and three women interviewed alone, either through choice or due to availability. The three partners who were not interviewed were present for the consultations recorded. We purposively sampled to reflect pregnancies affected by a range of anomalies, gestational age at diagnosis, ethnicity and socioeconomic status, and to include parents who continued with the affected pregnancy and those who made the decision to end the pregnancy. Further breakdown of the sample is not provided in order to protect participants' anonymity. The number of consultations recorded for each pregnancy ranged between one and seven, with a mode of three. This was reflected in the number of hours of recordings that ranged between 1.5 and 15 hours per pregnancy. Interviews were undertaken in the three-week period before the birth for women

who continued with the pregnancy, and six to eight weeks post-delivery for women whose pregnancy was terminated. Interview topics included discussion of pregnancy care prior to diagnosis of the anomaly, attitudes towards and perceptions of risk, breaking the news of the anomaly, and decision-making following diagnosis.

In addition, 18 clinicians working in fetal medicine in the participating clinics were interviewed. Sampling was purposive to reflect a range of clinicians, including fetal medicine consultants and midwives, fetal cardiology and neurology consultants, and neonatologists. Clinician interviews explored attitudes towards termination of pregnancy for fetal anomaly, national and local policies governing termination of pregnancy, and views on parental decision-making post-diagnosis.

Combining consultation recordings with the parent and clinician interviews gave over 80 hours of data. Heeding the warnings of Corbin, we do not claim data saturation, rather state that following analysis, data from the final three participants provided no new themes, with the data derived from the interviews and consultations supporting the categories already established.

Recordings were transcribed verbatim and analysis used a constant comparative-based approach, assisted by NVivo software. Memos and a reflective diary provided additional context. The resulting dataset was further analysed with reference to relevant theories of patient autonomy and decision-making, applying an empirical bioethics approach^{9-11,53}. Empirical bioethics analysis uses empirical evidence to interrogate, inform and refine existing bioethical theory. Examining these theories

through an empirical lens allows for the interrogation of that theory to determine its relevance to existing policy and practice, such that theory, policy, or practice can be changed or updated to reflect the social and clinical context of healthcare.^{9, 10, 38}

Findings

We present four themes, each reflecting a set of prominent characteristics and decision-making style identified during the decision-making process. The themes are entitled: analytical; absolute; assess/reassess; and delay/avoidance. The assess/reassess theme further splits into two distinct sub-themes: choice removed and choice disturbed.

Analytical

The 'Analytical' decision-making style reflects the systematic and analytical way in which decision-making was approached. The majority of these parents had received confirmed diagnosis of a chromosomal anomaly. This left little uncertainty attached to the diagnosis and prognosis following laboratory confirmation of chromosomal configurations incompatible with life. The maternal age range was marginally older than other participants, possibly reflecting the higher proportion of chromosomal anomalies identified. The time between suspicion of the anomaly and a confirmed diagnosis was typically three to four weeks. None of these women continued their pregnancy and the time from diagnosis to termination ranged between one and four days.

Women in this group all underwent early invasive testing. Parents perceived this to be responsible and essential for resolving any uncertainty and providing sufficient

information on which to make an informed decision, a necessary condition of autonomous decision-making.

The amnio was something we... saw it as part of our antenatal care. It was... our responsibility. (Mother06-Terminated)

Internalising the responsibility for decision-making was a prominent characteristic of this group. A failure to be informed was perceived negatively and seen as avoidance of responsibility.

If you are educated enough about something then you can make an informed decision... it's a lack of responsibility [not to do so]. (Mother09-Terminated)

These parents actively sought information in relation to their baby's diagnosis and prognosis, and sought to use this to inform their decision-making.

...we did a lot of research on the internet about what it was. (Mother09-Terminated)

...we had our rational heads on... you just think really rationally and the emotional side just came through afterwards. (Mother20-Terminated)

Consideration was also given to future consequences for the parents themselves, their family and their baby. In the excerpt below, the implications of placing the responsibility of long-term care onto existing or future children were contemplated.

... it's the long-term... in years to come if something was to happen to us and she was to live... I mean they [the other children] would then be responsible for her and

*that's a big responsibility for anybody. (Mother10-
Terminated)*

For the majority of these parents, the decision-making process appeared to be uncomplicated, with many highlighting that clinicians had commented on their preparation:

*We recently had our post-counselling consultation with
[clinician] and s/he did say how impressed s/he was with
how well informed we had both been... (Mother20 –
Terminated)*

This group of parents actively participated in a 'rational' decision-making process, characterised by their efforts to weigh up relevant information, consider likely future impacts, and balance risk in terms of likelihood as well as severity of the anomaly identified. This approach to decision-making broadly reflected the 'ideal' process envisaged by clinicians. As a result, the process was perceived by both parties to be 'successful', leading to a 'good' decision.

However, tensions could arise if clinicians misjudged when parents were ready to commit to a final decision. When this occurred, the support offered to the parents backfired and instead became a source of distress. Clinicians talked of 'leaning' towards an option in an effort to support parents' decisions.

*But without actually leaning any way until they've leant
and then try and facilitate sort of their enjoyment of that
decision. You know, it's difficult (Clinician18)*

'Leaning' in this way appeared responsive to the calls by some parents for clinicians to support them, by positively reinforcing the decision made. However, misreading the situation could result in added distress. An example of this is reflected by one

woman talking about her experience following a counselling session with one of the specialist clinicians:

And that [clinician] said that [the baby] wouldn't have known no different because he wouldn't have ever been no different. [Baby's] quality of life, it would have been just [his/her] life because [he/she] wouldn't have known no different. And that made me feel bad then. (Mother12 - Terminated)

In this scenario, a well-intentioned attempt by a clinician to support an initial decision to continue created an additional level of grief and guilt when the parents subsequently decided to terminate the pregnancy.

Absolute

The second decision-making approach was labelled 'Absolute', reflecting this group's belief that there was no additional information or understanding of the situation needed in order to make their decision. Instead, regardless of prognosis, a fundamental value or belief system directed these parents along a particular course. For some, this was determined by religion:

...it's big and it's scary and it's daunting and it's a lot to take on. But, God knows how big my shoulders are, and he wouldn't put anything too big on them. (Mother01-Continued)

For others, their strong belief systems were based on a personal moral code. Where these were aligned with a lack of acceptance of termination, ending the pregnancy was excluded. All the women in this group continued with the pregnancy.

Parents in this group did not actively seek information as the decision had, in essence, already been made, based on their personal ethical stance. However, they did choose to either forego or delay further invasive testing, with the majority opting for late amniocentesis (after 34 weeks) for postnatal planning rather than additional information to inform antenatal decision-making. This meant that some of the parents retained a high level of uncertainty relating to the diagnosis.

Where parents rejected accessible information in the form of invasive testing, clinicians deemed this illogical, effectively dismissing the authority and autonomous choice of the parents.

*I have concerns that they really don't understand what they are going to let themselves in for. I mean if it's [information] available, why wouldn't you want to know?
(Clinician17)*

The differing perspectives on the need for information between parents and clinicians created tension as clinicians believed the needs of the baby after birth could not be met without a full understanding of the diagnosis. This was at odds with parents' desire to protect their baby from the risks associated with gaining that information, and their own informational needs with regards to making an autonomous choice.

Assess/Reassess

For many parents, whilst their decision-making approach initially mirrored the 'ideal' characteristics associated with informed autonomous SDM outlined in the Analytical theme above, they subsequently entered a decision-making cycle. This style of decision-making was the most common and most complex within our data, with its cyclical nature ultimately playing out in one of two distinct ways. In all cases, information was highlighted, a tentative decision made, further information given and the parents' position re-evaluated. However, these parents seemed unable to move out of the cycle of gathering and assessing information. There appeared to be an endless search for information, associated with difficulty committing to a decision. The uncertainty associated with diagnosis and prognosis impacted significantly on this cycle.

... when we went to one of the scans we had a load of questions ready, [about the anomaly] And then this woman she just told us all this other stuff. Then she turned around and said have you got any questions? I was like this is something new now; we don't even know what it means, so we had to start all over again ... (Mother02-Terminated)

A number of the clinicians also highlighted this circular process.

I had to terminate the consultation. It was 2.5 hours ... Every question generated a question and every answer generated another question. And we were going in this big circle... (Clinician14)

Some of this process related to the uncertainty of the prognosis and the need to wait for the baby to grow in order to assess the severity of any structural anomalies that

had been identified. This waiting and uncertainty often proved particularly distressing.

...the chromosome tests came back all clear... then we had to wait till 16 weeks... that's when they picked up the heart defect. And then they wanted to compare everything with an MRI scan; so we had to wait to 20 weeks. The situation was always... we suspect this, we suspect that... dealing with the uncertainty, that was the worst bit really. (Mother02-Terminated)

Information seeking was a way of managing uncertainty. However, some clinicians highlighted the risk of tensions developing between clinician and parent, where parents were perceived as being 'too engaged'. Whilst clinicians perceived information seeking as a positive and essential attribute, they highlighted concerns when parents sought information outside of clinicians' control.

[with the internet] you won't be any wiser. So it's best if you stick to the tests ... have the MRI scan and let the consultants that are the experts on these conditions talk to you about it, or the paediatricians about the children they have seen, rather than put it into google because they come out with all sorts. (Clinician03)

Attempts to control access to information were highlighted by clinicians, including reporting findings in broad terms without committing to a specific diagnosis:

[clinicians] have got very much better at using words that are harder to google. It makes a difference. (Clinician07)

Tensions arose where clinicians expected parents to be informed, but not too informed; a confused approach that sees the requirement of sufficient information and understanding for autonomous decision-making as somehow contingent on the clinicians', and not the parents', opinion on what constitutes enough. Parents were encouraged to take ownership of the decision through accessing information, but only to the extent that clinicians felt was appropriate. This enabled clinicians to distance themselves from the decision itself, and protect them from accusations of eugenics. On the other hand, clinicians wanted to retain ownership of the information with the power to determine what, and how much, to provide to parents.

Although an ongoing assess/reassess cycle is potentially an inevitable process in response to uncertainty, issues arose when parents found difficulties moving from a tentative to a final decision. This cycle was ultimately broken by either the clinician becoming more directive in their advice, which we call 'choice removed', or by the parents becoming disengaged with the process, which we call 'choice disturbed'.

Choice Removed

Parents in this group eventually opted to terminate the pregnancy, although the time between suspicion of anomaly and termination ranged between 5-12 weeks, significantly longer than other parents and reflecting the prolonged decision-making period. The interaction between these parents and clinicians also differed from consultations with other groups, both in the way parents approached the clinicians and the way in which clinicians responded. Parents would seek clinicians' opinions and spent more time deliberating about their decisions. In turn, clinicians

responded by making the counselling more 'directive'. The following extract was taken from a counselling session with a couple and two clinicians:

Clinician31: *So this is usually the scenario for a [specific anomaly] patient. We have one dying horribly on [ward] at the moment... So to start with, this is what will happen if you carry on with the pregnancy, otherwise termination is always an option. I'm giving you all the facts here, I'm not swaying you towards any way.*

Mother02: *Crying*

Clinician31: *Yes you're sitting on a bomb...then of course they're that much older and you get attached and their loss is that much more painful. But they might not reach any of those points because with the [anomaly] there's not one surgeon who's going to want to... touch them.*

The clinician repeatedly expressed concerns for the suffering of the baby. In addition, they suggested the option for surgery would be removed, in essence closing the option for intervention. Finally they expressed concerns for the parents, stating that their grief would be greater if they continued. At this point a second clinician re-directed the consultation and re-established the boundaries and responsibility for the decision, while reiterating the need for a 'rational' informed decision.

Clinician27: *It's a lot to take in... we're not expecting any decisions from you [now]. All we're trying to say is how things are ... you just need to digest it ... Then you*

need to make a decision about what you want to do. And whatever you want to do we are here to support you.

The affected pregnancy was eventually terminated at 24 weeks' gestation. Five weeks after the termination, the parents stated their gratitude towards the "*straight talking*" clinician, as this helped them make their final decision. In this instance directiveness was perceived as "*good care*" by the parents.

...[the baby] was going to be that poorly that they probably wouldn't be able to operate on him, which would cause him to die anyway and um I think that was what helped us make our decision. (Father02-Terminated)

These parents had deliberated for a number of weeks and did not appear able to come to a decision unaided. Constant uncertainty was too hard for them to process, and more directiveness was desired.

Clinicians caring for parents characterised as 'indecisive' became increasingly insistent in their communication of the risks of continuing the pregnancy in ways that seemed at odds with the widely expressed statements of neutrality in their interviews. Where parents appeared unable to 'settle' on a decision, clinicians often responded by giving the 'push' they believed was required for a decision to be made. However, by acting in the perceived best interests of the parents, clinicians experienced a tension between this and the idealised requirements for non-directive counselling. One clinician encapsulated this tension stating:

Sometimes, [the parents] are asking you, "Is that right, is that wrong?" They are wanting you to be involved in their

decision-making... it's hard to see them like that and I want to call out and say "Well if it was me this is what I would prefer" (Clinician08)

This supports what has been found within the literature, where the difficulties in achieving non-directive counselling and informed choice have been well documented in relation to antenatal screening and testing.⁴⁰As became clear in these scenarios; *"situations of indecision emphasize the need for a collaborative, or even at times a delegated decision-making whereby the professional assumes a greater role"*.⁴¹

Choice Disturbed

For a small group of parents, a breakdown in the clinician – patient relationship resulted in them disengaging from the decision-making process. Unlike those described immediately above, these parents all continued their pregnancy. These parents often spoke of the need to *"prove clinicians wrong"*. The ongoing uncertainty around diagnosis and prognosis resulted in a disintegration of trust between clinicians and parents, and finally disengagement from the process.

Initial behaviours included the seeking of information from other sources, such as pursuing a second opinion. However, the consistent message provided during the second opinion resulted in further frustration, with parents assuming collusion rather than confirmation:

... it [second opinion] turned out basically pointless... it wasn't the fact ... for them to confirm that we've got a [diagnosis] because we've read it, we've seen it and we

know all about that ... The idea ... was to run some more tests ... but obviously we ended up that nothing was done because they've gone with your guys diagnosis; so in essence it was kind of a pointless exercise... (Father07-Continued-Consultation4)

In this instance, the parents had highlighted an issue pertaining to the invasive test and had become fixated on this. The father subsequently expressed his frustration that the clinicians to whom they had gone for a second opinion had not repeated the test and had instead 'colluded' with the other clinicians.

Events relating to issues of managing risk and uncertainty were prominent in the data from these parents. These parents discussed the impact of the imperfect science of antenatal diagnosis. The resulting changing landscape created issues of trust, where parents felt the option to terminate became questionable as they perceived the projected diagnosis and prognosis could not be guaranteed. Tensions developed as the parents became more aware of the difficulties of interpreting tests, and of the range of alternative approaches to problems. This is a well-rehearsed story within the risk management literature.⁴²

In the excerpt below, the parent discusses how the results of initial tests had been reported as negative before an unequivocal result was given. He also reiterates his understanding of the risk of a chromosomal anomaly being present:

... we were told that two of the tests came back and they were perfectly fine, there was a bit of an issue with the third result, then it went away for laboratory exploration At the moment, we were told that it was sort of a 66

percent likelihood of him having [severe chromosomal anomaly]. (Father07–Continued–Consultation4)

A clinical explanation of the results suggests a different picture, where rather than a 66% likelihood of the baby being affected, there was a 100% surety of the baby being affected, but only 66% of the cells:

It's unusual but it can happen that the FISH test comes back normal, but the full culture shows a mosaic [chromosomal anomaly]. It's because the full results give the cells time to grow... (Clinician04 – Consultation)

However, the parents interpreted this as the clinicians 'giving up' on the baby:

I mean this is like, it's like 66 percent, and it's not good enough to just say fuck it, is it really? (Father07–Continued–Consultation5)

This lack of a common understanding resulted in a number of noticeable changes in consultations. First was a move from maternally- to paternally-led discussion. Initial consultations were generally a 50/50 divide between mother and clinician, but this changed over the course of the pregnancy. In the final consultation between clinicians and one of the couples, the dialogue changed to 40% clinician, 60% father, with the mother contributing four words in the 45 minute consultation. This change in dynamics was acknowledged by one of the fathers, who explained that they had lost trust in the clinicians, and who felt his wife was being pushed to terminate the pregnancy:

I mean they got things wrong, like the first test [FISH test]. There were just too many 'what ifs'... they just tried to railroad [wife]..... (Father7 – Continued)

Second, changes in the way the clinicians approached the parents were noted. The couples were labelled as “*difficult to manage*”, and interactions became defensive. The extracts below were taken from a 30 minute pre-consultation meeting between clinicians from fetal medicine, genetics, neonatology, and paediatric surgery, with the intention of establishing a common and consistent line:

After they had gone for the second opinion they phoned back again and asked why the amniocentesis hadn't been repeated. It was explained that there was no point in repeating the test. He said they are a very difficult couple and we need to be very clear in what we are saying to them (Clinician09)

We need to make sure we complete an intra-partum care plan as I think she is likely to be quite difficult to manage (Clinician08)

A second extract, taken from the same pre-consultation meeting, highlighted additional mechanisms for managing the parents' expectations, namely supporting the worst case scenario and removing hope:

But I'm not happy to consider offering anything at this point [in terms of surgery] (Clinician35)

My worry is that we are giving them a ray of hope and that it is only going to make things worse. (Clinician06)

Ultimately the parents detached from the process, as this extract from the subsequent consultation demonstrates:

... I'd sooner not come here again, that's where I'm at, I'm fucked off with it, ... I know that you guys cannot tell us any more than you're telling us and you have to make us aware of this, that and the other, but I'd sooner not fucking know, like forget the lot, forget it, forget, forget it, ... (Father07- Continued - Consultation6)

The uncertainties created by the difficulties in interpretation and explanation of antenatal testing irrevocably damaged the relationship between parents and clinicians. Counselling became defensive and the directive tactics used successfully with parents described in the Choice Removed section above only served to increase tensions further.

Delay/Avoidance

The final group of parents drifted towards continuing the pregnancy without making an active decision. They perceived the clinicians' role as paternalistic, and expected direction. This category was the most difficult to define because a delay in coming to a decision could be seen as part of a rational decision-making process. However, four specific behavioural characteristics were identified suggesting these parents should be categorised separately, namely: avoidance of information; difficulty understanding or accepting the diagnosis; misinterpretation of risk and uncertainty; and lack of engagement with clinicians. Unlike the parents described as 'Choice Removed' the clinician-patient relationship revealed little engagement from the outset. Failure to establish a solid relationship was perhaps the underlying cause for the different approaches used by the clinicians with the two groups of parents.

The first characteristic exhibited by this group was an avoidance of information seeking.

To be honest I haven't really looked for any more information because my mind's not really in a place at the minute (Mother05-Continued)

Second, where a definitive diagnosis was given, a lack of understanding or acceptance of the information was often demonstrated. The extract below was taken from an interview with a parent whose baby had been identified as having a severe chromosomal anomaly with a number of associated structural problems also suspected.

They go on and on about this [anomaly] and all the problems and stuff ...I think they make it up half the time... (Mother13- Continued)

We asked all parents what advice they would give others facing a similar scenario. Most expressed regret at not having understood what was being said and not questioning.

I'm not good at explaining like. I think what went wrong really was like we didn't really say...when we didn't understand. (Mother04-Continued)

Generally a very passive stance was taken regarding the outcome, suggesting parents felt they had little control over the consequences.

Well there isn't much point in worry like... what happens, happens really. (Mother11-Continued)

Third, there was often a lack of understanding of the uncertainty that can come with antenatal diagnosis, as medicine was viewed as a perfect science. Parents interpreted clinicians' attempts to keep them informed of emerging potential problems as ineptitude in a field that should be precise. One father added a little humour to his analysis of the situation stating:

I'm sure he's got a book and just sticks a pin in it every time we come in. So next time ooh, green parrot disease today? (Father13- Continued)

Finally, interactions between clinicians and parents were often stilted. The lack of engagement and poor communication between the parties is clearly visible in the consultation abstract below.

Clinician01: *Have you got anything else to ask?*

Mother04: *No*

Mother04: *[Laughing with friends]*

Clinician01: *Sorry what did you say?*

Mother04: *I was speaking to my friends*

[Laughter and whispering]

Clinician01: *[Inputting details into computer]*

Clinician01: *So 4 weeks' time; any concerns with the baby's movements and you need to come in. And if you can reduce the smoking that will be great.*

Mother04: *[No response]*

During her interview the mother graphically described her feelings about the interactions with clinicians, and her perceived role as a patient.

Because doctors are so, you know they know such big words. And we're so young and stuff and we sit there and go, "What are you on about?"... I mean you know you... have to go and say "yeah, yeah"... (Mother04–Continued)

The authority divide was clearly demarcated in the relationship, with the clinician's role perceived as a paternalistic one. When asked about why she had decided to have an amniocentesis, one mother responded:

The doctor said to have it [amniocentesis]. (Mother13–Continued)

The behaviours by this group of parents led to high levels of concern amongst clinicians about their decision-making.

... there's concern that some people maybe, their default position is just to continue with the pregnancy because to actually go through that process of thinking, "Where might I be, what might the consequences be of this action or that action?" They're either not willing to do that, or they really just don't have the skills. (Clinician10)

The demographics of this group were often very different to those of the clinicians caring for them. Some clinicians suggested that this could be problematic.

... some younger people and some people who are less used to dealing with professionals.... They just want to be told what to do. (Clinician02)

Here again the ideals of non-directiveness and the needs of parents appeared to be at odds. Unlike the parents in the Choice Removed group, no relationship between clinicians and parents had been established. This may explain the subsequent difference in clinician behaviour, where a paternalistic pattern of behaviour was exhibited when caring for the Choice Removed Group and yet a rigid non-directive approach was adhered to when caring for this group of parents

In this and similar situations, the tension between patient and clinician was tangible. On one side the parents were looking for direction, but were potentially unable to express this. On the other side, clinicians were fearful of directing the discussions in a situation where a relationship had not been established.

Discussion

Considering different decision-making approaches against existing theories of patient autonomy and decision-making highlights how difficult it is for clinicians to accept and understand decisions made outside these prescribed approaches. In particular, the role that the clinician-parent relationship played across the styles in facilitating or hindering sufficient understanding illuminates how the persistent inequality of power and authority continues to influence patient autonomy, especially when the clinical understanding of autonomy is constrained to the principlist account.

The expectation of patients making decisions in the 'ideal' way was most obvious for those parents who adopted an analytical style of decision-making. These parents experienced no resistance or concern from their clinicians over how their decisions were made. The decision-making process exhibited by this group aligned well with clinicians' understandings of patient autonomy, informed by the principlist account,³ as these parents sought information from clinicians, demonstrated an understanding of this information and the consequences of their decision, and took informed and decisive decisions to terminate pregnancies that clinicians felt were free of any kind of constraint.³ In this way, parents in this group also fulfilled not only theoretical, but practical guidance for SDM.^{13, 43, 44} In turn, this supported the development of a positive relationship between parent and clinician, which further facilitated this information exchange. Parents in this group mirrored the clinicians' expectations that making decisions in this way was 'right' and 'responsible', thus reinforcing the clinicians' beliefs of engaging in 'good' decision-making, which subsequently justified the decision taken as the 'right' one.

Parents who engaged in an 'Assess/Reassess' style were constantly seeking information, and initially viewed by clinicians as engaging in the process of 'good' decision-making. The emergence of the two sub-groups (Choice Removed and Choice Disturbed) reflects clinician attitudes towards information-seeking behaviours that fall outside the patient-clinician relationship. In the Choice Removed category clinicians expressed concern and frustration that parents continually wanted more information from them than they were prepared to offer.

The Choice Disturbed group took the power struggle for information and authority to the other extreme. They actively sought information from sources outside their parent-clinician relationship, to establish their own expertise. This led to a self-reinforcing cycle for both sides with distrust in the clinician leading to information-seeking elsewhere, in order to establish further understanding of the situation. The clinicians dismissed any concerns brought up from outside sources in an attempt to regain their position as the expert in the dynamic, thus deepening the parents' concern that information was being withheld. While this group all continued their pregnancies, it is unclear whether this was a deliberate choice, or a rejection of clinical involvement. What is clear is that failure to acknowledge the parental information needs resulted in parents making decisions with little support from their clinicians, or not making a decision at all.

For parents who fell into 'Delay/Avoidance' style of decision-making, the importance of the clinician-parent relationship as the primary channel of information is starkly revealed. While the way in which this dynamic controls information access is deeply problematic for facilitating autonomy, it is still the prevailing framework for information

giving and receiving in medicine. When there was no relationship at all, parents were left without resources with which to make their decision. The parents in this group expected to be paternalistically guided through the decisions, while the clinicians expected 'active' information-seeking patients who would make clear decisions. When neither party behaved as expected, clinicians shied away from being overly directive, and parents were left without direction. As a result many continued their pregnancies for no other reason than they were waiting to be guided by their clinicians. In this group the fetishised process that ensures 'good' decision-making never seemed to start, as the first step of being responsive and information-seeking (on the parent's part) was never taken. The lack of a developing/developed relationship between the two parties meant that the usual avenues of information were closed. An expectation that the 'right' type of pathway to 'good' decision-making would be followed, meant that parents who wanted to be directed or led were left unsupported by clinicians who lacked the tools to facilitate parents' autonomous choice outside of the accepted process. As a result it is difficult to know whether parents felt their autonomy was facilitated in such a way that they made the best decision for themselves.

This apparent reliance on the principlist account of autonomy, as played out in existing guidance on patient decision-making, has been contested in recent years. It has been shown that some patients look to clinicians to be guided as an active choice they have made to transfer decisional responsibility to an expert, making it a valid part of the decision-making process⁸. Paton has described participants wanting to be led by someone with experience, and for clinicians to lead decision-making around fertility preservation before cancer treatment initiation.¹⁰ In a similar way, parents in our study

were arguably looking to be guided, but their clinicians were unable or unwilling to engage in this kind of relationship with them.

For all the decision-making styles presented, the clinician-parent relationship set the tone and the level of facilitation for sufficient understanding, and by extension, patient autonomy. Across the data, clinicians and parents did not come together as equals (as SDM advocates), nor did they engage in a client-provider relationship (as patient-centred care advocates): two increasingly dominant and influential paradigms in healthcare. Instead, the persistent power dynamics of expert and lay-person prevailed, to varying degrees, influencing the process by which parents achieved sufficient understanding and made autonomous decisions. In this unequal power dynamic paternalism remained a dominant force as clinicians dictated what and how much information was given, and how parents received it, as part of an expected, but rarely articulated, process that led to 'good' decision-making.

Parents who used an Analytic decision-making approach indicated that they had a strong and positive clinician-patient relationship. Similar to the findings from other studies, these patients demonstrated a better understanding, felt at ease asking for information from their clinicians, and described being comfortable with the decisions they made. Conversely, when the relationship was negative or non-existent, such as those parents from the choice disturbed group, patients viewed the doctor as an obstacle to accessing information. In both cases, what is striking is that the relevant information was felt to be held by the clinician; either to be benevolently given out when parents seek information

in the 'right' ways, or malevolently held back for those parents who looked to other sources of information, or interpreted the information given differently to the clinician.

Other studies have also highlighted how clinicians are still viewed as the 'keepers' of the information necessary for patients to make good, autonomous decisions, highlighting the level of power clinicians still hold in the contemporary clinical encounter. Parents often felt on the wrong side of this power relationship, struggling not just to access the level of information they felt necessary to make an informed choice, but also to have that information conveyed in an accessible way. Decisions not to access information at all resulted in parents being viewed as disengaged or even illogical; requests for more information were disregarded, and parents considered over-engaged or even delusional. The variety of decision-making styles articulated here stands as evidence that patients can and do make their own pathways towards valid, autonomous decisions. They should set the threshold for their own sufficient information, as the patient has to weigh up medical opinion with their own values and beliefs as part of the process of autonomous decision-making. Autonomy cannot be supported and autonomous decision-making cannot be facilitated when the bar for what constitutes a 'good' or 'right' choice is set by an outsider who never articulates that bar, and holds authority and power within the relationship as the provider of the information necessary to reach that bar.⁹

Whilst it may be impossible to fully remove the influence of power dynamics, it is possible to mitigate their negative effects on patient autonomy.⁹ Greater understanding of the value placed by patient and clinician on different sources of information, whilst

appreciating the expertise of both parties, may help to balance the power dynamics⁹ and allow for the SDM, as imagined in NHS policy and elsewhere, to be achieved in practice.

The different approaches to decision-making outlined in this paper acknowledge the validity of the information valued by those parents who made (or did not make) decisions outside the 'accepted' norms of the principlist account. Conceiving of decision-making as multi-faceted, as our findings show, recognises the different pathways that patients take to making decisions. By acknowledging parents as experts in their own lives, and at least discussing why different sources and interpretations of information are informing their decision-making approaches, clinicians would arguably have been able to better support the autonomy of those parents in the Absolute category (making decisive choices that went against clinical belief), and facilitate autonomous decision-making for those in the Assess/Reassess and Delay/Avoidance categories (by providing the necessary sufficient information in both type and amount that would lead the parent to make a decisive choice).

This is perhaps easier said than done, as it requires moving away from a principlist account of autonomy and its rigid expectations, and towards a more inclusive account of autonomy, such as relational autonomy. Given the diversity of the populations cared for by health service providers, it is reasonable to consider a more inclusive conception of autonomy, like relational autonomy, as the more appropriate theoretical scaffolding upon which to build policy and guidance on patient autonomy in the clinical encounter.

Conclusion

The empirical accounts of clinicians' reactions to the different forms of decision-making observed show how the prevailing power dynamic that prioritises an artificial set of criteria as a short cut to facilitating autonomous decision-making can serve to hinder that same autonomy in the clinical encounter. Even when parents took part in the process and articulated wanting more or less information, clinicians disregarded these requests and persisted in seeking to deliver the level of information *they* felt was necessary to make a good decision. It is thus no surprise that where parents attempted to hold their ground and their authority over how they made their decisions, the relationship became strained.

A better understanding of how the process of decision-making is uniquely personal is an area of clinical practice that remains under-researched. Despite evidence that patients consume and use information differently to reach decisions, these findings have yet to be operationalised in clinical practice in any way beyond a principlist account of the decision-making process. By ignoring the different ways that patients need and use information to make decisions, clinicians are inadvertently, and perhaps overtly, stifling patient autonomy in favour of formalised guidelines that disregard empirical evidence of how decisions are made in practice. In highlighting the many different ways that patients make decisions, we hope to contribute a new and more current understanding of the decision-making process.

References

1. Graffigna G, Barello S. Patient engagement in healthcare: pathways for effective medical decision making. *Neuropsychol Trends*. 2015; **17**: 53-65.
2. England N. Shared decision making. 2019.
3. Beauchamp T, Childress J. *Principles of Biomedical Ethics*. Oxford: Oxford University Press, 2009.
4. Paton A. "Being Guided": What oncofertility patients' decisions can teach us about the efficacy of autonomy, agency and decision-making theory in the contemporary clinical encounter. *International Journal of Feminist Approaches to Bioethics*. In press.
5. Padela AI, Malik AY, Curlin F, De Vries R. [R e] considering Respect for Persons in a Globalizing World. *Developing world bioethics*. 2015; **15**: 98-106.
6. Fried TR. Shared Decision Making — Finding the Sweet Spot. *New England Journal of Medicine*. 2016; **374**: 104-6.
7. Hargraves IL, A.; Shah, N.; Montori, V. Shared Decision Making: The Need For Patient-Clinician Conversation, Not Just Information. *Health Affairs*. 2016; **35**: 627-9.
8. Lotto R, Smith LK, Armstrong N. Diagnosis of a severe congenital anomaly: A qualitative analysis of parental decision making and the implications for healthcare encounters. *Health Expectations*. 2018; **21**: 678-84.
9. Paton A. No Longer "Handmaiden": The Role of Social and Sociological Theory in Bioethics. *IJFAB: International Journal of Feminist Approaches to Bioethics*. 2017; **10**: 30-49.
10. Paton A. 'It's Not Just About Having Babies': A Socio-bioethical Exploration of Older Women's Experiences of Making Oncofertility Decisions in Britain. *Philosophies and Sociologies of Bioethics*: Springer, 2018.
11. Paton A. About time: how time influences and facilitates patient autonomy in the clinical encounter. *Monash bioethics review*. 2018; **36**: 68-85.
12. Lotto R, Smith LK, Armstrong N. Clinicians' perspectives of parental decision-making following diagnosis of a severe congenital anomaly: a qualitative study. *BMJ Open*. 2017; **7**.
13. McMullen LM. Discourses of influence and autonomy in physicians' accounts of treatment decision making for depression. *Qual Health Res*. 2012; **22**: 238-49.
14. Council GM. Ethical Guidance. 2008.
15. Mackenzie C, Stoljar N. *Relational autonomy: Feminist perspectives on autonomy, agency, and the social self*. Oxford University Press, 2000.
16. Donchin A. Understanding autonomy relationally: toward a reconfiguration of bioethical principles. *The Journal of medicine and philosophy*. 2001; **26**: 365-86.
17. Fox R. Medical uncertainty revisited. In: Bird C, Conrad P, Freemont A, (eds.) *Handbook of Medical Sociology*. New Jersey: Prentice Hall., 2000.
18. Askelsdottir B, Conroy S, Rempel G. From diagnosis to birth: parents' experience when expecting a child with congenital anomaly. *Advances in Neonatal Care*. 2008; **8**: 348-54.
19. Lyus R, Creed K, Fisher J, McKeon L. Termination of pregnancy for fetal abnormality. *British Journal of Midwifery*. 2014; **22**: 332-7.
20. Budd J, Draper E, Lotto R, Berry L, Smith L. Socioeconomic inequalities in pregnancy outcome associated with Down Syndrome: a population based study. *Archives of Disease in Childhood, Fetal & Neonatal Edition*. 2015; **100**: F400-F4.
21. Care DoHaS. Abortion Statistics in England and Wales, 2017. 2017.

22. Best KE, Seaton SE, Draper ES, Field DJ, Kurinczuk JJ, Manktelow BN, et al. Assessing the deprivation gap in stillbirths and neonatal deaths by cause of death: a national population-based study. *Archives of Disease in Childhood - Fetal and Neonatal Edition*. 2019; fetalneonatal-2018-316124.
23. Chenni N, Lacroze V, Pouet C, Fraisse A, Kreitmann B, Gamberre M, et al. Fetal heart disease and interruption of pregnancy: factors influencing the parental decision-making process. *Prenatal Diagnosis*. 2012; **32**: 168-72.
24. France EF, Locock L, Hunt K, Ziebland S, Field K, Wyke S. Imagined futures: how experiential knowledge of disability affects parents' decision making about fetal abnormality. *Health Expectations*. 2012; **15**: 139-56.
25. Entwistle VA, France EF, Wyke S, Jepson R, Hunt K, Ziebland S, et al. How information about other people's personal experiences can help with healthcare decision-making: a qualitative study. *Patient Education And Counseling*. 2011; **85**: e291-8.
26. Locock L. The parents' journey: continuing a pregnancy after a diagnosis of Patau's syndrome. *BMJ : British Medical Journal*. 2005; **331**: 1430-.
27. Lotto R, Armstrong N, Smith LK. Care provision during termination of pregnancy following diagnosis of a severe congenital anomaly - A qualitative study of what is important to parents. *Midwifery*. 2016; **43**: 14-20.
28. McCoyd JLM. What do women want? Experiences and reflections of women after prenatal diagnosis and termination for anomaly. *Health Care for Women International*. 2009; **30**: 507-35.
29. Timmermans S, Buchbinder M. *Saving babies?: the consequences of newborn genetic screening*. University of Chicago Press, 2013.
30. Thomas G. *Down's Syndrome screening and reproductive politics: care, choice, and disability in the prenatal clinic*. London: Routledge Studies in the Sociology of Health and Illness, 2017.
31. Williams C, Sandall J, Lewando-Hundt G, Heyman B, Spencer K, Grellier R. Women as moral pioneers? Experiences of first trimester antenatal screening. *Social Science and Medicine*. 2005; **61**: 1983-92.
32. Sawyer SM, Cerritelli B, Carter LS, Cooke M, Glazner JA, Massie J. Changing Their Minds With Time: A Comparison of Hypothetical and Actual Reproductive Behaviors in Parents of Children With Cystic Fibrosis. *Pediatrics*. 2006; **118**: e649-e56.
33. Markens S, Browner CH, Mabel Preloran H. Interrogating the dynamics between power, knowledge and pregnant bodies in amniocentesis decision making. *Sociology of Health & Illness*. 2010; **32**: 37-56.
34. Hewison J, Green JM, Ahmed S, Cuckle HS, Hirst J, Hucknall C, et al. Attitudes to prenatal testing and termination of pregnancy for fetal abnormality: a comparison of white and Pakistani women in the UK. *Prenatal Diagnosis*. 2007; **27**: 419-30.
35. Popay J, Williams G, Thomas C, Gatrell T. Theorising Inequalities in Health: The Place of Lay Knowledge. *Sociology of Health & Illness*. 1998; **20**: 619-44.
36. Corbin J, Strauss A. *Basics of qualitative research: techniques and procedures for developing grounded theory*. London: Sage Publications, 2008.
37. Glaser BG, Strauss AL. *The discovery of grounded theory: Strategies for qualitative research*. New Jersey: Transaction Publishers, 2012.
38. Haimes E. What can the social sciences contribute to the study of ethics? Theoretical, empirical and substantive considerations. *Bioethics*. 2002; **16**: 89-113.

39. Lotto RS, L.; Armstrong, N. Diagnosis of a severe congenital anomaly: a qualitative analysis of parental decision-making and the implications for healthcare encounters. *Health Expectations*. 2017.
40. Schwennesen N, Koch L. Representing and intervening: 'doing' good care in first trimester prenatal knowledge production and decision-making. *Sociology of Health and Illness*. 2012; **34**: 282-98.
41. Madrigal VN, Carroll KW, Hexem KR, Faerber JA, Morrison WE, Feudtner C. Parental decision-making in the paediatric intensive care unit. *Critical Care Medicine*. 2012; **40**: 2876-82.
42. Taylor-Gooby P, Zinn JO. Current Directions in Risk Research: New Developments in Psychology and Sociology. *Risk Analysis*. 2006; **26**: 397-411.
43. Charles C, Gafni A, Whelan T. Shared decision-making in the medical encounter: what does it mean?(or it takes at least two to tango). *Social Science & Medicine*. 1997; **44**: 681-92.
44. Charles C, Gafni A, Whelan T. Decision-making in the physician–patient encounter: revisiting the shared treatment decision-making model. *Social Science & Medicine*. 1999; **49**: 651-61.
45. Kim J, Deal AM, Balthazar U, Kondapalli LA, Gracia C, Mersereau JE. Fertility preservation consultation for women with cancer: are we helping patients make high-quality decisions? *Reproductive biomedicine online*. 2013; **27**: 96-103.