Institution: University of Southampton

Unit of Assessment: 15 General Engineering

Title of case study: 15-04 Improved child hearing tests for the best start in life

1. Summary of the impact

The Institute of Sound and Vibration Research (ISVR) has played an influential role in transforming testing for child deafness in Europe, North America and elsewhere. In England, the NHS drew on its findings in deciding to replace traditional testing methods with universal newborn hearing screening programmes. This form of testing is more accurate, cost-effective and can be conducted at an earlier age. In England alone more than four million babies will be screened between 2008 and 2013, with around 6,000 identified as having hearing impairments. Earlier clinical intervention has benefited children's language development and overall quality of life.

2. Underpinning research

Permanent childhood hearing impairment (PCHI) affects more than 1 in 1,000 babies. It can have adverse effects on a child's neuronal development, language acquisition and educational outcomes. These effects can harm an individual's emotional wellbeing and employment opportunities later in life, with further negative consequences for the UK economy and society.

But if PCHI is detected at an early age, children can be provided with educational support, hearing aids and cochlear implants, transforming their future prospects. Prior to 2001, the standard test for PCHI in the UK was the health visitor distraction test (HVDT), based on observation of the baby's reaction to a distracting sound. This method has two significant drawbacks: the reliability of the test and the relatively late developmental age – seven months – at which it can be carried out.

In the mid-1990s, the ISVR trialled what they believed to be a more accurate identifier of PCHI **[4]**. Ears with no impairment emit low level sounds called transient evoked otoacoustic emissions (TEOAEs), which can be detected by a microphone in the ear canal, and are usually measurable within the first few days after birth.

The ISVR also contributed to the only controlled trial comparing universal screening based on TEOAEs with the HVDT. The Wessex Trial (1993-1996) **[1]** was conducted by a collaborative group comprising the ISVR, the Faculty of Medicine, and the Medicial Research Council's Institute of Hearing Research. ISVR researchers, Denise Cafarelli Dees (ISVR audiologist 1988-1997) and Roger Thornton (ISVR Visiting Reader 1991-1998), played substantial roles on the six-person steering committee, while Mark Lutman (ISVR Professor, 1995-2012) independently monitored the TEOAE data from the trial to ensure screeners had accurately interpreted it.

Around 25,000 newborn babies were screened using TEOAEs. Outcomes were then compared with those in a control group tested only using HVDT. The results published in 1998 **[1]** strongly suggested a worthwhile benefit of a universal Newborn Hearing Screening Programme (NHSP) based on TEOAEs in terms of age of detection, test accuracy and cost. The results of another large trial (1988-95) to estimate the field sensitivity of TEOAEs screening were published in 1997 **[2]**, receiving the Editor's prize for outstanding research that year. Though beginning the work in Nottingham, Lutman completed the analysis and final dissemination of this research at ISVR.

The results of these trials allowed several key aspects of screening to be assessed such as whether all babies or only those deemed "at risk" should be tested, how different protocols would affect the false-positive and false-negative rates, and what level of training the screeners would require. This analysis was crucial to the successful implementation an effective screening programme.

Lutman (in collaboration with the University of Milan) then played a key role in reviewing all the data, proposing changes in health policy, and successfully promulgating these proposals which

were later taken up both in the UK and overseas. Lutman achieved this as one of the main organizers of the Milan consensus conference on neonatal screening where he chaired the scientific committee, which produced the influential European consensus statement on NHSP in 1998 **[3]**.

In 1998, the UK Department of Health (DoH) commissioned a review of the data **[5]**, which drew heavily on the two trials **[1][2]**. It singled out the trials for their size, quality and design, and recommended that providers build on the experience of the trials **[5, pp.71-72]**. In a systematic review of the evidence by US epidemiologists **[6]**, the Wessex trial **[1]** was awarded the highest quality rating of all trials worldwide, being the only one to compare data against a control group.

3. References to the research

Selected ISVR publications

[1] Wessex Universal Neonatal Hearing Screening Trial Group[*] (1998) Controlled trial of universal neonatal screening for early identification of permanent childhood hearing impairment. Lancet. 352:1957-1964. [*] included D Cafarelli Dees, and ARD Thornton on the six-member writing and steering committees; ME Lutman on monitoring of TEOAE data. http://download.thelancet.com/pdfs/journals/lancet/PIIS0140673698063594.pdf?id=de2e5b4b1d461676:786693a9:13f09e197de:58911370265888397

[2] Lutman ME, Davis AC, Fortnum HM, Wood S, (1997). Field sensitivity of targeted neonatal hearing screening by transient-evoked otoacoustic emissions. Ear and Hearing;18:265–76. (Received the American Audiological Society Editors award for outstanding research in 1997.) http://journals.lww.com/ear-hearing/Abstract/1997/08000/Field_Sensitivity_of_Targeted_Neonatal_Hearing.1.aspx

[3] Grandori F, Lutman ME. (1998 & 1999) European Consensus Statement on Neonatal Hearing Screening (Milan, Italy, 15-16, May 1998). Widely published. Examples of publications carrying this statement are: American Journal of Audiology; 8:19-20; European Journal of Pediatrics. 158, 95-96; International Journal of Pediatric Otorhinolaryngology. 44:309-310; Scandinavian Audiology, 27(4), 259–260. http://informahealthcare.com/doi/pdf/10.1080/010503998420577 & http://link.springer.com/content/pdf/10.1007%2Fs004310051025.pdf & http://aja.asha.org/cgi/reprint/8/1/19

[4] Hunter MF, Kimm L, Cafarelli Dees D, Kennedy CR, Thornton AR. (1994) Feasibility of otoacoustic emission detection followed by ABR as a universal neonatal screening test for hearing impairment. British Journal of Audiology, 28: 47–51. http://www.ncbi.nlm.nih.gov/pubmed/7987271

Influence of ISVR research on setting up NHSP in UK

[5] Davis A, Bamford J, Wilson I, Ramkalawan T, Forshaw M, Wright S. (1997) A critical review of the role of neonatal hearing screening in the detection of congenital hearing impairment. Health Technology Assess 1997;1(10):1–176. http://www.hta.ac.uk/fullmono/mon110.pdf
Cites ISVR research **[1]** pp. 68-86, 111; **[2]** pp. 12, 21, 44, 58, 70-74, 111; **[4]** pp. 68, 69, 110.

Quality rating for ISVR research

[6] Thompson DC, McPhillips H, Davis RL, Lieu TL, Homer CJ, Helfand M (2001). Universal newborn hearing screening: summary of evidence. Journal of the American Medical Association; 286: 2000-10. http://jama.jamanetwork.com/article.aspx?articleid=194319
Cites ISVR research **[1]** pp. 2002, 2003, 2009; **[3]** pp. 2001, 2009.

4. Details of the impact

Newborn hearing screening services have been, and continue to be, revolutionised around the world This has directly led to the early detection of PCHI in thousands of babies, greatly improving their prospects for fulfilling their potential and eventually contributing to the economy.

The ISVR work **[1]**-**[4]** was among several studies internationally that have been influential in the establishment of NHSPs in the UK and overseas. However, the ISVR contributions were distinctive in their size and quality **[5][6][7][15]**. Without these specific contributions, it is likely that universal NHSPs would have been implemented later, in a different form and with a lower degree of confidence in their predicted performance.

Impact on health, language development, and quality of life in the UK

After reviewing the evidence **[5]**, the NHS began implementing a universal NHSP across the UK. The programme currently screens around 13,000 babies in England every week; an average of 265 of these are identified as requiring further audiological testing, with 21 then being identified as having PCHI **[11][10]**. Between 2008 and 2013, more than four million babies in England will have been screened, of whom around 5,000 will have been identified with PCHI. A respected study found that the introduction of NHSP reduced the median age for identifying PCHI from 60 to 10 weeks **[8, Fig. 1]**. The reduction for the upper quartile age was even more dramatic: from 140 to 25 weeks, greatly improving the prospects for these babies. The MRC currently uses the NHSP case study on its website **[7]** as a primary example of the social impact of its funded research, citing trials **[1][2]** as key drivers of the NHSP.

The significant benefits of TEOAE over HVDT – earlier screening, higher correct-detection rates, and lower false-positive rates – have had a huge social and economic benefit. Health providers have been able to intervene at an earlier age, for example by fitting hearing aids or cochlear implants, leading to improvements in language development **[9][16]**. In 2012, the Secretary of State for Health, Andrew Lansley, said:

> "Thanks to the NHS [NHSP], babies born with a hearing impairment can now be identified much earlier. This allows babies with hearing problems to receive the support they need earlier to give them the best possible start in life." **[10]**

Susan Daniels, Chief Executive of the National Deaf Children's Society also praised the NHSP:

> "If hearing loss is identified at birth, deaf children have every chance of achieving as well as other children." **[10]**

Economic Impact

It is likely that the NHSP has also led to economic savings for society between 2008-2013 in areas such as cost of education and lost parental earnings. Quantifying these savings is difficult, but one recent study estimated that the lifetime loss of earnings due to PCHI was between US$300,000 and US$610,000 per person. This figure, together with costs of welfare and education, is expected to reduce with early intervention in cases of PCHI **[11]**.

Impact Internationally

NHSPs continue to be reviewed and developed around the world. In 2009, the World Health Organisation (WHO) **[12]** recommended that a policy of universal (as opposed to targeted) newborn screening be adopted in all countries with available rehabilitation services, citing ISVR work **[3]**. And ISVR research continues to feature in the assessment of NHSPs by official committees in numerous countries tasked with steering health policy. Two examples are in the USA **[13]**, where they reviewed an existing NHSP in 2008, and in Australia **[14]**, where they considered the introduction of universal NHSP for the first time, citing **[1]** for its high quality.

Summary

In the UK, since 2008 the ISVR contributions have had an impact on many thousands of patients whose PCHI has been detected earlier than it would otherwise have been, with profound consequences both for the patient's quality of life, and for the economy of the UK. In addition, the ISVR research continues to influence overseas policy via periodic reviews of existing NHSPs, the establishment of new NHSPs, and in setting the WHO recommendations which guide future health policy overseas.

5. Sources to corroborate the impact

Influence of ISVR research on health policy in UK and of NHSP on society

[7] MRC website. (cites ISVR work **[1][2]**)
http://www.mrc.ac.uk/Achievementsimpact/Storiesofimpact/Hearingscreen/index.htm

Reduction in age of detection of PCHI due to NHSP

[8] Bamford J, Uus K, Davis A (2005) Screening for hearing loss in childhood: issues, evidence and current approaches in the UK. Journal of Medical Screening 12(3):119–124., (see Figure 1; also cites **[5][6]) .**http://msc.sagepub.com/content/12/3/119.full.pdf+html

Evidence that NHSP leads to improved child development

[9] Korver, AMH.; Konings, S; Dekker, FW.; et al. (2010) Newborn Hearing Screening vs Later Hearing Screening and Developmental Outcomes in Children With Permanent Childhood Hearing Impairment, JAMA Journal of the American Medical Association, 304, 1701-1708. (p.1 NHSP improves developmental outcomes)
http://jama.jamanetwork.com/article.aspx?articleid=186749

[10] Press release on benefits of NHSP and numbers screened
Press release in 2012. https://entuk.org/docs/prof/clinical_outcomes/110512_baby_screening

Effect of NHSP on economic costs to society of PCHI

[11]. Hjalte, F; Brannstrom, J; Gerdtham, UG (2012) Societal costs of hearing disorders: A systematic and critical review of literature. Int J Audiol, 51(9), pp. 655-662. (see p.660 for loss of earnings). http://informahealthcare.com/doi/pdf/10.3109/14992027.2012.690077

Influence of ISVR research on health policy around the world

[12] World Health Organization (2009) Newborn and infant hearing screening; Current issues and guiding principles for action. Geneva, Switzerland, 09–10 November 2009. (p.8 cites ISVR work **[3])**
http://www.who.int/blindness/publications/Newborn_and_Infant_Hearing_Screening_Report.pdf

[13] US Preventive Services Task Force (2008) Universal screening for hearing loss in newborns: US preventive services task force recommendation statement. Pediatrics (122) 143-148. (pp. 146, 147 cite ISVR work **[1])**
http://pediatrics.aappublications.org/content/122/1/143.long

[14] Universal Neonatal Hearing Screening: assessment report, Medical Services Advisory Committee, MSAC Reference 17, Commonwealth of Australia, 2008. (pp.57, 60, 65, 89, 91, & 141 cite ISVR work **[1]**). http://trove.nla.gov.au/work/36734798?q&versionId=47699799

Referee: Impact of ISVR research on NHS policy; Benefits of NHS programme

[15] Director NHS Newborn Hearing Screening and Physical Examination Programme

Referee: Benefits of universal screening from user's perspective

[16] Senior Policy, Campaigns and Research Officer, The National Deaf Children's Society.