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"Knowledge was clearly associated with education." epistemic positioning in the context of informed choice: a scoping review and secondary qualitative analysis



Niamh Ireland-Blake¹, Fiona Cram², Kevin Dew³, Sondra Bacharach⁴, Jeanne Snelling⁵, Peter Stone⁶, Christina Buchanan⁷ and Sara Filoche^{1*}

Abstract

Background Being able to measure informed choice represents a mechanism for service evaluation to monitor whether informed choice is achieved in practice. Approaches to measuring informed choice to date have been based in the biomedical hegemony. Overlooked is the effect of epistemic positioning, that is, how people are positioned as credible knowers in relation to knowledge tested as being relevant for informed choice.

Aims To identify and describe studies that have measured informed choice in the context of prenatal screening and to describe epistemic positioning of pregnant people in these studies.

Methods Online databases to identify papers published from 2005 to 2021. The PRISMA-ScR checklist guided data collection, analysis and reporting. Secondary analysis that considered hermeneutics (e.g., knowledge that was tested, study design) and testimony (e.g., population descriptors) developed a priori.

Findings Twenty-nine studies explored the measurement of informed choice. None reported that pregnant people were involved in the design of the study. Two studies reported pregnant people had some involvement in the design of the measurement. Knowledge tested for informed choice included technical aspects of screening, conditions screened and mathematical concepts. Twenty-seven studies attributed informed choice to population descriptors (e.g., race/ethnicity, age, education). Population descriptors were reified as characteristics of epistemic credibility for informed choice obtained. For example, when compared to a high school qualification, a tertiary qualification was a statistically significant characteristic of informed choice. When compared by race, white people were found to be significantly more likely to make an informed choice. Additional demographic descriptors such as age, language spoken, faith and previous pregnancies were used to further explain differences for informed choice obtained. Explanations about underlying assumptions of population descriptors were infrequent.

Conclusion Using population descriptors in the biomedical hegemony as explanatory variables for informed choice can position (groups of) people as more, or less, epistemically credible. Such positioning could perpetuate

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epistemic injustices in practice leading to inequitable access to healthcare. To better uphold (pregnant) people as credible knowers population descriptors should instead be contextual (and contextualising) variables. For example, as indicators of social privilege. Further, making room for ways of knowing that go beyond the biomedical hegemony requires the development of epistemically just 'measures' through intentional, inclusive design.

Keywords Informed choice, Epistemic injustice, Epistemic justice, Epistemology, Epistemic positioning, Equity

Introduction

Epistemology is the study of knowledge [1]. It examines how our views of knowledge are constructed through our worldviews (the lens through which we see and experience the world). To the extent that different people see and experience the world through different lenses, with different interests and cultural frameworks, the opportunity for epistemic injustice arises. An epistemic injustice is a harm done to someone in their position as a knower, that is, when people are judged as not being credible knowers or credible producers of knowledge [2, 3]. Epistemic injustices arise when there is a mismatch between the actual credibility of the knower and the credibility assigned to the knower as result of prejudices or stereotypes [2, 3]. There are two broad categories of epistemic injustice, testimonial and hermeneutical. Testimonial injustice occurs when prejudice causes a hearer to give a deflated, or inflated, level of credibility to the speakers' word [2, 3]. Hermeneutical injustices occur when someone is rendered unable to understand or express some important aspect of their own experience due to a gap in e.g., resources to understand them [2, 3]. Epistemic injustices are inextricably linked to injustices associated with race, gender and socioeconomic status [4]. In health, epistemic injustices affect equitable access to, and subsequent outcomes of, healthcare. For example, testimonies of pain by white patients in the United States lead to more diagnoses and treatment when compared to testimonies of pain reported by black patients [5, 6]. This arises when a testimony from a white patient is deemed more credible than a black patient because of stereotyping or identity prejudice against black patients. Epistemic (in)justice has real implications for healthcare [1, 2, 4].

Healthcare policy advocates that participation in healthcare should reflect "informed choice" [1-3]. An explicit policy aim in promoting informed choice is to enhance patient autonomy and to prevent people from being deceived or coerced. A broad definition of informed choice is one where people have been given the relevant information, and the decision to have, or not have, screening is consistent with their beliefs and values. How people are supported to make informed choices about their healthcare is therefore an epistemic consideration. For example, if assumptions are made about people's credibility as knowers, these may affect how, what and if information is shared, and as a consequence, equitable access to healthcare. So just as patients' testimony

about pain may be differentially validated by health care practitioners according to the race of the patient, perceptions of whether informed choice has been given by patients, or capability for informed choice, may be subject to similar epistemic injustices in practice.

Being able to measure informed choice represents a mechanism for service evaluation to monitor whether informed choice is achieved in practice. However, while professional recommendations are clear that a person's decisional autonomy needs to be upheld and their informed choice respected, there is a notable lack of agreement, consistency, and clarity about how informed choice should be measured [7, 8]. We have previously considered how epistemic (in)justices can affect (in)equitable implementation and uptake of new molecular technologies [9]. Epistemic considerations of measurements of informed choice have thus far not been considered. Approaches to measuring informed choice to date have been based in the biomedical hegemony, that is, they have set out to objectively measure people's biomedical knowledge (e.g., of the condition being screened for and/or technical aspects of the screen's accuracy, such as, positive predictive value) [10]. We are using the term epistemic positioning to refer to how, in studies that 'measure' knowledge (informed choice), people are positioned as credible or not credible knowers.

Prenatal screening for chromosomal conditions has been part of ante-natal care in many countries for over 40 years. Typically the screen includes three chromosomal conditions (Down's syndrome (trisomy 21), Edward's syndrome (trisomy 18) and Patau's syndrome (trisomy 13)). The screening is optional, with the ideal being that a pregnant person makes a voluntary and informed choice about whether, or not, to be screened. Prenatal screening is also an area where there continues to be rapid advances in genetic and genomic technology with the conditions and scope of screening increasing. Supporting informed choice is of paramount significance [11]. There has been some critique that information provided to prospective parents is biased towards a biomedical hegemony that does not consider experiential knowledge (e.g., from people with lived or living experience of these conditions) [12, 13]. Measuring informed choice for prenatal screening has been extensively explored. As such, we used prenatal screening for chromosomal conditions as a case example to explore epistemic positioning in the context of informed choice. Specifically, the aims of this scoping

review and secondary qualitative analysis are to describe measurements of informed choice and to describe the epistemic positioning of pregnant people in these studies. Understanding the effect of epistemic positioning in approaches that have measured informed choice to date could inform new approaches to measuring, and achieving, informed choice in practice in the future.

In all of the studies we reviewed, pregnant people were referred to as woman/women. We acknowledge that this descriptor may not reflect everyone's preferred gender identity. For the purpose of this critical scoping review and secondary qualitative analysis we use inclusive and additive terms, for example, woman/women and pregnant person/people.

Methods

Literature search

For the scoping review the search strategy was developed with a medical librarian. The Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) checklist guided the data collection, analysis and reporting [14] (Supplementary Table 1). The search was undertaken in Medline, Web of Science and Scopus for studies undertaken during 2005 to 2021 (Fig. 1). Prior to writing, we undertook another search of literature in 2022 which yielded no additional publications for inclusion. Our inclusion criteria were studies in English-language papers reporting empirical research that used qualitative interpretive or descriptive methodologies and quantitative studies that measured informed choice and decision making for prenatal screening for chromosomal and other conditions. Only peer-reviewed journals' were included. Full search strategy/search terms are included as a supplementary file (Supplementary Table 2). Broadly, the search terms included: prenatal screening (including antenatal screening, Down's syndrome, Trisomy 18 and 13, combined screening, sex chromosome aneuploidies); non-invasive prenatal screening (including cell-free fetal DNA); knowledge; information; informed choice/consent; Indigenous peoples).

Filter and refinement process

Potential articles to be included in the systematic scoping review were uploaded to EndNoteX9 (Fig. 1). The filter and refinement process consisted of excluding duplicate literature that had been acquired across different databases, and literature outside of the scope of the qualitative analysis. A total of 519 articles were initially identified by N I-B (Fig. 1). These were uploaded to a qualitative coding software (NVivo, Lumivero, Release 1.0, 2020) to support the charting and selection process. Abstracts and titles were reviewed independently by N I-B and SF. Full articles were reviewed by SF and cross-checked with N I-B through an iterative process comprising several rounds of assessment and review. This process also supported us to be reflexive in considering the selected articles for secondary analysis. Only studies that measured informed choice or knowledge were included. Intervention studies that used decision-aids to increase knowledge, and studies that explored decision making were excluded, but the references were reviewed for possible inclusion of other studies (Fig. 1).

Secondary qualitative analysis

A set of eight questions about epistemic positioning was developed by the named authors prior to the literature being read and analysed to help mitigate reviewer bias (Table 1).

We are positioning ourselves epistemically: as pragmatists around methodological considerations that enable better participant involvement, more reliable and more just approaches to informed choice, beyond non-positivistic. We are interested in supporting epistemic justice and empowerment for pregnant persons when it comes to making informed choices (either to accept or decline) and interested in expanding the ways in which knowledge is conceptualised or theorised. We believe that the kinds of information and experiences required to qualify as possessing knowledge varies (and not necessarily in ways that relate to population descriptors, like education and employment).

The questions were based on epistemic justice considerations that pertain to hermeneutics (e.g., what knowledge was tested, who was involved in the development of the measure/study design, relational aspects in the decision to accept or decline screening) and testimony (e.g., population descriptors such as educational attainment, socioeconomic status, race and/or ethnicity, parity, age) that informed how people are positioned as credible knowers, or not, in relation to the measurement of informed choice. Evidence of people's norms/preferences influencing the measure and/or reporting of informed choice was also reviewed (e.g., cultural norms, faith, spirituality). If information pertaining to any question was not discernible, e.g., if there was not an explicit statement describing how pregnant people were involved in the design of the study, the response to that question was recorded as 'not reported'. N I-B and SF undertook the analysis independently, and collated findings after three rounds of reviewing the texts. The analyses were shared and discussed with all authors.

A narrative summary was then developed to contexualise the findings. This summary included definitions of informed choice, types of screening and measures of informed choice, and how informed choice was described.



Fig. 1 Overview of the main steps for the selection of publications included in the systematic scoping review and secondary qualitative analysis

Table 1 Results from the secondary qualitative analysis of papers that explored measuring informed choice and/or knowledge (n=29)

| Questions for the secondary qualitative analysis | Not reported | Yes |
|---|--------------|------------------------------|
| 1. Were pregnant people involved in the design of the study? | 29 | - |
| 2. Were pregnant people involved in the development of the model that aimed to measure informed choice? | 24 | 5(15–19) |
| 3. Were pregnant people offered an opportunity to share their experiences of informed choice with the researchers/ other members of the team? | 24 | 5(16, 17, 20–22) |
| 4. Were pregnant people asked what information they considered to be essential, important, unhelpful? | 24 | 5(22–26) |
| 5. Were pregnant people asked if they received appropriate information before making a choice? | 24 | 4(21, 26–28) |
| 6. Was it recorded when was the decision to undergo screening, or not, made (e.g. on the same visit)? | 25 | 4(17, 23, 29, 30) |
| 7. Was it recorded who was involved in the decision making to undergo screening, or not? | 23 | 6(17, 19, 23, 24, 26, 31) |
| 8. Were pregnant people's decisions/scores correlated to SES, education, and/or Race/ethnicity? | 2(22, 32) | 27(15–21, 23–31, 33–43) |

Results

Twenty-nine studies measured informed choice (Table 2). Twenty-seven studies were quantitative and or/ mixed methods with qualitative interviews [15-41] and two studies were qualitative [42, 43]. The two qualitative studies, while not measuring informed choice, included sections in the results and discussion about people's knowledge and/or informed choice. Measurements of informed choice varied. Four studies measured knowledge [15, 21, 24, 28], six studies measured knowledge in relation to uptake of screening [19, 31, 32, 37, 41, 42], two studies measured knowledge in relation to levels of informed choice [16, 33] and 17 studies defined and measured informed choice [17, 18, 20, 22, 23, 25-27, 29, 30, 34-36, 38-40, 43]. The findings from the broader narrative review are described first to provide a context for the secondary qualitative analysis.

Types of screening and sample size

Types of screening included non-invasive prenatal testing (NIPT), maternal serum screening and/or nuchal translucency screening. The largest study comprised of 4111 people, and the smallest 38 (Table 2).

Definition of informed choice

Informed choice was often used interchangeably with informed consent and informed decision (making), within and between studies. Not all studies provided definitions of these terms.

Fifteen out of 29 studies used the multi-dimensional measure of informed choice (MMIC) that incorporates a definition of informed choice and the parameters that were measured according to that definition (Table 2):

The basis of the MMIC is that an informed choice to undergo a screening test occurs when an individual has a positive attitude towards undergoing a test, has relevant knowledge about the test and undergoes it. An informed choice to decline a test occurs when an individual holds a negative attitude towards undergoing a test, has relevant knowledge about the test and does not undergo it. The choices that occur when individuals do not have relevant knowledge or when their attitudes are not reflected in their behaviour are uninformed [38].

Description of informed choice and knowledge

Adjectives such as 'well', 'sufficient', 'mostly' and 'poor' were commonly used to describe levels of informed choice or states of being knowledgeable in relation to the test outcomes, for example:

Overall, the level of knowledge of the patients was moderate, with 227 of the 305 patients (74%) having a satisfactory knowledge score (> or = 10) [25].

As the above example shows, the level of knowledge, in this case described as 'satisfactory,' was based on a threshold of correctly answering 10 or more questions.

For studies that reported 'low' levels of informed choice, conclusions and future work included the need for development of health promotion strategies, and information to meet diverse learning needs.^{e.g.} [38]. Other studies focused on overcoming language barriers e.g. through the use of translated material and professional interpreters [34] and interventions to increase comprehension for women with low education. In a French study, the authors questioned the *"legitimacy of consent for first-trimester ultrasound scans"* because of low levels of informed choice that were ascertained [25].

Knowledge tested, and measurements used, for quantifying informed choice

The questions utilised in MMIC comprised eight questions about knowledge of the screen, possible consequences of a diagnostic test (e.g. amniocentesis), probability of a negative or positive screening result (based on the question "*If 100 women decided to have the screening test, about how many do you think would have a low-risk result*? With a multiple choice between:

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| Author | Date | Title | Country | Methodology | Type of measure | Number of participants | Research question/Purpose of study | Screening method | % In- formed choice |
|--|------|---|-----------|--|--|---------------------------|--|---|---------------------------|
| Chan LW, Chau MCM, Leung TY, et al. [23] | 2005 | Expectation and knowledge of women undergoing first- trimester combined screening for Down syndrome in a Chinese population | Hong Kong | Questionnaire | Knowledge of condition Recall Understand risk | 325 | Study the preference of pregnant women regarding the time taken to report the results of first-trimester combined screening for Down syndrome and their knowledge about it | Combined nuchal translu- cency and biochemical screen- ing (1st trimester) | |
| Constantine ML, Allyse M, Wall M, et al. [24] | 2013 | Imperfect informed consent for prenatal screening: Lessons from the Quad screen | SU | Quantitative Questionnaire Faden Beau- champ's IC Controlling influence | Knowledge | 226 | The study evaluated patient informed consent (IC) for the Quad screen and examined differ- ences in IC between test acceptors and test refusers | Maternal serum Quad screen (2nd trimester) | 43.8% |
| Dahl K, Hvidman L, Jørgensen FS, et al. [15] | 2011 | First-trimester Down syn- drome screening: pregnant women's knowledge | Denmark | Questionnaire | Knowledge Clinical | 4095 | Assess pregnant women's knowledge of first- trimester combined Down syndrome screening in a setting of required informed consent Identify relevant differences in knowledge level among subgroups of pregnant women, includ- ing those informed in different ways about prenatal examinations | Combined nuchal translu- cency and biochemical screen- ing (1st trimester) | |
| Dahl K, Hvidman L, Jørgensen FS, Kesmodel US. [33] | 2011 | Knowledge of prenatal screening and psychological management of test decisions | Denmark | Quantitative Questionnaire | Knowledge Clinical Decisional conflict | 411 | Study associations between pregnant women's knowledge of prenatal screening and decision- al conflict in deciding whether to participate in first-trimester screening for Down syndrome in a setting of required informed consent, and to study associations between knowledge and personal wellbeing, and worried in pregnancy | Combined nuchal translu- cency and biochemical screen- ing (1st trimester) | |
| Dormandy E, Michie S, Hooper R, Marteau TM. [29] | 2005 | Low uptake of prenatal screening for Down syndrome in minority ethnic groups and socially deprived groups: a reflection of women's at- titudes or a failure to facilitate informed choices? | ň | Questionnaire | Knowledge Attitude MMIC | 1499 | Assess whether the lower uptake of prenatal screening for Down syndrome in women from minority ethnic groups and socioeconomically disadvantaged groups reflects more negative attitudes towards undergoing the test (similar rates of informed choices) or greater inconsis- tency between uptake and attitudes (lower rates of informed choice) | | 20-56 |

| Author | Date | Title | Country | Methodology | Type of measure | Number of participants | Research question/Purpose of study | Screening method | % In- formed |
|--|------|--|-------------|--------------------------------------|---|---------------------------|--|---|-----------------|
| Farrell RM, Nutter B, Agatisa PK. [34] | 2011 | Meeting patients'education and decision-making needs for first trimester prenatal aneuploidy screening | S | Quantitative Questionnaire | Knowledge Select clinical information | 139 | Assess knowledge and decision-making of pa- tients who present for first trimester aneuploidy screening | Combined nuchal translu- cency and biochemical screen- ing (1st trimester) | |
| Farrell R, Hawkins A, Barragan D, et al. [16] | 2015 | Knowledge, understanding, and uptake of non-invasive prenatal testing among Latina women | S | Mixed Interviews Questionnaire | Knowledge Select clinical characteristics | 63 | Assess Latina patient understanding of non- invasive prenatal testing (NIPT) and identify what factors influence uptake/refusal to NIPT to adapt counselling to the needs and interests of this population | NIPT | |
| Favre R, Duchange N, Vayssière C, et al. [35] | 2007 | How important is consent in maternal serum screening for Down syndrome in France? Information and consent evaluation in maternal serum screening for Down syndrome: a French study | France | Quantitative Questionnaire | MMIC Knowledge of condition Risk associated with screen Select clinical information | 305 | Evaluate the level of information and informed consent for maternal serum screening (MSS) for Down syndrome (DS) in the second trimester of pregnancy and analyse the exercise of autonomy towards the test by the women concerned | Maternal serum screening | 40.3% |
| Favre R, Moutel G, Duchange N, et al. [27] | 2008 | What about informed consent in first-trimester ul- trasound screening for Down syndrome? | France | Quantitative Questionnaire | MMIC Knowledge of condition Rating of informa- tion provided by professional Select clinical information | 350 | To assess the level of information and informed consent before the first-trimester ultrasound scan and the nature of information given by medical professionals | | |
| Fransen MP, Essink-Bot ML, Vogel I, et al. [36] | 2009 | Ethnic differences in informed decision-making about prenatal screening for Down's syndrome | Netherlands | Mixed Interview | MMIC Knowledge | 270 | Assess ethnic variations in informed decision- making about prenatal screening for Down's syndrome and to examine the contribution of background and decision-making variables | Not stated | 5-71% |
| Gourounti K, Sandall J. [37] | 2006 | Do pregnant women in Greece make informed choic- es about antenatal screening for Down's syndrome? A questionnaire survey | Greece | Quantitative Questionnaires | MMIC Knowledge | 135 | Investigate the knowledge and attitudes towards Down's syndrome among pregnant women presenting for prenatal screening in Greece, in order to explore whether Greek women are able to make informed choices | Combined nuchal translu- cency and biochemical screen- ing (1st trimester) | 44% |

Table 2 (continued)

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| Author | Date | Title | Country | Methodology | Type of measure | Number of participants | Research question/Purpose of study | Screening method | % In- formed choice |
|---|------|---|-----------|--------------------------------------|--|---------------------------|---|--|---------------------------|
| Jaques AM, Sheffield LJ, Halliday JL. [28] | 2005 | Informed choice in women attending private clinics to undergo first-trimester screen- ing for Down syndrome | Australia | Questionnaire | MMIC Knowledge | 174 | Examining whether women having first-trimes- ter screening in a private clinic had made an informed choice | Combined nuchal translu- cency and biochemical ing (1st trimester) | 68-74% |
| Kuppermann M, Learman LA, Gates E, et al [43] | 2006 | Beyond race or ethnicity and socioeconomic status: predic- tors of prenatal testing for Down syndrome | USA | Quantitative Questionnaire | Knowledge | 819 | To further investigate the extent to which sociodemographic variation in prenatal test use can be explained by understanding of testing risks and benefits and preferences, values, and attitudes | | |
| Lewis C, Hill M, Chitty LS. [20] | 2016 | A qualitative study looking at informed choice in the con- text of non-invasive prenatal testing for aneuploidy | N | Qualitative Interviews | MMIC Knowledge Recall Deliberation | 45 | Explore women's attitudes towards non- invasive prenatal testing (NIPT) and determine factors influencing their decisions around uptake of NIPT | NIPT | |
| Lewis C, Hill M, Skirton H, Chitty LS. [17] | 2016 | Development and validation of a measure of informed choice for women undergoing non-invasive prenatal testing for aneuploidy | CK | Mixed Interviews Questionnaire | MMIC Knowledge Select clinical information Decisional conflict | 585 | Validate a modified MMIC instrument for NIPT following screening in a public health service | TqIN | 89% |
| Lewis C, Hill M, Chitty LS. [21] | 2017 | Offering non-invasive prenatal testing as part of routine clinical service. Can high levels of informed choice be maintained? | СK | Quantitative Questionnaire | MMIC Knowledge Decisional conflict | 220 | Assess rates of informed choice among women offered non-invasive prenatal testing (NIP) for aneuploidy as part of routine clinical care | TqIN | 75.6% |
| Lo TK, Chan KYK, Kan ASY, et al. [38] | 2017 | Informed choice and decision making in women offered cell-free DNA prenatal genetic screening | Hong Kong | Quantitative Questionnaire | MMIC Knowledge | 215 | Assess informed choice in at-risk women who were offered secondary screening by cf-DNA | NIPT | 80% |
| Piechan JL, Hines KA, Koller DL, et al. [25] | 2016 | NIPT and Informed Consent: an Assessment of Patient Understanding of a Negative NIPT Result | SU | Mixed Questionnaire | MSS Knowledge Questionnaire | 95 | Evaluate patients' basic understanding of NIPT, such as conditions assessed and accuracy. Investigated patient self-assessment of NIPT knowledge and satisfaction with the testing process | TqIN | |
| Pop-Tudose ME, Popescu- Spineni D, Armean P, Pop IV. [31] | 2018 | Attitude, knowledge and informed choice towards prenatal screening for Down Syndrome: a cross-sectional study | Romania | Quantitative Questionnaire | MMIC Knowledge | 530 | Investigate the knowledge and attitude towards prenatal Down syndrome screening in order to asses to what extent the Romanian women make informed choices in this area | Not stated | 14.2% |

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|--|--------|--|-------------|--|--|--|--|---|---------------------------|
| Author | Date | Title | Country | Methodology | Type of measure | Number of participants | Research question/Purpose of study | Screening method | % In- formed choice |
| Potter BK, O'Reilly N, Etchegary H, et al. [22] | 2008 | Exploring informed choice in the context of prenatal testing: findings from a qualitative study | Canada | Qualitative Interviews | MMIC Knowledge | 38 | Explored whether and how a sample of women made informed choices about prenatal testing for foetal anomalies; its aim was to provide insights for future health policy and service provision | Maternal serum screening | |
| Pruksanusak N, Suwanrath C, Kor-anan- takul O, et al. [39] | 2009 | A survey of the knowledge and attitudes of pregnant Thai women towards Down syndrome screening | Thailand | Quantitative Questionnaire | Knowledge Clinical information | 714 | Determine the knowledge and attitudes of pregnant Thai women towards Down syn-drome screening | Nuchal translucency | |
| Rowe HJ, Fisher JRW, Quinlivan JA. [40] | 2006 | Are pregnant Australian women well informed about prenatal genetic screening? A systematic investigation using the Multidimensional Measure of Informed Choice | Australia | Quantitative Questionnaire | MMIC Knowledge Psychometric Anxiety and Depression | τ 4 ε | To assess informed choice to participate in sec- ond trimester maternal serum screening (2MSS) in pregnant women using a validated measure and to compare anxiety levels in women who were well informed versus poorly informed | Maternal serum screen- ing (2nd trimester) | 37% |
| Schoonen M, Wildschut H, Essink-Bot M-L, et al. [18] | 2012 | The provision of information and informed decision-making on prenatal screening for Down syndrome: a question- naire- and register-based survey in a non-selected population | Netherlands | Quantitative Questionnaire | Knowledge MIMIC | 335 20 midwifery practices | Evaluating the information provision procedure about prenatal screening for Down syn- drome, using informed decision-making as a quality-indicator | | 75.5% |
| Seven M, Akyüz A, Eroglu K, et al. [19] | 2016 | Women's knowledge and use of prenatal screening tests | Türkiye | Quantitative Questionnaire | Knowledge Clinical information | 274 | Determine the rate of use of prenatal screening tests and the factors affecting the decision to have a prenatal screening test in pregnant women in Turkey | Combined nuchal translu- cency and biochemical screening | 34.3% |
| Sheinis M, Bensimon K, Selk A. [30] | 2017 | Patients'Knowledge of Prena- tal Screening for Trisomy 21 | Canada | Quantitative Questionnaire Health literacy | Knowledge Clinical information | 135 | Assess the knowledge of prenatal screening for Trisomy 21 in pregnant women in one institu- tion in Canada | Integrated prenatal screening | 74% |
| Skirton H, Barr O. [26] | 2009 | Antenatal screening and informed choice: a cross-sec- tional survey of parents and professionals | ň | Quantitative Questionnaire | | 189 women, partners, and midwives | Investigate knowledge of screening in both prospective parents and professionals offering screening in England, and to identify the ways in which pregnant women and their partners could be supported to make informed deci- sions about antenatal screening | Not stated | |

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|--|--|---------|--|-------------|--------------------------------|---|---------------------------|--|--|---------------------------|
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| van den Berg2006Informed decision makingNetherlandsQuantitativeKnowledge,962To construct a measure of informed decisionNuchalM, Timmer-in the context of prenatalQuestionnairedeliberation, andmaking that includes knowledge, deliberation,tanslucencymans DRM,screeningcreeningQuestionnairedeliberation, andmaking that includes knowledge, deliberation,tanslucencymans DRM,screeningvalue-consistencynad value-consistency, and to assess the levelorten Kate LP,etal [32]etal (32)of informed decision making about prenatalscreening, and value-consistency, and to assess the levelorWyter KH,2011Are Adolescents' decisionsAustraliaQuantitativeMMIC232Determine whether pregnant adolescents arcscreeningRowe HJ,about Prenatal Screening forQuestionnairesAnxiety andLoss and test decliners.screeningmaternalRowe HJ,about Prenatal Screening forQuestionnairesAnxiety andLoss and test decliners.screeningscreeningRowe HJ,about Prenatal Screening forQuestionnairesAnxiety andLoss and test decliners.screeningscreeningRowe HJ,about Prenatal Screening forQuestionnairesAnxiety andLoss and test decliners.screeningscreeningRowe HJ,about Prenatal Screening forQuestionnairesAnxiety andLoss and test decliners.screeningRowe HJ,about Prenatal Screening | van den Berg M, Timmer- mans DRM, ten Kate LP, et al. [41] | 2005 | Are pregnant women mak- ing informed choices about prenatal screening? | Netherlands | Quantitative Questionnaire | Knowledge Clinical information Decisional conflict Attitude Anxiety | 1159 | Assess to what extent pregnant women make informed choices about prenatal screening, and to assess the psychological effects of informed decision-making | Nuchal translucency or Maternal serum screening | 68% |
| Wynter KH,2011Are Adolescents' decisionsAustraliaQuantitativeMMIC232Determine whether pregnant adolescents areMaternalRowe HJ,about Prenatal Screening forQuestionnairesAnxiety andless likely to make informed choices about un-serumFisher JR, etDown Syndrome Informed? ADepressiondertaking this test than adult pregnant womenscreeningal. [42]Controlled, Prospective StudyKnowledgeNowledgescreening | van den Berg M, Timmer- mans DRM, ten Kate LP, et al. [32] | 2006 | Informed decision making in the context of prenatal screening | Netherlands | Questionnaire | Knowledge, deliberation, and value-consistency | 962 | To construct a measure of informed decision making that includes knowledge, deliberation, and value-consistency, and to assess the level of informed decision making about prenatal screening, and differences between test accep- tors and test decliners. | Nuchal translucency or Maternal serum screening | 51% |
| | Wynter KH, Rowe HJ, Fisher JR, et al. [42] | 2011 | Are Adolescents' decisions about Prenatal Screening for Down Syndrome Informed? A Controlled, Prospective Study | Australia | Quantitative Questionnaires | MMIC Anxiety and Depression Knowledge | 232 | Determine whether pregnant adolescents are less likely to make informed choices about un- dertaking this test than adult pregnant women | Maternal serum screening | 10-37% |

100, None, 50, 95, 5, Not sure"); and attitudes towards the screen, presented as: "For me, having the screening test for Down's syndrome when I am 15 weeks pregnant will be: (a) Beneficial 1 2 3 4 5 6 7 Harmful (b) Important 1 2 3 4 5 67 Unimportant (c) Bad thing 1234567 Good thing (d) Pleasant 1 2 3 4 5 6 7 Unpleasant".

Other measurements for quantifying informed choice comprised surveys and questionnaires with multiplechoice or true-false questions, or measures of selfreported knowledge with Likert-scale responses to various statements (e.g., the screening test aims to screen for disease or abnormalities, or the maternal blood tests and detailed ultrasonography can increase detection rate for Down syndrome screening). The number of questions that people were asked varied from 8 to 51. In none of these studies were respondents invited to make inferences or connections between facts, or to process any new information (that is, to measure 'understanding'); although in one study this was discussed and reflected in how the questions were developed [22].

Questions focused on risk, testing knowledge, (e.g., asking respondents if serum markers may also be used to screen for spina bifida), or technical knowledge (such as being able to know what the positive predictive value of the screen is). Mathematical concepts were also components of the tests of knowledge, such as "If a patient has an estimated risk of 1/50, what is the probability that the foetus has Down syndrome?" with a multiple choice of "20%, 10% or 2%" [33]. Six out of 29 studies also measured aspects such as depression, anxiety, decisional conflict and deliberation (Table 2).

Seventeen out of 29 studies provided an overall quantification of the level of informed choice (presented as a percentage; Table 2). The level of informed choice ranged from 10 to 89%. Other studies provided a range of percentages by question type, and not an overall level of informed choice. No study indicated whether the people were shown their results (e.g. levels of informed choice) to see whether this was a 'true' reflection of how they felt.

One of the qualitative studies explored informed choice through the MMIC [43]. In the other qualitative study, measures of knowledge were qualified in the text:

All the women interviewed were aware that NIPT tested for Down syndrome. Nevertheless, when asked about their understanding of the condition, this was found to be variable [42].

Epistemic positioning

Responses to the secondary qualitative analysis questions to explore pregnant people's epistemic positioning are shown in Table 1.

Study design, measure development and informational needs

No studies reported that pregnant people or women were involved in the design of the study. Five out of the 29 studies explicitly stated that pregnant people and women were involved in the development of the measure of informed choice [11–15]. Their level of involvement included being part of the piloting process^{e.g.} [15]. One paper described how people were consulted about the phrasing and the option to include questions [17]. One paper based specific knowledge components determined by people as being important for pregnant people to know about [18]. This process was ascertained by deriving content domains for knowledge about screening extracted from the literature in a prior study [44].

Five studies collected information about whether the type of information they were given met their needs [18–20, 42, 43]. The provision of information was sometimes discussed in relation to the screening guideline expectations of a particular country. In an Australian-based study *"all women having testing should be provided with written information, and have appropriate understanding of the test(s)"* but these expectations were not met, *"even in private clinical care"* [26].

Education, ethnicity, and race as population descriptors

Twenty-seven out of 29 papers used educational attainment as a population descriptor (Table 1), and were described using statements such as:

The only statistically significant predictor of making an informed choice pre-screening was tertiary (degree or higher) education level [26].

Or

Knowledge was clearly associated with education [31].

In one study, 89% of people were considered to have made an informed choice, with 65.9% of the participants having a tertiary degree or higher, highlighting the study's overrepresentation of educational privilege [20].

Ethnicity or race were frequently used to explain differences between people's uptake of screening, knowledge of screening or levels of informed choice (12 out of 29 papers). Ethnicity and race were used interchangeably within papers. Race was often reported as White, Black, Hispanic, or non-White. There were often no discussions of what the underlying assumptions were for any of these population descriptors, or whether people had the option of self-identifying ethnicity or race which were then categorised by the researchers. In terms of presenting the findings, racialised comparisons were common with descriptions:

Multivariable regression analysis demonstrated that ethnicity and NIPT uptake were significant predictors of informed choice when controlling for education, age, having a religious faith, parity and screening risk. White participants had almost three times higher odds of making an informed choice than other ethnic groups who were more likely to agree to testing despite their beliefs [17].

Or

No statistically significant association was identified between race and knowledge levels [32].

In the Discussion, results by ethnicity or race were often mapped to findings from other studies based in different countries:

The ethnic differences in informed decision-making found in our study are larger than those reported in the UK; in the latter study, 56% of the English, 20% of the South-Asians and 28% of the Black African Caribbean women made an informed decision whether or not to participate in prenatal screening. In our study, especially the Turkish women scored much lower on informed decision-making compared with the ethnic minority women in the UK [34].

As 27 out of 29 studies were quantitative, they included statistical analyses such as logistic regression. Statements such as 'significantly different [white vs.]', and 'controlling for [ethnicity, education...]' were frequent, conveying an epistemic authority of the measure, and resultant outcomes. When differences by ethnicities/race were observed, explanatory variables used included education, age, language (if not the dominant language of the country where the study was undertaken), faith and attitude towards screening, or having a child with a genetic condition.

Differences between Surinamese and Dutch women could largely be explained by differences in age and educational level. Differences between Dutch and Turkish women could mainly be explained by differences in gender emancipation and language skills [30].

Discussions about equity in access to healthcare was infrequent. There was also no, or little, discussion and interpretation of the findings in relation to social determinants of health, impacts of racism and discrimination in health and/or education.

Other frequently used population descriptors

Other frequently used population descriptors included annual income and type of work (e.g. blue collar). In the quantitative analyses, these were treated as explanatory variables often combined or discussed with educational attainment. Age, especially over 35 years old, of the pregnant person was sometimes used to stratify analysis. One paper specifically focussed on young people, with discussion acknowledging the complex information young people are expected to understand, and for healthcare providers to communicate clearly [40].

Relational aspects of informed decision making were reflected in the collection of marital or partner status, but rarely discussed as such. Experiential knowledge of screening was reflected through the collection of information about any previous pregnancy with a genetic condition and/or previous termination of pregnancy due to a chromosomal condition and number of previous pregnancies (parity) or children, although it was not evident how the previous pregnancy experience supported informed choice. Evidence of people's norms/preferences predominantly included religiosity and language. Religion or faith, often measured as 'very', 'somewhat' or 'not at all, was also collected as a values-based component of informed choice and/or testing decision. Language proficiency (if it was not the dominant language of the country) was used as a requirement for participation.

Reification of many of the above-described descriptors was evident as characteristics of epistemic credibility for making an informed choice.

Demographic characteristics such as older age (Schoonen et al. 2012 [18]) and being of a certain ethnicity (Dormandy et al. 2005; [27] Fransen et al. 2010 [34]), as well as social determinants of health such as higher educational level (Rowe et al. 2006 [38]) have been correlated with higher rates of informed decision making [28].

Two papers did not describe the findings by reference to any population descriptors. One did not state a reason [30], and one did, which was:

We did not capture data on either level of education (to avoid any perception that women were being 'tested') or religious affiliation (we felt it was more appropriate for women themselves to identify this if relevant) [43]0.18.

Discussion

The aim of this scoping review and secondary qualitative analysis was to describe how pregnant people and women have been positioned as knowers in relation to prenatal screening for chromosomal conditions. The analysis was undertaken based on defined questions that had been developed a priori, and a narrative summary of the included studies. To the best of our knowledge, this is the first report that has explored epistemic positioning in relation to informed choice, and more specifically of pregnant people and women. We are not making any assumptions about the belief systems of others working in the area. The goal of this work is to extend ways in which informed choice could be conceptualised and measured so that people's epistemic credibility is not undermined. In addition, this work aligns with guidelines about the use of population descriptors, in particular race and ethnicity, in genetic and genomic, epidemiological studies and in medicine more broadly [45, 46].

The use of population descriptors as explanatory variables had the effect of positioning (groups of) people to be more, or less, epistemically credible in relation to informed choice than others. Educational attainment, ethnicity, race and socioeconomic status in particular were often reified as characteristics of epistemic credibility for informed choice in the biomedical hegemony. The layering that was often applied in the analyses to control for certain and multiple population characteristics (e.g., education and race and age and socioeconomic status) compounds the effect of epistemic harm/privilege. Such epistemic positioning has the potential to perpetuate harm towards people in their respective positions as knowers, i.e., to perpetuate epistemic injustices [2, 12]. There are several potentially different ways that epistemic harm could be constituted in a healthcare context.

In Western society, education and employment are often viewed as indicators of intelligence and/or social status [4]. The findings from this review have shown that there were clear patterns of educational privilege and informed choice in the biomedical hegemony. Education and socioeconomic descriptors were frequently described as characteristics of informed choice. Such positioning risks pre-emptive testimonial injustices in practice [2, 47]. That is, such epistemic positioning could be conferred to people in practice, affecting what or how information is shared, or not, by a practitioner based on assumptions about someone's ability to 'understand' because of their educational attainment (or other characteristic). Education and employment status would better used as contextual variables for, indicators of, social privilege within the biomedical hegemony. Race is a social construct premised on colonial and imperial thinkings, and used as a, albeit poor, proxy for social class, culture and genes [45, 46]. Describing or reporting findings by, or to, ethnicity/race, also has the added potential to marginalise (racialise), positioning people as the 'problem' when the root causes of marginalisation lie elsewhere in a society's history (e.g., colonisation), social structure (e.g., privileging whiteness), or organisational racism that prevents equal access to health care, and education.

Other examples of how epistemic harm could be constituted in context relate to the presupposition, that in measurements of informed choice to date, biomedical hegemony dictates the forms and types of measures required. Biomedical knowledge is the dominant model of health and illness in the Western world. Our findings highlighted how biomedical knowledge was the expected norm for pregnant people to make an informed choice. This is not to say that biomedical knowledge is not relevant or important in this context, but not to the exclusion of other ways of knowing [13]. To continue considering informed choice (and measurements of) in terms of the biomedical hegemony places people at risk of hermeneutical injustices, that is, not enabling people to express the experience in a way that makes sense to them [2, 3]. In the measurements in this review, participants were not often given the opportunity to engage with research (and the resultant measure). It is likely that the information gathered was not comprehensive or reflective of their experience. Exclusion based on language of the dominant language of the country where the study was undertaken would have prevented the representation of women and pregnant people who may experience language barriers when receiving information (which would most likely impact their opportunity to make an informed choice). The exclusion of perspectives from patients speaking a non-dominant language also restricts the information that is gathered, and exhibits an expectation for patients to adapt to the needs of researchers to be heard and included (rather than having their language needs met to ensure participation). Epistemically just measures of informed choice could be achieved through co-design and approaches that prioritise other ways of knowing, and knowledge that go beyond the biomedical hegemony [12, 13, 47–50]. As any 'measure' comprises defined criteria, even if criteria are patient-defined metrics of knowing, future work would need to include how to mitigate the potential for epistemically (de)privileging certain groups of people by any such defined criteria. Whichever criteria are used, transparency around what they do, and do not, consider would contribute to a more epistemically just approach.

Population descriptors per se are not 'bad'. However, using population descriptors as contextual variables rather than explanatory variables would better uphold (pregnant) people as credible knowers beyond biomedical knowledge. Used as contextual variables, such as indicators of social privilege for example, that need contextualising [45, 46]. Future developments towards how informed choice is measured and reported could, as recommended for other aspects of genetic and epidemiological research, include explanations about the underlying assumptions for the population descriptors used, and/or decisions about how race/ethnic groupings were made. Additionally, as previously described, population descriptors could be developed to include, for example, measures of racism [46]. Such approaches could be incorporated into documentation about prenatal screening e.g., monitoring reports or service audits and evaluations. Where for example, if information about race/ ethnicity are collected describing these as indicators of risk of exposure to e.g., racism, discrimination and not as risk factors per se; or as contextual variables, which could include: accessibility of services (e.g., caseloads, number and type of ante-natal clinics in the community, consultation time); social-geographical measures of privilege (e.g. area decile rating); opportunity for self-determination in reporting (e.g., ethnicity); information (e.g., people's cultural values and preferences for information sharing, relational aspects). In-line with the findings and focus of this review and secondary analysis, would be the involvement of (pregnant) people, and communities, where for example, people with lived or living experience of prenatal screening and/or genetic conditions would be involved in the co-design, at each stage, of reporting and evaluation activities and initiatives. In particular, prioritising the inclusion of people who are deprivileged by the current model. Such a co-design approach could better meet local community contexts.

Strengths and limitations

We excluded studies that were not written in English, as we did not have capability to translate non-English texts; we acknowledge that this a limitation given the criticism of studies using only the dominant language, and we may have omitted more epistemically just research as a consequence. Although we have applied rigorous systematic review methods, there are some limitations to this study. As informed choice is defined in a number of ways, the search terms may not have included every definition and possible way that informed choice could be indexed in the databases. As the analysis progressed, it became apparent that there are other dimensions to supporting informed choice and epistemic justice which we did not include, such as: length of consultation, physical accessibility, frequency of consultations, and a more in-depth analysis of how the studies pertained to the political and social norms of the country it is based in. However, as this is the first time that epistemic positioning has been undertaken in the context of informed choice, the 'limitations' that we have identified are important findings in the context of developing and extending this methodology which

could be applied and used to strengthen future similar studies.

Conclusions

The use of population descriptors as explanatory variables can have the effect of positioning (groups of) people to be more, or less, epistemically credible for informed choice in the biomedical hegemony. The use of educational attainment, ethnicity, race and socioeconomic status in particular had an effect of reifying these as characteristics of epistemic credibility for informed choice. Using population descriptors as contextual variables (and contextualising) in the analyses (e.g., as indicators of social privilege) would better uphold pregnant people and women as credible knowers.

Many of the studies that have been included in this review are 10 years old. A measurement for informed choice may be an elusive goal. However informed choice is still a central tenet of healthcare and considering informed choice in a way that is epistemically just is needed. More epistemically just 'measures' of informed choice could be developed through inclusive design that intentionally makes room for forms of knowing that go beyond the biomedical hegemony.

Abbreviations

MMIC Multi-dimensional measure of informed choice NIPT Non-invasive prenatal testing

Supplementary Information

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Supplementary Material 1

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Author contributions

N I-B Project design, reference searches, analysis and manuscript preparation. FC Project design, analysis and manuscript preparation. KD Project design, analysis and manuscript preparation. SB Project design, analysis and manuscript preparation. JS Project design, analysis and manuscript preparation. PS Project design, analysis and manuscript preparation. CB Project design, analysis and manuscript preparation. SF Led project design, analysis and manuscript preparation.

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Data availability

The data that support findings are openly available from the associated publications. The questions for the secondary analysis are included in the publication for transparency.

Declarations

Ethics approval and consent to participate

As a secondary analysis of already published work no ethical approval was required for this research.

Consent for publication

Not applicable.

Competing interests

The authors declare no competing interests.

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