Institution: University of Leeds

Unit of Assessment: 2

Title of case study: Improving informed parental choice in prenatal screening services

Period when the underpinning research was undertaken: 2000-2014

Details of staff conducting the underpinning research from the submitting unit:

Name(s):	Role(s) (e.g. job title):	Period(s) employed by submitting HEI:
Louise Bryant	Professor of Psychological and Social Medicine	2002-present
Jenny Hewison	Professor of the Psychology of Healthcare	1984-present
Shenaz Ahmed	Associate Professor in Public Health	2001-present

Period when the claimed impact occurred: 2013-2020

Is this case study continued from a case study submitted in 2014? N

1. Summary of the impact (indicative maximum 100 words)

Studies led by academics at Leeds have been instrumental in developing evidence-based information for parents in the UK and Pakistan and the training of UK health professionals. Our research into psychosocial aspects of screening for conditions during pregnancy has materially benefitted the care of over 4.2 million women in England and around 12,000 in Pakistan. We identify four main impact points:

1. Development of Public Health England's (PHE) "Screening Tests for You and Your Baby" resource given to over 4.2 million pregnant women since 2014 (~700,000 per year).
2. Training of over 10,000 health professionals since 2018 in preparation for the NHS roll-out of non-invasive prenatal testing in England in June 2021.
3. PHE's policy decision to offer Down's syndrome screening separately from Edwards' and Patau's screening.
4. Provision of culturally appropriate decision-support for beta-thalassaemia major (β-TM) screening programmes in Pakistan.

2. Underpinning research (indicative maximum 500 words)

Bryant, Hewison and Ahmed have researched psychosocial aspects of prenatal testing and informed choice for over 20 years, with an overarching theme of incorporating parent and professional perspectives. Bryant's work considers representations and understanding of disability, Hewison considers the assessment and understanding of screening technology, Ahmed focuses on health inequalities and screening in lower- and middle-income countries.

Facilitating informed choice underpins all UK national screening programmes. It is increasingly possible to screen prenatally for many conditions in a single test, potentially reducing the opportunity for women to make choices informed by their own experience and values. Before the implementation of the UK National Down's syndrome Screening Programme in 2004 (now PHE Fetal Anomaly Screening Programme, FASP)), patient facing screening information was written by local clinicians. Our research includes 35 studies and 68 peer reviewed articles within the assessment period and features the following:

i) A Randomised Controlled Trial to assess the effect of a Down's syndrome screening information video on test uptake, knowledge, and psychological stress **[1].** The key finding was that good quality information, regardless of media, increases knowledge of screening without affecting the choices women make (assessed as uptake of testing) or increasing their anxiety. This allayed clinical concerns that increasing the content of screening information would raise anxiety about foetal anomaly in pregnant women.

ii) A content analysis of 80 Down's syndrome screening leaflets from UK maternity services **[2]** identified high variability in the quality of information, with a negative bias in content about Down's syndrome at odds with policy aims of non-directiveness.

iii) The first Systematic Review of research on the psychosocial aspects of prenatal and neonatal screening **[3],** identified a lack of screening knowledge in women and health professionals; a lack of evidence-based support for decision-making and a lack of research with minority ethnic groups.

iv) We surveyed attitudes to testing and termination for 30 different conditions in 420 women in the UK **[4]**. We identified that attitudes depended greatly on what was being tested for. In particular, women made significant distinctions between trisomy 21 (Down's syndrome) and the more serious conditions trisomy 13 (Patau's syndrome) and trisomy 18 (Edwards' syndrome), challenging prevailing clinical assumptions that women's attitudes are largely 'test for all or nothing'.

v) Stereotypical assumptions about the unacceptability of termination to Muslims have led to inequalities in the offer of screening. Our research **[4-6]**, provided evidence that Pakistani Muslim women can hold positive attitudes towards testing and termination for conditions they consider severe, including β-TM. Our research shows that when parents in the UK and Pakistan make decisions about prenatal screening or termination of pregnancy for foetal anomaly, they negotiate their religious beliefs alongside personal experiences of disability and pragmatic concerns **[5-6]**. Muslim parents may hold beliefs that termination of pregnancy is forbidden under Islamic law. With colleagues in Pakistan we identified a *fatwa* (religious ruling) that states termination of pregnancy is permissible under Islamic law in specific circumstances, including for a serious health condition, such as β-TM, to support parental decision making **[7]**.

3. References to the research (indicative maximum of six references)

1. Hewison J, Cuckle H, Baillie C, Sehmi I, Lindow S, Jackson F, Batty J. Use of videotapes to inform choice in Down's syndrome screening: A Randomised Controlled Trial. *Prenatal Diagnosis*, 2001; 21: 146-9. DOI: 10.1002/1097-0223(200102)21:2<146::aid-pd3>3.0.co;2-m

Used as evidence to support the giving of information about screened-for conditions in the current UK 'NICE Antenatal care for uncomplicated pregnancies' [CG62]' guidelines.

2. **Bryant LD,** Murray J, Green JM, **Hewison J**, Sehmi I, and Ellis A. Descriptive information about Down syndrome: a content analysis of serum screening leaflets. *Prenatal Diagnosis*. 2001; 21(12):1057-63. DOI: 10.1002/pd.179

In addition to impact on NHS screening resources, the paper provides evidence to support how genetic counsellors should communicate information to parents in "Practice Guidelines for Communicating a Prenatal or Postnatal Diagnosis of Down Syndrome" by the American National Society of Genetic Counsellors". These guidelines are themselves widely cited in other guidelines and handbooks for medical practitioners https://link.springer.com/article/10.1007/s10897-011-9375-8.

3. Green JM, **Hewison J**, Bekker HL, **Bryant LD,** & Cuckle, HS. Psychosocial aspects of genetic screening of pregnant women and newborns: a systematic review. *Health Technology Assessment*, 2004; 8:33. DOI: 10.3310/hta8330

Commissioned by the NIHR HTA https://fundingawards.nihr.ac.uk/award/93/56/99 this remains the best cited SR in the field, (in top 25% of all outputs as scored by Altmetric). HTA monographs are only published if they are of a sufficiently high scientific quality, as assessed by peer reviewers and editors. The SR is extensively referenced in the current UK 'NICE Antenatal care for uncomplicated pregnancies' [CG62]' guidelines.

4. 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. DOI: 10.1002/pd.1701

Funded as part of the ESRC/MRC 'Innovative Health Technologies' Programme, Social and ethnic differences in attitudes and consent to prenatal testing, GBP182,000; the final report was peer reviewed https://www.york.ac.uk/res/iht/projects/l218252013.htm. The findings have been *replicated in three other countries (Saudi Arabia, Pakistan and Ethiopia) and published in leading international per-reviewed journals.*

5. Ahmed S, Atkin K, **Hewison J**, Green J. The influence of faith and religion and the role of religious and community leaders in prenatal decisions for sickle cell disorders and thalassaemia major. *Prenatal Diagnosis*. 2006; 26(9):801-9. DOI: 10.1002/pd.1507

Research commissioned by the NHS Sickle Cell and Thalassaemia Screening Programme.

6. Bryant LD, **Ahmed S**, Ahmed M, Jafri SH, Raashid Y. 'All is done by Allah'. Understandings of Down syndrome and prenatal testing in Pakistan. *Social Science & Medicine*, 2011; 72 (8)1393-1399. DOI: 10.1016/j.socscimed.2011.02.036

This paper is in the top 25% of all research outputs as scored by Altmetric. It is cited by the British Islamic Medical Association's ***submission to the NICE call for evidence regarding termination of pregnancy guidelines (currently under review),*** *as evidence of the importance of understanding parental religious beliefs.*

7. Jafri H, **Ahmed S**, Ahmed M, **Hewison J,** Raashid Y, Sheridan E. Islam and termination of pregnancy for genetic conditions in Pakistan: implications for Pakistani health care providers. *Prenatal Diagnosis*. 2012; 32(12):1218-20. DOI: 10.1002/pd.3987

This is a published research letter based on the scholarship of the authors. The Journal website states that Research letters "will only be considered if they are of exceptional educational interest, value or novelty". Research letters are also peer reviewed.

4. Details of the impact (indicative maximum 750 words)

Our research, expert advice, and testimony has been central to the development of patient-centred, culturally appropriate, prenatal testing policies and information practice in the UK and Pakistan. The impact of our research has been through our expert roles, either in direct collaboration with policy makers, government bodies and health services, or via testimony on national committees. Four examples of our research impact are provided here with the citations linked to Section 3; sources of corroboration (Section 5) are identified [A – I].

1. National parental screening resource "Screening Tests for You and Your Baby"

In England, 700,000 pregnant women a year are offered screening for 16 congenital and genetic conditions as part of their antenatal care. Screening is optional and the programme aims to support personal informed choice (increasing screening uptake is not considered an appropriate goal of prenatal screening). Prior to 2014, each screening programme had a separate information resource. In 2013, FASP invited **Louise Bryant** and **Jenny Hewison** to join a working group to create a single integrated booklet called 'Screening Tests for You and Your Baby' (STFYAYB) **[A]**. Our research was instrumental in the evidence-based approach to the content and format of the whole resource **[1]**, and in relation to screening for Down's syndrome, structural anomalies and haemoglobinopathies. Recommendations from our research on how to balance information about conditions **[2]** and facilitate informed choice **[3]** were implemented in the resource https://www.gov.uk/government/publications/screening-tests-for-you-and-your-baby. The booklet (originally published in 2014) is the FASP endorsed screening information resource for parents (https://www.gov.uk/government/publications/fetal-anomaly-screening-programme-standards), with copies given to virtually all 700,000 pregnant women per year **[B]**. Updates occur to reflect pathway changes and three versions have been produced since 2014.

A 2019 PHE survey found that most (92%) services gave a physical copy to all pregnant women **[C].** Of the remainder, most gave a physical copy only to those who preferred not to access the resource online. Only one service said they gave no physical copies. The survey also identified that midwives use the booklet as a prompt and to aid their discussion with women about screening choices. Midwives reported high parent satisfaction with STFYAYB in informing screening decisions, for example, *"Most women when they attend for booking, having already received the booklet, have had a look at it, particularly the section on screening for Down's, Edwards and Patau's syndromes, and generally find it useful and easy to read and understand."* ***[C]***

The resource has been translated into 12 languages and is available online (over 71,000 views in 2020 alone) and in audio and Easy Read formats. The Covid-19 pandemic and the need for online antenatal appointments have accelerated the planned move to digital information. Key content from STFYAYB is provided in a short animation 'Screening Tests for You and Your Baby' (2018) https://www.youtube.com/watch?v=_afr5oIlpTM available on YouTube (viewed over 40,000 times) and shown in some antenatal clinics. A PHE evaluation identified high ratings of the animation by women including those with English as a second language, and those living in areas of social deprivation **[D]**.

2. **Education and training for health professionals in facilitating informed choice**

Our research has had significant impact on the content of education and training around informed choice for NHS staff who deliver screening **[A]**. New online training introduced in 2017 'Screening and informed choice' describes the principles of screening and offering informed choice to parents considering antenatal and/or new-born screening. PHE state "*The module features interviews with a variety of experts and healthcare professionals"*; **Bryant's** interview within the module provides evidence-based advice on how to facilitate informed choice. An online evaluation by 1,055 users rated the module 4.4 out of 5. PHE recommends that all health professionals involved in offering screening should complete the module every 24 months. The film was also the focus of a 2019 PHE blog (**Bryant** described as a '*national screening expert'*), which has been viewed ~1,000 times **[E].**

The PHE FASP Non-invasive Prenatal Testing (NIPT) Information and Education Group chaired by **Bryant** are responsible for overseeing the development, delivery, and evaluation of NIPT education and training to midwives and other screening professionals **[A].** Around 10,000 screening professionals received face-to-face or online cascade training during 2017/ 2018 to improve understanding of informed choice and first-hand knowledge of Down's syndrome, Edwards syndrome and Patau's syndrome based on recommendations in our research. PHE evaluated the training using an approach developed with **Bryant** based on our research **[F]**, with 92% of respondents rating it 'good' or 'excellent' (62%). The evaluation **[G]** identified "*improved reported levels of confidence in discussing a diagnosis of Down's syndrome, Edwards' syndrome and Patau's syndrome. The number of people reporting low levels of confidence post training decreased 7-fold when compared to pre-training*". Attendee quotes included:
"I went with very limited knowledge regarding Down's syndrome, Edwards', Patau's and the NIPT but came away feeling I had learnt so much" (Midwife).

3. **Screening policy for Trisomy 21, Trisomy 18, and Trisomy 13**

Our findings **[4]** led to PHE's 2015 policy to offer screening tests for trisomy 18 and trisomy 13 separately from screening for trisomy 21 (Down's syndrome) instead of in a single test. In 2014, FASP aimed to introduce trisomy 18 and trisomy 13 to the first trimester 'Down's syndrome' screening pathway. The initial plan was to offer women a single 'trisomies test' that would produce a combined screening result, based on assumptions that women would accept or reject all tests in line with their attitudes towards termination of pregnancy. As a FASP Advisory Group member, **Hewison** provided evidence to the 'Task and finish laboratory outputs sub-group' that women do not hold 'all or nothing' attitudes towards testing, an important argument against implementing a single 'trisomies' test. The sub-group subsequently recommended that two different risk results be calculated, one for trisomy 21 only, and one for trisomy 13 and 18 together. This policy recommendation was accepted by the FASP Advisory Group in July 2014 and was implemented across England between April 2015 and March 2016 **[A]:** women can choose a) no screening, b) screening for all three conditions, c) screening for trisomy 21 only or d) screening for trisomy 13/18 only. The importance of differentiating between conditions was endorsed by representatives from the Down's Syndrome Association, Down Syndrome Research Foundation, SOFT UK (Support Organisation for Trisomy 13 and 18), and parent support organisation Antenatal Results and Choices. PHE figures show that since 2016 over 11,000 women have chosen to have screening for trisomy 21 only, or trisomy 13/18/ only. The policy recommendation was subsequently also implemented by Public Health Scotland on 28th September 2020 **[H]**.

4. **Development of culturally appropriate information for β-TM screening in Pakistan**

Our research **[4-6]** has supported the development of culturally appropriate information about screening for haemoglobinopathies in the NHS. In Pakistan, it is being used to evidence the need for a parent-focused approach within the Punjab's beta-thalassaemia major (β-TM) screening programme. Pakistan has a population of around 180 million people and a β-TM carrier frequency of more than 5% - over nine million carriers. Children born with β-TM in Pakistan have an average life-expectancy of around 10 years and require regular blood transfusions and chelation therapy. **Ahmed** leads a research network of academic clinicians in Pakistan, which includes the Provincial Minister of Punjab for Health and the Deputy Director of the Punjab Thalassaemia Prevention Programme (PTPP). **Ahmed** has been a formal advisor to the PTPP since 2016 **[I]**.

Our research findings have been used to develop training for genetic counsellors on providing culturally appropriate information to support informed screening choices. Parent information provided via the PTPP now incorporates the *fatwa* we identified **[7]**, which allows healthcare professionals to deliver a comprehensive service by enabling parents to opt for termination of an affected pregnancy in a Muslim country if they wish. Since 2016 this information resource has been given annually to around 1,500 couples (~7,500 by end 2020) identified via carrier screening programmes as being at higher risk of having a pregnancy affected by β-TM.

5. Sources to corroborate the impact (indicative maximum of 10 references)

A. Testimonial from Programme Director, FASP on expert evidence provided by **Bryant** and **Hewison** in information development and training activities, and to support claim that the trisomy screening policy decision was based on expert advice by **Hewison** supported by our research evidence, 26/11/2020

B. Letter from Director of UKNSC confirming the contribution of **Bryant** to the development of STFYAYB, 10/10/2014

C. Results of PHE survey about STFYAYB

D. PHE blog on value of translated STFYAYB and parent satisfaction with screening animation https://phescreening.blog.gov.uk/2019/09/25/digital-screening-information/;
PHE blog on 40,000 views of animation
https://phescreening.blog.gov.uk/2021/02/08/seven-new-nhs-antenatal-and-newborn-screening-animations-published/

E. Film of **Bryant** providing evidence-based information for health professionals about informed choice https://phescreening.blog.gov.uk/2019/08/07/helping-health-professionals-support-personal-informed-choice-in-screening/

F. Paper on which PHE based NIPT training evaluation: **Bryant LD**, Puri SC, Dix L, **Ahmed S**. Tell it Right, Start it Right: An evaluation of training for health professionals about Down syndrome. British Journal of Midwifery. 2016; 24(2):110-7.
DOI: 10.12968/bjom.2016.24.2.110

G. Blog on evaluation of NIPT training https://phescreening.blog.gov.uk/2018/09/19/nipt-implementation-how-professionals-felt-about-our-face-to-face-training-and-development-events/

H. Letter from Interim Chief Medical Officer, Public Health Scotland confirming trisomy policy "Important changes to the Scottish pregnancy screening programme", 13 August 2020

I. Testimonial from Deputy Director of the Punjab Thalassaemia Prevention Project, Sir Ganga Ram Hospital, Lahore, Pakistan. Testimonial to support impact of research and scholarship by **Ahmed & Hewison** to prenatal screening information for parents at high-risk of having a child with β-TM, 24/09/2020