Institution: Institute of Education

Unit of Assessment: 25

Title of case study: Neonatal screening: educating parents and health professionals to improve children's health

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

IOE researchers have helped the NHS to maximize the number of babies in the UK who are screened for a range of serious but treatable conditions when they are about a week old. This means that more children with one of the screened-for conditions can start a course of treatment quickly. The fruits of the IOE team's work – a suite of training materials for healthcare staff and information leaflets for parents – have had another important consequence. They have enabled parents in the UK and in other countries to make much more informed decisions on screening than they could in the past.

2. Underpinning research (indicative maximum 500 words)

Context: Midwives collect blood spots from the heels of almost all newborn babies in the UK to establish whether they have an inherited health condition. In 2011-12, more than 810,000 babies were screened and 1,481 were found to have one of the five disorders that they were tested for: cystic fibrosis, phenylketonuria, congenital hypothyroidism, sickle cell disorders and medium-chain acyl CoA dehydrogenase deficiency (MCADD). As prompt treatment can often prevent disability or even death, it is essential to learn as much as possible about the best ways of informing, and consulting, parents about the tests and their results. It is also vital that healthcare staff are expertly trained in best-practice approaches. The UK has had a national neonatal screening programme since 1969 but both the programme and the ethical issues it raises have changed markedly over the past 40 years. Parents' consent to neonatal tests used to be assumed and commercial companies often sponsored leaflets on screening. Recent years have witnessed a growing culture of transparency and informed consent. One way in which the Department of Health reacted to these developments in 2002 was by commissioning the UK Newborn Screening Programme Centre (UKNSPC), a collaboration between Great Ormond Street Hospital for Children NHS Trust, the Institute of Child Health and the IOE. The UKNSPC was given responsibility for developing, implementing and maintaining a high quality screening programme for all newborn babies. It has since created a broader evidence base, largely thanks to the IOE research featured here.

R&D methods: The IOE team conducted two systematic reviews of international research into communication about newborn screening that were published in 2003 and 2004, the latter **(R1)** answering questions raised by the former. Primary research included a survey of current practice in England and Wales, a qualitative study of parents' and health professionals' experiences of blood spot screening **(R2 & R3)**, and a survey of more than 100 screening leaflets used in the UK, Australia and the USA **(R4)**. Most were aimed at parents, others at health professionals. The researchers then applied the insights they had gleaned from their inquiries. They wrote their own leaflets in collaboration with parents and healthcare professionals (one is issued to parents before screening, others are given to parents if it is suspected that their baby has one of the five conditions). They produced a handbook and training materials for midwives and other staff involved in screening. The researchers also convened an advisory group: the Parent Information and Communication Working Group represented all four UK nations and included midwives and parents, a GP, health visitor, specialist nurse, geneticist and counsellor. Under the researchers' leadership, this 24-member group developed guidelines on communicating with parents about screening – during pregnancy and after birth. The guidelines were based largely on the researchers' findings but also on the group members' own experience as professionals or parents.

Key findings: The researchers established that parents were being offered little information about blood spot tests and even less choice. They also found that:

- Parents rarely refused screening but there was no uniform procedure for inviting informed consent.

- Leaflets portrayed screening in a wholly positive light and rarely addressed the difficulties that it can entail – such as occasional 'false-positive' results and the identification of family members who are carriers of genetic conditions.
- Poor communication of test results could cause parents needless anxiety.
- Many health professionals lacked experience or confidence in giving screening results that indicated a possible problem.
- Many parents of unaffected babies were not being told the results at all.

The qualitative study concluded that clear, brief and accurate parent information and effective communication between health professionals and parents were needed if informed choice and public health screening for children were to co-exist successfully. The survey of leaflets recommended criteria for judging parent information.

Researchers: Professor Sandy Oliver, Dr Katrina Hargreaves and Dr Ruth Stewart took the leading roles in this work. Oliver is the IOE's Public Engagement Champion. At the time of the research discussed in this case study Oliver was Reader in Public Policy while Hargreaves and Stewart were research officers.

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

R1: Oliver, S., Lempert, T., Stewart, R., Kavanagh, J, & Dezateux, C. (2004) *Disclosing to parents newborn carrier status following routine blood spot screening*. London: EPPI-Centre, Social Science Research Unit, IOE.

R2: Stewart, R., Hargreaves, K., & Oliver, S. (2005) Evidence informed policy making for health communication. *Health Education Journal,* 64(2), 120-128.

R3: Hargreaves, K. Stewart, R. & Oliver, S. (2005) Informed choice and public health screening for children: the case of blood spot screening. *Health Expectations* 8(2), 161-171.

R4: Hargreaves, K., Stewart, R., & Oliver, S. (2005) Newborn screening information supports public health more than informed choice. *Health Education Journal*, 64(2), 110-119.

R5: Hargreaves, K., Sinclair, J. & Oliver, S. (2007) *Evaluation of UK Newborn Screening Programme Centre Information Resources*. SSRU, IOE.

Grants: The work was funded by the Department of Health (DH) in England, on behalf of the four UK nations. The DH provided £441,132 to the IOE, through Great Ormond Street Hospital, between April 2002 and March 2010. Oliver was the grant-holder.

Indicators of quality: Oliver is an editor of the Cochrane Consumers and Communication Review Group. She is a member of the World Health Organisation Expert Advisory Panel on Clinical Practice Guidelines and Research Methods and Ethics, and the NICE Accreditation Advisory Committee, and co-directs the IOE's Research Councils UK-funded programme on public engagement with research.

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

Principal beneficiaries: Children with one of the screened-for conditions have gained most because their health needs are likely to be assessed more accurately and quickly than in the past. Parents of newborns have benefited too because they have been treated in a more considerate manner. Arguably, there is also a substantial financial saving for the NHS as the IOE team's work has reduced the likelihood of costly medical treatment and legal cases resulting from mistakes in the screening process. **Dates of impact**: The benefits have been felt throughout the REF period (2008-13) and are accumulating year-on-year.

Reach and significance: The IOE team have persuaded policy makers, practitioners and parents to help them translate their research into better evidence-based policy and practice. In doing so, they have improved the quality of information resources and given parents a bigger input into the screening process. They have also helped to ensure a higher quality of life for many children with the screened-for conditions, not only in the UK but around the world. They have had three forms of impact[1]: instrumental (influencing policy /practice), conceptual (enhancing general understanding and informing debate) and capacity-building.

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[1] Using Evidence: How Research can Inform Public Services (Nutley, S., Walter, I., Davis, H. 2007)

How the impact was achieved: The immediate impact of the IOE team's work was on the UKNSPC. As the Centre's parent support research director, Oliver directly influenced the implementation of national policy. The parents' leaflets and information for health professionals that the IOE researchers produced were distributed through regional antenatal and child health screening co-ordinators, public health directors, heads of midwifery, health visitors, laboratory directors and child health record departments. This process benefited from the researchers' inclusive approach. Not only did those involved ensure that the materials were appropriate, they also encouraged their subsequent use. The materials were also made publicly available via the UKNSPC website. Their use was then promoted in articles that the researchers wrote for voluntary sector publications and practitioner journals. For example, in 2005 they co-authored an article for the Royal College of Midwives magazine, describing the new standards and policies **(S1)**. Practitioners and parents were invited to co-author several of these articles to ensure they spoke directly to these readerships.

Devolved administrations and overseas: Soon after the UKNSPC was set up, its directors, including Oliver, visited devolved government departments in Belfast, Cardiff and Edinburgh to engage them in its work. In 2009, Oliver visited Edinburgh again to share with Scottish policy-makers the learning that had accrued since 2002. The researchers also delivered presentations on their work to practitioners and policy-makers in Australia (e.g. the Victorian Health Promotion Foundation, Melbourne, and Queensland Health, Brisbane) in order to demonstrate how stakeholders could be involved in evidence-informed policy-making.

Capacity building: In addition to designing training materials for use by others **(S2)**, the researchers contributed to CPD for health professionals around the country (e.g. for neonatal nurses in Bradford, screening co-ordinators in Oxford, counsellors in Manchester and midwives at hospitals in the East and South East of England).

Instrumental impact: *Communication with parents*: Before 2005, parents were not provided with impartial information on newborn screening. By 2011-12, largely because of the IOE team's work, clear, unbiased information was available to the parents of the 810,000 babies screened and the 1,481 who needed further diagnostic tests. It had been feared that if parents were told that screening could have disadvantages as well as advantages, many more would choose not to have their baby screened. However, this has not happened. Screening was declined by the parents of only 6 in 10,000 newborns in 2011-12 **(S3)**.

Discussions with midwives: As the researchers recommended, parents can now discuss blood spot screening with midwives during pregnancy and immediately before the test. This enables them to make an informed choice. At the time of the test, midwives explain the various research and public health uses of blood spots to mothers – an approach the IOE team advocated. The Supervisor of Midwives (**S4**) for the East Kent Hospitals University NHS Foundation Trust (one England's largest hospital trusts, serving around 759,000 people) said that they had been using the IOE researchers' leaflets for five years. "Having the leaflets makes it much easier for the midwives to discuss screening and for parents to understand the information about the range of conditions tested, the procedure itself and the risks and benefits", she said. "When providing mandatory training for all midwives on the screening and conditions we draw on the health professional's handbook and training materials [which the IOE team produced]. They are valuable resources to signpost if we have midwives who struggle to provide consistently good quality blood samples." A spokesperson for the Cambridge University Hospitals NHS Trust (**S5**) also confirmed that its midwives have been using the leaflets for parents since they were introduced. "They are well used and valued by both women and midwives", she said. Healthcare staff in Wales and Scotland use adapted versions of the leaflets.

Minority ethnic mothers: Screening leaflets have been translated into minority languages, another proposal that came from the researchers. In England, information is now available in 18 of the most commonly used languages other than English.

The changes referred to above have helped to bring about the following improvements:

Timely sample collection: Most areas of the UK have seen a year-on-year improvement (the proportion of children screened at between 5 and 8 days now ranges from 96% in Wales to 98% in Scotland and Northern Ireland). In England, the rate rose from 91% to 97% between 2006-7 and 2011-12.

Timely dispatch of blood spot cards: Again, the overall trend is upwards. In 2011-12, 96% of samples taken in England were received in the screening laboratory within four working days, an improvement of about 10 percentage points since 2006-7. The number of samples reaching the laboratory late (five working days or more after sample taken) has reduced from 14% to 4% between 2006-7 and 2011-12 **(S6)**.

Phenylketonuria (PKU): In 2011-12, 96 babies in England screened positive for PKU, which prevents the proper breakdown of an amino acid called phenylalanine. The great majority (95%) of these 96 samples were processed within three working days (compared with 90% in 2005) and 97% were dealt with within four working days (92% in 2005). Babies referred after 14 days now tend to have been born abroad or are very rare cases where there was a screening error.

Cystic fibrosis: Dr Kevin Southern **(S7)**, chair of the European Cystic Fibrosis Society neonatal screening working group, said it is "difficult to overstate" the impact that the UK screening programme and its information materials have had on cystic fibrosis care. "Children are receiving prompt and appropriate early care … [the parent information documents] were developed in collaboration with the IOE (Oliver's team) and provide parents with clear information on very difficult concepts. It is a credit to the UKNSPC that these freely available leaflets have been used as a resource by many programmes around the world."

Conceptual impact: *International influence*: The IOE team's work is also referred to on health information websites throughout the English-speaking world. The International Society of Nurses in Genetics **(S8)**, for example, cites their research in its 'position statement' on the nurse's role in newborn screening. It refers readers to **(R5)** after stating: "Though the risk of harm from the screening procedures is low, it cannot be assumed that the individual and societal benefits justify the assumption that parents should have no role in the screening process unless results are abnormal". The Health Issues Centre, a Melbourne-based body that provides a voice on health issues for communities and consumers, also cites their research **(S9)**. Their work has been referred to in overseas medical journals that are read by doctors and other health professionals **(S10)** and has been translated into other languages. For example, the Cochrane Library offers Spanish and Chinese **(S11)** translations of their paper on the problematical issue of notifying parents that a child is a carrier of an inherited condition.

Increased collaboration: In addition to their other achievements, Oliver and her colleagues have also demonstrated the value of adopting a collaborative approach to such R&D work and being committed to genuine user-engagement. They have helped to change not only policy and practice but the culture of blood spot screening.

5. Sources to corroborate the impact[2] (indicative maximum of 10 references)

S1: Judge, B., Pepper, J., Stewart, R. & Hargreaves, K. (2005) *Newborn blood spot screening.* RCN Midwives, 8(5), 216-218. http://www.rcm.org.uk/midwives/features/newborn-blood-spot-screening/

S2: Presentations prepared by the IOE team which support trainers in delivering training about newborn blood spot screening http://newbornbloodspot.screening.nhs.uk/education

S3: Morgan T and Coppinger C (2013) Data Collection and Performance Analysis Report. Newborn Blood Spot Screening in the UK 2011-12. UK Newborn Screening Programme Centre, London. See Fig. 39 and accompanying text http://newbornbloodspot.screening.nhs.uk/performance

S4: Supervisor of Midwives, East Kent Hospitals University NHS Foundation Trust

S5: Antenatal and Newborn Screening Midwife, Cambridge University Hospitals NHS Trust

S6: Morgan and Coppinger, Fig. 10

S7: Kevin Southern, Reader and Hon. Consultant in Paediatric Respiratory Medicine, Univ. of Liverpool, and Chair, European Cystic Fibrosis Society Neonatal Screening Working Group. http://newbornbloodspot.screening.nhs.uk/cms.php?folder=2716

S8: http://www.isong.org/pdfs2013/PS_Newborn_Screenings.pdf

S9: http://www.healthissuescentre.org.au/documents/items/2008/04/204877-upload-00001.pdf

S10: www.samj.org.za/index.php/samj/article/download/1082/545

S11: http://onlinelibrary.wiley.com/doi/10.1002/14651858.CD003859.pub2/abstract

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[2] All web links accessed 22/10/13