Evaluation of the newborn hearing screening programme (NHSP) in England

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Report of the Evaluation of the first phase of implementation of the NHSP

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ABSTRACT

The first phase of the Newborn Hearing Screening Programme (NHSP) in England involved twenty-three sites with a total birth-rate of approximately 120,000 births per year. The aim of the evaluation of NHSP first phase was to appraise the benefits, effects, costs and practical implications of the pilot implementation of the national model for newborn hearing screening. The evaluation focused on six domains: (i) audit of screen performance and follow-up; (ii) satisfaction and anxiety; (iii) impact on services; (iv) outcomes; (v) cost and cost effectiveness. The results enable the future direction and fine tuning of newborn hearing screening and associated services within modernised paediatric audiology services. The analyses allow the National Screening Committee to assess the quality of the programme against agreed targets and to ensure that its remaining concerns (e.g. levels of and ways of minimising maternal anxiety, appropriate information materials, authoritative progress to case identification, time to audiological certainty, and involvement of education services in management and support of true cases etc) are appropriately addressed.

ABBREVIATIONS

AABR	automated auditory brainstem response
ABR	auditory brainstem response
AOAE	automated otoacoustic emissions
BATOD	British Association of Teachers of the Deaf
BC	bone-conduction
CHSWG	Children's Hearing Services Working Group
CI	confidence interval
CR	clear response
dB HL	decibel hearing level
DSL	Desired Sensation Level
DfES	Department for Education and Skills
DoH	Department of Health
ESP	Early Support Programme
eSP	electronic Screener Plus
GP	general practitioner
HF	high-frequency
HTA	Health Technology Assessment
HV	health visitor
ICER	incremental cost effectiveness ratio
IDT	infant distraction test
IHR	Institute of Hearing Research
IQR	interquartile range
kHz	kilohertz
LEA	Local Education Authority
LREC	Local Research Ethics Committee
MCHAS	Modernising Children's hearing Aid Services
MRC	Medical Research Council
MREC	Multicentre Research Ethics Committee
NAL	National Acoustic Laboratories of Australia
NC	not completed
NCR	no clear response
NDCS	National Deaf Children's Society
NHS	National Health Service
NICU	neonatal intensive care unit
NSC	National Screening Committee
NUD*IST	Non-numerical unstructured data indexing searching and theorizing software
NW	North-West
OAE	otoacoustic emissions
PCHL	permanent childhood hearing loss
PCT	Primary Care Trust
PPV	positive predictive value
PTM	probe tube microphone
QALY	quality adjusted life year
QCM	quality weighed detected child month
QSR	qualitative solutions and research
RNID	Royal National Institute for the Deaf People
SCBU	special care baby unit
SPSS	Statistical Package for the Social Sciences
SLT	speech and language therapist
ToD	teacher of the deaf
UNHS	universal newborn hearing screening
WBN	well-baby nursery

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EXECUTIVE SUMMARY[1]

Background

The decision to implement a national newborn hearing screening programme and to phase out the existing 8-month infant hearing screen was taken in 2000, following the HTA review (Davis *et al* 1997). Implementation began in 2001 and is expected to be complete for England in 2005/6. A concurrent evaluation of the national Newborn Hearing Screening Programme (NHSP) took place between May 2001 and June 2004. The evaluation was based exclusively on the first phase of implementation, which covered 23 'sites' or service areas in England. This represents an annual birth cohort of about 120,000 births or about a fifth of the national birth cohort. Implementation of NHSP in the first phase sites began in January 2002, with the last of the sites starting screening by September 2002. Eighteen of the first phase sites used the hospital-based screening model (a new cadre of screeners trained to carry out the screen in maternity units before discharge), four the community-based model (existing Health Visitors trained to carry out screening at an early home visit), with one site a hybrid model based on a small cadre of specialist Health Visitors carrying out all screens in a community setting.

The evaluation was directed at screen performance, assessment and follow-up, psychological evaluation of the NHSP (including assessment of maternal anxiety), experience of the parents of true cases identified by the screen, the impact of the screen on related services, and costs and cost-effectiveness of the screen. The following paragraphs summarise the findings from each domain, and are presented as short summary statements for clarity. Further detail can be found by referring to the relevant chapter.

Screen performance in NHSP

1) A user-friendly tailored screening-management system is vital for managing and auditing the screening programme; eSP seems to fulfil that need, while the original systems did not.

2) 99.5% of all target babies were offered a screen; the draft minimum quality standard is 99%.

[1] Based upon Chapter 8 of the Report.

3) 97.5% of all target babies entered the screen; the draft minimum quality standard is 95%.

4) 96.0% of all target babies completed the screen; the draft minimum quality standard is 95%.

5) Refer rate decreased consistently from the beginning of the screen in 2002 to 2.7% averaged across sites by September 2003; the draft minimum standard is 3%.

6) 9.6% (95% CI 5.9-13.3%) of all referred babies had not been followed up by 6 months after referral; there is no direct minimum standard for 'lost-to-follow-up' although the draft minimum standard that 95% of referred babies should start assessment within four weeks of screen applies indirectly.

7) 11.5% (95% CI 8.7-14.3%) of all referred babies were identified with hearing loss.

8) Yield per 1000 babies screened is 1.64 (95% CI 1.27-2.01): 1.00 (95% CI 0.78-1.22) per 1000 screened for bilateral permanent hearing loss—this is similar to published prevalence rates; and 0.64 (95% CI 0.37-0.91) per 1000 screened for unilateral permanent hearing loss.

9) Aggregated screen performance data across all first phase sites were good, and met most of the current NHSP draft minimum standards; however, within these data were individual sites not performing at acceptable levels. Action is being taken by the implementation team; explicit process and procedures need to be in place to manage such under-performing sites.

Follow-up of true cases identified by NHSP

10) Based on data from true cases, median age at first follow up after screen referral was five weeks of age. Some 64% of well babies are likely to have their first audiological follow-up by 4 weeks of age. Ninety-five per cent of cases had had the first follow-up by 11 weeks of age. Reasons for the longer delays for well babies are mainly service-related and suggest the need for improvements in aspects of paediatric audiology services; efforts should be made to prioritise follow-up of screen referrals in order to shorten the waiting period to no more than four weeks, and clear explanations of the reason for the wait should be given; mothers of referred babies should be given an appointment date and time before discharge if at all possible.

11) The median age at identification of permanent bilateral hearing loss was 10 weeks which marks a major improvement compared to 18 months of age before the implementation of newborn hearing screening. Ninety per cent of the true cases identified via the screen were identified before six months of age; the draft minimum standard is 80%. Age of identification was independent of the severity of the hearing loss.

12) Age at follow-up and age of identification were not dependent upon severity of the hearing loss.

13) The median age of children who were fitted with hearing aids was 4 months which is a very considerable improvement compared to around 2 years of age before the implementation of newborn hearing screening. Eighty per cent of well babies were fitted with hearing aids by 6 months of age; including NICU babies, 90% were fitted by about 30 weeks of age (the draft minimum standard is 6 months of age). Babies with moderate hearing loss tended to be fitted later than those with severe or profound loss, often because of parental choice. Efforts should be made to fit hearing aids, where appropriate, within four weeks of identification of hearing loss.

14) The very early fitting of hearing aids requires considerable skill and knowledge, particularly with the advent of DSP (digital signal processing) hearing aids. Systems for ensuring the quality of hearing aid fitting and management in very young infants need to be strengthened.

15) There were significant numbers of babies with unilateral hearing loss identified by the screen. Evidence-based guidelines for management are urgently needed.

16) 54% of all cases with permanent bilateral hearing loss are from an 'at-risk' population. 3/4 of these 'at-risk' babies have spent 48 hours or more in the neonatal intensive care unit. 36% of children identified with permanent bilateral hearing loss have additional conditions and/or disabilities.

17) It is not appropriate to screen babies with unilateral or bilateral meatal atresia; such cases should be automatically referred; this is now in the national protocol.

18) About 10% of the cases with bilateral hearing loss were cases of auditory neuropathy. Research into the causes, management and outcomes of auditory neuropathy is urgently needed.

Psychological evaluation of NHSP

19) Referral for diagnostic tests has a small but significant effect on mothers' emotional well-being in the first three weeks after screening; the effect is below the cut-off for clinical concern. This small but significant emotional distress following recall for diagnostic tests after newborn hearing screening is no longer evident at six months.

20) Ensuring good knowledge of possible reasons for referral seems to be protective against anxiety and thus suggests a potentially effective yet simple intervention to minimize the adverse emotional impact of this screening programme.

21) The results provide evidence to support the hypothesis that mothers of babies receiving a referral for diagnostic tests after screening experience less emotional distress if the screening is conducted in the community compared with the screening conducted in the hospital. This hypothesis awaits testing.

22) Newborn hearing screening does not cause more emotional distress than a test conducted some months later in infancy.

23) As well as its advantages in terms of sensitivity and specificity, newborn hearing screening is associated with higher levels of maternal satisfaction. Such satisfaction may help facilitate attendance for follow-up tests.

24) Hospital-based dedicated screeners expressed more job satisfaction than community-based Health Visitor screeners. Although the two groups differed in overall levels of job satisfaction, their satisfaction was influenced by similar factors. These factors need to be taken into account in continuing the effective implementation of newborn hearing screening. Evaluation of the long term job satisfaction of hospital-based screeners is needed.

The true cases study—the experiences of parents whose children were correctly identified as deaf through the screen

25) For parents, the defining experience of screening is how to interpret and how to respond to the inconclusive message that each stage of the process delivers. For about half of the parents in the sample, the inconclusive message gives little or no concern. This lack of concern is assisted by two main factors: the totally reassuring manner of the screener and the content of the explanation offered. Positive appraisal of screener manner was not just made on grounds of what they said, but also how they seemed as people – their character and their sensitivity.

26) The offering of an explanation why the baby had not passed the screen was important in reducing anxiety. Where explanations were vague parents were more worried. For some parents, an important element in that explanation must be an acknowledgement that deafness might be one of the range of explanations why the baby was not passing. This was of particular importance in situations where there were potentially other signs that the baby may be at higher risk (e.g. NICU history).

27) An explanation that set the screen outcome in a wider context was considered vital i.e. one that showed that few babies that were referred actually had a hearing loss. Where parents were told this, it was very helpful, where parents were not, it added to their growing concerns. There was evidence of the importance of checking that parents really have understood what the screen result implies rather than simply assuming that the reassuring message will of itself be adequate explanation.

28) A waiting time between the end of screening and the first appointment with audiology that was short was helpful for many families. In addition the possibility of receiving the appointment date immediately at the end of screening was especially reassuring. Knowing exactly why they were required to wait (e.g. giving time for fluid to clear from baby's ears) was also helpful. When the appointment followed on quickly it tended to be positively perceived as being part of the same process that was being handled efficiently by professionals who knew what they were doing. This routineness was linked by parents to helping to reduce stress/worry.

29) There were some examples of poor practice, and two cases raise particular concern: (i) the family who during the waiting time felt unsure whether they should communicate with their baby and if so how; (ii) the family who had received no information in their preferred language, an appointment letter in English that they

could not understand and who waited three months for an audiology appointment without being sure if that was a usual period of time to wait or not.

30) Families made good suggestions about how to improve the transition to audiology for follow-up assessment; e.g. by setting aside slots of time on a regular basis for those who had been referred so that there were no unnecessary service-linked barriers to their progression through the system.

31) A minority of families would have appreciated active support during this waiting time.

32) Good explanations at follow-up assessments were a key component of what parents perceived to be good professional communication. In order for parents to positively appraise an explanation, it had to be thorough, using appropriate register or using examples that were connected to a reality with which they were familiar. Parents identified that being made a partner in the process was a key feature of good communication. One way of achieving partnership with parents is by engaging them in the testing procedures. Being approachable was identified as an essential component of professional manner. Those professionals described as unapproachable were generally those seen at the first audiological assessment.

33) The practicalities of the diagnostic process could be challenging for many families. However, having a professional that was accommodating helped to counter this. One way that professionals could be accommodating was by notifying parents of the duration of appointments so that they could prepare themselves and the baby appropriately.

Impact of NHSP on services

34) The advent of NHSP was seen to help improve inter-agency working between health (audiology services) and education (LEA support services for deaf children). Examples of improvements included increased frequency of contact, the use of IT to enable fast referral, the joint development of protocols to redefine roles and responsibilities, the inclusion of education staff at the point of disclosure, the establishment of joint care pathways, and the joint development of web-based resources.

35) Other national initiatives relating to young deaf children—MCHAS (Modernising Children's Hearing Aid Services) and ESP (Early Support Programme)—were noted to be having a significant impact on joint working.

36) Social Services rated their relationship with audiology to be good (65 per cent of services interviewed stated they were extremely satisfied with their links), but usually this is linked to their work with older deaf children, young people or adults, as opposed to deaf children 0-2 years of age. Some Social Services have no links with audiology or education services. Perceived reasons for this include Social Services workloads, lack of resources, the difficultly in establishing a specific contact point or person within Social services, lack of clarity about the role of Social services with young deaf infants and families, and strategic level barriers.

37) All three service groups (audiology, education, social care services) identified the need for appropriate training opportunities and linked this to their ability to provide a high quality service for very early identified deaf children and their families.

38) Out of the three groups of health professionals studied which have an awareness role in the NHSP programme (Health Visitors, midwives and GPs), HVs are the most knowledgeable and GPs are the least knowledgeable about NHSP. Efforts are needed to improve awareness in these groups.

39) Almost all the Health Visitors and midwives who responded to questionnaires expressed some degree of satisfaction with the changes brought upon by NHSP; the views of non-respondents may of course differ.

40) The focus groups with D/deaf professionals indicated that these professionals have had little involvement in NHSP and it has had little impact on their working practices. Consideration needs to be given as to how to change the situation, and thus affirm D/deaf professionals as active and valued members of the early years team.

Costs and cost-effectiveness

41) The NHS costs of NHSP (universal newborn hearing screening) and IDT (the Infant Distraction Test screen at 8 months of age) in those NHSP first phase sites studied (16 sites for NHSP and 10 sites for IDT) ranged from £26,384 to £55,874 (average £34,315) and £10,042 to £48,074 (average £25,170) respectively.

42) NHSP appears to be a cost effective strategy for hearing screening when compared to IDT screening with an average additional health service cost of £12,500 per additional case detected. Including family costs, NHSP is the dominant policy option: cost saving and more effective (higher case detection rate). These findings support the findings of the UK study of Davis *et al* (1997) and recent US cost effectiveness analyses.

43) Based on the data from first phase NHSP sites, modelling indicates the costs and effects (i.e. yield) of community-based and hospital-based newborn hearing screening to be equivalent. However, further data are required to confirm this finding.

Overview issues

44) The evidence from the evaluation points to a highly-competent implementation, delivering in the first phase sites good information for parents (via video and leaflets), well-trained screeners, an effective screen meeting most of the draft minimum quality standards. Within this aggregate picture, some screening teams (which tend to be urban with social and other challenges) have been under-performing; the implementation team is aware of these and has put procedures in place to manage the transition to acceptable screen performance. The general processes guiding such intervention need to be made explicit.

45) The Newborn Hearing Screening Programme in England is regarded as a model of good practice, especially because it has been developed with a top-down public health perspective and on a whole-population basis, because a team has been funded to manage the implementation, because appropriate IT systems to support the screen have been developed, because the implementation covers intervention with health, education and social services as well as the screen itself, and because there has been a separate evaluation exercise.

46) The evaluation of the first phase implementation has demonstrated, broadly speaking, that maternal anxiety is likely to be within acceptable limits, and that maternal satisfaction with the screen is generally high. However, there is evidence that not all parents are receiving or able to access the information materials.

47) There are doubts about the quality of some paediatric audiology services in England, particularly with regard to post-screen assessment and the fitting and management of digital signal processing hearing aids; such services need to be identified, and support and training systems put in place.

48) The funding of the ESP programme by DfES is to be welcomed, and should help to secure appropriate support from education services for families and children identified via the newborn hearing screen. Concerns remain about the impact of this and other initiatives in sites early into NHSP, and about underlying issues of workforce numbers and training.

49) The lack of involvement of social care services has been borne out by the evaluation, and this is being addressed by the NHSP implementation team: a study has been commissioned and draft recommendations made to develop the role of social care services, although resource issues represent a crucial barrier to progress in this area.

50) The eSP screening management system for NHSP has met user expectations and is the first national system to be integrated with the central issuing system for NHS numbers (NN4B); it is important that eSP is fully integrated with future systems and is not undermined by the introduction of the new NHS IT systems.

51) Changes to screen protocol should be based upon robust evidence of gains (cost-effectiveness, increased benefits, reduced harm etc), and should be agreed nationally and implemented across all sites so that IT systems, and training and information to parents can be brought into line with the changes. Such changes should be based on robust evidence—the source of such evidence will be the national implementation itself, obtained through the ongoing quality monitoring and via agreed sub-trials of protocol changes (which should only be undertaken after full implementation has been achieved).

52) On the basis of limited findings on screen performance, maternal anxiety, and cost-effectiveness it could be argued that either model of screening (hospital-based, community-based) can be implemented successfully. However, the evaluation was not designed as a controlled trial and generalisability is uncertain. Furthermore, other considerations (set-up costs, quality assurance/IT issues) would argue against the community-based model, and also against running two models side-by-side.

53) The draft Quality Assurance (QA) specification is central to the future success of NHSP, and requires the appropriate infrastructure and staffing.

54) Research is needed on the outcomes associated with mild hearing loss and babies identified with unilateral hearing loss, and on the appropriate management; this will have implications for the case definitions for NHSP.

55) Surveillance systems need to be implemented in order to remain alert to children with progressive, late onset and acquired hearing loss; guidelines are now available from the implementation team.

56) Work is needed on how best to provide families of children with hearing loss with informed choices.

57) There is a significant shortage of specialised staff to work in audiology, deaf education and social care, and strategies need to be in place to address this; how to provide appropriate training for audiology, education, social, and D/deaf workers active with families of young deaf babies is a related issue.

58) The factors relevant to job satisfaction for screeners need to be taken into account in continuing the effective implementation of newborn hearing screening.

1. INTRODUCTION AND BACKGROUND

1.1 Background

In 1994, the then Minister for Health, John Bowis, announced that he was seeking a review of the screening arrangements leading to the identification of children with permanent childhood hearing loss (PCHL) in the UK, in particular to examine the possible role for newborn hearing screening.

Shortly thereafter, the Health Technology Assessment (HTA) arm of the National Health Service Research and Development effort commissioned a systematic review of screening for PCHL, including some primary research on current practice.

The review was completed and published some two years later (Davis *et al* 1997). Among a number of recommendations for service development and research in the field of paediatric audiology and early intervention for families of children with PCHL, the review recommended the introduction of universal newborn hearing screening, and the phasing out of the existing 8-month infant distraction test (IDT) screen, performed (usually) by Health Visitors (HVs) with children at about 8-months of age.

The case upon which these recommendations were based can be summarised in eight key points:

- Currently outcomes in communication, educational achievements, mental health, and quality of life for children with PCHL tend to be less than optimal.

- About 800 children are born each year in England with a permanent bilateral hearing loss (≥40 dB HL) that could be identified at birth. Current screening services identify only a small proportion of the children by one year of age.

- There is emerging evidence that intervention in the first six months of life improves at least some of the outcomes.

- The evidence suggests that more precise and detailed neural connections depend upon appropriate early stimulation; myelination of auditory pathways by 6 months of age is delayed by almost any chronic insult.

- Earlier identification allows earlier assessment of progress, with earlier management decisions; the starting point for intervention is not, therefore, from a position of developmental deficit.

- Costs are broadly acceptable, and probably less than the 8-month screen.

- Parents have the right to be informed as early as possible about factors likely to affect their child's development.

- Evidence from parents of children with PCHL indicates that they would have welcomed identification as soon as possible after birth.

The review led to a recommendation from the National Screening Committee (NSC), and its Child Health Screening Sub-Group, for a national programme of newborn hearing screening to be introduced across the country. At the same time, however, the NSC expressed concern about the potential maternal anxiety engendered by a newborn hearing screen, particularly for the parents of those babies referred by the screen, and about the ability of services in health (paediatric audiology) and education (LEA Support Services for Hearing Impaired Children) to assess accurately and manage effectively children identified very young, and about the role (or lack of) of Social Services with families of true cases.

The Davis *et al* (1997) recommendations were based largely upon published evidence from hospital-based screening programmes: that is, where each baby is screened before leaving the maternity or birthing hospital. The NSC recommendation was, therefore, for a hospital-based newborn screening programme. At the same time, however, the Committee recognised that although there were few studies on the relative effectiveness of community-based screens, there were some practitioners who argued strongly for a screen based upon community-health systems; specifically, a screen carried out by HVs at home as part of the statutory ten-day visit. In terms of technology and equipment, both systems (hospital-based and community-based) appear to be viable.

The procedure to be followed when the research evidence suggests that a new screening programme should be introduced is usually to complete a pilot implementation, followed by full implementation if the results of the pilot are satisfactory. Pilots are '*a useful mechanism for testing the feasibility, public acceptability and cost-effectiveness of new screening programmes in practice*' (NSC 1998). However, the case for introducing newborn hearing screening, and for phasing out the existing poorly-performing 8-month IDT screen, was so strong that the NSC recommended immediate national implementation (on a phased timescale) in parallel with an evaluation of phase one of the implementation such that some of the details of how the screen should be introduced could be modified, if necessary, during the implementation.

In June 2000 a decision was made by Yvette Cooper, Minister for Public Health, to accept the NSC advice with respect to England, and the Department of Health (DoH) commissioned a team led by Professor Adrian Davis at the MRC Institute of Hearing Research in Nottingham to manage all aspects of the implementation. The remit was encouragingly wide, since it was

recognised that early screening delivers only the potential for significant health, educational, and social gains: for that potential to be realised, paediatric audiology services, education services, and social services each have a crucial role to play. Furthermore, it was known that there was considerable practice variability and many examples of poor service provision in this field (Bamford *et al* 2001). Thus the implementation of the Newborn Hearing Screening Programme (NHSP), which began in 2001/2 and will be completed in 2005/6, assumed a key place in the wider modernisation agenda for paediatric audiology services, and prompted significant activity funded by the DfES to develop training and innovations to support early intervention with children and families (see e.g. www.espp.org.uk).

At the same time, the DoH tendered for an evaluation of the first phase of the NHSP. The brief was to 'evaluate the benefits, effects, costs and practical implications of the implementation of a national model of newborn hearing screening recommended by the NSC in order to identify best practice for:

- the implementation of hearing screening of newborn babies before they are discharged from hospital or as soon as possible thereafter;
- the implications for the phasing out the IDT screen;
- the development of paediatric audiology services to meet the needs of very young babies;
- the promotion of the role of education and social services in the delivery of services for deaf and hard of hearing babies.'

The NHSP implementation identifies two screen protocols, one for well babies and one for babies who have been in a neonatal intensive care unit (NICU). Both protocols involve two test procedures within the one screen. In the former, representing the great majority of screenings, babies are tested first using transient evoked Automated OtoAcoustic Emissions (AOAEs, Kemp 1978) and, if the results are not clear, they undergo Automated Auditory Brainstem Response (AABR) testing. Thus for well babies, the AABR test is contingent upon the lack of clear AOAE responses. For NICU babies both OAE and AABR are used, with screen refer if a clear response is missing on either. The rationale for this difference is that NICU babies are known to be at risk of auditory neuropathy (Rance *et al* 1999), which would be missed by AOAE testing alone. For both of the tests used in the screen, equipment is available[2] which gives a pass-refer decision that does not require interpretation by an audiologist.

The detailed decision routes for the protocols (e.g. whether to pass a baby with a clear response on one ear only, or what combination of AOAE and AABR testing to use) involves a trade-off between sensitivity, specificity, and costs. Sufficient data are available in the

[2] See appendix for a listing of the screen testing equipment purchased for the NHSP implementation.

literature to allow justified decisions on these details, and the implementation team settled on the national protocols (see appendix for details).

In view of the strong arguments put forward by some for a community-based model of the newborn screen, but in the light of the lack of evidence on the performance of such a model, the NSC and the DoH agreed to implement a community model of the well-baby protocol at a number of sites. The protocol is modified only in terms of timing—see appendix.

Thus the first phase of the NHSP implementation involved 23 sites[3] of which four adopted the community-based screen, 18 the hospital-based model and one site continued to use a hybrid model in which a small cadre of Health Visitors were dedicated to screening and carried out all the screens in a community setting.

It is important to note that even in the hospital-based screen 'community-based screening services' may be involved in order to increase coverage of those missed in the maternity hospitals/birthing units; and of course babies from community sites who have been in NICU are nevertheless screened in hospital using the NICU baby protocol. Note also that the term 'site' should not be taken necessarily to imply a single birthing unit; a 'site' is roughly equivalent to an old 'District Health Authority'. Total annual birth rate in the hospital sites was 102,569, and 19,246 for the community sites in year 2003. Selection of sites was done against an invitation from DoH to bid to be in the first phase; selection aimed to achieve a spread of geographic, demographic, social, urban and rural, and size features, and was not restricted to those with the best health and education services for deaf children.

The evaluation was designed to cover five domains:

- Screen and follow-up: audits of performance
- Parental satisfaction and anxiety
- Impact of the screen on services
- Early outcomes, or surrogates of
- Health economics and cost effectiveness of the screen

Within each evaluation domain a number of studies were designed to answer specific comparisons or performance questions. The following tables and notes give summary details of these. Ethical approval for all aspects of the evaluation was sought and obtained via the North West Multicentre Research Ethics Committee (MREC), and from the Local Research Ethics Committees (LRECs) covering all sites. The evaluation was planned to run from September 2001 to February 2003, but delays in the implementation of NHSP (largely due to IT system difficulties) resulted in aspects of the evaluation being extended to the end of June

[3] See appendix for the list of sites and annual birth rates.

2004. The matrix that follow summarises the studies that were undertaken in each domain; it is provided as background information only. The tables that follow were prepared for the Ethics Committees in order to summarise the studies that were undertaken in each domain; they are provided here as a convenient summary of the work undertaken. More detail will be given in the relevant chapter of this Report: Screen performance in NHSP (Chapter 2), Follow-up of true cases identified by NHSP (Chapter 3), Psychological evaluation of NHSP (Chapter 4), True cases study—the experience of parents whose children have been correctly identified as deaf through NHSP (Chapter 5), Impact of NHSP on services (Chapter 6), Cost and cost effectiveness (Chapter 7), and Summary and recommendations (Chapter 8).

1.2 Domain One: Screen performance and follow-up of true cases

Most of the data needed for the audit of the performance of the NHSP will be obtained from the Screening Management System via the amalgamated database based in IHR. However, some is collected by hand.

Basic demographic data (e.g. number of residential and non-residential births in the Health Authority, homebirths etc) and preliminary data on 'true cases' and unilateral hearing losses (e.g. risk indicators; estimated degree of hearing loss; date of confirmation of true case etc) are collected via proformas to Team Leaders or nominated representatives.

Using similar proformas, IDT screen performance and follow-up data are also collected. The data are collected retrospectively on the cohort of children born between 1st May 2000 and 30th April 2001. The screen performance data is in some areas available from the Child Health Database (e.g. number of infants due for IDT, number starting, completing, referred by screen). We also ask for basic data of true cases identified from the IDT screen (e.g. risk indicators; estimated degree of hearing loss; date of confirmation of true case etc).

1.2.1 Performance of newborn hearing screening

Aim	To collect screening data on each individual newborn
Procedure	Data sent from the sites via IHR to the Evaluation Team
Instrument	Screening Management System
Timelines	On weekly basis. Data will be collected till the end of June 2004.
Comments	Data from HiTrack and SIMs is amalgamated in IHR and sent to the Evaluation Team in the amalgamated form. Data Protection Act is strictly followed.

Aim	To collect baseline demographic data
Procedure	Team Leader asked to fill in the proforma (Proforma 1.1)

Instrument	Proforma 1.1
Timelines	Each month starting from the beginning of the NHSP in the area. Data will be collected till the end of June 2004.
Comments	The proforma sent to the Team Leader in electronic form and for electronic return to the Evaluation Team

1.2.2 Newborn hearing screening true case data

Aim	To collect preliminary data on each 'true case' identified via the newborn Hearing Screening Programme
Procedure	The Head of Paediatric Audiology Service (or a person nominated by them) is asked to fill in the proforma for every 'true case' (Proforma 1.2)
Instrument	Proforma 1.2
Timelines	'True case packages' sent to all sites (using a staggered approach) from March to June 2002. Expected to be completed every time a 'true case' is confirmed and to be sent back to the Evaluation Team URGENTLY as this form will trigger further action (see Domain 2 Experiences of parents of true cases). Data will be collected till the end of June 2004.
Comments	By 'true case' we mean a child having a permanent bilateral hearing loss with hearing threshold ≥ 40 dB HL based on the estimated average in the better hearing ear at 0.5, 1, 2 and 4 kHz and who has been identified via the Newborn Hearing Screening Programme

Aim	To collect preliminary data on every child identified with unilateral hearing loss who has been identified via the Newborn Hearing Screening Programme
Procedure	The Head of Paediatric Audiology Service (or a person nominated by him/her) will be asked to fill in the proforma for every unilateral hearing loss (Proforma 1.2A)
Instrument	Proforma 1.2A
Timelines	Forms sent to all sites (using a staggered approach) from April to July 2002; Expected to be completed every time a case of unilateral hearing loss is confirmed. Data will be collected till the end of June 2004.
Comments	By permanent unilateral hearing loss we mean a hearing loss with hearing threshold of ≥ 40 dB HL in one ear and <40 dB HL in the other ear (based on the average threshold at 0.5, 1, 2 and 4 kHz) (Proforma 1.2A) and who has been identified via the Newborn Hearing Screening Programme

1.2.3 Performance of IDT screen

Aim	To collect basic screening data (e.g. coverage, referral rate etc) on the IDT screen for infants born between 1st May 2000 and 30th April 2001
Procedure	A person nominated by the team leader is asked to retrospectively fill in the proforma (Proforma 1.3) on IDT screen for infants born between 1st May 2000 and 30th April 2001
Instrument	Proforma 1.3
Timelines	Sent out on 17th April 2002 and expected back by 1st November 2002

Comments	Sent only areas with full IDT

1.2.4 IDT true case data

Aim	To get preliminary data on each 'true case' i.e. permanent bilateral hearing loss of ≥ 40 dB HL based on the estimated average threshold in the better hearing ear at 0.5, 1, 2 and 4 kHz who was born between 1st May 2000 and 30th April 2001 and who was identified via IDT screen; to assess the NHS costs of audiological follow-up of true cases
Procedure	A person nominated by the team leader retrospectively fills in the proforma (Proforma 1.4) on IDT screen for infants born between 1st May 2000 and 30th April 2001
Sample size	Every known 'true case' i.e. permanent bilateral hearing loss with hearing threshold ≥ 40 dB HL based on the estimated average in the better hearing ear at 0.5, 1, 2 and 4 kHz
Instrument	Proforma 1.4
Timelines	Sent out on 17th April 2002 and expected back by 1st November 2002
Comments	Only phase one areas with full IDT

1.3 Domain Two: Anxiety and satisfaction

1.3.1 Psychological evaluation of NHSP

Questionnaires will be used to assess satisfaction and anxiety associated with the screen. About 200 mothers per area will be sampled according to the stage at which their baby passed or was referred by the screen. Samples of mothers from the IDT have also been approached. Mothers are asked to fill in questionnaires 1 week and 6 months following the screen.

Additionally, screeners will be asked to fill in a short questionnaire assessing their satisfaction with NHSP.

In depth interviews are carried out with a sample of mothers and families of babies identified via the NHSP with permanent bilateral hearing loss. The timing of these interviews is handled carefully by the team, in consultation with local service providers. The approach is made through the Team Leader.

1.3.1.1 Comparison of maternal satisfaction between hospital-based NHSP, community-based NHSP and IDT screen

Aim	To explore the impact of the type of hearing screening upon maternal anxiety and satisfaction, comparing hospital-based NHSP, community-based NHSP and IDT screen; to examine predictors of maternal anxiety and satisfaction.
Procedure	A sample of mothers are sent a questionnaire (Questionnaire 2.1) 3 weeks after the screen and an identical questionnaire 6 months after the screen

Sample size	N=1692 Hospital-based NHSP: Group 1 (passed AOAEs): 182; Group 2 (passed AABR): 182; Group 3 (pass AABR only in one ear): 182; Group 4 (fail AABR bilaterally): 200 Community-based NHSP: 182 (pass AOAE); 182 (pass AABR) and 200 (referred) IDT: 182 (pass IDT) and 200 (referred)
Instrument	Questionnaire 2.1
Timelines	IDT: July 2002 to July 2003 Community-based and hospital-based NHSP: from March 2003 till October 2004
Comments	Mothers who do not read English and Mothers of NICU babies are not included. Information from the Child Health person re: deceased babies Chesterfield; Whipps Cross and Camden & Islington are not included.

1.3.1.2 Satisfaction of screeners

Aim	To describe screeners' satisfaction with the NHSP
Procedure	The screeners are asked to fill in a questionnaire (Questionnaire 2.5)
Sample size	N=250 150 (hospital-based) and 100 (community-based) screeners
Instrument	Questionnaire 2.5
Timelines	1 year from the NHSP start date (January -March 2003)

1.3.2 Experiences of parents of true cases

Aim	To consider the impact of screening process, and of very early identification, from the perspective of parents of true cases
Procedure	In-depth interviews with parents of true cases identified via NHSP are carried out by a member of the Evaluation Team. By 'true case' we mean a child having a permanent bilateral hearing loss with hearing threshold ≥ 40 dB HL based on the estimated average in the better hearing ear at 0.5, 1, 2 and 4 kHz
Sample size	N=40 (positive sampling among parents who have volunteered to participate)
Instrument	Interview
Timelines	Interviews done when appropriate (from December 2002 to March 2004)
Comments	A member of the Evaluation Team approaches the Team Leader (triggered by Proforma 1.2) and the parents of true cases are then contacted via Team Leader. Interviews carried out a venue chosen by parent.

1.4 Domain Three: Impact on Services

Newborn Hearing Screening will have an impact on various related services. In order to explore this, the pre-NHSP state of the services has to be described – questionnaires were sent to the Paediatric Audiology and Education Services followed by short phone interviews.

Questionnaires were also sent to the appropriate persons from the Social Services (following approval from the ADSS).

Approximately 1 year after the start of the NHSP, the above-mentioned Services are asked to fill in a repeated questionnaire followed by another short phone interview. In addition, a sample of HVs, GPs and midwives are asked to fill in a questionnaire after implementation to describe the impact of Newborn Hearing Screening on their services.

1.4.1 Prescreen State of Paediatric Audiology Services

Aim	To describe the pre-NHSP state and planning of the Paediatric Audiology services
Procedure	Heads of Paediatric Audiology were asked to fill in a questionnaire and contacted for a phone interview
Instrument	Questionnaire 3.1

1.4.2 Prescreen State of Education Services

Aim	To describe the pre-NHSP state and planning of the Education Services.
Procedure	Heads of Education Services were asked to fill in a questionnaire and contacted for a phone interview
Instrument	Questionnaire 3.2

1.4.3 Prescreen State of Social Services

Aim	To describe the pre-NHSP state and planning of the Social Services.
Procedure	Nominated persons from the Social Services were asked to fill in a questionnaire and contacted for a phone interview
Instrument	Questionnaire 3.3

1.4.4 Impact of NHSP on Paediatric Audiology Services

Aim	To explore the Impact of NHSP on Paediatric Audiology services
Procedure	Paediatric Audiology is asked to fill in the questionnaires and that will be followed up by a phone interview
Instrument	Questionnaire 3.4
Timelines	1 year from the start of NHSP using staggered approach. Data will be collected till the end of December 2003.

1.4.5 Impact of NHSP on Education Services

Aim	To explore the Impact of NHSP on Education Services
Procedure	Education is asked to fill in the questionnaires and that will be followed up by a phone interview
Instrument	Phone interview
Timelines	1 year from the start of NHSP using staggered approach. Data will be collected till the end of December 2003.

1.4.6 Impact of NHSP on Social Services

Aim	To explore the Impact of NHSP on Social Services
Procedure	Social Services are asked to fill in the questionnaires and that will be followed up by a phone interview
Instrument	Phone interview
Timelines	1 year from the start of NHSP using staggered approach. Data will be collected till the end of December 2003.

1.4.7 Impact of NHSP on HVs

Aim	To explore the Impact of NHSP on HVs.
Procedure	A sample of HVs (400) are asked to fill in the questionnaires.
Instrument	Questionnaire 3.8
Timelines	1 year from the start of NHSP using staggered approach. Data will be collected till the end of December 2003.
Comments	Is sent to all apart from Whipps Cross, Chesterfield and C&I

1.4.8 Impact of NHSP on GPs

Aim	To explore the Impact of NHSP on GPs.
Procedure	A sample of GPs (150) are asked to fill in the questionnaires.
Instrument	Questionnaire 3.9
Timelines	1 year from the start of NHSP using staggered approach. Data will be collected till the end of December 2003.
Comments	Is sent to all apart from Whipps Cross, Chesterfield and C&I

1.4.9 Impact of NHSP on Midwives

Aim	To explore the Impact of NHSP on Midwives.
Procedure	A sample of Midwives (150) are asked to fill in the questionnaires.
Instrument	Questionnaire 3.10
Timelines	1 year from the start of NHSP using staggered approach. Data will be collected till the end of December 2003.
Comments	Is sent to all apart from Whipps Cross, Chesterfield, C&I, Shropshire, Wiltshire and East Sussex

1.5 Domain Four: Outcomes

The introduction of newborn hearing screening for a large population provides an important opportunity to monitor communicative, educational, social, family, emotional and other outcomes. The current evaluation is limited in time and resources; so further funding is being sought for a long-term prospective study. Meanwhile, the current Evaluation will collect data on the most obvious outcome-mediating variables for all true cases as an interim step.

Aim	To collect the outcome mediating variables for future analysis of outcomes in true cases identified via NHSP
Procedure	Persons nominated by the Team Leader from the Audiology Services are asked to fill in a detailed proforma (Proforma 4.1) on every 'true case'
Instrument	Proforma 4.1
Timelines	When appropriate information is available (when the child with a permanent bilateral hearing loss has been fitted with hearing aids). Data will be collected till the end of June 2004.
Comments	By 'true case' we mean a child having a permanent bilateral hearing loss with hearing threshold ≥ 40 dB HL based on the estimated average in the better hearing ear at 0.5, 1, 2 and 4 kHz.

1.6 Domain Five: Cost and cost effectiveness

We are assessing the relative cost effectiveness of both NHSP and IDT screens. We are looking at the NHS cost, training cost and societal cost associated with the screen and audiological follow-up. Data is collected through proformas filled in by Team Leaders (or a persons nominated by Team Leaders) and questionnaires/activity sheets filled out by a sample of screening staff and parents. IDT data is collected on children born between 1st May 2000 and 30th April 2001.

1.6.1 NHS costs of associated with Newborn Hearing Screening

1.6.1.1 Screening costs associated with NHSP

Aim	To assess NHS costs associated with Newborn Hearing Screening Programme
Procedure	Team Leader (or a person nominated by the Team Leader) is asked to fill in a proforma (Proforma 5.1)
Instrument	Proforma 5.1
Timelines	To be filled in and returned 12 months from the start of the NHSP.
Comments	Modified versions of the proforma were sent to all 4 community sites and all the hospital-based sites that had started NHSP before 1st May 2002 for comparison between costs of hospital-based and community-based screening. Modelling of this comparison has been prioritized by the Steering Group, and is being carried out in collaboration with health Economists in Manchester and the German UNHS Modelling Group, based in Munich. Report to the Steering Group in September 2003

1.6.1.2 Training costs of Newborn Hearing Screening

Aim	To assess the costs associated with training for the Newborn Hearing Screening Programme.
Procedure	The providers of the training; trainees and organizers of the training event are asked to fill in the proformas after every training event
Instrument	Proforma 5.6; Proforma 5.7 and Proforma 5.8

1.6.1.3 Family costs of Newborn Hearing Screening

Aim	To assess the family costs associated with attending the screen.
Procedure	Sampling will be done by the Evaluation Team.
Sample size	N=950 (150 from the community-based NHSP sites and 800 from the hospital-based NHSP sites)
Instrument	Questionnaire 5.9
Timelines	Data collection will be completed by the end of December 2003
Comments	We only sample among parents who have had to travel (whose babies have not completed or have missed the hospital-based NHSP or screening was done outside their home in the community-based NHSP). Families involved in 'Maternal Anxiety' study will be excluded.

1.6.1.4 NHS Costs of Follow-Up of referrals from the Newborn Hearing Screening

Aim	To assess the NHS costs of audiological follow-up of babies referred by the NHSP
Procedure	Those carrying out audiological assessments are asked to prospectively fill in a Proforma (Proforma 5.11)
Sample size	10 referrals per area starting from month 9 after starting NHSP

Instrument	Proforma 5.11
Timelines	Forms sent out October 2002 – April 2003. Data will be collected till the end of December 2003.
Comments	Overseen by nominated person from the Audiology Service

1.6.1.5 NHS Costs of Follow-Up of TRUE CASES referred from Newborn Hearing Screening

Aim	To assess the NHS costs of audiological follow-up of babies referred by the NHSP and identified as true cases
Procedure	A nominated person from the Audiology Service is asked to retrospectively fill in a Proforma (Proforma 5.11A) on the procedures done from the referral to the confirmation of true case and from then on prospectively from the confirmation of true case to hearing aid fitting.
Sample size	Every true case
Instrument	Proforma 5.11A
Timelines	When appropriate. Data will be collected till the end of December 2003.

1.6.1.6 Family costs associated with audiological follow-up of referrals from Newborn Hearing Screening

Aim	To assess the family costs associated with attending the audiological follow-up clinics
Procedure	A nominated person from the Audiology Service who fills in a Proforma (Proforma 5.11 gives the family the questionnaire (Questionnaire5.12)
Sample size	10 referrals starting from month 9
Instrument	Questionnaire 5.12

1.6.2 Costs associated with IDT screening

1.6.2.1 Screening costs associated with IDT screening

Aim	To assess NHS costs associated with IDT screen
Procedure	A person nominated by the Team Leader (we suggest HV manager) is asked to fill in proforma (Proforma 5.13) about the IDT screen for the cohort born between 1st May 2000 and 30th April 2001
Instrument	Proforma 5.13
Comments	Study undertaken only in areas with full IDT

1.6.2.2 Family costs associated with IDT screening

Aim	To assess the family costs associated with IDT screen
Procedure	In each HA 10 families are given a questionnaire.
Sample size	N=150 (10 questionnaires distributed by one HV; if an area is mixed then 5 by an urban HV and 5 by a rural HV)
Instrument	Questionnaire 5.14
Timelines	When appropriate
Comments	Only families who have to travel to get their infant screened are included.

1.6.2.3 NHS Costs of follow-up of referrals from IDT screen

Aim	To assess the NHS costs of audiological follow-up of babies referred by the IDT
Procedure	A person nominated by the Team Leader (we suggest either Community Paediatrician or somebody from Audiology Service) is asked to fill in proforma (Proforma 5.15) prospectively
Sample size	Consecutive 25 referrals
Instrument	Proforma 5.15
Comments	This study looks at the NHS costs associated with audiological follow-up of babies referred by the IDT screen prospectively

2. SCREEN PERFORMANCE IN NHSP

2.1 Background

Coverage, referral rate, and yield are three of the key performance markers for screening programmes. Davis *et al* (1997) reported in their critical review of the evidence that high coverage of over 90 per cent was possible with hospital-based newborn hearing screening programmes. Overall referral rates for those services early in the field were about 5-8%. The referral rates have been shown gradually but significantly to reduce as advances in technology, use of two-stage screening (i.e. incorporation of two tests) and training methodology have evolved (Maxon *et al* 1995, Mehl *et al* 1998). The yield of the early studies of newborn hearing screening in the UK has in general been encouragingly high (e.g. Watkin 1996, Kennedy 1999).

2.2 Aims

The extent to which the research results on screen performance could be achieved in a national programme is addressed by this part of the evaluation of the first phase of the NHSP in England.

2.3 Methods

2.3.1 Sources of data

Screen performance, as with other aspects of the NHSP evaluation, was based on data from the 23 first phase NHSP sites. Four of these sites were permitted to implement a 'community-based' screen, while 18 sites used the 'hospital-based' model. In the community-based model, the screen is administered by Health Visitors at home or community clinic as part of their routine 10-day visit, and alongside their other duties. In the hospital-based model screening is performed by a new cadre of specially-trained screeners whose job is solely to carry out the screening (with associated duties); if uncompleted before the baby and mother are discharged home, the screen may have to be completed at outpatients or in the community. One site continued to use a hybrid model in which a small cadre of Health Visitors were dedicated to screening and carried out all the screens in a community setting.

Three sources of data were used: (i) amalgamated data from Hi*Track and OZ SIMS screening management systems; (ii) hand-collected data from the sites; (iii) data from the eSP screening management system introduced during the first phase of screening implementation.

Detailed data on coverage and referral rates were provided in part by the screening management systems Hi*Track and OZ SIMS. An early plan was to report screen performance data with regard to the babies born from the 1st January 2003 to 31st December 2003 in all twenty-three first phase NHSP sites via the amalgamated data from Hi*Track and OZ SIMS. However, difficulties with data entry at screening sites, difficulties with the amalgamation of data at a national level from two different systems, and loss of data (due to local IT problems) undermined the reliability and comprehensiveness of this source of data. This data uncertainty should be borne in mind when examining results from this source.

Thus, further data on coverage, referral rates and cases identified with permanent bilateral and unilateral hearing loss were collected by hand from site managers using Proforma 1.5 and its modifications (see appendix) for July 2002, November 2002, March 2003 and September 2003.

And finally, much higher quality more comprehensive data from the eSP screening management system introduced to Phase 1 sites from April 2003 (following the difficulties with the Hi*Track and OZ SIMS systems) were used as a further source of coverage and referral rate data. The data from this source reported here refer to babies whose records were created during the period from 1st November 2003 to 29th February 2004.

2.3.2 Definitions of the performance measures used

Coverage is the proportion of the target population who undergo the screen. The target population was defined by the Primary Care Trust (PCT) residency, which in turn was derived from the address of the baby's current general practitioner (GP)[4]. We will define the term at three different levels of coverage:

(i) the proportion of target babies whose parents are offered a screen *('offered coverage')*;

(ii) the proportion of target babies who entered the screen (i.e. at least one test attempt is carried out) *('entered screen coverage')*;

(ii) the proportion of target babies who completed the screen *('completed screen coverage').*

Referral rate is the number of babies referred by the screen expressed as a proportion of the number of babies completing the screen; it is applied to those showing no clear response on AABR on one ear (unilateral screen referrals) and to those showing no clear responses on both ears (bilateral screen referrals).'

[4] The resident population was used as base for performance measurement even though teams had to screen out of area babies. The issue of the efficiency with which teams screen out of area babies, and the efficiency with which they were then followed up if they were referred by the screen was not addressed by the present evaluation. This is an important issue for future evaluation.

Yield is defined here as the number of babies with a follow-up outcome that meets the definition of the target case, per 1000 babies screened. The original target case for NHSP was permanent hearing loss of 40dB HL or greater (averaged across 0.5, 1, 2 and 4 kHz detection thresholds) in the better hearing ear. However, early in the programme implementation it became clear that unilateral permanent cases were being identified and could not be ignored (despite a lack of evidence on appropriate management and cost effectiveness of intervention), and therefore permanent unilateral cases with a hearing threshold of 40dB HL or greater in one ear and <40 dB HL in the other ear are also regarded as target cases.

Other performance measures also reported here are:

AOAE fail rate: the proportion of babies who entered the screen and showed no clear response on one or both ears on AOAE tests (the first of the tests in the screen)

'Lost-to-follow-up' rate: the proportion of screen-referred babies who are not followed up for one of several possible reasons

Positive predictive value of the screen is the proportion of screen referrals that are identified by follow-up assessment with the condition.

2.3.3 Quality standards

Screen performance outcomes were assessed against the initial NHSP quality standards set for implementation. The opportunity exists, of course, to modify the standards on the basis of experience with the implementation or data from the evaluation (see appendix for listing of quality standards).

2.4 Results

2.4.1 Coverage

Table 2.1 shows 'completed screen coverage' figures (proportion of target babies who completed the screen) for all babies by NHSP site, using data derived from the Hi*Track and Oz SIMS screening management systems.

Figure 2.1 shows the number of target babies entered into the screening management systems expressed as a percentage of the number of target babies (obtained from Child Health data systems). There are clear weaknesses in the data, suggesting that this particular source for data audit is unreliable. Apart from Site 23 where no data were entered to the management system, the percentage of target babies entered ranges from 24% (Site 19) to 103% (Sites 1 and 14). Furthermore, the particularly low coverage rates at sites 6, 19 and 22 reflect the extra difficulties the community-based screening sites faced in using the Hi*Track and Oz SIMS screening management systems.

Site	Total number of babies in the database	Babies completing the screen	
		Number	%
1	10404	8660	83.2%
2	2227	2183	98.0%
3	5106	4925	96.5%
4	3439	3238	94.2%
5	4542	4449	98.0%
6**	2805	982	35.0%
7	2954	2931	99.2%
8	2230	1758	78.8%
9*	2702	2648	98.0%
10	8773	7529	85.8%
11	1925	1877	97.5%
12	2451	2406	98.2%
13	5031	4847	96.3%
14	3005	2947	98.1%
15	7636	7096	92.9%
16	6206	5480	88.3%
17	2572	2440	94.9%
18	5444	5264	96.7%
19*	1236	740	59.9%
20	6604	6416	97.2%
21	2956	2658	89.9%
22*	3000	1644	54.8%
23*	No data	No data	No data
Total	**93248**	**83118**	**89.1%**

Table 2.1. Percentage 'completed coverage' for all babies (WBN and SCBU) by NHSP 1st phase site in 2003 (based on data from Hi*Track and OZ SIMS systems). Sites marked with (*) are community sites and (**) hybrid model

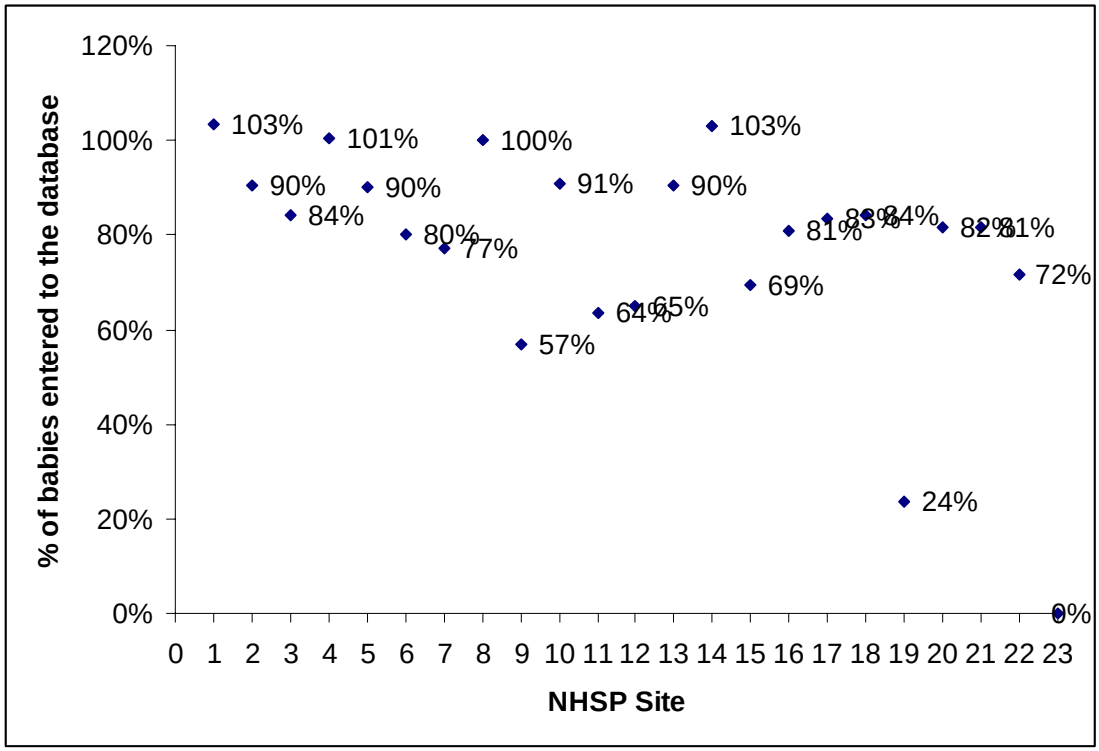


Figure 2.1. Percentage of babies born in 2003 and eligible for screening who entered to the Hi*Track and OZ SIMS systems.

The coverage data in table 2.2 refers to 'entered screen coverage' (the proportion of target babies who entered the screen) and is derived from hand-collected data obtained direct from screen managers for the four specified months.

The data collected from the Team Leaders and Local Coordinators in the main show achieved coverage rates to be within the required standard. By the last time period, when all sites had been running the programme for at least nine months, only Sites 8 and 10 had coverage rates below 90%. These sites have particular local issues (e.g. early discharge, high proportion of non-residential births, large ethnic minority population) which they are addressing with the help of the implementation team. Using the data in table 2.2 there is a significant improvement in coverage across all sites from July 2002 to September 2003 (p=0.034).

NHSP Site	July 2002	November 2002	March 2003	September 2003
1	60%	100%	97%	97%
2	100%	97%	99%	100%
3	90%	91%	96%	98%
4	91%	100%	99%	100%
5	97%	98%	98%	100%
6**	97%	No data	No data	No data
7	100%	100%	100%	100%
8	90%	No data	69%	86%
9*	100%	99%	100%	100%
10	47%	88%	89%	84%
11	100%	98%	98%	100%
12	80%	96%	98%	100%
13	100%	100%	100%	100%
14	96%	100%	100%	100%
15	86%	96%	93%	97%
16	97%	99%	100%	99%
17	No data	No data	No data	No data
18	93%	96%	99%	97%
19*	99%	99%	100%	100%
20	89%	100%	89%	100%
21	95%	100%	93%	100%
22*	100%	99%	98%	98%
23*	NA[5]	98%	95%	99%
Total	**91%**	**98%**	**96%**	**98%**

Table 2.2. Coverage figures for all babies (WBN and SCBU) by NHSP 1st phase site (based on hand-collected data from the Team Leaders/Local Coordinators).

Tables 2.3-2.6 present data derived from the eSP screening management system for the months from November 2003 to February 2004. These data include coverage data for all three definitions: offered coverage, entered screen coverage, and completed screen coverage.

There are no eSP data for Sites 6, 14, 19, 22 and 23. This is due to eSP not having been in use in these sites. These sites were using their respective local data management system and, with an exception of Site 6, had in general very good overview of the state of their screen performance.

[5] Site 23 had not started NHSP by July 2002.

Site	N	Offered	%	Entered	%	Completed	%
1	716	709	99.0	702	98.0	691	96.5
2	195	195	100.0	189	96.9	187	95.9
3	404	404	100.0	394	97.5	392	97.0
4	252	251	99.6	247	98.0	246	97.6
5	363	363	100.0	358	98.6	351	96.7
6**	No data						
7	198	198	100.0	197	99.5	197	99.5
8	174	157	90.2	149	85.6	140	80.5
9*	229	229	100.0	228	99.6	228	99.6
10	443	442	99.8	404	91.2	404	91.2
11	185	185	100.0	185	100.0	183	98.9
12	234	234	100.0	223	95.3	218	93.2
13	356	355	99.7	352	98.9	352	98.9
14	No data						
15	531	531	100.0	513	96.6	510	96.0
16	490	490	100.0	488	99.6	486	99.2
17	258	258	100.0	257	99.6	223	86.4
18	387	387	100.0	377	97.4	371	95.9
19*	No data						
20	658	658	100.0	650	98.8	640	97.3
21	261	260	99.6	256	98.1	254	97.3
22*	No data						
23*	No data						
	6348	**6320**	**99.6**	**6182**	**97.4**	**6086**	**95.9**

Table 2.3. Coverage figures for all babies (WBN and SCBU) by NHSP 1st phase site (based on eSP) for November 2003.

Site	N	Offered	%	Entered	%	Completed	%
1	829	792	95.5	776	93.6	768	92.6
2	178	178	100.0	173	97.2	172	96.6
3	389	389	100.0	385	99.0	385	99.0
4	249	249	100.0	243	97.6	243	97.6
5	378	378	100.0	373	98.7	370	97.9
6**	No data						
7	176	176	100.0	174	98.9	174	98.9
8	119	109	91.6	101	84.9	97	81.5
9*	187	187	100.0	185	98.9	185	98.9
10	452	448	99.1	413	91.4	406	89.8
11	157	157	100.0	151	96.2	147	93.6
12	243	243	100.0	234	96.3	233	95.9
13	355	355	100.0	354	99.7	354	99.7
14	No data						
15	576	575	99.8	565	98.1	560	97.2
16	491	487	99.2	479	97.6	480	97.8
17	285	283	99.3	282	98.9	244	85.6
18	375	375	100.0	363	96.8	353	94.1
19*	No data						
20	1035	1035	100.0	1016	98.2	1013	97.9
21	229	229	100.0	222	96.9	217	94.8
22*	No data						
23*	No data						
	6738	**6680**	**99.1**	**6520**	**96.8**	**6432**	**95.5**

Table 2.4. Coverage figures for all babies (WBN and SCBU) by NHSP 1st phase site (based on eSP) for December 2003.

Site	N	Offered	%	Entered	%	Completed	%
1	815	805	98.8	798	97.9	784	96.2
2	232	232	100.0	230	99.1	229	98.7
3	417	417	100.0	416	99.8	414	99.3
4	257	257	100.0	254	98.8	252	98.1
5	424	424	100.0	416	98.1	408	96.2
6**	No data						
7	187	187	100.0	187	100.0	184	98.4
8	211	207	98.1	186	88.2	179	84.8
9*	349	349	100.0	349	100.0	342	98.0
10	441	436	98.9	404	91.6	402	91.2
11	196	195	99.5	192	98.0	191	97.4
12	261	261	100.0	252	96.6	253	96.9
13	369	369	100.0	368	99.7	368	99.7
14	No data						
15	587	586	99.8	570	97.1	568	96.8
16	582	581	99.8	575	98.8	570	97.9
17	316	315	99.7	312	98.7	271	85.8
18	492	492	100.0	486	98.8	481	97.8
19*	No data						
20	630	630	100.0	619	98.3	610	96.8
21	229	229	100.0	223	97.4	220	96.1
22*	No data						
23*	No data						
	7017	**6994**	**99.7**	**6859**	**97.7**	**6748**	**96.2**

Table 2.5 Coverage figures for all babies (WBN and SCBU) by NHSP 1st phase site (based on eSP) for January 2004.

Site	N	Offered	%	Entered	%	Completed	%
1	637	636	99.8	634	99.5	632	99.2
2	166	166	100.0	161	97.0	159	95.8
3	401	401	100.0	398	99.3	398	99.3
4	216	216	100.0	215	99.5	212	98.1
5	408	408	100.0	403	98.8	399	97.8
6**	No data						
7	185	185	100.0	184	99.5	182	98.4
8	199	188	94.5	167	83.9	163	81.9
9*	208	208	100.0	207	99.5	204	98.1
10	425	423	99.5	405	95.3	403	94.8
11	150	150	100.0	150	100.0	145	96.7
12	227	227	100.0	225	99.1	224	98.7
13	356	356	100.0	356	100.0	356	100.0
14	No data						
15	393	393	100.0	386	98.2	385	98.0
16	470	470	100.0	467	99.4	466	99.1
17	248	248	100.0	248	100.0	194	78.2
18	471	471	100.0	468	99.4	464	98.5
19*	No data						
20	588	588	100.0	579	98.5	574	97.6
21	239	239	100.0	229	95.8	227	95.0
22*	No data						
23*	No data						
	5998	**5984**	**99.8**	**5893**	**98.2**	**5798**	**96.7**

Table 2.6. Coverage figures for all babies (WBN and SCBU) by NHSP 1st phase site (based on eSP) for February 2004.

2.4.2 Refer rates

The NHSP quality standards state that the proportion of babies who are referred by the screen for audiological follow-up assessment should not exceed three per cent of those completing the screen. There is no standard for the AOAE fail rate (the first of the two tests in the screen protocol). Tables 2.7 and 2.8 present the AOAE fail rates and the screen refer rates respectively as recorded by the Hi*Track and Oz SIMS screening management systems. As would be expected, there is a strong positive correlation between AOAE fail rate and screen refer rate (Spearman's rho=0.683, $p<0.001$).

NHSP site	**N having AOAE**	**NCR in one ear only**		**NCR in both ears**		**Total**
		N	%	N	%	%
1	9944	1024	10.3	935	9.4	19.7
2	1925	71	3.7	31	1.6	5.3
3	4705	284	6.0	83	1.8	7.8
4	3162	95	3.0	22	0.7	3.7
5	4326	113	2.6	73	1.7	4.3
6**	2774	39	1.4	47	1.7	3.1
7	2800	128	4.6	82	2.9	7.5
8	2171	106	4.9	59	2.7	7.6
9*	2531	54	2.1	27	1.1	3.2
10	8218	689	8.4	478	5.8	14.2
11	1827	73	4.0	22	1.2	5.2
12	2286	227	9.9	45	2.0	11.9
13	4853	330	6.8	165	3.4	10.2
14	2950	100	3.4	18	0.6	4.0
15	7250	370	5.1	152	2.1	7.2
16	3900	184	5.5	89	1.5	7.0
17	2448	141	5.8	72	2.9	8.7
18	5103	173	3.4	123	2.4	5.8
19*	841	30	3.6	7	0.8	4.4
20	6125	118	1.9	78	1.3	3.2
21	2792	106	3.8	28	1.0	4.8
22*	1781	40	2.2	17	1.0	3.2
23*	No data					
TOTAL	**84712**	**4633**	**5.3**	**2653**	**3.1**	**8.4**

Table 2.7. AOAE fail rate by NHSP 1st phase site in 2003 (based on Hi*Track and OZ SIMS systems)*; NCR= no clear response.*

NHSP Site	N completing the screen	NCR in one ear only		NCR in both ears		Total
		N	%	N	%	%
1	9667	138	1.4	36	0.4	1.8
2	1892	49	2.6	21	1.1	3.7
3	4638	173	3.7	45	1.0	4.7
4	3125	27	0.9	23	0.7	1.6
5	4000	12	0.3	4	0.1	0.4
6**	3200	13	0.4	3	0.1	0.5
7	2778	17	0.6	8	0.3	0.9
8	2167	9	0.4	4	0.2	0.6
9*	2625	15	0.6	6	0.2	0.8
10	8083	219	2.7	72	0.9	3.6
11	1818	16	0.9	4	0.2	1.1
12	2313	55	2.4	19	0.8	3.2
13	5588	79	1.4	16	0.3	1.7
14	3167	12	0.4	7	0.2	0.6
15	7583	75	1.0	16	0.2	1.2
16	5667	44	0.8	24	0.4	1.2
17	2385	16	0.7	15	0.6	1.3
18	5091	31	0.6	25	0.5	1.1
19*	1000	2	0.2	0	0	0.2
20	7500	43	0.5	17	0.3	0.8
21	2714	17	0.6	2	0.1	0.7
22*	1800	5	0.3	4	0.2	0.5
23*	No data					
TOTAL	**88800**	**1044**	**1.2**	**371**	**0.4**	**1.6**

Table 2.8. Screen refer rates by NHSP 1st phase site in 2003 (based on Hi*Track and OZ SIMS systems).

The data in tables 2.7 and 2.8, from the OZ SIMS and Hi*Track systems, is again of doubtful quality since, for example, some sites apparently completed screens on more babies than had the AOAE test, which if the national protocol is being followed, is not possible. The screen refer rates from hand collected data are shown in table 2.9 (AOAE fail rates are not available from this source).

NHSP Site	July 2002		November 2002		March 2003		September 2003	
	One ear	Both ears	One ear	Both ears	One ear	Both ears	One ear	Both ears
1	1.1%	0.3%	1.8%	0.3%	1.5%	0.4%	1.6%	0.5%
2	3.8%	1.5%	3.2%	1.0%	1.2%	0.3%	2.6%	0.9%
3	8.9%	3.8%	8.3%	3.6%	7.1%	2.5%	3.8%	1.8%
4	5.8%	3.3%	4.7%	2.3%	3.7%	No data	3.0%	1.7%
5	1.6%	0.3%	1.3%	0.5%	1.0%	No data	1.0%	0.4%
6	0.1%	0.1%	No data					
7	2.1%	0.5%	1.2%	0.4%	1.8%	1.2%	0.7%	0.6%
8	5.4%	3.6%	No data	No data	No data	No data	4.3%	5.9
9	0.5%	1.3%	0.3%	0.2%	No data	No data	1.0%	0.9
10	8.7%	1.9%	4.9%	1.5%	4.8%	3.8%	3.5%	1.1
11	1.7%	0.2%	2.3%	0.8%	No data	No data	2.0%	0.5
12	7.2%	5.2%	1.1%	0.3%	5.1%	1.6%	1.9%	0.6%
13	3.4%	0.7%	1.1%	0.3%	2.4%	0.8%	2.1%	0.5
14	0.7%	0.3%	0.5%	0.2%	No data	No data	0.5%	0.3
15	0.9%	0.3%	1.0%	1.3%	1.0%	0.5%	3.0%	1.8%
16	2.2%	0.5%	1.1%	0.4%	No data	No data	1.5%	0.7%
17	No data							
18	0.7%	0.5%	No data	No data	3.3%	1.1%	No data	No data
19	0.4%	0.1%	0.4%	0.2%	No data	No data	0.4%	0.2
20	0.6%	0.3%	0.6%	0.3%	0.6%	0.3%	0.7%	0.4
21	2.0%	0.5%	1.4%	0.2%	0.9%	0.2%	0.7%	0.2
22	0.1%	0.1%	0.1%	0.2%	0.3%	0.2%	0.3%	0.2
23	N/A	N/A	0.8%	0.6%	No data	No data	0.3%	0.5
Total	**4.0%**	**2.8%**	**2.2%**	**0.9%**	**2.5%**	**1.1%**	**1.7%**	**1.0%**

Table 2.9. Screen refer rates (=NCR on AABR) for all babies (WBN and SCBU) by NHSP 1st phase site (based on hand-collected data from the Team Leaders/Local Coordinators).

There is a significant reduction in the unilateral refer rate across the time periods (p=0.013), largely because of the (relatively) high refer rate in the first month period, and no significant change in bilateral referral rate which remained below three per cent throughout. (p=0.053). From the (more reliable) hand-collected data it can be seen that by September 2003 the overall referral rate was 2.7%. This is in agreement with the results from the eSP system, where the average refer rate over the four month period (November 2003—February 2004) was 2.8%. Tables 2.10-2.13 show the refer rate data from the eSP system for the months from November 2003 to February 2004.

Site	N completed	Bil referral	%	Uni referral	%
1	691	4	0.6	17	2.5
2	187	2	1.1	6	3.2
3	392	6	1.5	3	0.8
4	246	3	1.2	5	2.0
5	351	0	0.0	1	0.3
7	198	3	1.5	0	0.0
8	228	2	0.9	1	0.4
9	140	4	2.9	2	1.4
10	404	4	1.0	16	4.0
11	183	1	0.5	1	0.5
12	218	3	1.4	7	3.2
13	352	4	1.1	9	2.6
15	510	1	0.2	6	1.2
16	486	5	1.0	5	1.0
17	223	2	0.9	0	0.0
18	371	1	0.3	4	1.1
20	640	0	0.0	3	0.5
21	254	1	0.4	6	2.4
	6086	**46**	**0.8**	**92**	**1.5**

Table 2.10. Screen refer rates (=NCR on AABR) for all babies (WBN and SCBU) by NHSP 1st phase site (based on data from the eSP) for November 2003. Data from NHSP Sites 6**, 14, 19*, 22* and 23* missing.

Site	N completed	Bil referral	%	Uni referral	%
1	768	8	1.0	13	1.7
2	172	1	0.6	2	1.2
3	385	2	0.5	10	2.6
4	243	3	1.2	6	2.5
5	370	1	0.3	5	1.4
7	174	0	0.0	3	1.7
8	97	3	3.1	2	2.1
9	185	0	0.0	4	2.2
10	406	8	2.0	19	4.7
11	147	1	0.7	2	1.4
12	233	3	1.3	3	1.3
13	354	0	0.0	6	1.7
15	560	1	0.2	5	0.9
16	480	9	1.9	6	1.3
17	244	4	1.6	2	0.8
18	353	1	0.3	10	2.8
20	1013	3	0.3	5	0.5
21	217	1	0.5	2	0.9
	6432	**49**	**0.8**	**105**	**1.6**

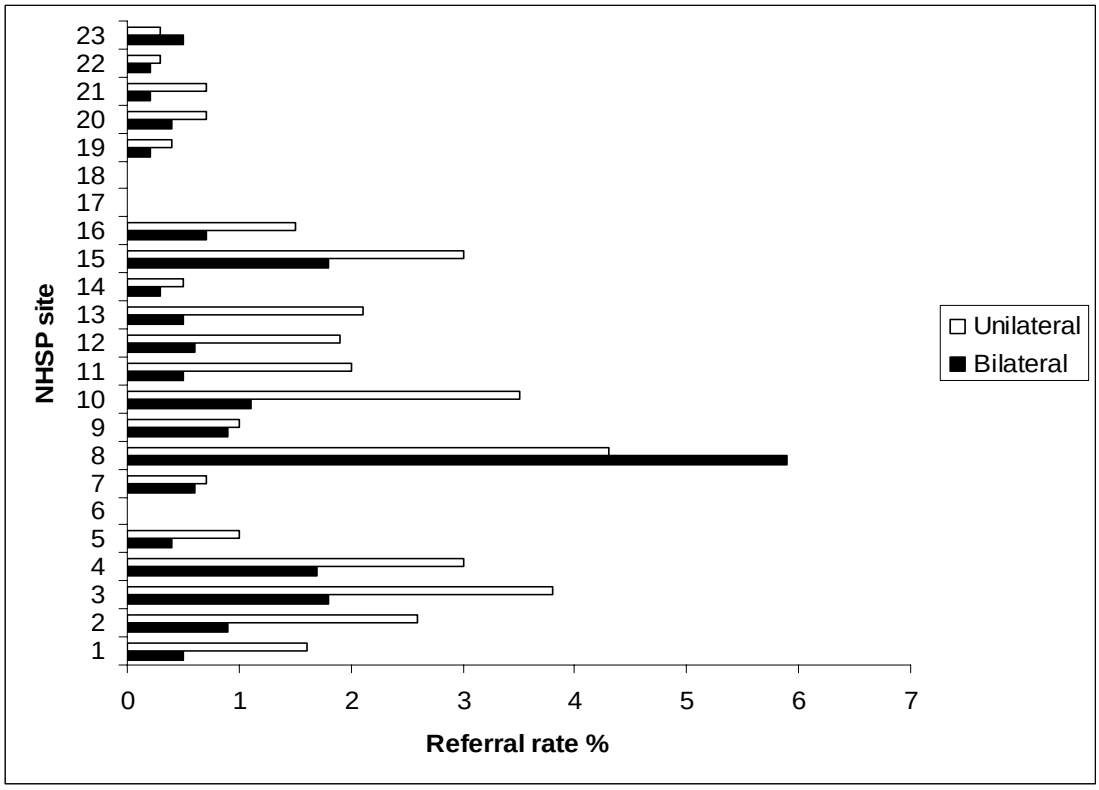
Table 2.11. Screen refer rates (=NCR on AABR) for all babies (WBN and SCBU) by NHSP 1st phase site (based on data from the eSP) for December 2003.

Site	N completed	Bil referral	%	Uni referral	%
1	784	6	0.8	16	2.0
2	229	3	1.3	7	3.1
3	414	5	1.2	18	4.3
4	252	2	0.8	5	2.0
5	408	1	0.2	4	1.0
7	184	0	0.0	2	1.1
8	179	7	3.9	8	4.5
9	342	4	1.2	5	1.5
10	402	5	1.2	11	2.7
11	191	0	0.0	5	2.6
12	253	1	0.4	8	3.2
13	368	4	1.1	10	2.7
15	568	6	1.1	6	1.1
16	570	9	1.6	6	1.1
17	271	3	1.1	2	0.7
18	481	1	0.2	11	2.3
20	610	4	0.7	10	1.6
21	220	1	0.5	4	1.8
	6748	**62**	**0.9**	**138**	**2.0**

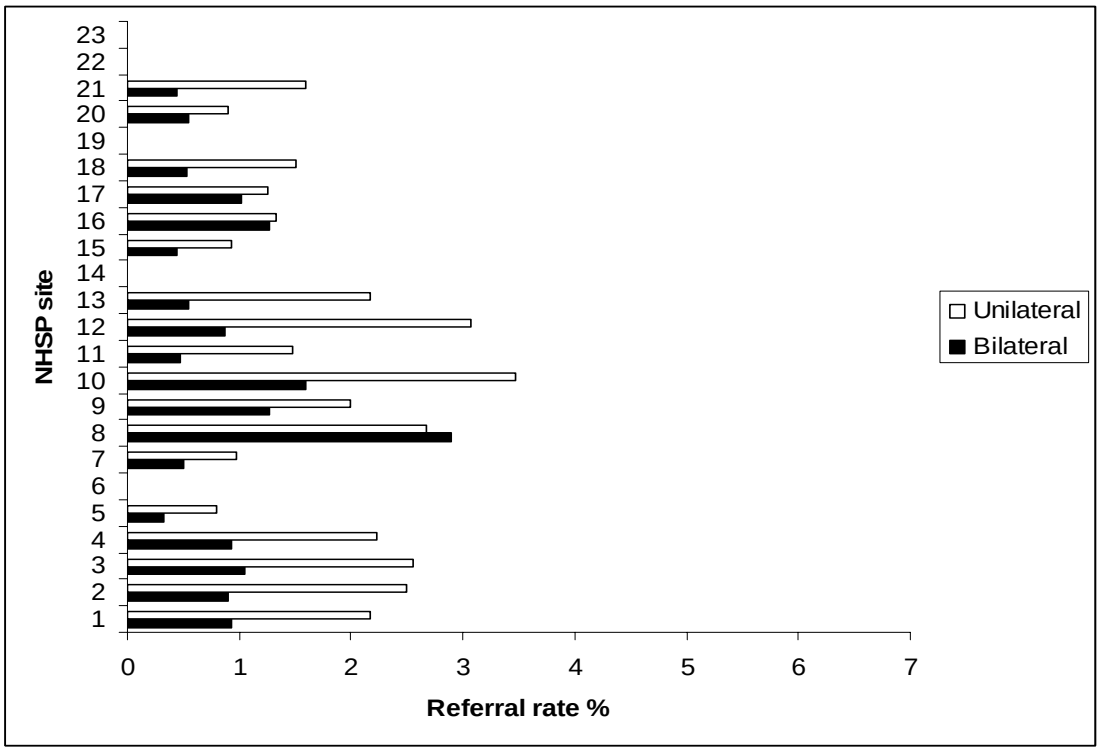
Table 2.12. Screen refer rates (=NCR on AABR) for all babies (WBN and SCBU) by NHSP 1st phase site (based on data from the eSP) for January 2004.

Site	N completed	Bil referral	%	Uni referral	%
1	632	2	1.3	16	2.5
2	159	1	0.6	4	2.5
3	398	4	1.0	10	2.5
4	212	1	0.5	5	2.4
5	399	3	0.8	2	0.5
7	182	1	0.5	2	1.1
8	163	6	3.7	6	3.7
9	204	2	1.0	6	2.9
10	403	9	2.2	10	2.5
11	145	1	0.7	2	1.4
12	224	1	0.4	10	4.5
13	356	0	0.0	6	1.7
15	385	1	0.3	2	0.5
16	466	3	0.6	9	1.9
17	194	1	0.5	5	2.6
18	464	6	1.3	8	1.7
20	574	7	1.2	6	1.0
21	227	1	0.4	3	1.3
	5798	**50**	**0.9**	**112**	**1.9**

Table 2.13. Screen refer rates (=NCR on AABR) for all babies (WBN and SCBU) by NHSP 1st phase site (based on data from the eSP) for February 04.



a)



b)

Figure 2.2. Bar chart summarising the mean referral rates a) from hand-collected data from Team leaders and b) based on data from the eSP..

Figure 2.2 summarises the bilateral and unilateral referral rates. Figure 2.2 a) is based on data from the hand-collected data from the Team Leaders that reflects the cumulative referral rate from the beginning of NHSP by September 2003. The mean bilateral rate is 1.0% and unilateral referral rate is 1.7%. Figure 2.2 b) summarises the referral rates across four months from November 2003—February 2004 based on data extracted from eSP. The mean bilateral referral rate is 0.9% and 1.9% for unilateral referral.

Figure 2.3 shows the flow chart of screen performance for well babies in hospital-based sites based on all hospital-based sites except Site 14 (which did not have eSP data available). The flow chart also shows the numbers of babies that were screened in hospital as 'in-patients' in maternity units and those screened as 'out-patients'. The data are from the hospital-based sites only, so those screened as out-patients would have been discharged home before the screening tests were undertaken.

The data are aggregated to show test and screen outcomes for the 14,898 well babies who were screened during the month of February 2004. Results are shown for the first and second AOAE tests (AOAE1 and AOAE2), and for the AABR test. Outcomes of each test are classified by clear response in both ears (CR), no clear response in one or both ears (NCR), or not completed for one of a number of possible reasons (NC).

Figure 2.4 shows similar data for babies who came from NICU, where the screen protocol involves testing with both AOAE and AABR, rather than contingent AABR with well babies.

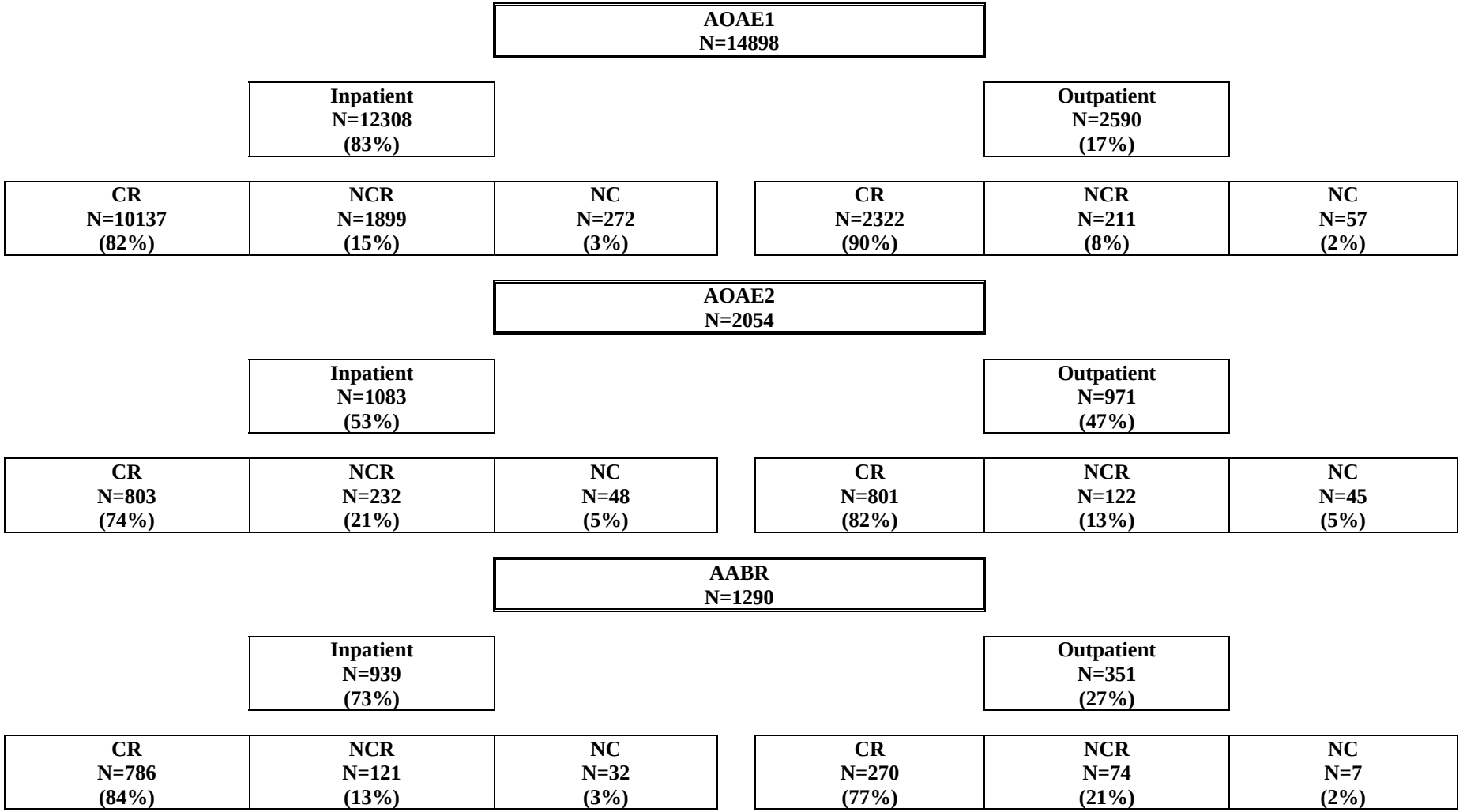


Fig 2.3. Test and screen results for well babies who entered the screen in all hospital-based sites except site 14 for the month of February 2004. CR = Clear Response; NCR = No Clear Response, and NC = all other test result outcomes.

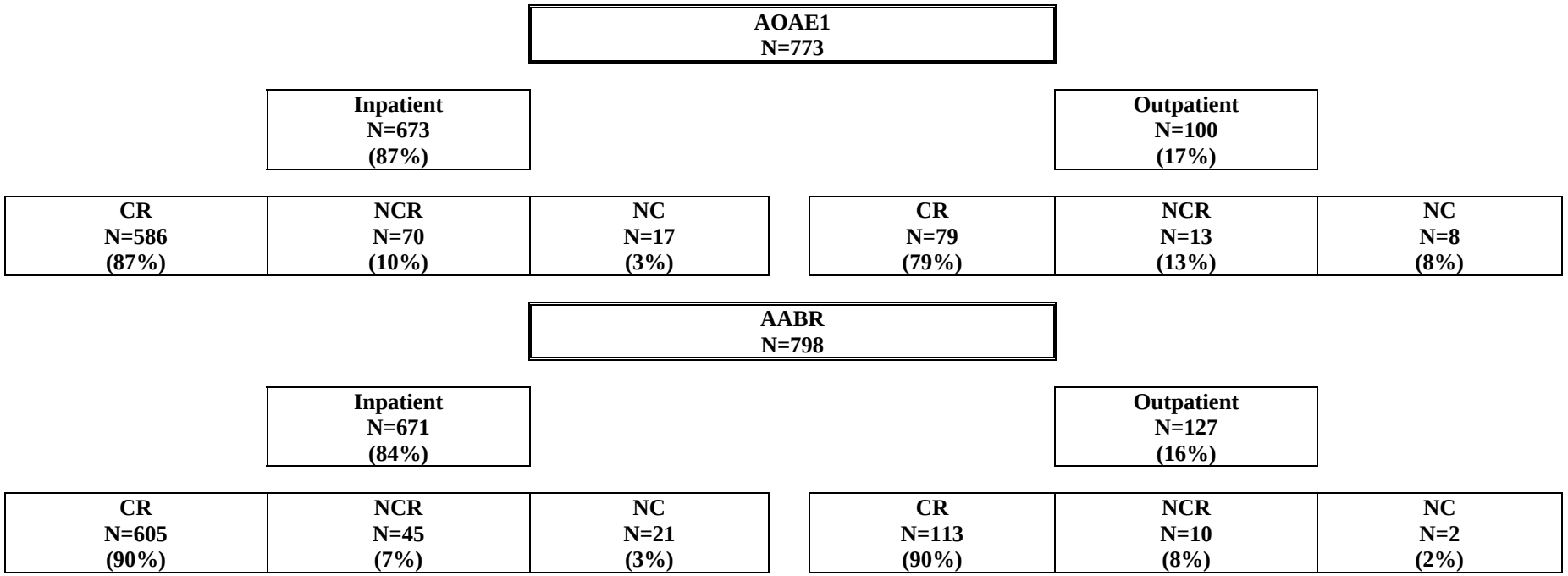


Fig 2.4 change caption: 'Fig 2.4. Test and screen results for NICU babies who entered the screen in all hospital-based sites except site 14 for the month of February 2004. CR = Clear Response; NCR = No Clear Response, and NC = all other test result outcomes.

2.4.3 Lost-for-follow-up rate

The probability of not identifying a child who is hearing-impaired increases at the point of referral. This has a direct impact on programme sensitivity and consequently the yield. Unacceptably high lost-for-follow-up rates of 40-50% in newborn hearing screening programmes have been reported previously in countries outside the UK (e.g. McPherson *et al* 1998, Aidan *et al* 1999, Mehl *et al* 2002, Gorga & Neely 2003).

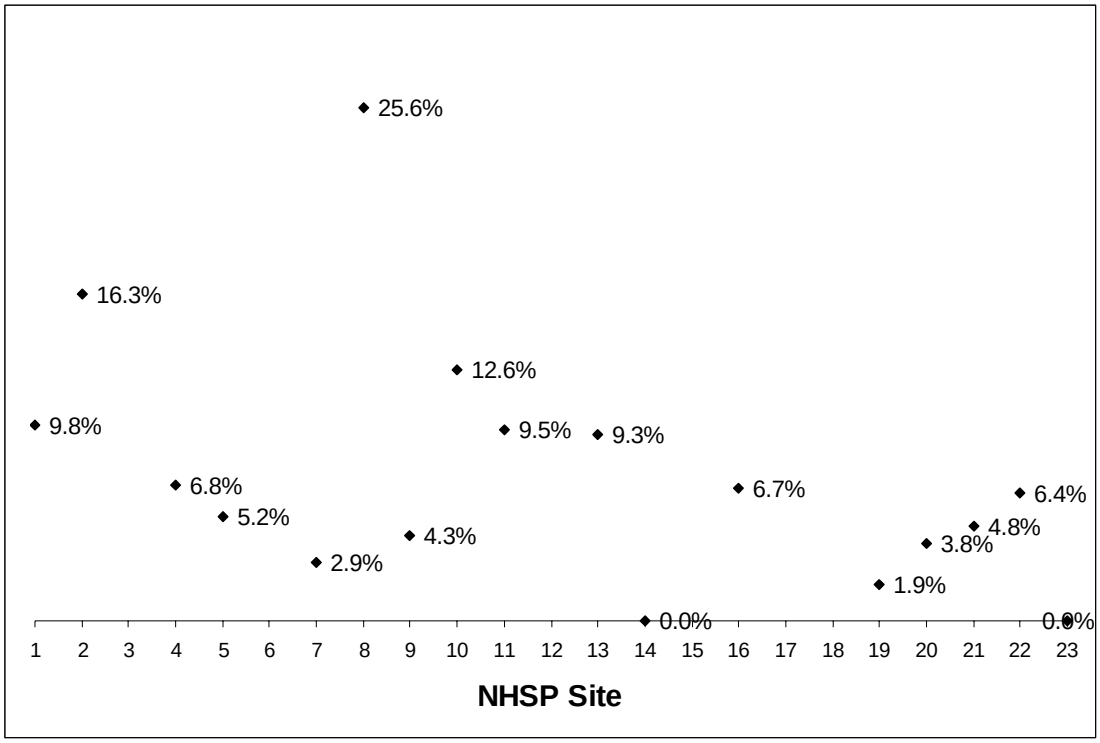


Figure 2.3. Lost for follow-up rate in 17 first phase NHSP sites based on hand-collected data from sites.

In the present evaluation, 9.6% (95% CI 5.9-13.3%) of babies referred by the screen had not been followed up six months after referral. Figure 2.3 displays the lost-for-follow-up rate in 17 NHSP sites based on hand-collected data from the Team Leaders for the 12 month period of screening (data missing from 6 sites). Considerable between-site variability is apparent. Whereas in some sites high lost-to-follow-up rate may be the result of small numbers, in Site 8 (25.8% of referrals are lost for follow-up) it is a symptom of a genuine problem and is possibly having an impact on yield.

In the community-based sites the mean 'lost-for-follow-up' rate is significantly lower, just 3.3% as opposed to 10.1% in the hospital sites (p=0.031).

2.4.4 Positive predictive value (PPV) for screen referral

PPV (the proportion of referred cases which are found to be true positives) is dependent on the prevalence of the condition, as well as on the sensitivity/specificity of the screen and the values and risks associated with the various categories of screen outcome.

The PPV across all sites in the period to 31.5.03 for bilateral hearing loss was 6.7% (95% CI 4.9-8.5%) and PPV for bilateral and unilateral hearing loss combined was 11.5% (95% CI 8.7-14.3%).

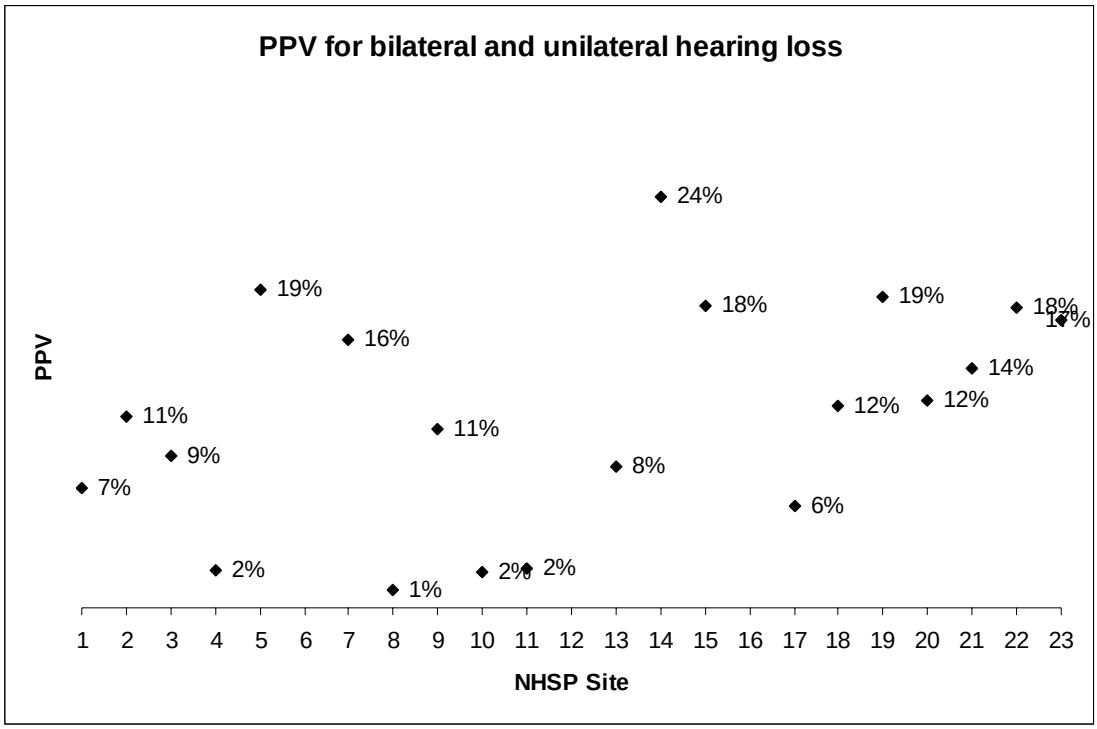


Figure 2.4. Positive predictive value for screen referral for unilateral and bilateral hearing loss combined.

Figure 2.4 shows the variation across sites of PPV for unilateral and bilateral hearing loss combined. The figures are based on hand-collected data from the Team Leaders and refer to a period from the start of NHSP at each site to 31st May 2003 (data missing from 3 sites). There is large inter-site variation, the reasons for which are not clear. Possibilities are the pattern (timing) of discharges from the maternity ward, with more false positives the earlier the screen is completed, differences in equipment performance, differences in lost-to-follow-up rates, and the skills of the screening team. More work is needed to understand the optimal PPV in newborn hearing screening and the factors affecting it.

2.4.5 Yield

The yield for the unilateral and bilateral case definitions combined was 1.64 per 1000 screened (95% CI 1.27-2.01) from the start of NHSP to 30 September 2003 in 21 NHSP first wave sites (data missing from 2 sites). Yield for bilateral hearing loss was 1.00 (95% CI 0.78-1.22) per 1000 screened and for unilateral hearing loss 0.64 (95% CI 0.37-0.91) per 1000 screened. In absolute numbers of true cases identified, this represents 154 bilateral cases and 99 unilateral cases. The yield at each site is too low in a one year period to report as meaningful figures.

2.4.6 Summary of screen and follow-up data

Figure 2.5 uses the eSP data to summarise the aggregated data for the overall screen journey for the target babies from all first wave sites for the four months from November 2003 to February 2004. If we compare the figures in Fig 2.5 against the original NHSP standards, we can see that on major measures of screen performance the standards are met at an aggregated level, although as we saw from individual site data this is not always the case. According to NHSP standards ≥95% of all babies entering the screening programme, should complete the programme. From the aggregated data it is apparent that 96% of all target babies completed the screen. As for referral, NHSP standards state that the referral rate should not be over 3%. The average referral rate across November 2003—February 2004 combining both bilateral and unilateral referrals was 2.8% thus meeting the set target.

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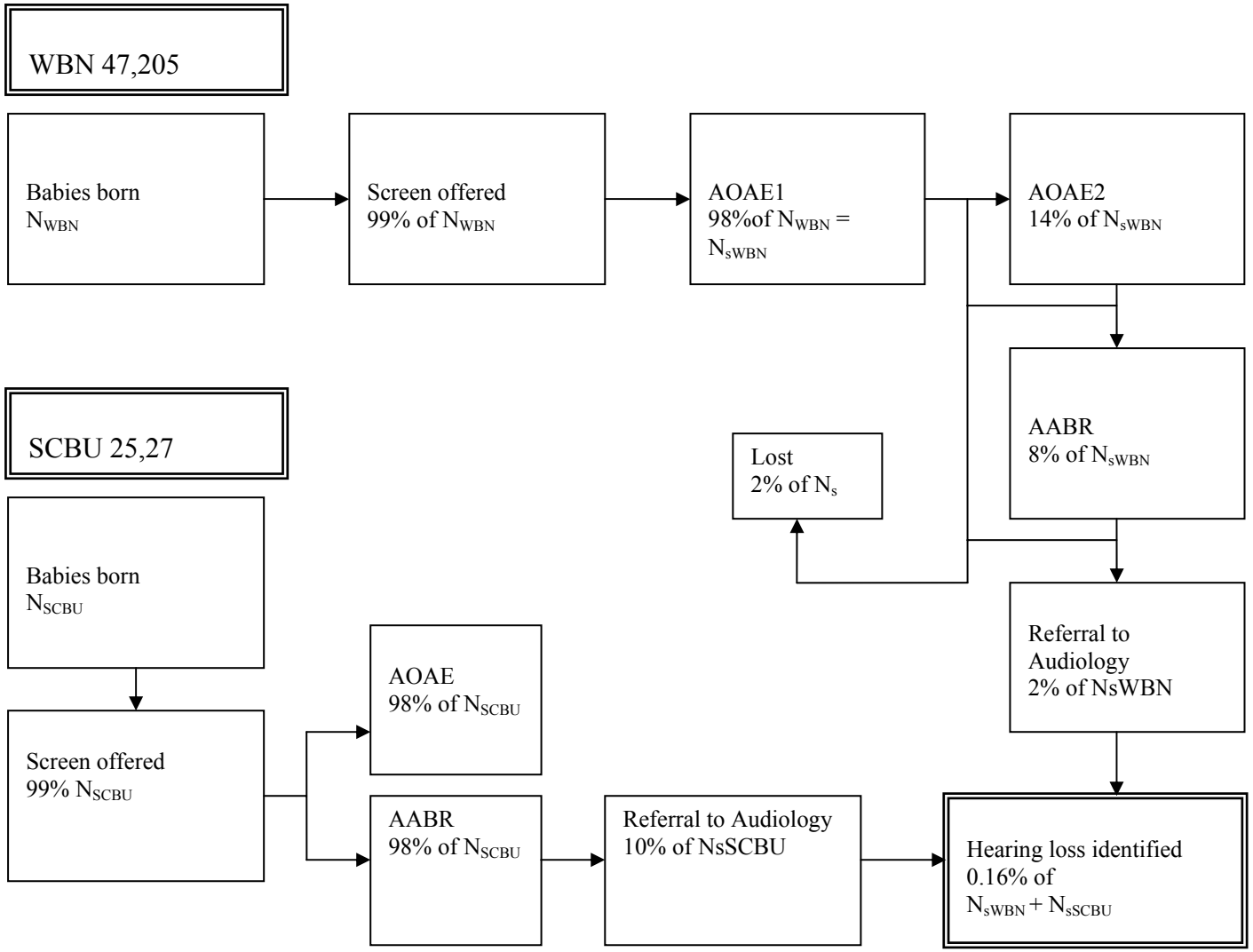

Figure 2.5. Flowchart of the aggregated screen data for all first wave sites (excl. Sites 6, 14, 19, 22, 23) based on 4 month data (November 2003 – February 2004) from eSP.

2.5 Discussion

Drawing on the most reliable data source, 99.6% of mothers of new babies were offered the newborn screen across all first phase sites between the months of November 2003 and February 2004. This time frame was selected as recent, the screen having been in place for well over a year in all sites, and there is no reason to suspect that it is not representative. All but three sites offered the screen to over 99% of mothers; of the three sites who did not achieve this level, there were four one-month-periods when the offer rate was between 95% and 99%, and three one-month-periods when it was between 90% and 94.9%.

Thus, with occasional exceptions, almost all mothers who should have been offered the screen were indeed offered it. This is important to know, and implies that mechanisms are in place even in sites which have high very early discharge rates and which are using the hospital-based model to make sure that babies are screened.

The two percent difference between the numbers of *offered coverage* and *entered coverage* was mainly due to practicalities such as early discharge right after the parent had been offered the screen. Only a very small minority actually refuse to consent to the screen. According to the NHSP Population Report produced on 11.10.2004 parents of just 0.27% of the babies born between 01.01.—31.05.2004 declined to sign the consent and a further 0.04% withdrew their consent midway. There is anecdotal evidence to suggest that the refusals group is likely to contain a high proportion of babies with parents from ethnic minority backgrounds. If this were so, it may be that interpreters are not as available to mothers as they should be, or that requiring written consent for this specific procedure (rather than for a raft of routine procedures including hearing screening) is discriminatory. Research is needed into which groups decline to sign the consent from, and why.

The encouraging entered coverage rate of 97.5% over all sites for the four month period apparently took time to achieve for some sites. There was a significant improvement in mean entered coverage reported by team leaders across the first year of screening, taking data from July 2002, November 2002, March 2003, and September 2003 (n = 21 sites, data not available from two sites). In July 2002, when all sites had been running for less than a year (and only a month or two in some cases), nine sites had entered coverage of less than 95%. For later phases of the implementation of the screen, the implementation team has developed much more explicit procedures and a workbook for sites preparing for the screen, and this is likely to reduce the 'learning curve', during which sites fail to achieve screen performance targets, to a minimum. Despite the overall trend for coverage to improve during year one, some sites remained cause for concern with entered coverage below the required standard of 95%. This pattern of some sites consistently performing below standard is one which is repeated again with other screen performance data.

The screen completed rate was over 95% in all but eight sites for all four of the November 2003 to February 2004 months, and over 90% for all but three sites for all four months. There were five one-month-periods at five different sites when completed coverage fell to between 90 and 95%; these appeared to be 'blips' and did not indicate consistent underperformance. However, there were three sites (numbers 8, 10 and 17) that had consistently poor completed coverage; site 8 averaged 81.5%, site 10 averaged 91.3%, and site 17 averaged 83.5%. All these sites represent large urban areas with high proportion of non-residential births and a

significant ethnic minority population coupled with early discharge from the maternity ward. Site data are monitored on a weekly basis by the implementation team and action taken should standards not be met. The nature of the action taken will vary according to circumstances but in the case of consistent under-performance the challenges may be longterm and difficult. Clear and transparent procedures need to be in place (after the implementation programme is complete as well as during implementation) to manage and remedy such consistent under-performance.

The standard set by the Newborn Hearing Screening Steering Committee for screen refer rate is <3% of screened babies. Taking the most reliable data source, mean refer rates for the four months from November 2003 to February 2004 across all sites were 2.3%, 2.4%, 2.8% and 2.8%. The mean bilateral refer rate over all sites and all four months was 0.85%; the unilateral refer rate across the same period was 1.75%. Data reported by team leaders for the period July 2002 to September 2003 gave similar rates, with some evidence of higher rates in the month just after implementation of the screen.

This encouraging overall pattern again hides consistent under-performance from some sites. Site 8 had >3% referral for bilateral alone for three of the four months; there were 26 one-month-periods in which the combined unilateral and bilateral refer rates were >3%, within which one site had rates above 3% for all four months (site 10), and three sites for three of the four months (sites 8, 2 and 12).

The mean lost-to-follow-up rate at six months of age for those referred by the screen was approximately 10%. Again, site 8 performed poorly with 25% lost to follow-up; site 2 had a 16% loss. If these two outliers are removed the lost-to-follow-up rate was in the order of 4%, which is certainly more reasonable. However, every effort should be made to reduce the lost-to-follow-up rates. Adequate information to parents before, during and after the screen combined with involvement from other primary care professionals, especially health visitors, are the key to attendance at appointments. Seeking support from the voluntary organisations and local community as well as improving the interpreting services could be essential in promoting the screening programme in families for whom English is not their preferred language. Flexibility in offering appointments suitable for the young family is conducive to attendance. An efficient screening management system coupled with well-organised administration are vital in supporting attendance at follow-up appointments. A minimum standard for loss-to-follow-up should be set, perhaps at 10%, with an aspirational standard of 5%.

The Positive Predictive Value (PPV) of screen referral for true case status (unilateral or bilateral permanent hearing loss of moderate or greater degree) was highly variable across sites, ranging (in the period assessed—from screen start to 31.5.03 for all sites) from 1% to 24%, with a mean of 11.8%. This high level of practice variability would represent potential cause for concern if it were to continue or be a feature of later phase sites. It is partly a refection of high referral numbers (sites 8 and 10) and could also involve possible poor assessment practice leading to low yields. On the one hand measures should be taken to lower the referral rates in areas where they are over the quality standard. The Implementation team has acted at such sites by changing equipment if necessary and by providing extra (remedial) training to teams. On the other hand, the low PPV of screen referral for hearing loss may be the function of diagnostic difficulties (particularly in case of moderate hearing loss). From the study of the impact of NHSP on Paediatric Audiology services (Chapter 6) it is apparent that

the services feel an urgent need for further training in advanced diagnostic procedures used in very young infants.

The yield of true bilateral cases from the first wave sites from the time they started screening to the end of September 2003 was 1.0 per 1000 babies screened (95% CI 0.78-1.22), which accords well with expected congenital prevalence rates (Fortnum *et al* 2001). The corresponding yield for true unilateral cases in the same time period was 0.64 (0.37-0.91), which (although fewer data on prevalence rates are available) also accords well with estimates from other sources.

Thus, the overall pattern on screen performance from first wave sites is encouraging, with high coverage, referral rates within the 3% standard, and yield suggestive of high programme sensitivity. There are, however, some concerns: an overall lost to follow up rate of about 10%, and some sites with high referral rates. In fact, three sites (2, 8 and 10) showed clear evidence of consistent underperformance across all or most measures. This points to the need for a detailed and on-going Quality Assurance (QA) system, with standards which if not met would trigger appropriate action.

2.5.1 Community-based and hospital-based screen performance

Early discussions with the proponents of the community-based screening model suggested that one possible advantage of this approach might be an increased probability of meeting the standards for screen performance, compared with the hospital-based model (the other possible advantages of lower levels of maternal anxiety and better cost effectiveness are dealt with elsewhere). In fact, the evidence from the first phase sites suggests that both models can meet the necessary screen performance targets. Table 2.14 summarises the key screen performance data for both models. The data are extracted solely from the hand-collected dataset since for only one community-based site were the data available from eSP.

Performance measure	Hospital-based screen	Community-based screen
Entered-screen coverage (%):	97	99
Bilateral referral rate (%):	1.1	0.4
Unilateral referral rate (%):	1.9	0.5
Lost-to-follow-up (%):	10.1*	3.3

Table 2.14. Mean screen performance measures in the hospital-based and community-based sites from the start of screening until 30 September 2003 (*If the two outliers are excluded, i.e. sites 2 and 8, this figure falls to 6.5%).

While both models meet the necessary standards for coverage and refer rates, it is also clear that the community-based model as delivered by the four sites in the first phase of NHSP implementation exhibit particularly high performance levels. The four community-based sites in the first phase were arguably especially committed to delivering the community-based model, and the extent to which these levels of performance are emulated by the community-based sites in the later phases of NHSP will be of interest.'

2.6. Summary points

- A user-friendly tailored screening-management system is vital for managing and auditing the screening programme, eSP seems to fulfil that need, while the original systems did not
- 99.5% of all target babies were offered a screen
- 97.5% of all target babies entered the screen
- 96.0% of all target babies completed the screen
- Refer rate decreased consistently from the beginning of the screen in 2002 to 2.7% averaged across sites by September 2003
- 9.6% (95% CI 5.9-13.3%) of all referred babies had not been followed up by 6 months after referral
- 11.5% (95% CI 8.7-14.3%) of all referred babies were identified with hearing loss
- Yield per 1000 babies screened is 1.64 (95% CI 1.27-2.01): 1.00 (95% CI 0.78-1.22) per 1000 screened for bilateral permanent hearing loss and 0.64 (95% CI 0.37-0.91) per 1000 screened for unilateral permanent hearing loss
- Aggregated data across all first phase hospital-based sites were good, and exceeded the current NHSP standards; however, within these data were individual sites not performing at acceptable levels. Action is being taken by the implementation team; procedures need to be in place to manage such under-performing sites
- On the basis of the limited data available, it appears that both screening models (hospital-based and community-based) can meet the screening standards set

3. FOLLOW-UP OF TRUE CASES IDENTIFIED BY NHSP

3.1 Aims

The aim of this part of the evaluation was to provide a profile of true cases identified through the NHSP first phase and to determine the main proxy outcomes: age at first audiological follow-up; age at identification of hearing loss and age at hearing aid fitting.

3.2 Method

3.2.1 Outcome measures

Age at first audiological follow-up is the chronological age of the baby at the first audiological follow-up assessment after 'screen refer' that was triggered by no clear response on either ear after *the* AABR *test* (i.e. according to the national screen protocol). The data on first audiological follow-up appointment presented here are based on the cases that were eventually identified with hearing loss.

Age at identification is the chronological age of the baby when, using age-appropriate testing, there is good clinical evidence to suggest that the baby has a permanent bilateral hearing loss with hearing threshold ≥ 40 dB HL based on the average in the better hearing ear at 0.5, 1, 2 and 4 kHz. At this stage the exact degree and configuration of the hearing loss may still remain uncertain.

Age at hearing aid fitting is the chronological age of the baby at the first hearing aid fitting.

Where possible, the measures were compared against the NHSP quality standards according to which referred babies should start the assessment procedure within 4 weeks of the screen completion; have audiological confirmation by the age of 5 months and in appropriate cases be fitted with hearing aids within 4 weeks of audiological certainty (diagnosis). See appendix for details.

3.2.2 Procedure

The 'True Case Packs' were sent to the Team Leaders of the 23 first phase NHSP sites shortly after their site had started the screening programme. True Case Packs consisted of a covering letter from the Evaluation Team to the Team Leader, a detailed instruction chart and 4 Proformas that the Team Leader was instructed to send to the paediatric audiology service(s)

in their area. Three of the Proformas (yellow Proforma 1.2; pink Proforma 4.1 and blue Proforma 5.11A—see appendix) were completed by the paediatric audiology service and returned directly to the Evaluation Team. These proformas were anonymous, displaying only the child's local unique identifier and date of birth, and giving information on the three main outcome measures, as well as gender, the degree and type of hearing loss, risk factors and comorbidity data. The green form, detailing the name, address and local unique identifier of each true case, was sent by paediatric audiology services to the Team Leaders and retained by them.

Data were collected for all children identified through first phase NHSP sites who were born before 1 January 2004.

3.2.3 Data Analysis

Analyses were conducted using SPSS for Windows version 10. The main analyses consisted of one-way analysis of variance comparing degrees of hearing loss. Independent t-tests were conducted for comparing age at first audiological follow-up assessment, at identification of hearing loss and age at hearing aid fitting between babies from WBN and NICU.

3.3 Results

3.3.1 Bilateral hearing loss cases

The number of babies identified with permanent bilateral hearing loss with hearing threshold ≥ 40 dB HL based on the average in the better hearing ear at 0.5, 1, 2 and 4 kHz through first phase of NHSP that the Evaluation Team was informed about was 169. Considerable effort was put into reminding services to pass the information on true cases back to the evaluation team, and this figure is therefore likely to be close to the 'true' number identified. It also gives a yield figure per thousand screened babies which is close to published prevalence figures for congenital bilateral permanent hearing loss of moderate or greater degree (e.g. Davis *et al* 1997, Fortnum *et al* 2001). Of course, some cases may still have not been identified due to (for example) assessment difficulties with mild-moderate losses or temporary conductive hearing loss overlay, non-attendance at follow-up etc. Nevertheless, the data presented in this section are likely to be robust and close to the 'true' situation.

3.3.1.1 Profile of cases of permanent bilateral hearing loss.

Figures 3.1-3.4 provide the basic information on the profile of the 169 babies identified with permanent bilateral hearing loss.

Figure 3.1 demonstrates the gender distribution: more boys (57%) than girls (43%) were identified with permanent bilateral hearing loss. Figure 3.2 provides details of the distribution of degrees of hearing loss: moderate hearing loss 40-69 dB HL; severe hearing loss: 70-94 dB HL and profound hearing loss ≥95dB HL. As expected sensorineural hearing loss was the most predominant type of hearing loss (Figure 3.3). Auditory neuropathy was defined as a

condition characterised by the absence or severe abnormality of auditory brainstem response (ABR) in the presence of evoked otoacoustic emissions (OAEs).

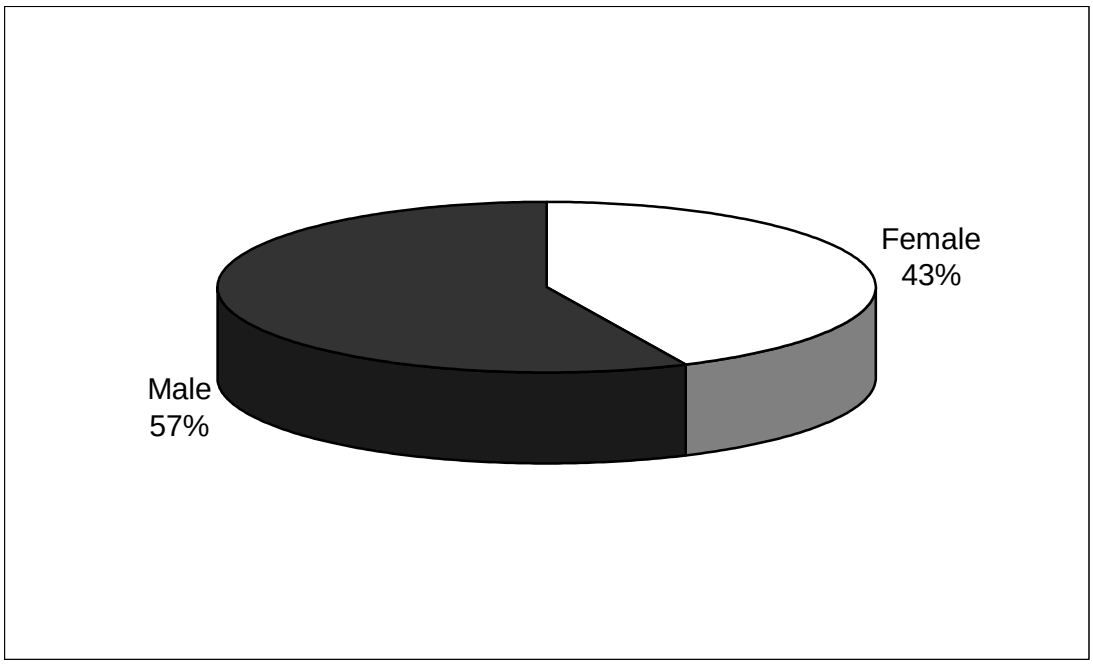

Figure 3.1. Distribution of cases identified with permanent bilateral hearing loss by gender.

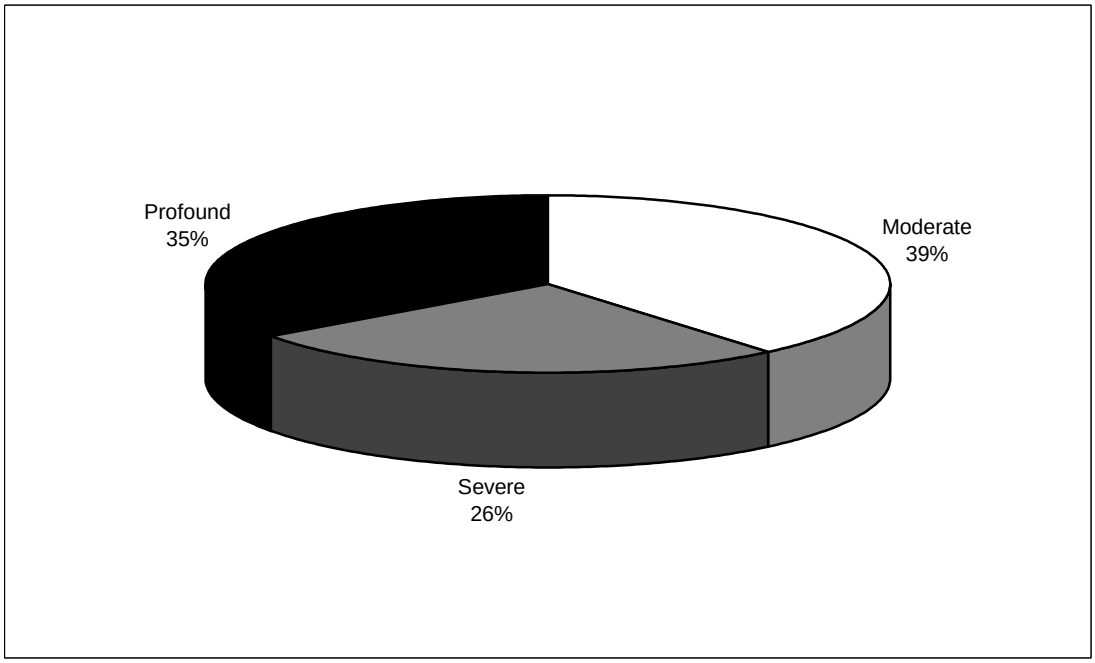

Figure 3.2. Distribution of cases identified with permanent bilateral hearing loss by degree of hearing loss.

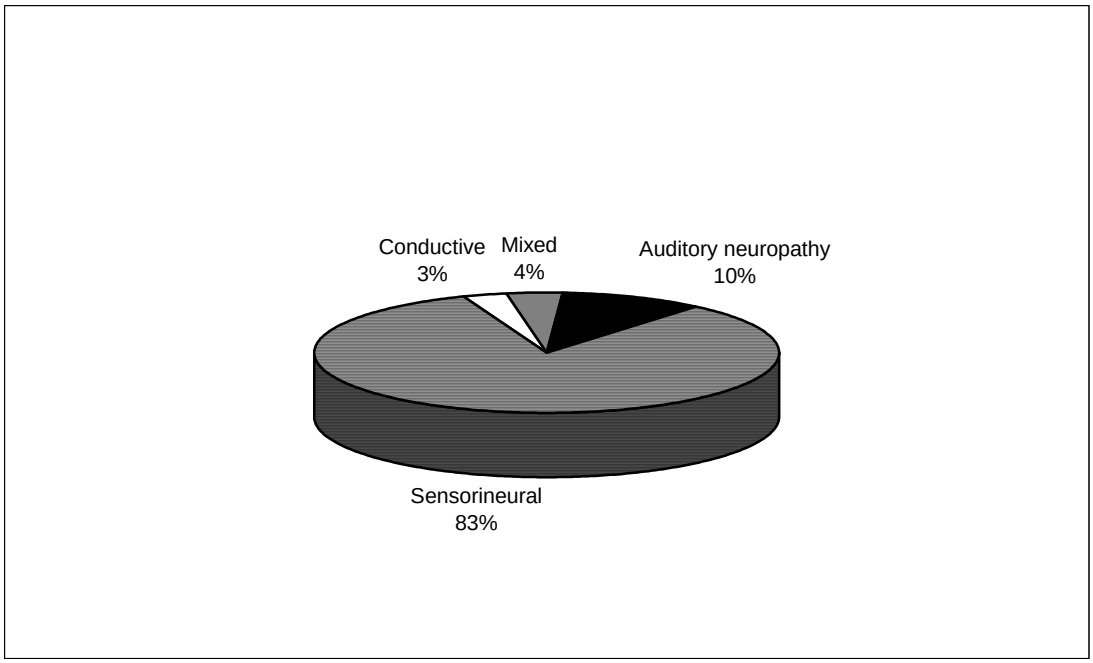

Figure 3.3. Distribution of cases identified with permanent bilateral hearing loss by type.

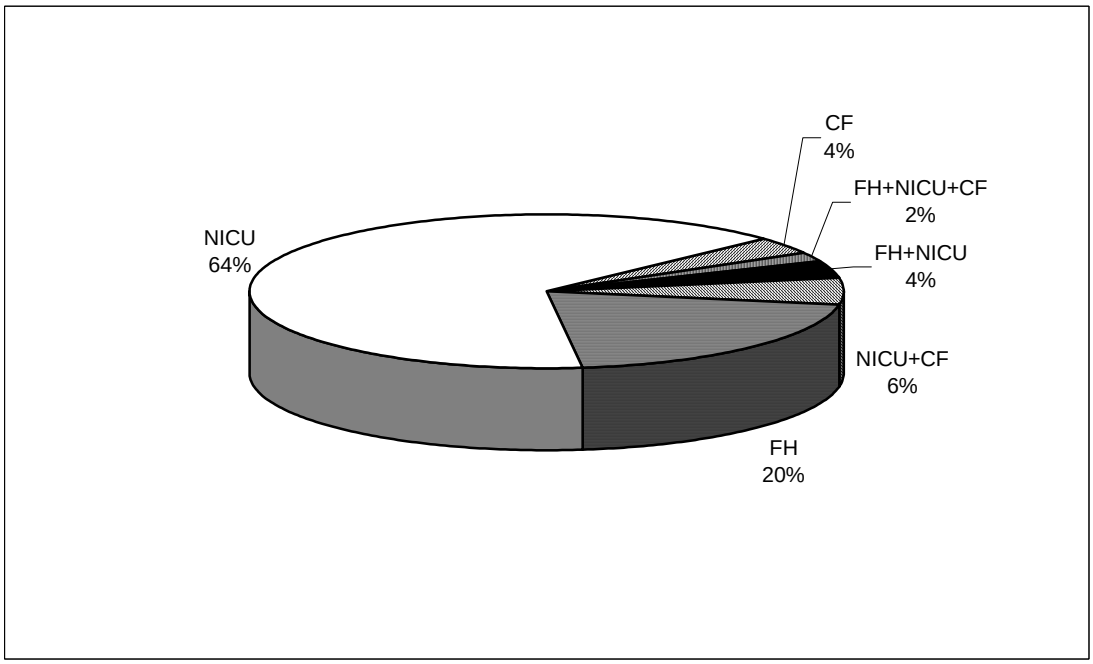

Figure 3.4. Distribution of risk factors in 91 high-risk babies identified with permanent bilateral hearing loss. NICU: a history of admission to NICU for more than 48 hours; FH: a family history of early childhood permanent deafness*;* and CF: a craniofacial anomaly (e.g. cleft palate) associated with permanent hearing loss

Risk factors for permanent congenital hearing loss are well established. The three major risk factors are (i) a history of admission to NICU for more than 48 hours; (ii) a family history of early childhood permanent deafness*;* and (iii) a craniofacial anomaly (e.g. cleft palate) associated with permanent hearing loss (Davis *et al* 1992, Fortnum *et al* 1997). Ninety-one of the 169 true cases (54%) had one or more of these risk factors, which is similar to published estimates (e.g. Fortnum & Davis 1997). Figure 3.4 details the distribution of risk indicators. The most common risk indicator was spending 48 hours or more in NICU, occasionally

combined with family history of childhood hearing loss or craniofacial anomaly or even both: 75.8% (N=69) of high-risk babies are from NICU population.

No association was found between the presence of a risk-factor and the degree of hearing loss (NICU: χ^2=1.404, df=2, p=0.495, FH: χ^2=4.679, df=2, p=0.096, CFA: χ^2=0.190, df=2, p=0.910).

36.4% (N=55) of children identified with bilateral hearing loss had additional conditions and/or disabilities (see table 3.1).

Condition	**N**
Congenital heart defect	**6**
Cleft lip and /or palate	**5**
Multiple unspecified problems	**5**
Visual problems	**4**
Cerebral palsy	**4**
Marked developmental delay	**4**
Waardenburg's Syndrome	**3**
Down Syndrome	**3**
Unspecified gastrointestinal problems	**2**
CHARGE Syndome	**1**
Cornelia De Lange Syndrome	**1**
Dandy Walker Syndrome	**1**
Pierre Robin Syndrome	**1**
Wolf Hirschhorn Syndrome	**1**
Treacher-Collins Syndrome	**1**
Chronic respiratory problems	**1**

Table 3.1. Additional conditions found in babies identified with permanent bilateral hearing loss (in the order of frequency).

3.3.1.2 Age at the first audiological follow-up assessment[6]

The quality standards state that at least 95% of those requiring assessment (i.e. referred by the screen) should start the audiological follow-up assessment within 4 weeks of the screen completion. It was not possible with the data collection systems available at the time to obtain the dates of all the screening episodes for all babies identified with hearing loss. Therefore the data presented here are based on the chronological ages of the babies.

Table 3.2 summarises the descriptive statistics for the chronological age at the first audiological follow-up assessment for all babies who were eventually identified with hearing loss. Median age at the first follow-up was 5.0 weeks.

		Age at first audiological assessment (in weeks)
N	Valid	143
	Missing	9[7]
Range	Min	0
	Max	31
Mean		7.5
SD		6.2
Percentiles	25	3.0
	50	5.0
	75	10.0

Table 3.2. Descriptive statistics of age at first audiological assessment.

Figure 3.5 shows the cumulative percentage for chronological ages by which the first audiological follow-up assessment was carried out. By 4 weeks of age 64% of WBN babies had had their first audiological follow-up assessment. The target of following up ≥95% of WBN babies was achieved by 11 weeks of age.

Figure 3.6 is a non-parametric representation of the age at the first audiological follow-up session by degree of hearing loss. The box shows median and 1st and 3rd quartiles; whiskers show tails to largest and smallest acceptable values. (o) represents outliers (1.5 x IQR from 1st and 3rd quartile) and (*) stands for extremes (3 x IQR from 1st and 3rd quartile). Post hoc testing using Tukey B test indicated that there was no significant difference in age between the groups ($F(2,130)=2.641$, $p=0.075$).

Figure 3.7 represents the distribution of age at the first audiological follow-up assessment by well baby or NICU baby. Independent t-test showed a significant difference in the age at the first audiological follow-up assessment between these two subpopulations ($t=6.516$, $df=139$, $p<0.001$). Table 3.3 separates the descriptive statistics for these two groups. The median age at the first audiological follow-up assessment for WBN and NICU babies is 4.0 and 9.0 weeks respectively.

[6] From this point onwards cases with auditory neuropathy have been excluded.
[7] The audiology services failed to provide the Evaluation Team with a date of first audiological assessment for 9 cases.

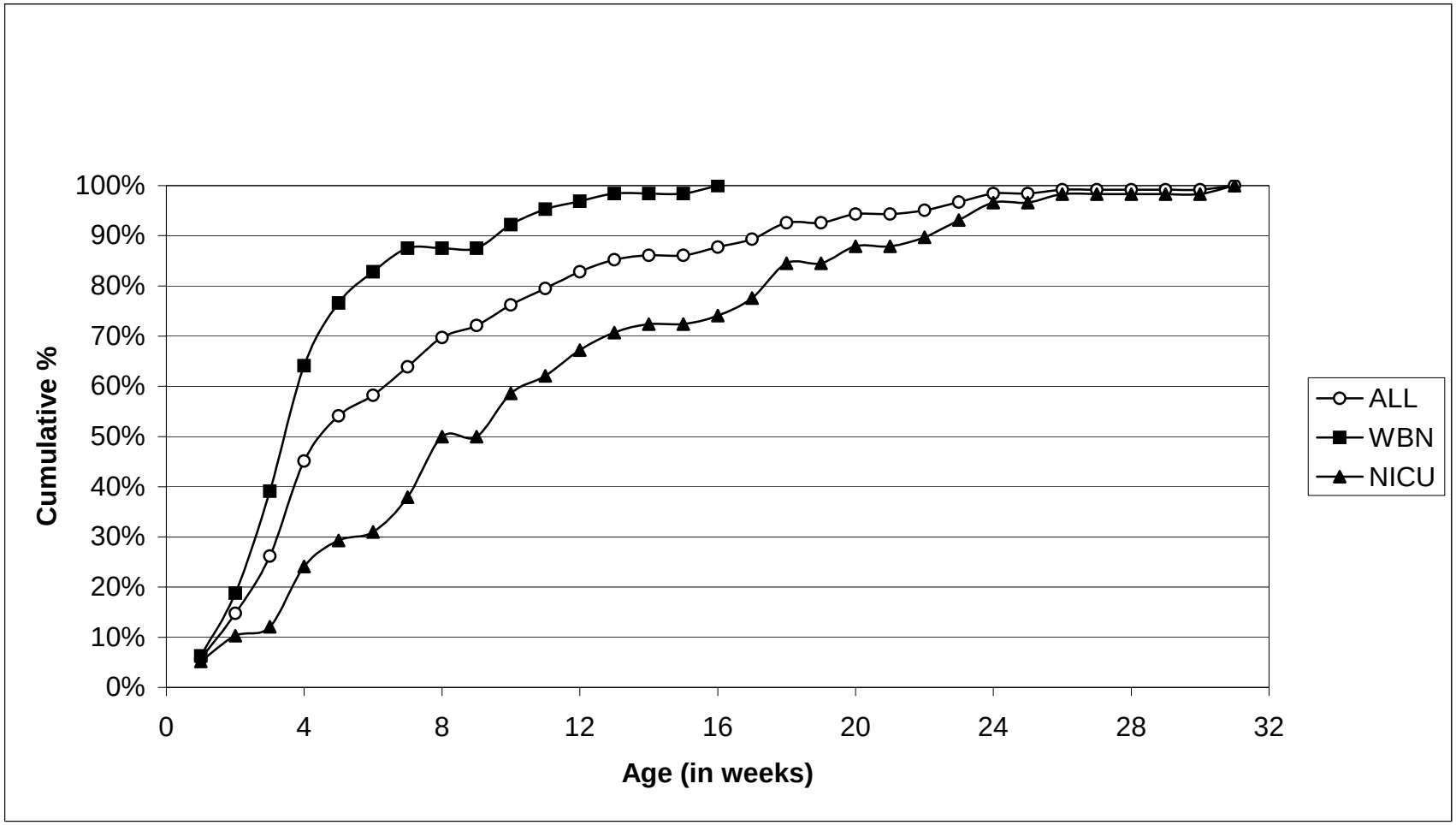

Figure 3.5. Cumulative percentage for chronological ages by which first audiological follow-up assessment was carried out.

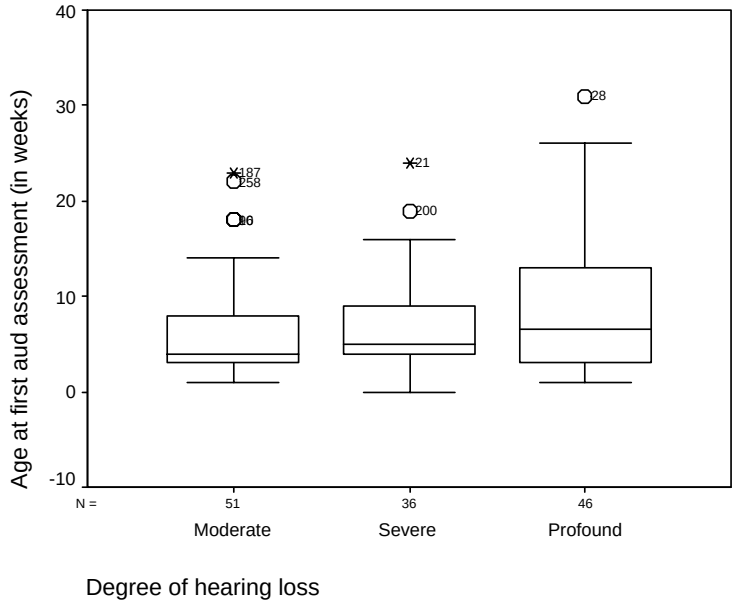

Figure 3.6. Age at the first audiological follow-up assessment by degree of hearing loss.

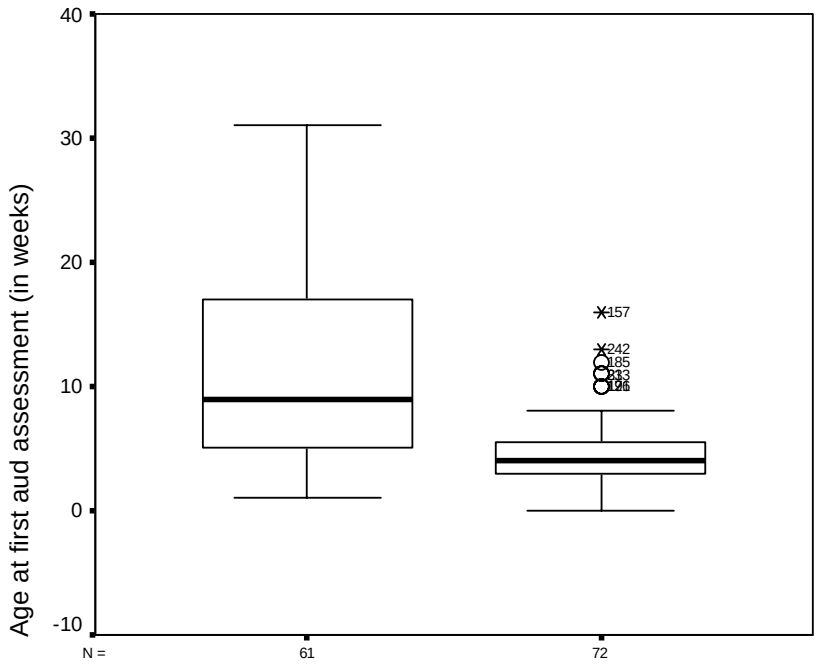

Figure 3.7. Age at the first audiological follow-up assessment by nursery: NICU (N=61) and well-baby nursery (N=72).

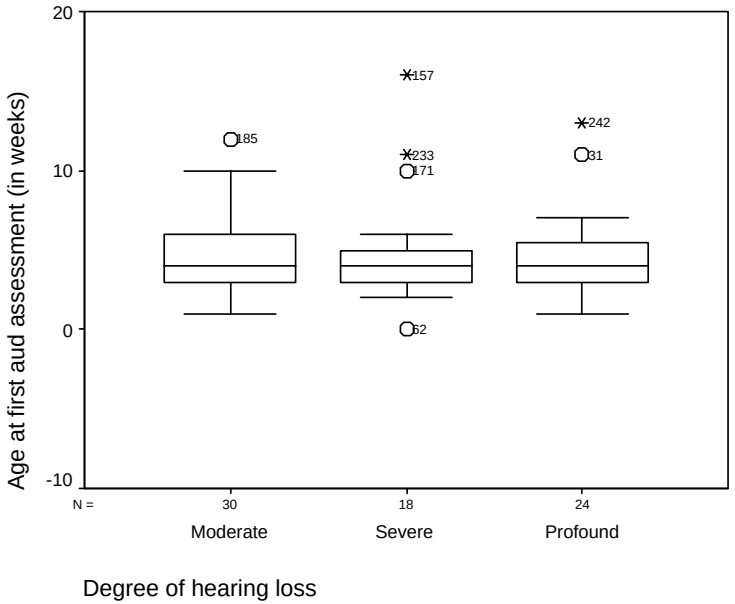

Figure 3.8. Age at the first audiological follow-up assessment by degree (WBN population).

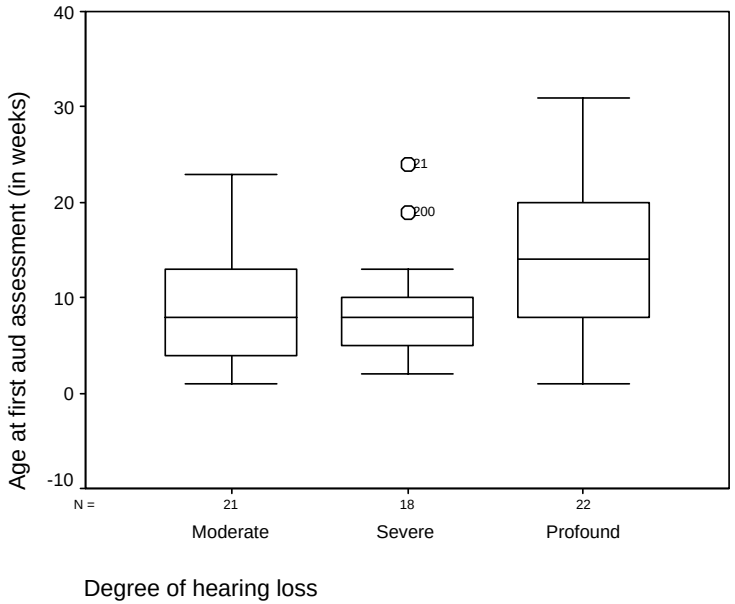

Figure 3.9. Age at the first audiological follow-up assessment by degree (NICU population).

		Age at first audiological assessment (in weeks)	
		WBN	NICU
N	Valid	77	66
	Missing	5	4
Range	Min	0	1
	Max	16	31
Mean		4.7	10.8
SD		3.0	7.3
Percentiles	25	3.0	4.5
	50	4.0	9.0
	75	5.8	17.0

Table 3.3. Descriptive statistics for age at audiological assessment (WBN and NICU separately).

In the WBN population there were no significant differences in age between babies who were eventually identified with different degrees of hearing loss ($F(2,69)=0.114$, $p=0.892$).

Table 3.4 details the reasons for the delay for the first audiological follow-up for babies who emerged as 'outliers' and 'extremes' on the box-and-whisker plot in Figure 3.8.

Case	Age at the first audiological follow-up assessment	Explanation for the delay
12	8 weeks	Service-related delay (long waiting time)
31	15 weeks	Missed the appointment (reason unknown)
126	12 weeks	Non-resident; administrative difficulties
157	16 weeks	Bereavement in the family
172	10 weeks	Service-related delay (long waiting time)
185	12 weeks	Non-resident
233	11 weeks	Service-related delay (long waiting time)
242	13 weeks	Administrative difficulties

Table 3.4. Details of reasons for delay of the first audiological follow-up assessment (WBN population).

In the NICU population, Tukey B test revealed a significant difference in age between babies who were eventually identified with different degrees of hearing loss ($F(2,58)=4.132$, $p=0.021$). Babies with profound hearing loss fall into a separate subset for $\alpha=.05$.

Table 3.5 shows the explanation of the delay for the first audiological assessment for NICU babies who appeared as outliers on box-plot in figure 3.9.

Case	Age at the first audiological follow-up assessment	Explanation for the delay
21	24 weeks	Severe respiratory distress; prolonged stay in NICU
200	19 weeks	Very premature (25/40), prolonged stay in NICU. The baby was 44 weeks gestational age at the first audiological assessment hence meeting the NHSP targets.

Table 3.5. Details of reasons for delay of the first audiological follow-up assessment. (NICU population).

3.3.1.3 Age at identification of hearing loss

Age at identification of hearing loss is considered one of the most important proxy outcomes. Table 3.6 summarises the descriptive statistics for age of identification, and figure 3.10 presents the cumulative distribution of age of identification. It is apparent that 92% of the

WBN babies (and 83% of all babies, both WBN and NICU) in the present cohort had their hearing loss identified by 5 months of age.

		Age at first identification of HL (in weeks)
N	Valid	152
	Missing	0
Range	Min	1
	Max	62
Mean		13.2
SD		11.5
Percentiles	25	5.1
	50	10.0
	75	16.4

Table 3.6. Descriptive statistics for age at identification.

Figure 3.11 illustrates the distribution of age at identification of hearing loss by degrees of hearing loss. Tukey B test indicated that there was no significant difference in age between the groups ($F(2,130)=0.046$, $p=0.955$).

Figure 3.12 and table 3.7 compare the age at identification of hearing loss for babies from WBN and NICU. Median age at identification for WBN babies was 7.0, and 13.0 weeks for NICU babies. As expected, independent t-test revealed a significant difference in the age at identification between babies from the WBN and NICU ($t=2.638$, $df=148$, $p=0.009$).

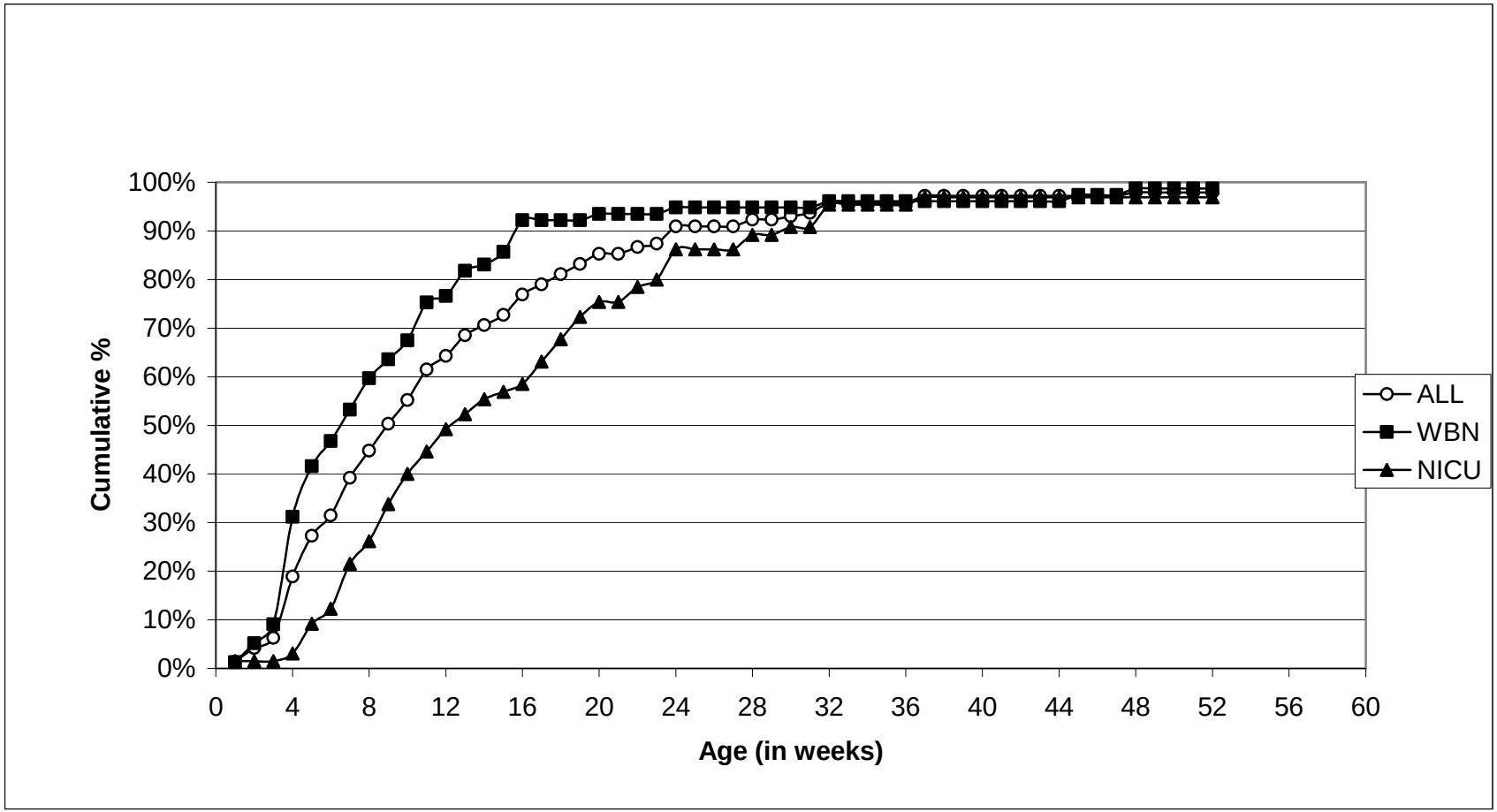

Figure 3.10. Cumulative percentage for chronological ages (in weeks) by which hearing loss has been identified.

		Age at first identification of HL (in weeks)	
		WBN	NICU
N	Valid	83	68
	Missing	0	0
Range	Min	1	1
	Max	62	56
Mean		11.7	16.0
SD		3.0	10.7
Percentiles	25	4.0	8.3
	50	7.0	13.0
	75	13.0	21.0

Table 3.7. Descriptive statistics for age at identification of hearing loss (separately for WBN and NICU population).

Figures 3.13 and 3.14 show age of identification by degree of hearing loss for WBN and NICU babies respectively. There was no statistically significant difference in the age at which hearing loss was identified for different degrees of hearing loss for both WBN babies ($F(2,80)=2.076$, $p=0.132$) and NICU babies ($F(2,64)=1.601$, $p=0.210$).

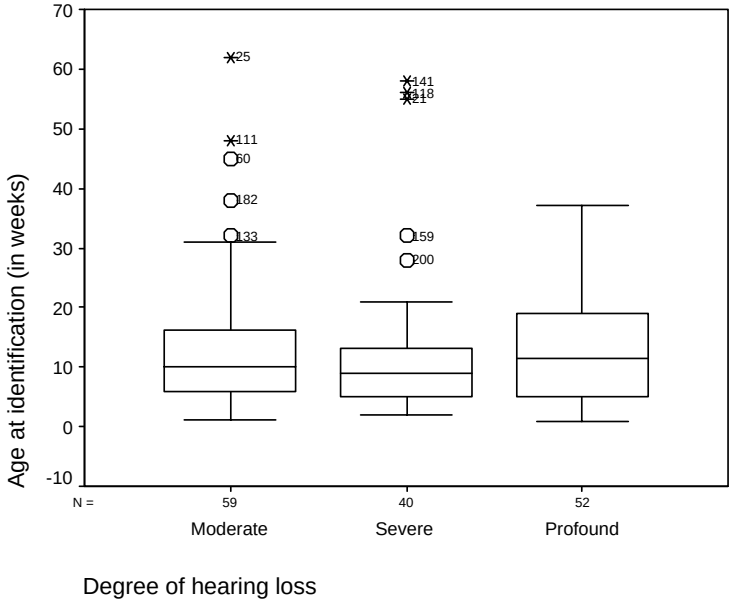

Figure 3.11. Age at identification of hearing loss by degree.

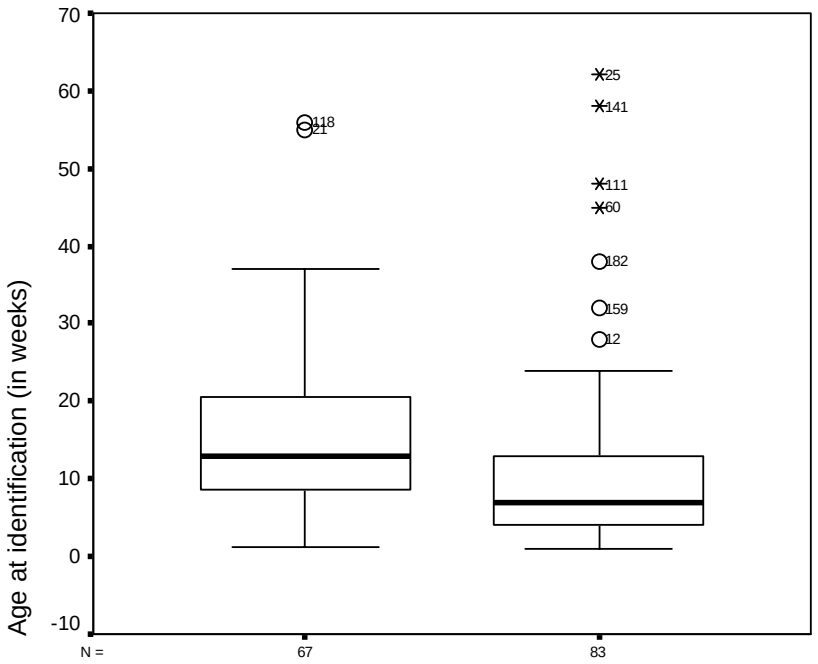

Figure 3.12. Age at identification of hearing loss by nursery: NICU (N=67) and WBN (N=83).

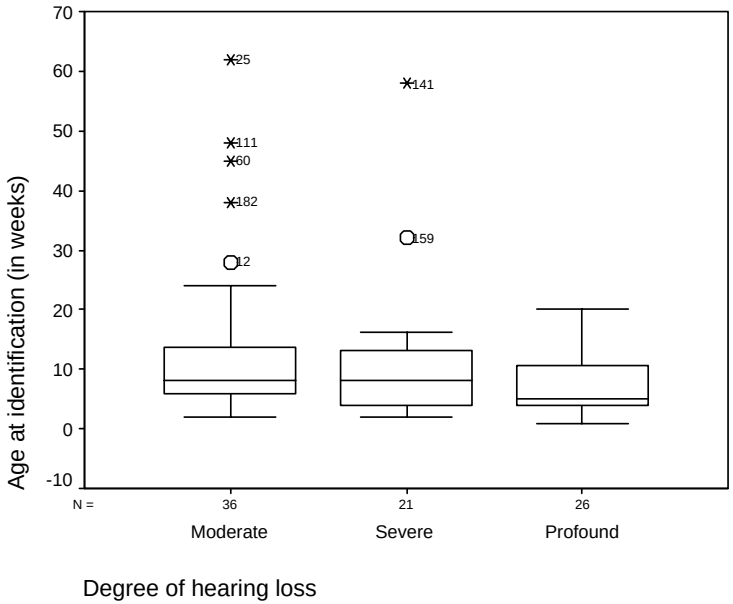

Figure 3.13. Age at identification of hearing loss by degree (WBN population).

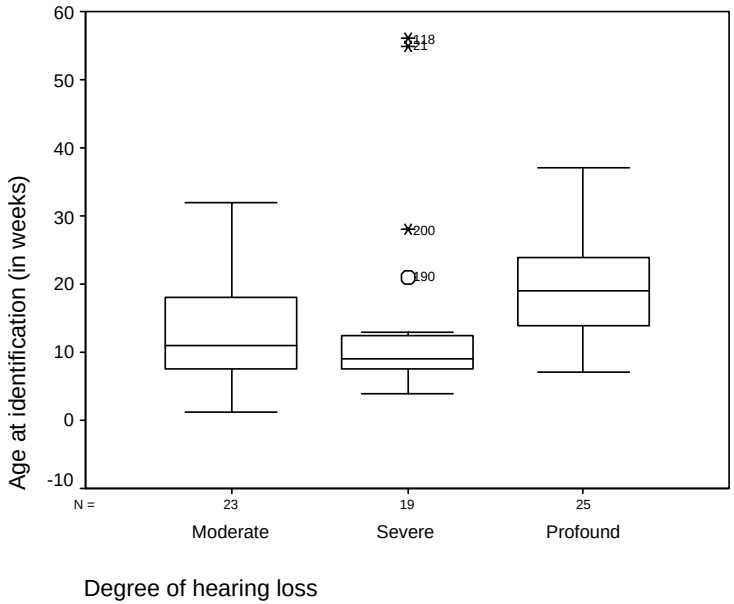

Figure 3.14. Age at identification of hearing loss by degree (NICU population)

Tables 3.8 and 3.9 provide summary reasons for the delay of hearing loss identification for cases that fall out of the 1.5 x IQR from 3rd quartile.

Case	Age at identification of hearing loss	Explanation for the delay
12	28 weeks	Diagnosis was delayed until behavioural thresholds could be obtained
25	80 weeks	Diagnostic difficulties (conductive hearing loss)
60	45 weeks	Diagnosis was delayed until behavioural thresholds could be obtained
111	48 weeks	Diagnostic difficulties (conductive hearing loss); diagnosis was delayed until behavioural thresholds could be obtained
141	58 weeks	Missed the appointments (family difficulties)
159	32 weeks	Non-resident
182	38 weeks	*A*dministrative error

Table 3.8. Details of reasons for delay of identification of hearing loss (WBN population)

Case	Age at identification of hearing loss	Explanation for the delay
21	55 weeks	Severe respiratory distress; prolonged stay in NICU; further delay due to administrative oversight
118	56 weeks	Severe dysmorphic features; delayed on parental request
190	21 weeks	Severe developmental and neurological problems; delayed on parental request
200	28 weeks	Very premature (25/40), prolonged stay in NICU

Table 3.9. Details of reasons for delay of identification of hearing loss (NICU population)

3.3.1.4 Age at hearing aid fitting

Sixty nine per cent of the first phase babies identified with bilateral hearing loss who were fitted with hearing aids received their amplification by 6 months of age. Figure 3.15 shows the cumulative distributions. It is apparent from the graph that 80% of WBN babies had hearing aid fitted by 6 months of age. The implications of very early fitting and management of digital hearing aids requires particular skills, knowledge and understanding; systems for ensuring quality of this aspect of provision need to be in place otherwise much of the potential benefit of newborn hearing screening will not be realised.

Table 3.10 provides the descriptive statistics for the age at hearing aid fitting. The median age at hearing aid fitting was 16.0 weeks. Median delay from identification of hearing loss to hearing aid fitting was 4.9 weeks.

		Age at HA fitting (in weeks))	Time between identification of hearing loss and hearing aid fitting (in weeks)
N	Valid	118	
	Missing	34	
Range	Min	3	0
	Max	92	76
Mean		13.2	97
SD		17.0	12.7
Percentiles	25	9.8	2.0
	50	16.0	4.9
	75	30.0	12.0

Table 3.10. Descriptive statistics for age at hearing aid fitting

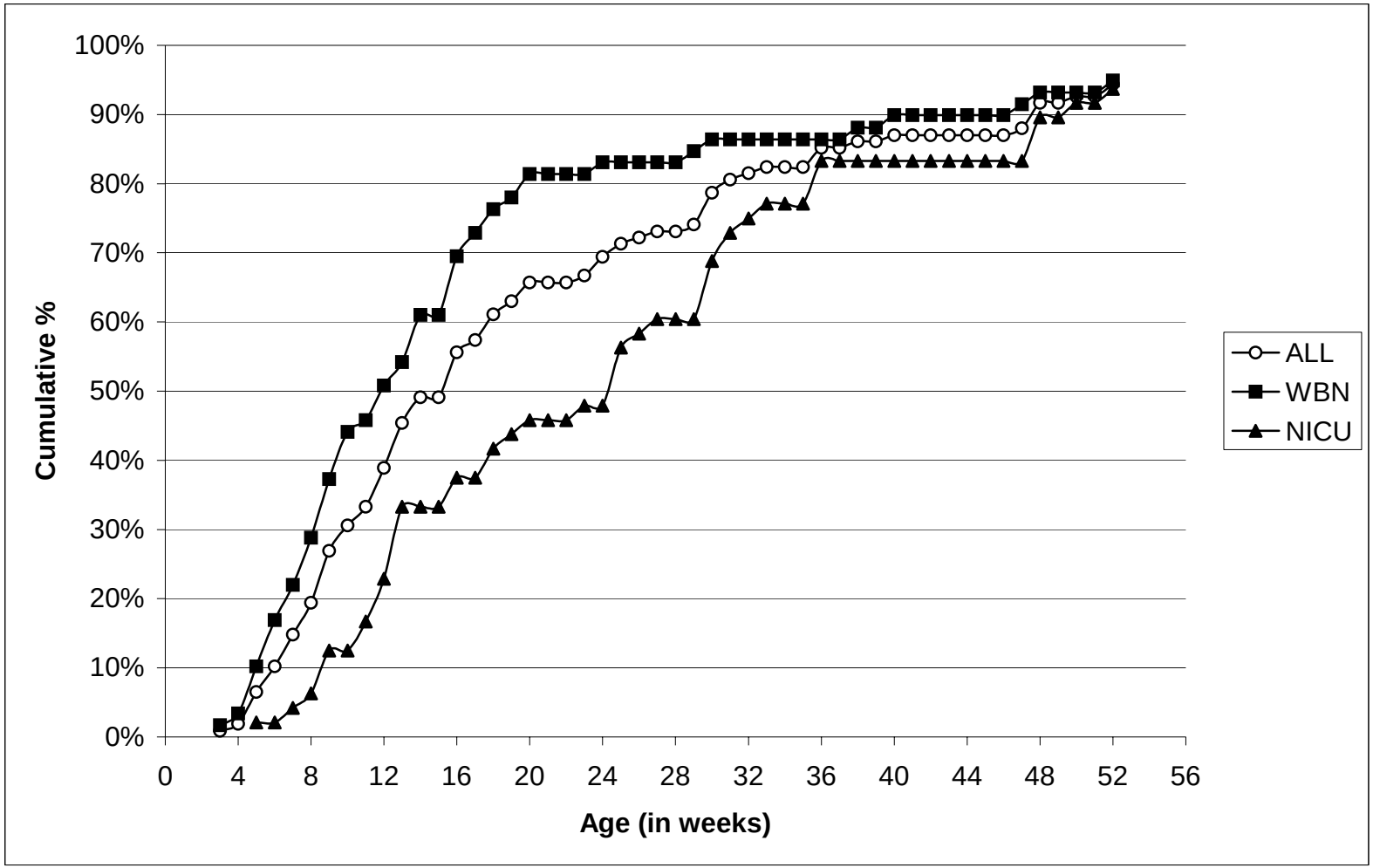

Figure 3.15. Cumulative percentage for chronological age at hearing aid fitting.

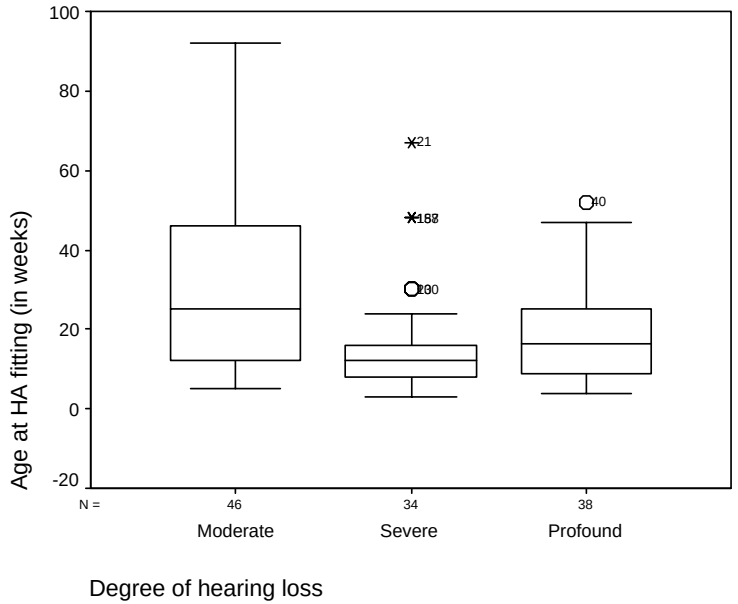

Figure 3.16. Age at hearing aid fitting by degree of hearing loss.

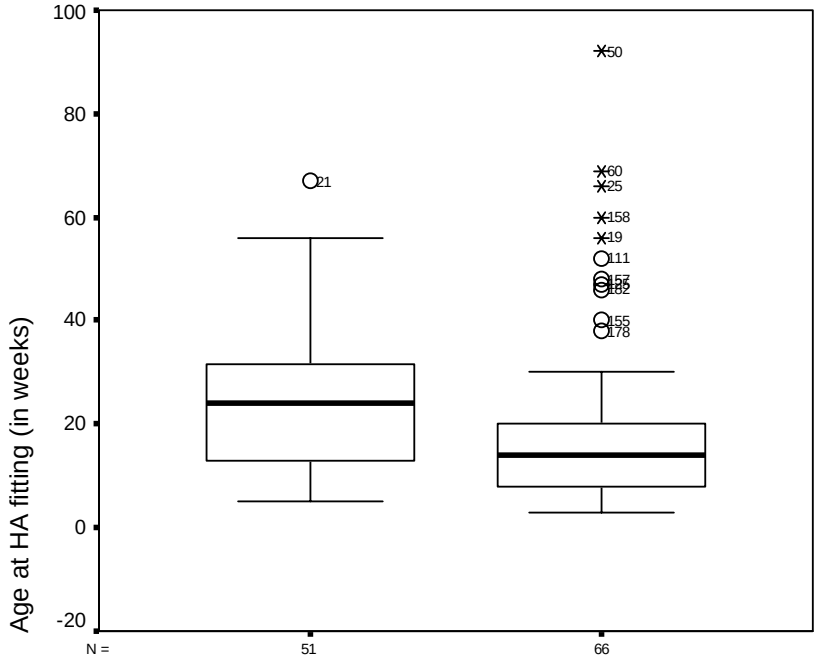

Figure 3.17. Age at which hearing aids were fitted by nursery: NICU (N=51) and WBN (N=66).

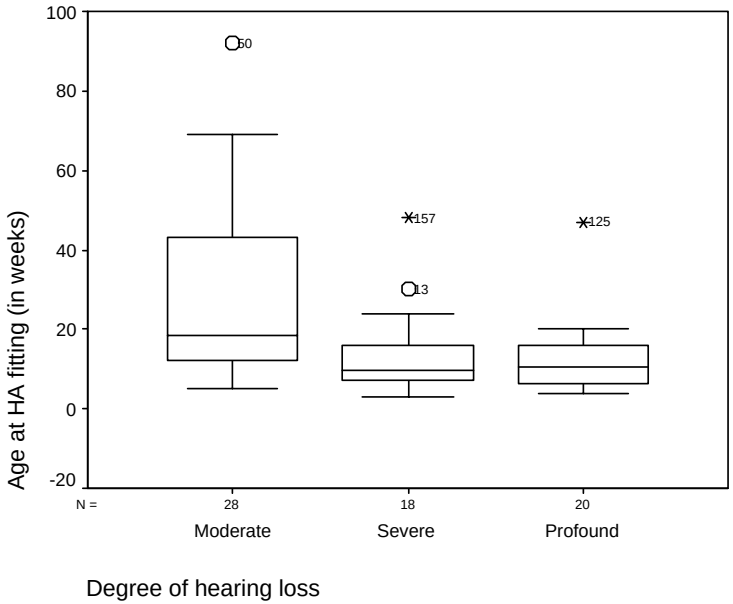

Figure 3.18. Age at which hearing aids were fitted by degree (WBN population).

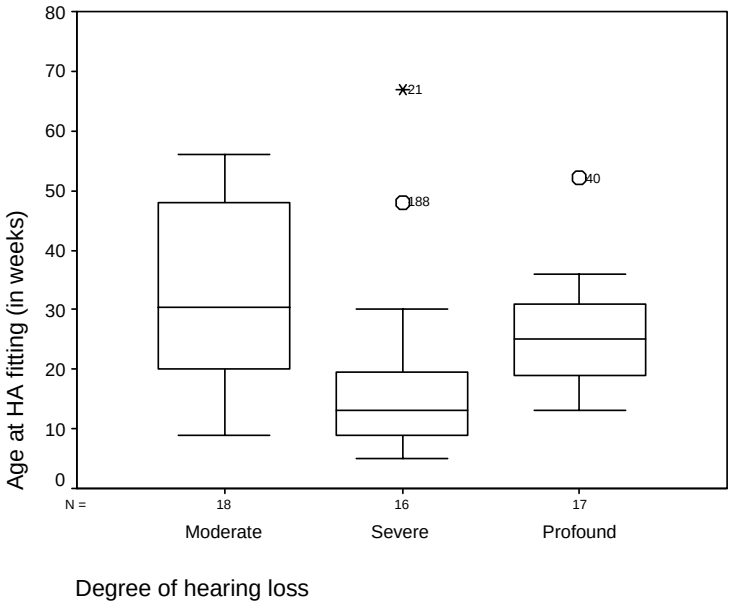

Figure 3.19. Age at which hearing aids were fitted by degree (NICU population).

Figure 3.16 shows the distribution of age of hearing aid fitting by degree of hearing loss. Tukey B test indicated that there was *a* significant difference in age between the groups ($F(2,130)=8.667$, $p<0.001$). Infants with moderate hearing loss fall into a separate subset for $\alpha=.05$.

Figure 3.17 and table 3.11 show data for age of hearing aid fitting for WBN and NICU separately. Independent t-test showed no significant difference in the age at hearing aid fitting in babies from WBN and NICU ($t=1.868$, $df=115$, $p=0.064$). Neither was there any difference in the delay from the identification of hearing loss and hearing aid fitting ($t=0.455$, $df=115$, $p=0.650$).

		Age at HA fitting (in weeks)	
		WBN	NICU
N	Valid	66	51
	Missing	17	17
Range	Min	3	5
	Max	92	67
Mean		19.8	25.7
SD		18.3	14.8
Percentiles	25	8.0	13.0
	50	14.0	24.0
	75	21.0	32.0

Table 3.11. Descriptive statistics for hearing aid fitting (WBN and NICU separately).

Figures 3.18 and 3.19 show age of identification by degree of hearing loss for WBN babies and NICU babies respectively. The Tukey B test showed a significant difference between the WBN groups ($F(2,63)=6.555$, $p=0.003$), with moderate hearing loss babies in a separate subset for $\alpha=.05$. Similarly, the Tukey B test indicated that there were significant differences between the NICU groups ($F(2,48)=3.798$, $p=0.029$); in this case, the severe hearing loss babies fall into a separate subset for $\alpha=.05$ from babies with moderate and profound hearing loss, with the severe group being fitted earlier.

Tables 3.12 and 3.13 detail the explanations for the delay of fitting hearing aids for cases that emerged as outliers and extremes both for WBN and NICU infants.

Case	**Age at hearing aid fitting**	**Explanation for the delay**
13	30 weeks	Delay due to parental request
0	92 weeks	Moderate HL; parents were not convinced about HL and postponed all habilitation for months
125	47 weeks	Home language BSL; parents were not keen on early amplification
157	48 weeks	Delay due to parental request

Table 3.12. Details of reasons for delay of the hearing aid fitting (WBN).

Case	**Age at hearing aid fitting**	**Explanation for the delay**
21	67	Delay due to parental request
40	52	Multiple developmental problems
188	48	Multiple developmental problems

Table 3.13. Details of reasons for delay of the hearing aid fitting (NICU).

In 15 cases (9.9% of the cohort) the decision was made not to fit hearing aids at the present time: 7 moderate, 2 severe and 6 profound hearing loss. For all these cases the decision was made on the basis of parental choice.

For comparison purposes, Figure 3.20 presents the cumulative distributions for the three outcome events used in these analyses: age at first follow-up appointment, age at identification of hearing loss, and age of hearing aid fitting.

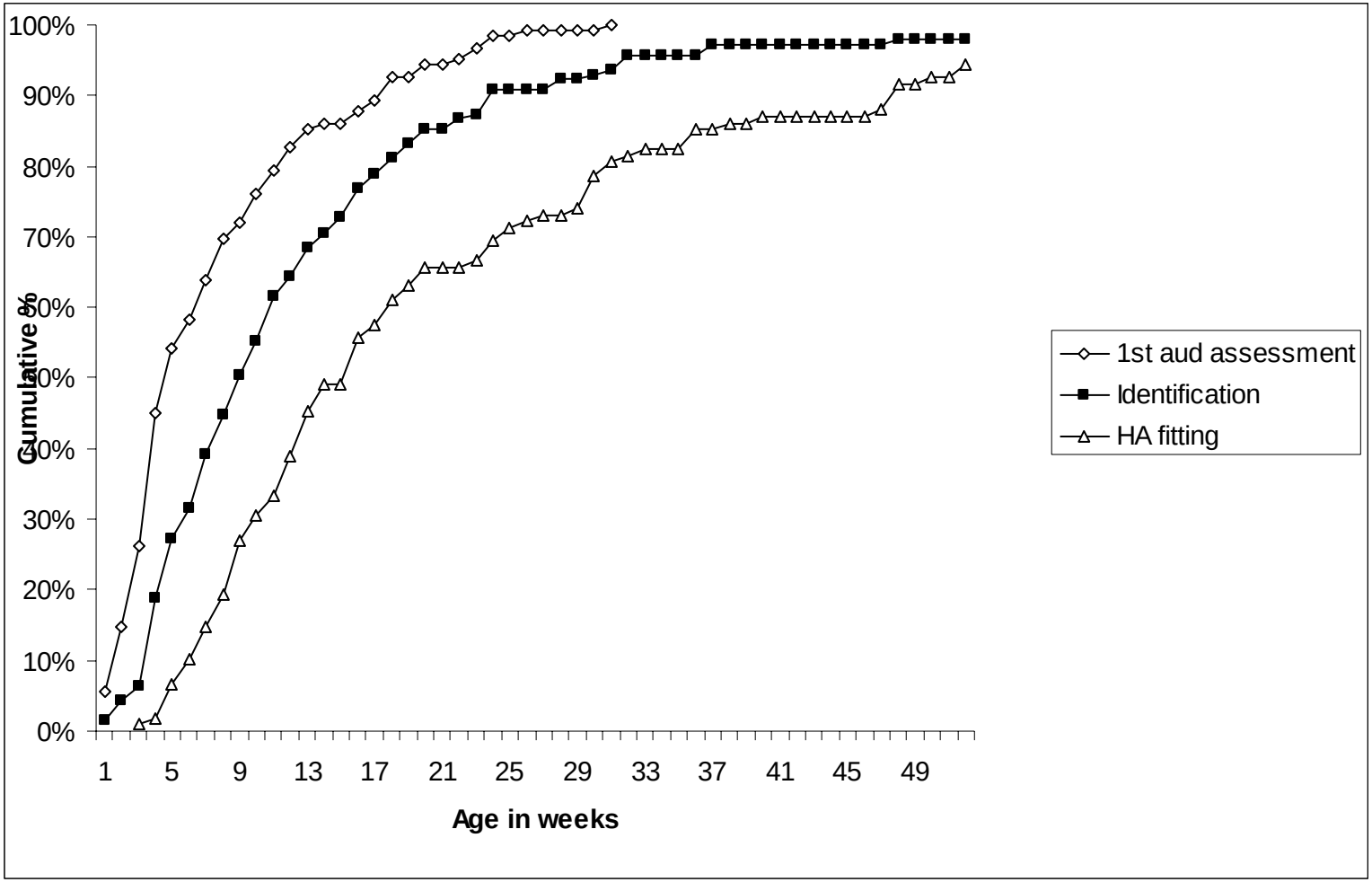

Figure 3.20. Summary of cumulative percentage for chronological age at the main events.

3.3.2 Unilateral hearing loss cases

The number of babies identified with permanent unilateral hearing loss with hearing threshold ≥ 40 dB HL in one ear and <40 dB HL in the other ear (based on the average threshold at 0.5, 1, 2 and 4 kHz) was 93 which represents 35.5% of all cases (bilateral and unilateral).

3.3.2.1 Profile of cases of permanent unilateral hearing loss

As with bilateral hearing loss, unilateral hearing loss was more frequent in male (58%) than in females (42%).

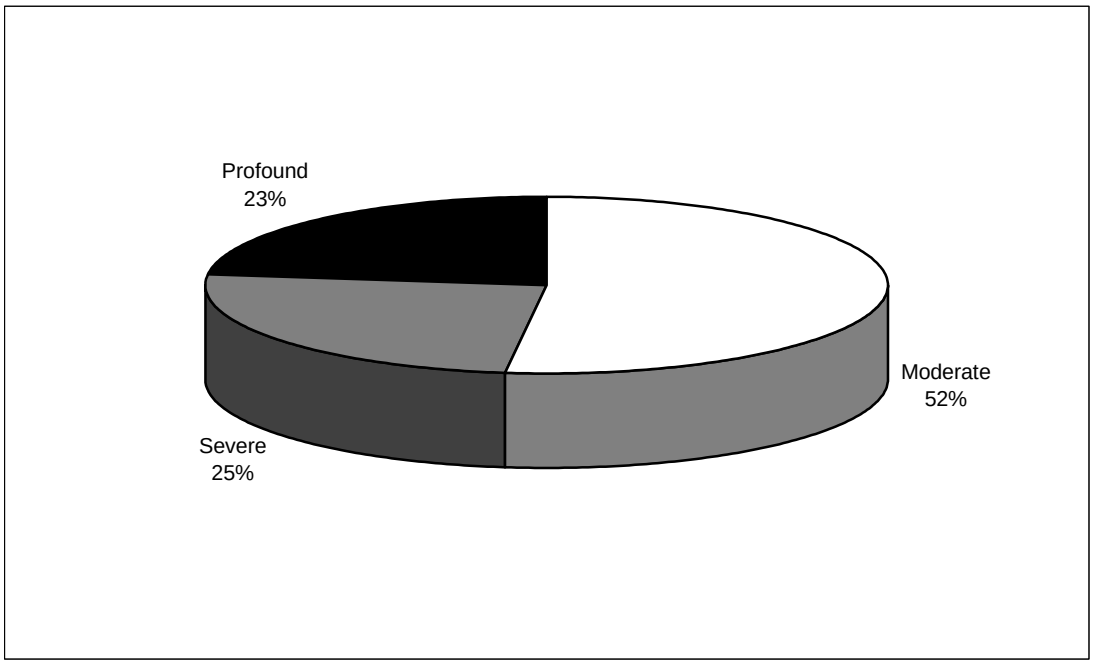

Figure 3.21. Distribution of cases with permanent unilateral hearing loss by degree.

More than half of all unilateral hearing loss cases were moderate (Figure 3.21). This 2:1:1 ratio is similar to that expected and usually found for bilateral hearing loss. As expected, permanent unilateral hearing loss was predominantly sensorineural, in 71% of cases, but much less than bilateral. Permanent conductive hearing loss is more common in unilateral cases than in bilateral cases (Fig 3.22).

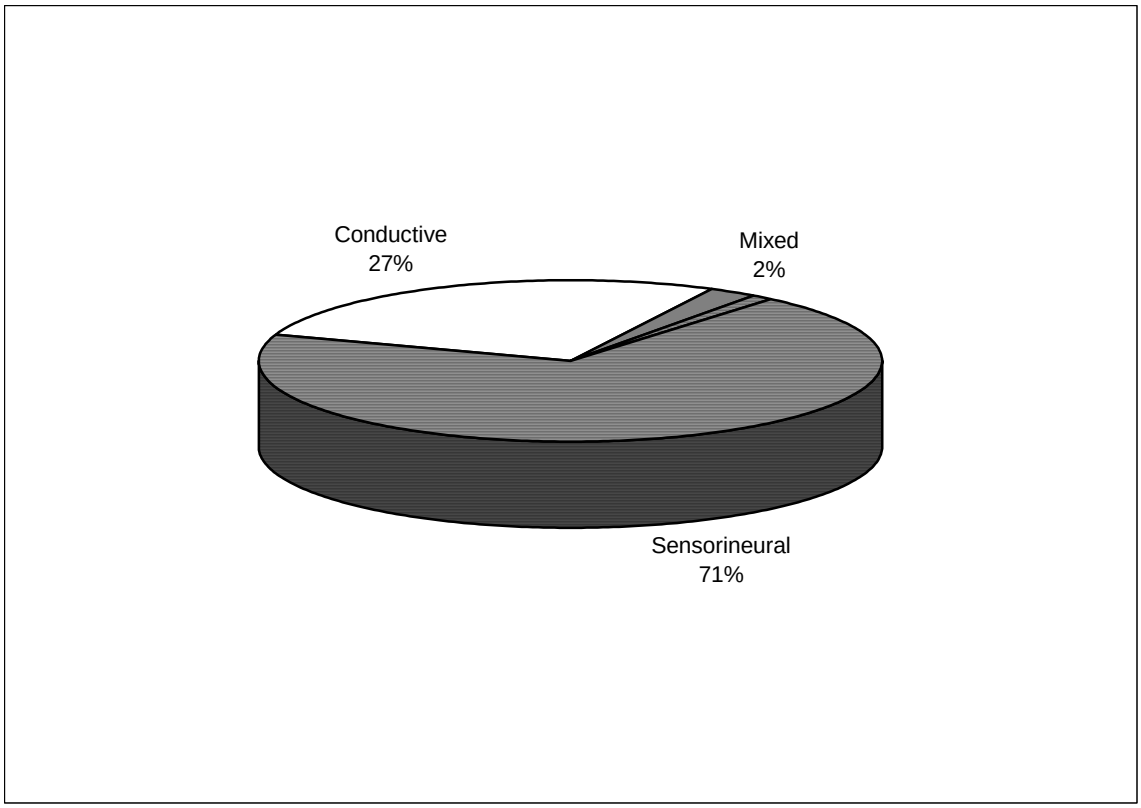

Figure 3.22. Distribution of the cases with permanent unilateral hearing loss by type.

50.5% (N=47) had at least one risk factor. Figure 3.23 shows that the risk factor pattern was very different from permanent bilateral hearing loss: 64% (N=30) of all unilateral cases who had one or more risk indicators, presented a craniofacial abnormality (in some cases combined with other risk factors). Just over a third (N=17) came from NICU population. There was no association between the presence of a risk-factor and degree of hearing loss (NICU: χ^2=0.707, df=2, p=0.702, FH: χ^2=3.228, df=2, p=0.199, CFA: χ^2=4.059, df=2, p=0.131).

Additional conditions were present in 10.8% (N=10). Table 3.14 details the conditions in the order of frequency. Additionally, 19.4% (N=18) babies presented with auricular malformations of various degree. See table 3.15 for details. Note that 12 babies were identified via the newborn screen as having a unilateral hearing loss who also had unilateral meatal atresia. This prompted the evaluation team to alert the implementation team to the issue, since there is no point (and arguably potential harm done) in screening a baby for whom it is certain that the outcome will be refer. This message has been re-emphasised to teams such that unilateral meatal atresia would trigger an automatic referral without a screen.

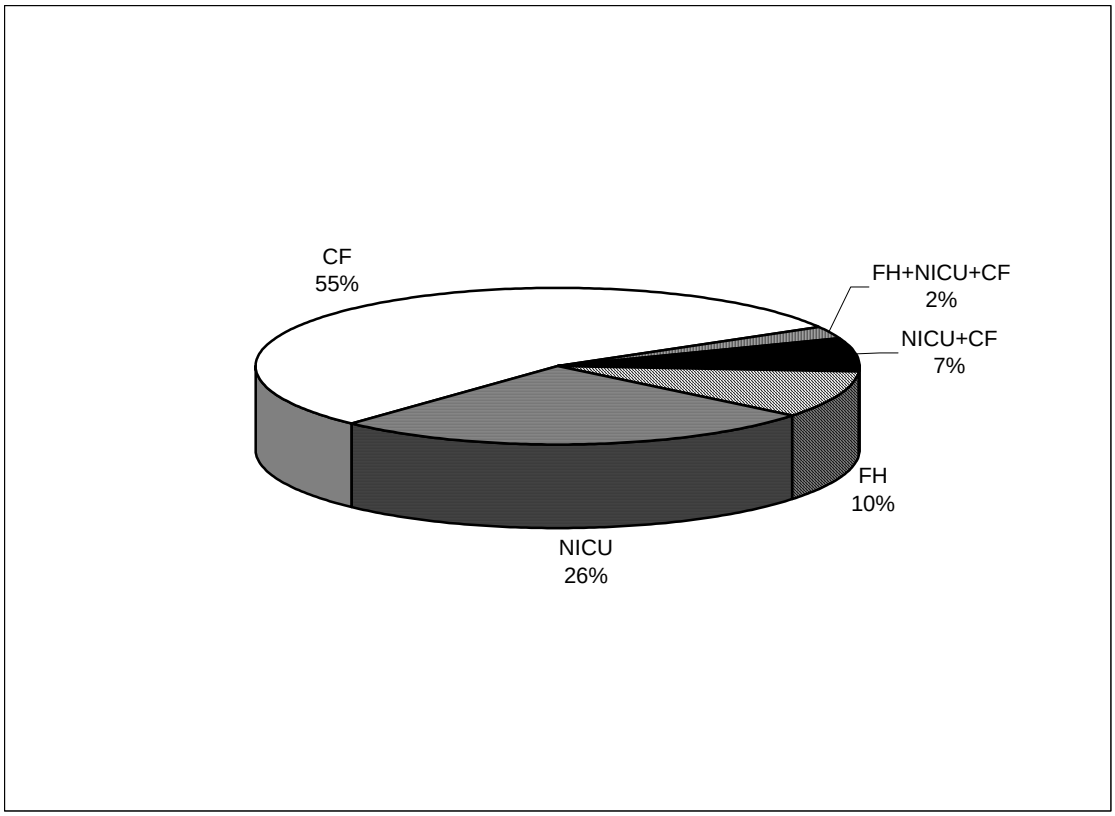

Figure 3.23. Distribution of risk factors in 47 high-risk babies identified with permanent unilateral hearing loss.

Condition	N
Lip and or palate cleft	2
Multiple unspecified problems	2
Di George Syndrome	1
Down Syndrome	1
Edwards' Syndrome	1
Lacrimo-auriculo-dento-digital syndrome	1
Visual problems	1
Motor delay	1

Table 3.14. Additional conditions found in babies identified with permanent unilateral hearing loss.

Condition	N
Atresia	12
No pinna	3
Microtia	2
Pre-auricular fistula	1

Table 3.15 Auricular malformations found in babies identified with permanent unilateral hearing loss.

3.3.2.2 Age at identification of unilateral hearing loss

Table 3.16 provides data on distribution of age at identification of unilateral hearing loss. The median age was 6.1 weeks.

		Age at first identification of HL (in weeks)
N	Valid	93
	Missing	0
Range	Min	0
	Max	49
Mean		9.0
SD		8.8
Percentiles	25	4.4
	50	6.1
	75	9.0

Table 3.16. Descriptive statistics of age at identification of unilateral hearing loss.

Independent t-test indicated significant difference in the age of identifying hearing loss in bilateral and unilateral hearing loss (t=2.970, df=240, p=0.003). Unilateral hearing loss was identified at a significantly earlier age (see Figure 3.24). Tukey B test showed that there was no significant difference in age between degrees of hearing loss (F(2,89)=1.305, p=0.260) (Figure 3.25).

Presence of risk factors played a significant role in the age at which unilateral hearing loss was identified. Independent t-test revealed significant difference for babies with and without craniofacial abnormalities (t=–3.043, df=90, p=0.003) (Figure 3.28) and babies from WBN and NICU (t=3.441, df=90, p=0.001) (Figure 3.29).

No standards currently exist for the management of unilateral hearing loss although it is worth noting that 53.1% of cases with unilateral hearing loss were not referred to Education services.

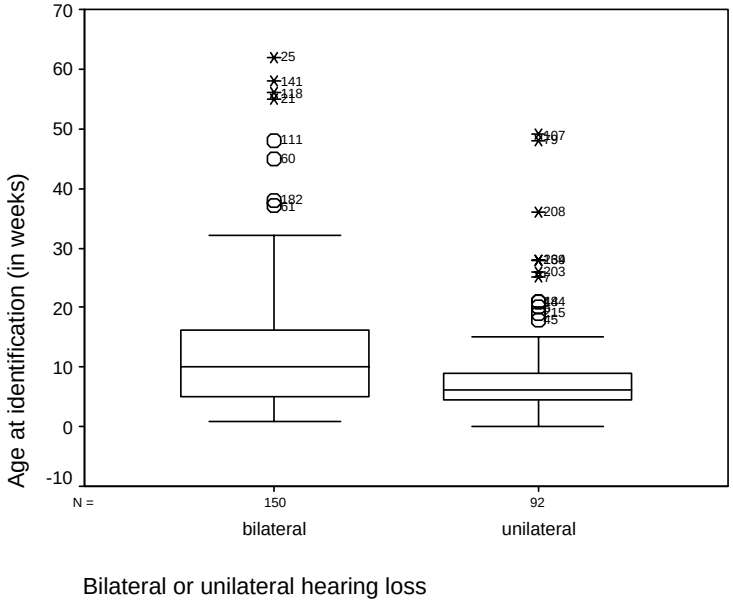

Figure 3.24. Age at identification of hearing loss: bilateral versus unilateral.

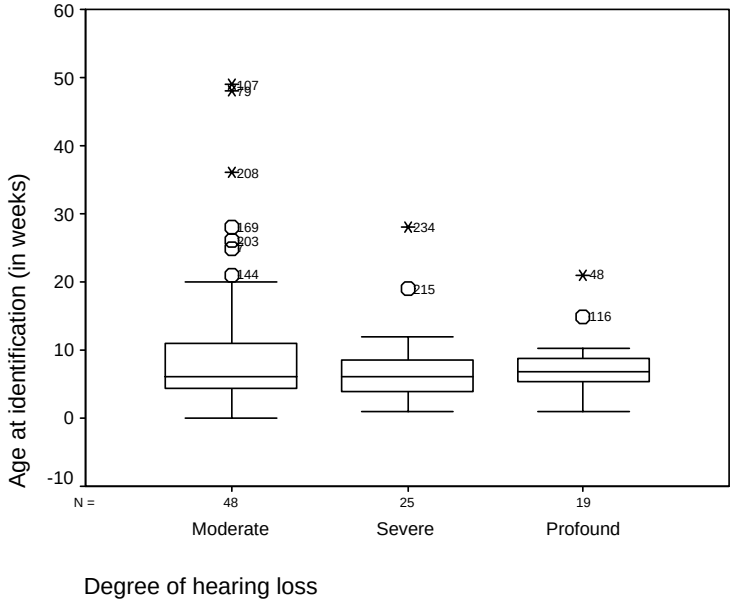

Figure 3.25. Age at identification of unilateral hearing loss by degree.

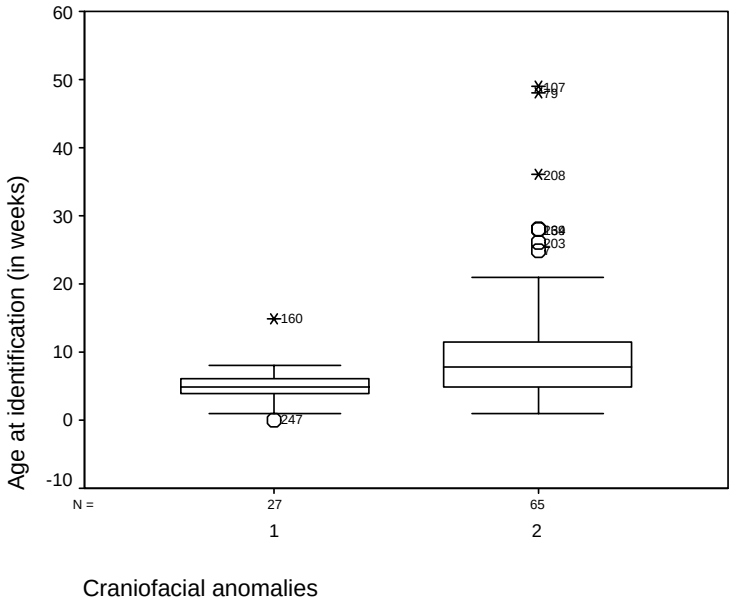

Figure 3.28. Age at identification of unilateral hearing loss in babies with (1) and without (2) craniofacial abnormalities .

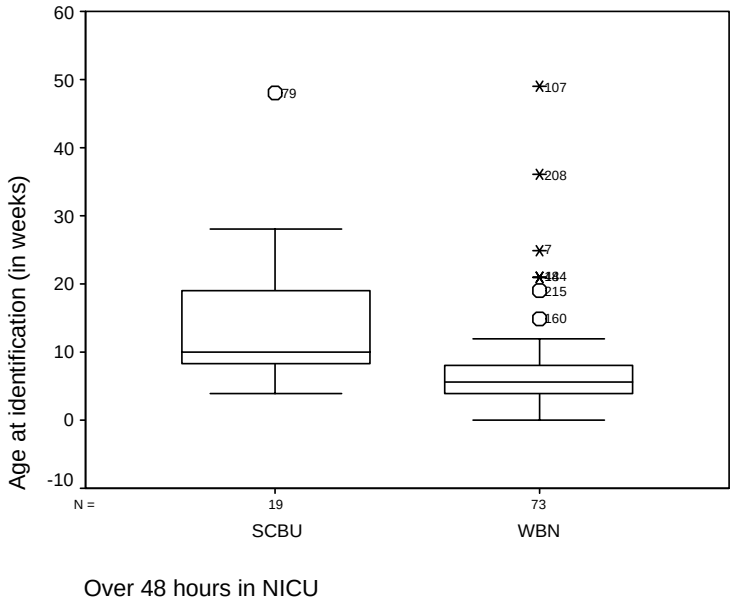

Figure 3.29. Age at identification of unilateral hearing loss by nursery.

3.4 Discussion

Median age at the first audiological follow-up for babies subsequently identified as having a bilateral hearing loss was five weeks of chronological age. There is no evidence that this is any different for all screen referrals (i.e. including false positives), but it seems unlikely to be different since the hearing status of babies is unknown at the point of referral. Babies from the well-baby nursery tended to be followed up earlier. Babies from NICU are often too ill and may have serious health problems which take immediate priority over possible hearing problems. Babies from NICU who will eventually be identified with profound hearing loss are followed up significantly later than babies who have lesser degrees of hearing loss. This may well be because these babies are most severely ill.

The reasons for exceptional delays to follow-up were different in WBN babies and NICU babies. In NICU babies these were related to baby's health and long hospitalisation. In WBN babies, the reasons were service-related, mainly due to long waiting times for audiological assessments and administrative slip-ups. Some families could not or would not attend appointments for a range of reasons (e.g. bereavement in the family).

The newborn screening programme first phase sites identified and informed the evaluation team of 169 babies with bilateral permanent hearing loss between their starting date (staggered from December 2001 to September 2002) and 1st January 2004. The yield for the period was 1.0 per thousand babies screened, which is close to expected figures based on prevalence data (e.g. 1.12 per 1000 from Fortnum and Davis 1997).

The distribution of degree of hearing loss is somewhat different from previous studies. Fortnum and Davis (1997) found that moderate hearing loss was more prevalent than severe and profound hearing loss put together. In the present cohort profound hearing loss was more common than expected. It should be noted that the category of hearing loss for any particular child was determined in the early stages of the assessment/diagnostic procedures, and that these may change as more accurate information emerges. On the other hand, if guidelines for assessment from the implementation team are followed properly, then accurate assessment should be possible before six months of age in most cases; furthermore, in all cases, the clinicians maintained the reported degree of hearing loss when they fitted the hearing aids, suggesting a high degree of certainty about degree of loss. Thus, the reasons for the somewhat different distribution of degrees of hearing loss in the current cohort are unclear; assessment difficulties for moderate hearing loss may play a part, but an increase in the proportion of profound hearing losses (for example in the increasing population of NICU babies) is also a possibility. Such questions should be easily resolved as the newborn screening programme rolls out and the number of true cases increases.

Not surprisingly, sensorineural hearing loss was the most common type of hearing loss. About 10% of the cohort of true cases were identified with auditory neuropathy; this is in line with other studies (Rance *et al* 1999, Mehl *et al* 2002, Sininger 2002). All babies identified with auditory neuropathy were from NICU population due to the protocol adopted for NICU babies;

any cases from the WBN population (thought to be a much lower prevalence) will have to emerge over time. Auditory neuropathy is likely to be a heterogeneous disorder with different possible sites of lesion and a variety of aetiological factors. Patients with auditory neuropathy present behavioural hearing thresholds ranging from within normal limits to profound hearing loss; typically they have poorer speech discrimination than would be predicted from the behavioural audiogram, and show a poor relationship between physiological measures of hearing sensitivity and hearing function. In patients with auditory neuropathy, the benefit from amplification remains controversial. The heterogeneity of the condition leads to pronounced diagnostic and management challenges. Guidelines for the assessment and management of auditory neuropathy have now been developed by a working group initiated by the Implementation team.

The proportion of high-risk babies of all permanent bilateral hearing loss cases is in line with Davis & Wood (1992) and Fortnum & Davis (1997): around 60% of all congenitally-impaired children had one or more risk factors. However, there may be a shift towards the increase in the proportion of NICU babies, with 44% in this cohort as opposed to 29% found in both above-mentioned studies. The proportion with a family history of permanent childhood deafness was just 19% which may reflect the complexity of history taking at such an early stage. Craniofacial abnormalities were present in 9%, which is between the 4% reported by Davis & Wood (1992) and the 12% reported by Fortnum & Davis (1997). The high proportion of cases with risk factors led, in the early 1990s, to the widespread introduction of 'at risk' newborn screening in which attempts were made to screen all those babies (just under 10% of the birth cohort) with risk factors. However, in practice, due to the difficulty experienced by maternity services in reliably identifying a family history of permanent childhood hearing loss, the proportion of the target population identified by at risk screening was rarely above 40 per cent. Therefore it is unsafe to assume that all these 91 babies with risk indicators identified with hearing loss would have been found through targeted newborn hearing screen.

Additional conditions were found in 36% of babies identified with bilateral hearing loss which is slightly higher than in previous reports on congenital hearing loss (Fortnum & Davis 1997). That is even more surprising considering that the reported conditions were picked up at a very early infancy and thus were likely to be the more serious ones. Whether this shift is a sign of improvement in aetiological diagnostics or a real increase in comorbidity, is too early to tell. The most common additional condition was congenital heart defect, followed by cleft lip and palate (either in combination or in an isolated form), unspecified multiple problems, visual problems, cerebral palsy and developmental delay.

The median age of identification for those screened neonatally has been shown in research studies to be of the order of two months (Watkin 1996, McClelland *et al* 1999, Dalzell *et al* 2000). The findings from this evaluation of the first phase of the national programme are in line with that: the median age of identification was 10 weeks. Though it only applies to the limited cohort and does not take into consideration potential false negatives or babies who missed the screen, compared to the situation before NHSP was introduced when the median age at identification of bilateral hearing loss was 18 months (Fortnum & Davis 1997), this is a massive improvement.

Importantly, given that intervention has been argued and shown to be beneficial for degrees of hearing loss down to at least moderate levels (Davis *et al* 1997), age at identification was independent of the degree of hearing loss. This was not the case with the previous national 8-month hearing screen, the sensitivity of which was severity-dependent.

Hearing-impaired babies from the healthy baby population were identified significantly earlier than those from NICU. Where there were delays in identification these were often due to assessment and service difficulties in the well-baby population, while in NICU babies the delay was more likely to be associated with the health of the baby.

Median age at hearing aid fitting has also improved considerably to a median age of four months, as opposed to 26 months before newborn hearing screening (Fortnum & Davis 1997). Of course, a few screen false negatives may yet be identified, and not all cases identified had hearing aids fitted, and this median age may therefore change, but it is unlikely to be by significant amount.

For the whole cohort, age at hearing aid fitting is the first event where degree of hearing loss plays a role: infants with moderate hearing loss are fitted with hearing aids significantly later than those with severe or profound hearing loss; possible reasons are parental reticence (and the exercise of parental choice) in the light of some auditory responsiveness; assessment uncertainty as the distinction between mild and moderate hearing loss in a particular baby becomes less clearcut. For NICU babies, those with profound (as well as moderate) hearing loss are fitted significantly later than those with severe hearing loss, presumably due to more involvement of other difficulties.

The newborn screening programme first phase sites identified and informed the evaluation team of 93 babies with unilateral permanent hearing loss between their starting date and 1st January 2004. There are few if any reliable studies on the prevalence of congenital unilateral permanent hearing loss in preschool children, and this is the first good evidence that the condition exists in significant numbers at or very soon after birth.

The profile of risk factors and additional conditions is very different in unilateral hearing loss cases. First, not unexpectedly, the predominant risk indicator is craniofacial anomaly present in 32% of all babies with unilateral hearing loss. Only 11% of all unilateral hearing loss cases presented with an additional condition. It is impossible to distinguish at this point whether this reflects the lack of effort invested into the diagnostic process or real absence of additional conditions in these babies with unilateral hearing loss.

Interestingly, even though the national protocol recommends that bilateral referrals should be given the priority, unilateral hearing loss was actually identified earlier than bilateral. There are no standards or guidelines for management of babies with early identified congenital unilateral hearing loss. More than half of the babies with unilateral hearing loss were not referred to Education Services; other studies (e.g. Reeve 2003) have pointed to the lack of information and support received by parents of those with unilateral hearing loss. Unilateral hearing loss cases are not routinely provided with hearing aids, the benefits of which are uncertain in these cases, and research and eventually better service guidelines are urgently needed into the management options for congenital unilateral hearing loss.

3.5 Summary points

- Based on data from true cases, median age at first follow up after screen referral was five weeks of age. Some 64% of well babies are likely to have their first audiological follow-up by 4 weeks of age. Ninety-five per cent of cases had had the first follow-up by 11 weeks of life. Reasons for the longer delays for well babies are mainly service-related and suggest the need for improvements in aspects of paediatric audiology services.
- The median age at identification of permanent bilateral hearing loss was 10 weeks which marks a major improvement compared to 18 months of age before the implementation of newborn hearing screening. Age of identification was independent of the severity of the hearing loss.
- The median age of children who were fitted with hearing aids was 4 months which is a massive improvement compared to around 2 years of age before the implementation of newborn hearing screening. Eighty per cent of well babies were fitted with hearing aids by 6 months of age. Babies with moderate hearing loss tended to be fitted later than those with severe or profound loss, often because of parental choice. The very early fitting of hearing aids requires considerable skill and knowledge, particularly with the advent of DSP (digital signal processing) hearing aids. Systems for ensuring the quality of hearing aid fitting and management in very young infants need to be strengthened.
- Age at follow-up and age of identification were not dependent upon severity of the hearing loss.
- There were significant numbers of babies with unilateral hearing loss identified by the screen. Evidence-based guidelines for management are urgently needed.
- 54% of all cases with permanent bilateral hearing loss are from ‘at-risk’ population. 3/4 of these ‘at-risk’ babies have spent 48 hours or more in the neonatal intensive care unit. 36% of children identified with permanent bilateral hearing loss have additional conditions and/or disabilities.
- It is not appropriate to screen babies with unilateral or bilateral meatal atresia; such cases should be automatically referred.
- About 10% of the cases with bilateral hearing loss were cases of auditory neuropathy. Research into the causes, management and outcomes of auditory neuropathy is urgently needed.

4. PSYCHOLOGICAL EVALUATION OF NHSP

4.1 Introduction and summary of studies

In this chapter we report on four questionnaire-based studies, presented in five papers. These papers are prepared for publication but are still in draft form; the format of each has been modified where appropriate to suit the context of the whole NHSP evaluation report.

This aspect of the evaluation of the NHSP centred on the psychological impact of newborn hearing screening, and particularly the impact, on the emotional-well being of mothers whose babies are referred for diagnostic testing following screening. The evaluation also considered issues relating to the ways in which the screening programme is implemented, specifically comparing aspects of the hospital and community modes of delivering newborn hearing screening, aspects that have the potential to influence mothers' experience of screening.

The first two papers compare the impact on maternal anxiety of receiving different results following hospital-based newborn hearing screening. The first of these papers describes maternal anxiety in the short-term (3 weeks after the screening tests were completed) and the potentially moderating effect of knowledge on anxiety among mothers of babies who received a referral for diagnostic testing. The second paper evaluates the impact 6 months following completion of the screening test.

The third and fourth papers both compare hospital-based newborn hearing screening with alternative, community-based, models of hearing screening. The first of these compares the impact of hospital-based and community-based newborn hearing screening on anxiety in mothers whose babies had clear responses at the first stage of the newborn hearing screening programme. The second of these studies compared the impact on maternal anxiety and satisfaction of hospital-based newborn hearing screening and the community-based Infant Distraction Test (IDT) which is now being replaced by the newborn hearing screening tests. Finally we present a study comparing the job satisfaction of two groups who conduct the screening test, hospital-based dedicated screeners and community-based Health Visitor screeners.

In the remainder of this Introduction we provide a summary of each of the five studies. This is followed by each of the papers in full.

We would like to acknowledge the help of the mothers who participated in these studies.

4.1.1 Maternal anxiety following newborn hearing screening: the moderating role of knowledge

One of the main areas of concern in relation to the implementation of newborn hearing screening has been the impact of screening babies in the emotionally demanding neonatal period on the mother's psychological well-being.

4.1.1.1 Aims

- To describe the possible adverse emotional effects on mothers of newborn hearing screening, and particularly of referral for further tests within one month of the completion of the hearing screening tests.

- To describe any moderating effects of knowledge of the screening test on mothers' emotional well-being.

4.1.1.2 Design

A prospective descriptive study was conducted comparing the responses of four groups of mothers whose babies had different hearing test results.

4.1.1.3 Main Findings

- Levels of maternal state anxiety were in the normal range but there was a significant linear trend for anxiety to increase as the number of tests the baby required increased.

- Levels of maternal worry and uncertainty about the baby's hearing increased significantly as the number of tests that the baby had also increased.

- Mothers whose babies required a referral for possible bilateral hearing loss were less anxious, worried and uncertain if they understood that an unclear response was unlikely to mean that their baby had a hearing loss.

4.1.1.4 Conclusions

- Referral for diagnostic tests has a small but significant effect on mothers' emotional well-being in the first three weeks after screening.

- Ensuring good knowledge of possible reasons for referral seems to be protective against anxiety and thus suggests a potentially effective yet simple intervention to minimize the adverse emotional impact of this screening programme.

4.1.2 Evaluation of long-term maternal anxiety following newborn hearing screening

Having ascertained that there are effects of newborn hearing screening on the emotional well-being of mothers in the first three weeks after screening we wanted to ascertain whether these effects persisted in the longer term. Follow-up questionnaires were therefore sent to mothers six months following the completion of screening.

4.1.2.1 Aim

To assess the impact of the newborn hearing screening tests on mothers' emotional well-being six months following completion of the screening tests.

4.1.2.2 Design

A prospective descriptive study was conducted comparing the responses of four groups of mothers whose babies had different hearing test results.

4.1.2.3 Main finding

There were no significant differences between the groups six months following the completion of screening in maternal state anxiety, worry or certainty about the baby's hearing.

4.1.2.4 Conclusion

- The small but significant emotional distress following recall for diagnostic tests after newborn hearing screening is no longer evident at six months.

4.1.3. A comparison of anxiety between mothers of babies who had hospital-based screening and mothers of babies who had community-based newborn hearing screening

A central element of the evaluation of the implementation of newborn hearing screening was the comparison of the two different models of implementation, the hospital-based model and the community-based model. A full evaluation of the effect on mothers' emotional well-being of the receipt of different hearing test results was not possible due to a lack of data, but we were able to complete a comparison of the impact of receiving clear responses at the first stage of screening in those screened in hospital and those screened in the community.

4.1.3.1 Aim

To compare the effects on mothers' well-being of having their babies screened in hospital or community, having received a clear response at the first stage of screening.

4.1.3.2 Design

A prospective descriptive study was conducted to compare the emotional responses of the two groups of mothers.

4.1.3.3 Main findings

- Overall, there were low levels of state anxiety and worry about the baby's hearing, and high levels of certainty about the babies hearing and knowledge of the hearing screening tests.

- Although there were no differences between the groups in relation to state anxiety, certainty and knowledge, mothers of babies who had their hearing screened in the community were marginally less worried about their babies hearing.

4.1.3.4 Conclusion

- The results provide evidence to support the hypothesis that mothers of babies receiving a referral for diagnostic tests after screening experience less emotional distress if the screening is conducted in the community compared with the screening conducted in the hospital. This hypothesis awaits testing.

4.1.4 A comparison of maternal anxiety and satisfaction with newborn hearing screening following the IDT and newborn hearing screening

Despite the apparent advantages of newborn hearing screening over the IDT in terms of accuracy and age of identification, concerns about the replacement of the IDT with newborn hearing screening were expressed in that a test conducted in the neonatal period might generate greater levels of emotional distress than one conducted later in childhood. However, to our knowledge there have been no studies comparing the emotional impact of the two types of screening. This study fills this gap and compares the emotional impact of the two screening tests.

4.1.4.1 Aims

- To compare the emotional impact on mothers of referral following newborn hearing screening and the IDT.

- To compare the acceptability of the two screening tests for mothers.

4.1.4.2 Design

A prospective descriptive study was conducted to compare the two screening programmes.

4.1.4.3 Main findings

- There were no differences between mothers of babies undergoing the two tests in terms of maternal anxiety, worry and certainty about the baby's hearing.
- Those whose babies had undergone newborn hearing screening were significantly more satisfied with the test that the baby had received.
- Among those who received a satisfactory result, those whose babies had undergone newborn hearing screening had significantly more positive attitudes to the test.

4.1.4.4 Conclusions

- Newborn hearing screening does not cause more emotional distress than a test conducted some months later in infancy.
- As well as its advantages in terms of sensitivity and specificity, newborn hearing screening is associated with higher levels of satisfaction. Such satisfaction may help facilitate attendance for follow-up tests.

4.1.5. Job satisfaction in newborn hearing screeners: a comparison of hospital-based screeners and community-based Health Visitors

The health care professionals who conduct a screening test have a key role in influencing participants' experiences of that screening programme including their emotional responses to it. Under the premise that the job satisfaction of screeners would affect the way they conducted the screening, this study compared the job satisfaction of hospital-based dedicated screeners with that of community-based Health Visitor screeners.

4.1.5.1 Aim

To describe and compare levels of job satisfaction in hospital and community-based screeners and to identify the factors associated with it.

4.1.5.2 Design

A descriptive study comparing job satisfaction of the two types of screener.

4.1.5.3 Main findings

- Both hospital-based dedicated screeners and community-based Health Visitor screeners expressed high levels of job satisfaction, although overall, hospital–based dedicated screeners expressed higher levels of job satisfaction.

- For both groups, satisfaction was predicted by the extent to which people felt listened to at work, their job met career aspirations, and they were satisfied with their salaries. Among hospital screeners, feeling part of the team at work was also predictive of satisfaction.

- Hospital-based dedicated screeners expressed considerable dissatisfaction with their salaries.

4.1.5.4 Conclusions

- Hospital-based dedicated screeners expressed more job satisfaction than community-based Health Visitor screeners
- Although the two groups differed in overall levels of job satisfaction, their satisfaction was influenced by similar factors. These factors need to be taken into account in continuing the effective implementation of newborn hearing screening.
- However, compared with community-based Health Visitor screeners, hospital-based dedicated screeners had relatively little experience of working in a healthcare setting giving a shorter time scale over which dissatisfaction might develop. Evaluation of the long term satisfaction of these screeners is needed.

4.2 Maternal anxiety following newborn hearing screening: the moderating role of knowledge

Abstract

Objectives: To describe the impact upon maternal anxiety of newborn hearing screening and examine the possible moderating role of knowledge.

Methods: Questionnaires assessing maternal state anxiety, worry and certainty about the baby's hearing, and knowledge about screening, were sent to four groups of mothers three weeks after screening: Group 1: mothers whose babies had clear responses on a first or second screening test (n=103); Group 2: mothers whose babies had clear responses on the final screening test (n=81); Group 3: mothers whose babies did not have clear responses in one ear at the final screening test and were referred for audiological assessment (n=105); Group 4: mothers whose babies did not have clear responses in either ear at the final hearing test and were referred for audiological assessment (n=55).

Results: Although mean anxiety levels were in the normal range, there was a significant trend for anxiety to raise as testing increased ($F(3,327)=4.280$, $p<0.05$). Worry ($F(3,34)=25.282$, $p<0.001$) and uncertainty ($F(3,347)=9.738$, $p=0.001$) were significantly raised as the number of tests increased. Although total knowledge did not have a significant moderating effect on anxiety ($R^2=0.016$, $p=0.096$), there was a significant interaction between mothers' group and one knowledge item, understanding that the receipt of no clear responses was unlikely to mean that the baby had a hearing loss: group 4 mothers who understood this had lower anxiety ($F(3,323)=4.791$, $p=0.01$) and lower worry ($F(3,332)=3.565$, $p<0.01$) compared with those who did not.

Discussion: These findings suggest that knowledge about the meaning of being recalled following screening may avert some of the adverse psychological effects of being recalled.

4.2.1 Background

4.2.1.1 Psychological Effects of Screening

Screening programmes to allow early diagnosis and treatment of disease have been widely implemented for a range of conditions. Along with benefits, there are costs to any screening programme, including the psychological costs of anxiety particularly among those who screen positive and require further tests (Meystre-Agustoni *et al* 2001, Parker *et al* 2002). A systematic review (Shaw *et al* 1999) found that receiving a positive test result was associated with depression and anxiety in the short term (within a month of receiving the results), although these did not persist in the longer term. This review also found that interventions could be implemented to reduce the adverse psychological consequences of receiving a positive test results.

4.2.1.2 Knowledge as a moderator of anxiety

An intervention that might be implemented to reduce anxiety is that of the provision of knowledge about the screening test. Among mothers of babies who were recalled following newborn hearing screening, there was a non-significant trend for the women to describe themselves as being less anxious if they had understood the meaning of possible results on the

hearing screen (Clemens *et al* 2000). However, studies that have tried to increase knowledge of screening tests have yielded mixed evidence as to its effects on anxiety. In one study (Marteau *et al* 1993) women were given an information intervention, anxiety management training, or both, prior to undergoing routine prenatal testing. Neither intervention resulted in lower anxiety following the receipt of a false positive result compared to a standard care intervention group. However, this finding may be a consequence of the absence of raised anxiety in this sample following the receipt of a result indicating possible foetal abnormality. In another study (Cope *et al* 2003) women who received either an audiotaped recording or a non-technical letter about their prenatal diagnostic consultation were less anxious than routine care control group. However there were no differences between the groups in how well they recalled the information suggesting that the reduced anxiety was not moderated by increased knowledge.

In contrast, a further study (Marteau *et al* 1996), albeit of adult screening, provides evidence that knowledge can moderate anxiety The study compared the impact of two types of information leaflet sent to women prior to attending an appointment for colposcopy, a follow-up test after cervical screening. A "simple" leaflet gave information about the procedure and ways of coping with it as well as about the high rates of cervical abnormalities and the low probability of cancer and high success rates following treatment. A "complex" leaflet gave information about the aetiology of cervical abnormalities and their treatment and the likely outcome. Receipt of either of the leaflets increased knowledge about the screening, but only the "simple" leaflet resulted in decreased anxiety. This suggests that not only can knowledge reduce the impact of screening on psychological well-being, but that it is likely to be specific types of information that are important. In the above study it may be that the information present in the "simple leaflet" but not the "complex" leaflet concerning the low probability of cancer following an abnormal cervical screening test result may have been particularly important.

Based on these studies, further research into the effects of knowledge on anxiety following screening is warranted, particularly relating to the types of information that may be effective in reducing psychological distress.

4.2.1.3 Newborn hearing screening

Given that newborn hearing screening can involve a number of tests before a baby is either discharged or referred, there is particular potential for anxiety to be provoked among parents. While some studies have suggested that there is no emotional impact of newborn hearing screening on mothers (Watkin *et al* 1998), others have found raised levels of anxiety among mothers of babies who receive a false positive result and require more than one screening test (Clemens *et al* 2000, Magnuson & Hergils 2004, Vohr *et al* 2001).

However this research has a number of limitations. There is a reliance on small samples of mothers of recalled babies, with only 20 mothers in one case (Vohr *et al* 2001). Only one study (Magnuson & Hergils 2004) differentiated between mothers of babies recalled because of possible unilateral and possible bilateral losses. Even in this study the very different implications of the two types of recall were not highlighted. While a baby with a unilateral loss will have normal hearing in one ear, a baby with a hearing loss in both ears may have very little functional hearing. In two studies anxiety was assessed after the result of the screen was known (Clemens *et*

al 2000, Magnuson & Hergils 2004); in one case, 9–12 months after the screen (Magnuson & Hergils 2004). Single item measures of anxiety were used in all but one study (Watkin *et al* 1998). Such single item measures of a higher order construct, such as anxiety, lack validity. To the best of our knowledge there have been no prospective studies powered to assess maternal anxiety and concern in response to screening, particularly following recall.

The aim of the present study was to describe the possible adverse emotional effects of newborn hearing screening and particularly the effects of receiving a referral for diagnostic tests. The possible moderating effects that knowledge of the test might have on these emotional consequences were also assessed. There was a particular focus on the impact of understanding the meaning of the test result given previous research suggesting that reassuring mothers referred for further tests that their baby would probably be all right was effective in reducing anxiety (Watson *et al* 2002).

4.2.2 Method

4.2.2.1 Design

A prospective descriptive study was conducted comparing the responses of four groups.

4.2.2.2 Measures

Four outcomes were measured by questionnaire:

- *Maternal state anxiety:* assessed using the short form of the state scale of the Spielberger State-Trait Anxiety Inventory (Marteau & Bekker 1992). Scores on this measure range from 20 to 80 with a normal score of 35 and a clinical range indicated beyond 49. The reliability of this measure in this sample is indicated by a Cronbach's alpha of .81.

- *Worry about the babies' hearing:* assessed using one item asking "How worried do you feel at the moment about your baby's hearing?" Mothers were asked to indicate their worry on a seven point scale anchored by *"not at all worried"* and *"extremely worried"*.

- *Certainty about the babies hearing:* assessed using one item asking "How certain do you feel at the moment that your baby is normally hearing?" Mothers were asked to indicate their certainty about their baby's hearing on a seven point scale anchored by *"not at all certain"* and *"very certain"*.

- *Knowledge about the newborn hearing screening programme:* assessed using a multiple-choice measure, similar to ones developed for assessing knowledge of prenatal screening

tests (Marteau *et al* 2001). This comprised eight items (see appendix) concerning what happens at different stages of the screen, possible results of the hearing screen, reasons for the receipt of no clear responses and numbers of babies referred for diagnostic tests who will be found to have a hearing loss. The alpha for this scale was 0.57.

4.2.2.3 Participants

A total of 342 mothers whose newborn babies had received hospital-based newborn hearing screening at one of the NHSP first phase sites participated in the study. These mothers comprised four groups:

- Group 1: Mothers of babies who had clear responses in both ears on the first or second (OAE) screening test.

- Group 2: Mothers of babies who did not have clear responses in one or both ears at the first or second screening test, but did at the final (AABR) one.

- Group 3: Mothers of babies who did not have clear responses in one ear at the final screening test and who were referred for follow-up assessment

- Group 4: Mothers of babies who did not have clear responses in either ear at the final screening test and were referred for follow-up assessment.

Babies who had been admitted to the Special Care Baby Unit were excluded from the study because of the likelihood of raised anxiety levels in mothers of these babies.

4.2.2.4 Sample size calculations

The study was designed to have 80% power to detect an effect size of $f = 0.17$ of screening result group upon state anxiety at the 0.05 level of significance. This required 100 respondents in each of the study groups.

4.2.2.5 Procedure

The screening process was begun prior to discharge from the Maternity Unit. On the morning of their baby's hearing screening, screeners routinely gave mothers a leaflet to read entitled 'Your Baby's Hearing Screen'. This leaflet included information on (i) reasons for screening; (ii) details of the screening test; (iii) when screening is undertaken; (iv) the meaning of screening test results; and (v) who to contact for further information. Mothers may also have been given this leaflet and viewed an explanatory video at prenatal classes as part of the routine NHSP implementation. Immediately before screening, screeners gave mothers a brief verbal

explanation of the screen. Women were informed about the study and the possibility of being asked to participate in the questionnaire-based evaluation when consenting for their babies to undergo the screen.

Sampling took place once a week and was dependent upon the type of screening test results. Because of the relatively small number of cases receiving a bilateral referral, all such cases were sampled. The number of cases in this group was used as a guide to the number of cases to be randomly sampled from the other three, more numerous, groups. Sampling targets were generated, based on an average of four cases from group 4 being identified each week. If there were insufficient cases in group 4 to meet the sampling targets, sufficient cases were still sampled from the other groups to ensure that the sampling targets were met for those groups. The names of all mothers whose babies had received newborn hearing screening, and had received a result other than bilateral referral, were entered into a Microsoft Access database and queries used for random sampling.

Questionnaires were sent three weeks following completion of the screen. If a completed questionnaire or a decline form had not been received three weeks later, a reminder was sent. Questionnaire packs included information about the study, a decline form and a freepost envelope.

4.2.2.6 Analysis

Analyses were conducted using SPSS for Windows version 10. The main analyses consisted of one-way analysis of variance comparing levels of anxiety, worry, certainty and knowledge across the different hearing test results group. Linear trend analysis was conducted to ascertain whether, as predicted, anxiety and worry increased with the number of tests the baby had, while certainty decreased. Pearson correlations were conducted on data from groups 3 and 4 to explore possible relationships between worry, certainty and knowledge. Hierarchical multiple regression was used to explore the moderating effect of knowledge on anxiety of mothers in the different groups. Three analyses of variance using a 2 (correct or lack of understanding of specific information) x 4 (hearing test result group) were used to examine the effect of understanding that the most likely reason for referral was not hearing loss on anxiety, worry and certainty.

4.2.3 Results

Overall, a return rate of 53 % (384/722) was achieved comprising return rates of 65% in group 1, 57 % in group 2, 48% in group 3 and 41% in group 4. The demographic characteristics of respondents are shown in table 4.1. A one-way analysis of variance of age by group was significant ($F(3,339)=3.029$ $p<0.030$). However post hoc testing using Tukey's b test indicated that there were no significant pairwise differences between the groups. As there were no significant correlations between outcome variables, age was not controlled for in the main analyses. The numbers of non-white respondents were too small to conduct a χ^2 test to identify any differences between the groups in ethnicity. In order to identify whether there were differences between the groups in educational level a Chi square test was conducted. There was a marginally significant difference between the groups in educational level ($\chi^2(6)=10.977$, p =

0.089). Given this result, the analyses were also run controlling for educational level, but this made negligible differences to the analyses and did not affect the key outcomes.

	Group 1 (n=103)	Group 2 (n=81)	Group3 (n=105)	Group 4 (n=55)	All Groups (n=343)
Age Mean(SD)	30.64 (6.17)	28.48 (6.79)	28.31 (5.64)	28.91 (5.74)	29.14 (6.16)
Ethnic Background %(n)					
White	97 (99)	91 (74)	91 (96)	89 (49)	93 (318)
Non white	3 (3)	9 (7)	9 (9)	11 (6)	7 (25)
Education* %(n)					
Up to 16 years	50 (51)	42 (33)	38 (40)	40 (22)	43 (146)
Post 16 up to degree	21 (21)	38 (30)	36 (37)	42 (23)	33 (111)
Degree or higher	29 (29)	20 (16)	26 (27)	18 (10)	24 (82)

Table 4.1. Demographic characteristics of the 343 respondents (%(n)). * 5 cases missing (2 from group 1, 2 from group 2 and 1 from group 3).

Table 4.2 indicates that maternal anxiety was in the normal range. Although the omnibus ANOVA on maternal anxiety was not significant ($F(3,327)=1.486$, $p=0.218$), there was a significant linear trend for maternal anxiety to increase across the four groups ($F(3,327)$ 4.280, $p<0.05$), being highest in mothers of babies recalled with possible bilateral loss (group 4) and lowest in those who received clear responses on the initial screen (group 1). Overall, worry about the babies' hearing was low and certainty was high. However, there were significant difference between the groups in levels of worry ($F(3,337)=26.415$ $p<0.001$) and certainty about babies' hearing ($F(3,339) = 10.109$ $p=0.001$). Linear trend analysis showed that there were significant trends for worry to increase ($F(3,337)=70.342$, $p<0.001$) and certainty to decrease ($F(3,339)=27.474$, $p<0.001$) as the number of tests that the baby needed increased.

4.2.3.1 Relationship between knowledge and anxiety

Average knowledge scores were between 5 and 6 out of a possible 8 across the sample and there were no significant differences between groups ($F(3,339)=1.726$ $p=0.726$).

There were no significant correlations between knowledge and anxiety variables among mothers of babies requiring assessment for a possible unilateral hearing loss (table 4.3). However, among mothers of babies referred for a possible bilateral loss higher knowledge was associated with lower state anxiety (rho=-.297, n=53, $p<0.05$), and greater certainty that the baby was normally hearing (rho=.266, n=53, $p<0.05$). The moderating effect of knowledge on anxiety was examined using multiple regression (see table 4.4). On the first step the variables knowledge and group, dummy coded with group 1 as the reference group, were entered. On the second step the product terms knowledge x group were entered. The first step of the model did not significantly explain variance in state anxiety ($F(4,326)=1.355$, $p=0.249$, adjusted $R^2=0.004$). The addition of the product terms on the second step marginally improved the prediction of anxiety (F change$(3,323)=2.259$, $p<0.09$, R^2 change=0.02, adjusted $R^2=0.016$). However, none of the individual predictors on the model was conventionally or marginally significant.

4.2.3.2 Specific knowledge and reactions to screening

Given that knowledge in general did not moderate the effect of test result on anxiety, the potential moderating effects of specific knowledge were investigated. Analysis of variance showed there were significant main effects of understanding that referral for diagnostic testing was unlikely to mean that the baby had a hearing loss on state anxiety ($F(1,323)=6.810$, $p<0.01$), worry ($F(1,332)=24.020$, $p<0.01$) and a marginal effect on certainty ($F(1,334)=3.559$, $p=0.060$). As would be expected from previous analyses, there were also significant main effects of hearing test result group on state anxiety ($F(3,323)= 5.064$, $p<0.01$), worry ($F(3,332)=24.020$, $p<0.001$ and certainty ($F(3,334)=9.742$, $p<0.001$). There were significant interactions of hearing test result group and of understanding that referral is unlikely to mean that the baby has a hearing loss on state anxiety ($F(3,323)=4.791$, $p<0.01$) and worry ($F(3,332)=3.565$, $p<0.05$) and a marginally significant interaction on certainty ($F(3,334)=2.451$, $p=0.063$). These interactions (figures 4.1-4.3) show that anxiety and worry were higher and certainty lower among mothers in group 4 who did not understand that the receipt of no clear responses was unlikely to indicate hearing loss, compared with those mothers who did not understand this.

	Group 1: Stage 1 of Screening-clear response	Group 2: Stage 1 of screening-no clear response, stage 2-clear response	Group 3: Suspected unilateral hearing loss-referral for audiological assessment.	Group 4: Suspected bilateral hearing loss-referral for audiological assessment.	Planned trend analysis F	Omnibus ANOVA F
Maternal state anxiety	31.99 (11.08)	32.68 (12.07)	33.95 (9.44)	35.72 (12.80)	4.280*	1.486
Worry about baby's hearing	1.34 (0.96)	1.41 (1.04)	2.71 (1.85)	3.07 (2.20)	70.342***	26.415***
Certainty about baby's hearing	6.40 (1.25)	6.14 (1.70)	5.33 (1.72)	5.31 (2.01)	27.474***	10.109***
Knowledge	5.81 (1.57)	5.40 (1.74)	5.79 (1.65)	5.35 (1.98)	0.989	1.726

*** $p < 0.05$ *** $p < 0.001$**

Table 4.2. Mothers' state anxiety, worry and certainty about their babies' hearing and knowledge about the newborn hearing screening test. Mean (SD)

	State Anxiety	Worry about babies hearing	Certainty about babies hearing	Knowledge
State anxiety		.455**	-.376**	.080
Worry about babies hearing			-.572**	-.048
Certainty about babies hearing				.110

(b) mothers of babies referred to audiology for suspected bilateral hearing loss (n=53)

	State Anxiety	Worry about babies hearing	Certainty about babies hearing	Knowledge
State anxiety		.668**	-.454**	-.297*
Worry about babies hearing			-.403**	-.129
Certainty about babies hearing				.266*

*** $p < 0.05$ ** $p < 0.01$**

Table 4.3. Correlations between state anxiety, worry and certainty about the baby's hearing and knowledge about newborn hearing screening mothers of babies referred to audiology for suspected unilateral hearing loss (n=102).

Variable	Partial Correlation	B	Standard error B	95%Confidence Interval for B	
				Lower Bound	Upper Bound
Knowledge	-0.050	-0.656	0.728	-2.087	0.775
Bilateral referral	0.090	3.101	1.904	-0.644	6.846
Unilateral referral	0.060	1.714	1.580	-1.394	4.822
AABR	0.020	0.620	1.696	-2.718	3.957
Bilateral referral x knowledge	-0.066	-1.255	1.060	-3.341	0.831
Unilateral referral x knowledge	0.062	1.135	1.011	-854	3.125
AABR x knowledge	0.058	1.082	1.034	-0.953	3.117

Table 4.4. Results of hierarchical regression examining whether knowledge moderates the impact of baby's group on mother's state anxiety.

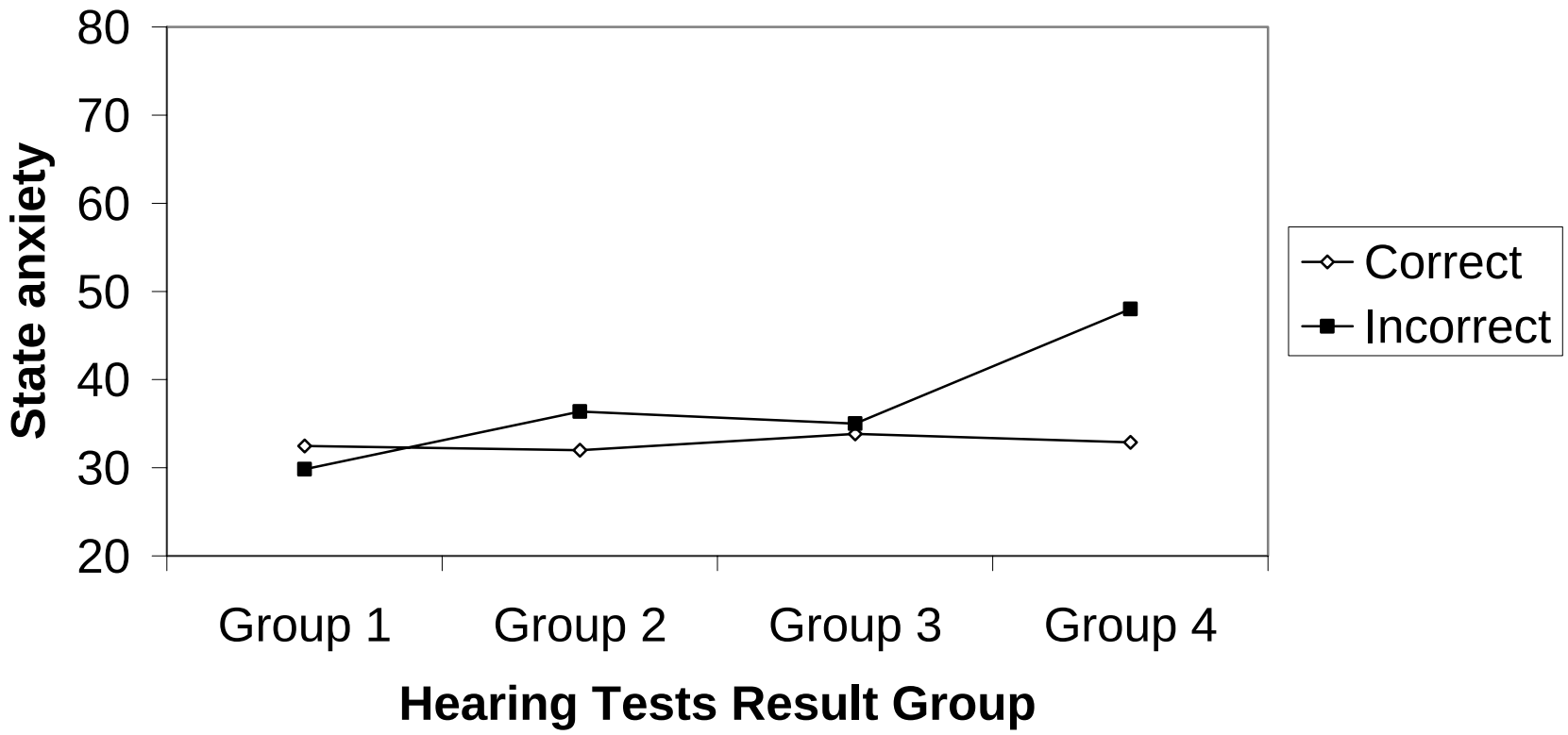

Figure 4.1. State anxiety among mothers who correctly understood that the most likely reason for the receipt of no clear responses was not hearing loss and those who did not understand this.

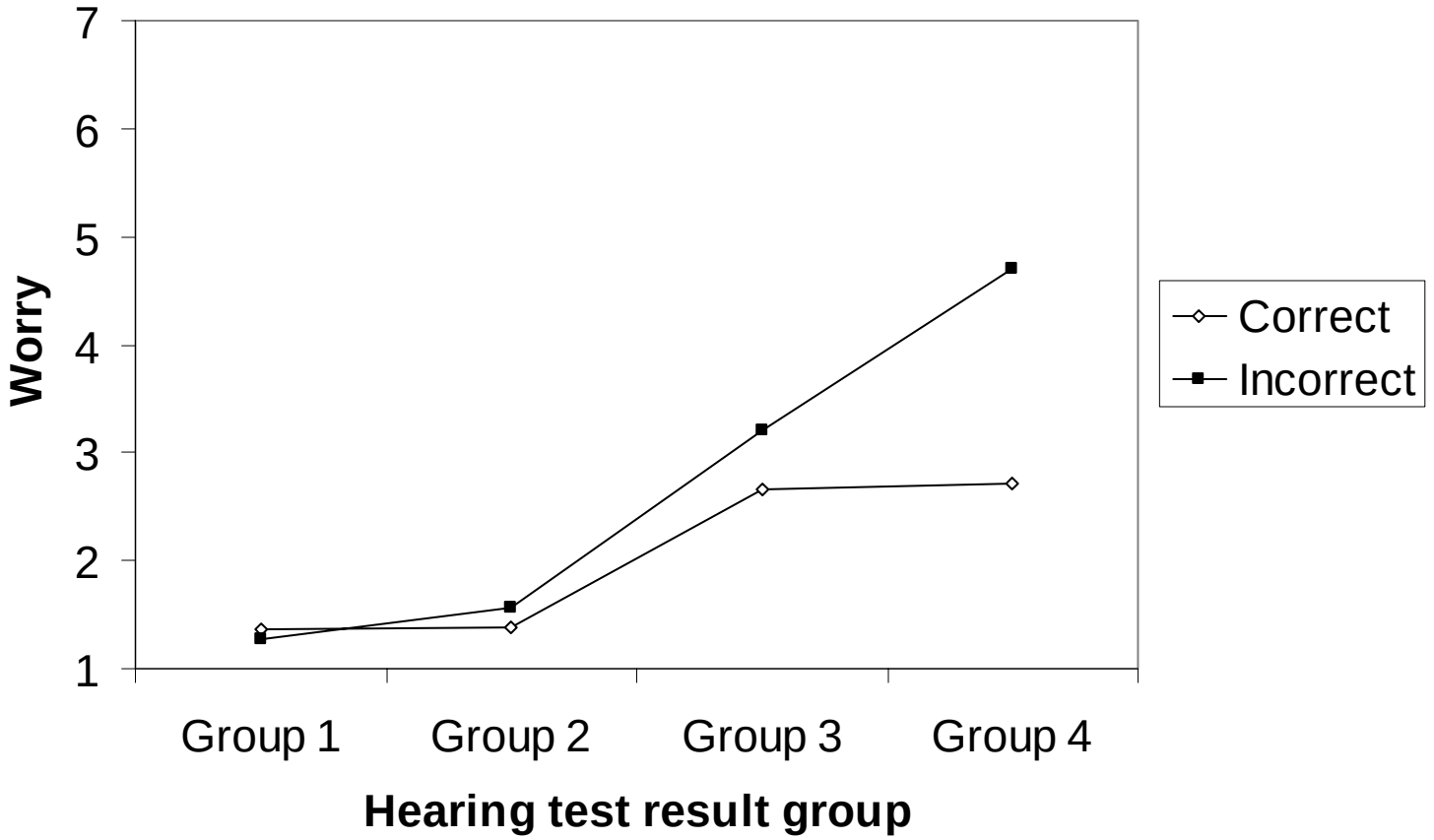

Figure 4.2. Worry about the baby's hearing among mothers who correctly understood that the most likely reason for the receipt of no clear responses was not hearing loss and those who did not understand this.

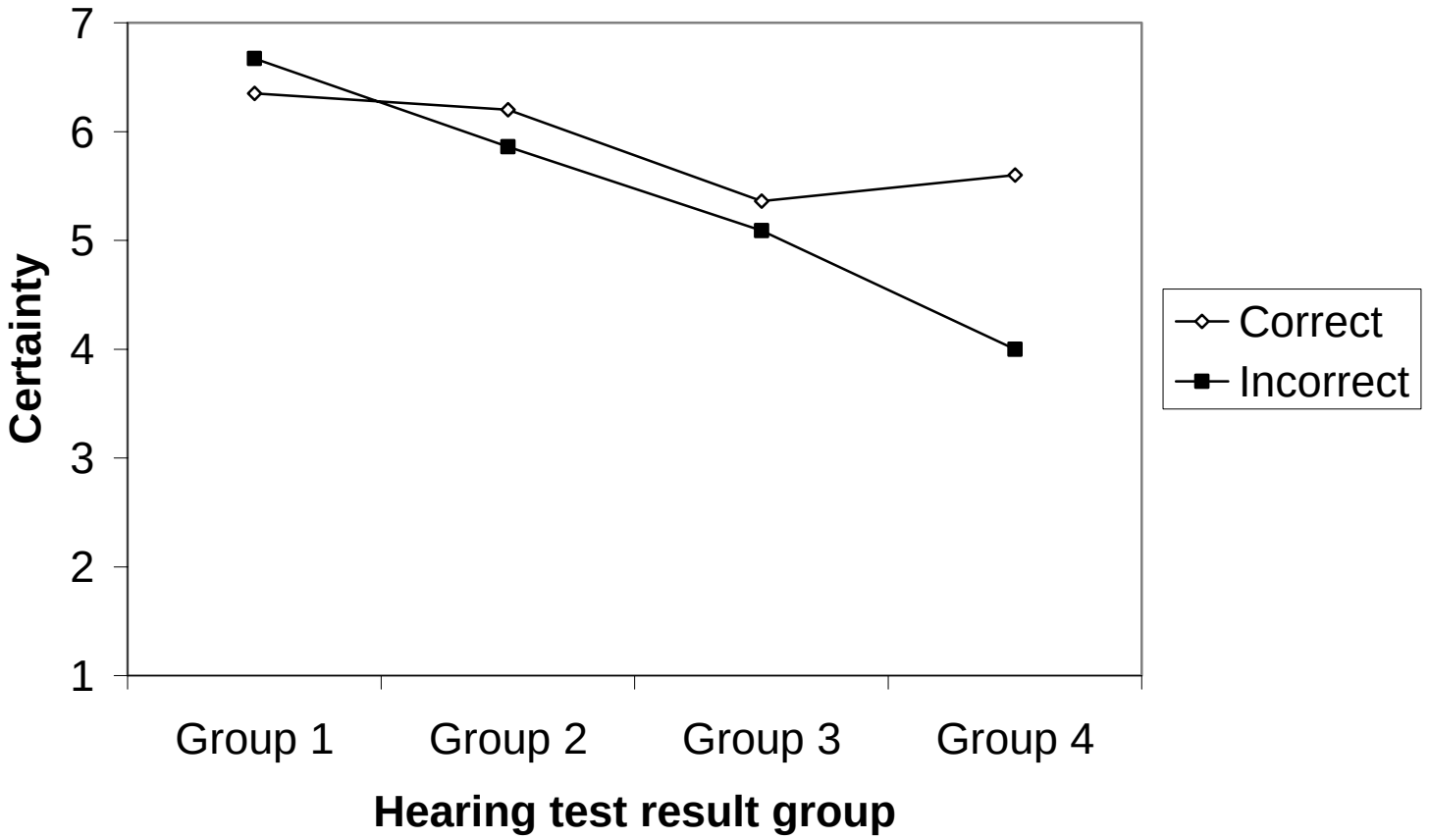

Figure 4.3.Certainty about the baby's hearing among mothers who correctly understood that the most likely reason for the receipt of no clear responses was not hearing loss and those who did not understand this.

4.2.4 Discussion

Although levels of state anxiety across the whole sample were in the normal range there was a significant linear trend for anxiety to increase across the four groups as the number of tests that the baby received increased. While there were low levels of worry and high levels of certainty about the baby's hearing, mothers of babies referred for audiological assessment were significantly more worried and less certain about their babies' hearing. Overall, mothers showed good knowledge of the newborn hearing screening programme and there were no significant differences in knowledge between the groups. Among mothers of babies who received a bilateral referral, knowledge was correlated with lower state anxiety and greater certainty about the baby's hearing. Although general knowledge about newborn hearing screening did not have a significant moderating effect on maternal anxiety, the specific knowledge that the most likely reason for "no clear responses" was not hearing loss moderated the impact of bilateral referral on anxiety worry and certainty.

Two factors may have resulted in an underestimation of anxiety generated by NHSP: the timing of the questionnaires, and bias in respondents. Although the questionnaires in this study were sent out within four weeks of the completion of the screen, by the time mothers received, completed and returned the questionnaire it was more than four weeks since completion of the screen. The review of the evidence regarding the psychological impact of screening (Shaw *et al* 1999) found that that distress tended to be highest in the period immediately following the completion of the screen, but that these effects quickly started to fade and were no longer apparent one month following screening. Anxiety levels, therefore, may well have been higher closer in time to the screen. The remnants of this anxiety may be reflected in the significant trend for anxiety to increase with the number of tests that the baby underwent. A further factor that would lead to an underestimation of anxiety is if anxiety was greater in non-responders than in responders. There is good evidence from other studies of the psychological impact of health risk assessments to show that those not responding are more distressed and anxious than those responding (Maissi *et al submitted*, Timman *et al* 2004). It therefore seems likely that our results underestimate the anxiety caused by recall following newborn hearing screening.

The correlation between knowledge and anxiety among mothers of babies recalled for possible bilateral loss suggests that understanding the reasons for recall may protect against anxiety. Given the cross-sectional study design, an alternative explanation for this association is that mothers who are generally less anxious may have been better able to process and remember information about the screening test. Thus the resulting correlation may be an end result of anxiety levels, not a cause of them. Establishing the nature of this relationship requires prospective experimental designs to ascertain whether, by ensuring high knowledge before screening, anxiety can be avoided or reduced in those whose babies are recalled for further tests. A further explanation would be that those who were less anxious had greater personal and material resources indicative of higher levels of education which were significantly correlated with knowledge in this sample. However, anxiety, worry and concern were not associated with educational level in this sample and so this is unlikely to be an explanation for our findings in this study.

This study has a number of important strengths. It provides the most robust assessment to date of the emotional impact of newborn hearing screening. It assessed the emotional distress and knowledge of mothers using established measures as close to the end of the screen as was

practicable. The research was comprehensive in considering the differential effect on mothers of the number of screening tests needed, and in distinguishing between the effects of a unilateral and a bilateral referral. However, some limitations should be acknowledged. First, the response rate is relatively low. Although it is not untypical of the response rates often observed in response to questionnaire-based postal surveys (Asch *et al* 1997), it limits the strength of conclusions that can be made. Second, the use of a cross-sectional design means that the causal direction of the observed relationships cannot be inferred. Third, the numbers of mothers participating whose babies required a bilateral referral did not reach the number required to detect a medium effect. This was partly due to the relatively small numbers of infants receiving this result, but also to the lower response rates of mothers in this group. This lower response rate may in itself be indicative of these mothers having a less positive experience of the screening programme.

This study suggests that newborn hearing screening causes emotional distress in mothers whose children require a number of tests, at least in the short term. Furthermore, the findings suggest that knowledge, particularly about the meaning of the results, may reduce the impact on anxiety of a referral for possible bilateral hearing loss. Further studies are now needed to test the validity of these results in this and other screening programmes. In particular attempts should be made to explain the association between understanding and anxiety and in particular whether this is due to understanding protecting against anxiety or due to anxiety inhibiting information processing and hence understanding.

4.3 Maternal anxiety following newborn hearing screening: 6 months follow-up

Abstract

Objectives: To assess whether the impact of newborn hearing screening upon maternal anxiety, worry and certainty about the babies hearing three weeks after screening is evident six months later.

Methods: Questionnaires assessing maternal state anxiety, worry and certainty about the baby's hearing, were sent to four groups of mothers three weeks and six months after screening: Group 1: mothers whose babies had clear responses on a first or second screening test (n=79); Group 2: mothers whose babies had clear responses on the final screening test (n=49); Group 3: mothers whose babies did not have clear responses in one ear at the final screening test and were referred for audiological assessment (n=70); Group 4: mothers of babies referred for audiological assessment because no clear responses were received in either ear at the final screening test (n=30).

Results: At the six-month follow-up there were no significant differences between the groups in maternal state anxiety, worry or certainty about the baby's hearing

Discussion: The minor short-term adverse psychological impact of referral following newborn hearing screening is no longer evident at six months.

4.3.1 Background

We have found in study 1 that within four weeks of screening mothers of babies who require referral for diagnostic testing experience increased worry and uncertainty about their babies' hearing, with general levels of state anxiety rising as the number of tests that the baby needs increases. We report here on the longer term outcomes in this group of mothers.

Current evidence is inconclusive about the length of time after screening anxiety persists. A systematic review of screening in adults (Shaw *et al* 1999) found little evidence one month after screening of an effect on anxiety following a positive test. In keeping with this it has been suggested that raised anxiety levels return to normal rapidly after the receipt of a negative result on follow up tests (Ekeberg *et al* 2001, Parker *et al* 2002).

Two studies which have investigated the effects of false positive results on mothers' well-being following newborn hearing screening also suggest that there is little lasting impact, but these studies have limitations. In one (Clemens *et al* 2000) a small sample of 49 mothers responded. They were asked to indicate their anxiety levels on a single item, yet anxiety is a complex variable that is unlikely to be validly measured by a single item. There was no comparison group and follow up varied from 2 to 13 months after the screening tests had been completed. In the other study (Kennedy 1999) questionnaires were sent to a sample of 100 mothers of babies who screened positive and 100 who screened negative following newborn hearing. A 75% return rate was achieved in each group. Anxiety was measured using a reliable multi-item measure, the Spielberger State Trait Anxiety Inventory (Marteau & Bekker 1992). However, follow up again varied between 2 and 12 months after completion of the screening tests and it is not clear whether mothers of true cases were included in the analysis alongside false positives. Neither study distinguished between referral for possible unilateral and bilateral hearing loss.

The aim of the present study therefore was to assess the impact of the newborn hearing screening tests on mothers' well-being six months following completion of the screening tests. Based on the most comprehensive review of the evidence thus far relating to the psychological effects of screening, albeit in adults (Shaw *et al* 1999), we hypothesized that there would be no significant differences between the groups in state anxiety, worry and certainty about the baby's hearing at follow-up.

4.3.2 Method

4.3.2.1 Participants

A total of 228 mothers whose newborn babies had received hospital-based newborn hearing screening participated in the study. These mothers comprised four groups:

- Group 1: Mothers of babies who had clear responses in both ears on the first or second (OAE) screening test.

- Group 2: Mothers of babies who did not have clear responses in one or both ears at the first or second screening test, but did at the final (AABR) one.

- Group 3: Mothers of babies who did not have clear responses in one ear at the final screening test and referred for follow-up assessment.

- Group 4: Mothers of babies who did not have clear responses in either ear at the final screening test and were referred for follow-up assessment.

Mothers who returned a questionnaire at both assessment points were included in the sample. Overall, of those who returned their questionnaires at time 1, 66 % returned a questionnaire at time 2 (260/391): 72% in group 1 (78/109), 62% in group 2 (63/101), 70% in group 3 (81/116) and 58% in group 4 (38/65). However, not all these cases were included in the final analysis. Reasons for exclusions included admission of the baby to the NICU (n=23), identification of a hearing loss following follow-up assessment (n=2), birth of twins (n=2) and screening at community site (n=5).

Responders at six months were compared with non-responders on demographic characteristics (age, education and ethnicity) and outcome measures (state anxiety, worry and certainty about the baby's hearing) at time 1. Responders were significantly older (mean(SD)30.21(5.95)) than non responders (mean(SD)26.99(5.99):t(342)=4.721, $p<0.001$), and had significantly higher levels of education (education up to 16: responders 38 % vs non responders 54%; education after 16: responders 33% vs non responders 31%; degree level: responders 29% vs non responders 15%: $\chi^2(2)=10.012$, $p<0.01$) but there were no significant differences in ethnicity. In relation to the outcome variables, responders differed from non responders in terms of being less anxious at

time 1 (mean(SD) 32.32(10.57) vs 35.49(12.09): t(329)=-2.444, $p<0.05$) and less worried about the baby's hearing (mean(SD) 1.87(1.53) vs 2.42(1.94): t(339)=-2.851, $p<0.01$). There were no significant differences between the two groups in relation to certainty about the baby's hearing.

4.3.2.2 Measures.

The three outcome measures detailed in Study 1 were used:

Maternal state anxiety: assessed using the short form of the state scale of the Spielberger State-Trait Anxiety Inventory (Marteau & Bekker 1992);

- *Worry about the baby's hearing:* assessed using one item "How worried do you feel at the moment about your baby's hearing?" Mothers were asked to indicate their worry about their baby's hearing on a seven point scale anchored by *"not at all worried"* and *"extremely worried"*.

- *Certainty about the baby's hearing:* assessed using one item "How certain do you feel at the moment that your baby is normally hearing?" Mothers were asked to indicate their certainty about their baby's hearing on a seven point scale anchored by *"not at all certain"* and *"very certain"*.

4.3.2.3 Procedure

Details about the information given to mothers about the screen and the study can be found in the report on Study 1, as can details about sampling procedures.

Questionnaires were sent three weeks and six months following completion of the screen. If a completed questionnaire or a decline form had not been received three weeks later, a reminder was sent. We report here on the six month questionnaires, having reported in Study 1 on those returned at three weeks.

4.3.2.4 Analysis

Analyses were conducted using SPSS for Windows version 10. Differences in the demographic characteristics of the four groups were assessed using ANOVA and χ^2 tests. For comparisons of the data at three weeks after completion of the screening tests, ANOVA was used with a priori linear contrasts. Since the four groups were not expected to differ at 6-month follow-up, comparisons were made using ANOVA with Tukey's b post hoc test to identify between group differences.

4.3.3 Results

The characteristics of the study sample are shown in table 4.5. Although the ANOVA comparing the mean ages of the four groups was close to significant ($F(3,227)=2.593$, $p=0.054$) post hoc testing with Tukey's b test indicated that there were no significant differences between the individual groups. There were no significant differences between the groups in ethnicity ($\chi^2(3)=4.983$, $p=0.173$) but there were in educational level ($\chi^2(3)=13.532$, $p=0.035$). In group 1 there was a higher than expected number of mothers who did not have education beyond 16 years and in groups 2 and 4 there were higher than expected numbers of mothers who had education beyond 16 years but not up to degree level. Oneway ANOVA comparing the outcome variables between the different educational levels was therefore conducted. None was significantly different, indicating that education was not a covariate of the outcome variables, and thus did not need to be controlled for in the main analyses.

	Group 1 (n=79)	**Group 2 (n=49)**	**Group3 (n=70)**	**Group 4 (n=30)**	**All Groups (n=228)**
Age Mean(SD)	31.58 (5.54)	29.63 (7.24)	28.99 (5.75)	30.37 (4.57)	
Ethnic Background* %(n)					
White	99 (77)	90 (44)	93 (65)	93 (28)	94 (214)
Non white	1 (1)	10 (5)	7 (5)	7 (2)	6 (13)
Education %(n)**					
Up to 16 years	47 (36)	31 (15)	36 (25)	30 (9)	38 (85)
Post 16 up to degree	19 (15)	45 (22)	34 (24)	50 (15)	33 (76)
Degree or higher	34 (26)	24 (12)	30 (21)	20 (6)	29 (65)

*** 1 case missing**

*** 2 cases missing**

Table 4.5. Demographic characteristics of the 228 respondents (%(n)).

Table 4.6 shows the means and standard deviations for this sample at both assessment points. At follow-up there were no significant differences between hearing test result groups in relation to state anxiety ($F(3.222)=1.191$, $p=0.314$) and, although analysis of variance indicated a significant model for worry ($F(3,223)=3.377$, $p<0.05$) and certainty about the baby's hearing ($F(3,226)=3.111$, $p<0.05$), Tukey's b tests indicated that there were no significant between-group differences.

4.3.4 Discussion

As predicted the emotional effects on mothers of receiving a referral for diagnostic testing following newborn hearing screening six months earlier were no longer evident at the time of follow-up. These results not only provide support for existing findings (Shaw *et al* 1999) that adverse emotional effects of recall in screening tend to dissipate over time but also extends research in to the area of child health. Research in this area is important because concerns have been expressed about the possibility that raised maternal anxiety following neonatal screening programmes might interfere with the development of the mother-child relationship and therefore with the psychological well-being of the child (Paradise 1999).

This study has a number of strengths. It distinguished between mothers of babies who received different results on the screening tests, used standardised measures and was sufficiently powered to detect small to medium effects. However the study does have two limitations. Firstly, the response rate was low which, although not unusual for response rates in postal questionnaires (Asch *et al* 1997), limits the strength of the conclusions that can be drawn from these results. A further limitation of this study is that those responding differed from those who did not in being less anxious immediately after the screening. This has been reported in other studies (Maissi *et al submitted*) and it is thus possible that these results underestimate the emotional distress present in mothers six months following the screening process.

4.3.5 Concluding comment

Newborn hearing screening brings benefits in terms of early detection and intervention for children born with a hearing loss. The results of the current study suggest that the adverse psychological consequences of recall for follow-up assessments do not persist in the long-term. This fact, together with our previous finding from Study 1 that the understanding of the meaning of referral following the screening tests may ameliorate the short-term effects on mothers' emotional well-being, support the continuing implementation of newborn hearing screening given it is provided in a way that ensures mothers have a good understanding of the tests their babies are undergoing.

	Group1: Clear responses at OAE test	Group 2: Clear responses at AABR test	Group 3: Referral for diagnostic testing of possible unilateral hearing loss	Group 4: Referral for diagnostic testing of possible bilateral hearing loss	A priori linear trend analysis F(*p*)	Omnibus analysis of variance F(*p*)
State anxiety						
3 weeks post test	31.52 (11.21)	31.27 (11.02)	33.00 (9.27)	34.37 (11.24)	1.851 (0.175)	0.753 (0.522)
6 months post test	32.72 (10.98)	29.79 (9.61)	31.62 (9.96)	29.43 (9.51)		1.191 (0.314)
Worry about the baby's hearing						
3 weeks post test	1.20 (0.70) a	1.31 (0.74)a	2.62 (1.88)b	2.67 (1.90)b	50.040 (0.001)	19.839 (0.001)
6 months post test	1.09 (0.37)	1.41 (0.61)	1.47 (1.20)	1.43 (0.94)		3.377 (0.019)
Certainty about the baby's hearing						
3 weeks post test	6.44 (1.23)a	6.22 (1.63)a	5.26 (1.82)b	5.73 (1.46)b	16.203 (0.001)	8.074 (0.001)
6 months post test	6.72 (0.75)	6.53 (1.32)	6.07 (1.73)	6.40 (1.28)		3.111 (0.027)

Table 4.6. Means and standard deviations of psychological outcomes 3 weeks and 6 months following completion of screening tests.

4.4 A comparison of anxiety between mothers of babies who had hospital and community based newborn hearing screening

Abstract

Objectives: To compare the impact upon mothers' emotional well-being of hospital-based and community-based newborn hearing screening.

Methods: Postal questionnaires assessing maternal state anxiety, worry and certainty about the babies' hearing, and knowledge of screening, were sent to two groups of mothers, those whose babies had undergone hospital-based newborn hearing screening (n=94) and those whose babies who had undergone community-based newborn hearing screening (n=114) three weeks following screening. All had received clear test results.

Results: There were low levels of state anxiety and worry and high levels of certainty about the babies' hearing which were similar in the two groups, although mothers of babies screened in the community were marginally less worried about their baby's hearing.

Conclusion: The marginal difference in worry may suggest that community-based screening has a less negative impact on mother's emotional well-being. This effect may be greater among mothers of babies who need more than one test and for whom greater emotional distress is generated. Further research is needed to compare the effects of the two types of screening on the emotional well-being of those whose babies need more hearing tests.

4.4.1 Background

The newborn hearing screening programme (NHSP) is being implemented in two different settings. In hospital-based programmes the screening is conducted on the maternity unit by a new cadre of dedicated screeners, trained and employed specifically to conduct newborn hearing screening prior to discharge from the Maternity Unit. In community-based programmes, Health Visitors conduct the screening at their routine postnatal home visit, usually ten days after birth. One of the areas of interest for the evaluation of the NHSP pilot was the comparison of hospital-based and community-based newborn hearing screening including the differential effects on the psychological well-being of mothers. The period immediately after birth is one in which mothers will be recovering from both the physical and emotional demands of labour. It is possible, therefore, that having their babies' hearing tested at this time might be more stressful than having it screened in the community a few days later when the mother is back at home and has had time to recover from the demands of the child birth.

Due to the very small numbers of cases referred by the screen for follow-up assessment at the community-based pilot sites we were not able to sample a sufficient number of such cases within the time-scale available to enable a comparison of emotional outcomes among mothers of babies referred following both types of screening. Because the previous studies suggested that there is very little impact on mothers emotional well-being of the baby having clear responses at the first stage of screening, we did not make any predictions about differences in emotional distress between the two groups. The aim of this study was therefore to compare emotional outcomes among mothers of babies having clear responses at the first stage of screening following hospital and community-based newborn hearing screening. In addition, because our previous research has suggested that increased knowledge of the screening programme is protective against raised anxiety, we compared knowledge of the screening programme following hospital and community-based screening.

4.4.2 Method

4.4.2.1 Design

A prospective descriptive study was conducted using a two-group between-subjects design.

4.4.2.2 Measures

Three of the four outcome measures used in Studies 1 and 2 were used in this study, as well as a further measure:

- *Maternal state anxiety:* assessed using the short form of the state scale of the Spielberger State-Trait Anxiety Inventory (Marteau & Bekker 1992).

- *Worry about the babies' hearing:* assessed using one item asking "How worried do you feel at the moment about your baby's hearing?"

- *Certainty about the babies hearing:* assessed using one item asking "How certain do you feel at the moment that your baby is normally hearing?"

- *Knowledge about the newborn hearing screening programme:* assessed using a multiple-choice measure, similar to ones developed for assessing knowledge of prenatal screening tests (Marteau *et al* 2001). This comprised eight items concerning what happens at different stages of the screen, possible results of the hearing screen, reasons for the receipt of no clear responses and numbers of babies referred for diagnostic tests who will be found to have a hearing loss.

4.4.2.3 Participants

The sample comprised 208 mothers whose newborn babies had had newborn hearing screening and whose babies had received clear responses at the first stage of screening. These mothers comprised two groups: 94 mothers of babies who had had hospital–based newborn hearing screening and 114 mothers of babies who had had community-based newborn hearing screening. It was not possible to do a sample size calculation as we did not make any predictions regarding differences between the two groups. A target of 100 in each group was set to allow detection of a medium effect on the main outcome variables.

4.4.2.4 Procedure

The place of screening varied between the two groups, being conducted either on the maternity unit prior to discharge, or in the baby's home at the Health Visitor's routine postnatal visit. Mothers were informed about the study and the possibility of being asked to participate in the questionnaire-based evaluation when consenting for their babies to undergo the screen.

In the case of hospital-based screening, sampling took place once a week over the course of three weeks. The names of all mothers whose babies had received newborn hearing screening were entered into a Microsoft Access database and queries used for random sampling. Because of the smaller numbers of cases being received from community sites, all the mothers of babies who received a clear response at the first stage of screening at community sites were recruited until the sample size was reached.

Questionnaires were sent three weeks following completion of the screen. If a completed questionnaire or a decline form had not been received three weeks later, a reminder was sent. Questionnaire packs included information about the study, a decline form and a freepost envelope.

4.4.2.5 Analysis

Analyses were conducted using SPSS for Windows version 10. Preliminary analyses to check for between-groups differences were conducted using t-tests and χ^2 test and where necessary correlation using Pearson's *r* was used to ascertain whether the demographic variable was associated with any outcome variables. The main analyses consisted of t-tests, comparing levels of anxiety, worry, certainty and knowledge across the different hearing test results group.

4.4.3 Results

There was an overall response rate of 58% (209/363) comprising a response rate of 52% (94/181) among mothers of babies screened in the hospital-based programme and 63% (115/182) among mothers of babies screeners in the community-based programme. One mother of a baby who received community-based newborn hearing screening was excluded from the study as she did not return a questionnaire until 6 months after completion of screening, giving a sample of 114 in that group.

The demographic characteristics of the two groups of mothers are shown in table 4.7. There were no significant differences between the two groups in relation to mother's age ($t(206)=-0.733$, $p=0.464$) or educational level ($\chi^2(2)=3.020$). The age of the baby at the completion of the screening tests did vary between groups with babies screened in the community being significantly older ($t(204)=-6.275$, $p<0.001$). The age of the baby did not correlate with any of the outcome variables and therefore was not controlled for in the main analyses. The numbers of non-white participants were too small to compare.

	Mothers of hospital screened babies (n=94)	Mothers of community screened babies (n=114)
Age of mothers in years (Mean(SD))	30.54	31.14
Age of babies in days (Mean(SD))	7.68 (11.39)	15.60 (6.40)
Education of mothers (%(N))		
Up to 16 years	40 (38)	30 (34)
Beyond 16 but not degree	27 (25)	27 (31)
Degree and beyond	33 (31)	43 (49)
Ethnicity of mothers (%(N))		
White	93 (87)	97 (111)
Non white	7 (7)	3 (3)

Table 4.7. Demographic characteristics of participants.

The means and standard deviations for the outcome variables are shown in table 4.8. Overall, there were low levels of maternal state anxiety and worry about the baby's hearing and high levels of certainty about the baby's hearing and knowledge of the screening test. There were no significant differences between the groups in relation to state anxiety ($t(197)=-435$, $p=0.664$), certainty ($t(205)=-0.677$, $p=0.499$) or knowledge ($t(2,206)=0.143$, $p=0.886$). There was, however, a marginally lower level of worry about the baby's hearing among mothers of babies screened in the community ($t(204)=1.922$, $p=0.056$).

	Mothers of hospital screened babies (n=94)	Mothers of community screened babies (n=114)	t (*p*)
State anxiety	30.07 (10.08)	30.65 (8.66)	-0.435 (0.664)
Worry	1.56 (1.10)	1.31 (0.74)	1.922 (0.056)
Certainty	6.34 (1.28)	6.46 (1.10)	-0.677 (0.499)
Knowledge	5.98 (1.68)	5.95 (1.47)	0.143 (0.886)

Table 4.8. Comparison of outcome variables between hospital and community mothers (Mean(SD)).

4.4.4 Discussion

There were low levels of state anxiety and worry about the baby's hearing and high levels of certainty about the baby's hearing among mothers of babies who had a clear response at the first stage of screening. In addition, there was good understanding of the screening tests. Although there were no differences between the groups in relation to state anxiety, certainty about the baby's hearing and knowledge, mothers of babies screened in hospital were marginally more worried about their baby's hearing.

The marginally lower levels of worry among mothers of babies screened in the community which this study identifies suggests that community-based screening may evoke less emotional distress. One possible explanation for this is it reflects the mothers gaining confidence with their babies' health as the baby grows. The age of the baby was, however, unrelated to any of the outcomes. Another possible explanation is that mothers are more reassured by tests conducted by a Health Visitor or by tests conducted in their own home. It is possible that the marginal difference in worry we observed in the current study between the hospital and community mothers would become more pronounced with the need for more

testing given that emotional distress increased with the need for more tests in hospital-based screening. There is, therefore, a need for further research to determine whether community-based screening may ameliorate some of the adverse emotional consequences of referral for follow-up assessment following referral from hospital based newborn hearing screening.

This study has important strengths in terms of using a reliable and well validated measure of anxiety and of measuring mothers' emotional distress as close as was possible to the completion of the screening tests. The conclusions that can be generated from it are, however, limited by the relatively low response rate, although these rates are not unusual in questionnaire-based studies (Asch *et al* 1997).

In conclusion, the benefits that newborn hearing screening brings in terms of the early identification and treatment of hearing loss can be furthered by reducing the negative impact on mothers' emotional well-being that referral following newborn hearing screening can evoke. These results provide evidence to support the hypothesis that mothers of babies receiving a referral for follow-up assessment after screening experience less emotional distress if the screening is conducted in the community compared with the screening conducted in hospital. This hypothesis awaits testing.

4.5 A comparison of the IDT and newborn hearing screening: maternal anxiety and satisfaction

Abstract

Background: Newborn hearing screening is currently replacing the IDT, conducted at 8 months. Our previous research indicates that recall for further tests following newborn hearing screening can have a negative impact on the emotional well being of mothers but it is not known if this is greater than that caused by recall following the distraction test.

Objective: To compare the impact on maternal anxiety and satisfaction of recall following newborn hearing screening and the IDT.

Methods: Four groups participated: 27 mothers of babies receiving a satisfactory result and 21 mothers of babies recalled after the distraction test; and 26 mothers of babies receiving a satisfactory result and 16 mothers of babies recalled after newborn hearing screening. Questionnaires assessing maternal anxiety, worry and certainty about the babies' hearing, satisfaction with, and attitudes towards the screening test were sent to mothers 3 weeks and 6 months following screening.

Results: Comparison of the effects of receipt of different results showed no significant differences in maternal anxiety, worry and certainty between the two tests. Those mothers whose babies had newborn hearing screening were significantly more satisfied, regardless of the result received. Those who received a satisfactory result on the newborn hearing screening programme also had more positive attitudes towards that screening test than those receiving a satisfactory result following the IDT.

Conclusion: These results suggest that newborn hearing screening does not have a more negative emotional impact than the IDT.

Until January 2002 the universal screening programme of infant hearing in England was the IDT screen. The IDT was a behavioural hearing test which relied on the ability of infants from the age of six months to locate sounds by turning their heads towards it (Weir 1985). The screen was typically carried out between 6 and 8 months of age by two trained personnel (McCormick 2002). The test was conducted in a quiet room, usually at a Health Visitor clinic (Davis *et al* 1997). While the baby sat on its caregiver's lap one professional, out of the infant's view, would present sounds of various known frequencies to both the infant's left and right ear while a second professional situated to the front of the baby would observe the baby's response to those sounds (McCormick 2002).

4.5.1 Background

The IDT screen had a number of limitations including poor sensitivity and specificity. Sensitivity rates have been found to vary from 36% to 88% (Davis *et al* 1997) while Johnson and Ashurst (Johnson & Ashurst 1990) found a specificity of 97% for the IDT following two distraction tests. A further problem with the IDT was that confirmation of deafness was delayed with a median age of identification of between 13 and 20 months following the screening test (Davis *et al* 1997). Yet children who are identified before 6 months of age have significantly better language development at ages 13 to 36 months than those who are identified after 6 months of age (Yoshinaga-Itano *et al* 1998). Such early identification is not possible with a screen that cannot be conducted until the infant is at least six months of age.

Newborn hearing screening has apparent advantages over the IDT. Sensitivity rates vary between 80 and 100% (Davis *et al* 1997) while, during the first phase of NHSP, a specificity in the region of 97-99% has been achieved. A median age of identification at 2-3 months is possible if babies are screened in the neonatal period (Davis *et al* 1997). However, our previous research suggests that there are negative emotional consequences of newborn

hearing screening on mothers of babies who are referred for follow-up assessment following the screening programme but, to our knowledge, there have been no studies of the impact of the distraction test on maternal anxiety. The purpose of this study was therefore to compare the emotional impact on mothers of referral on the two screening programmes and the acceptability of the two programmes to mothers.

4.5.2 Method

4.5.2.1 Design

This is a descriptive, between subjects design study.

4.5.2.2 Measures

Five outcomes were measured by the questionnaire.

- *Maternal state anxiety:* assessed using the short form of the state scale of the Spielberger State-Trait Anxiety Inventory. .

- *Worry about the baby's hearing:* assessed using one item asking mothers to indicate their worry about their babies' hearing on a seven point scale anchored by "not at all worried" and "extremely worried".

- *Certainty about the baby's hearing:* assessed using one item asking mothers to indicate their certainty about their babies' hearing on a 7 point scale anchored by "not at all certain" and "very certain".

- *Satisfaction with the screening test:* assessed using four 7 point rating scales anchored by "not at all satisfied" and "extremely satisfied" which assessed satisfaction with the screening programme in general and the way screening was conducted. Together the four items formed a scale with an alpha in this sample of .90.

- *Attitude to the screening test:* assessed using three 7 point rating scales. One item was anchored by "beneficial" and "harmful", one was anchored by "important" and "unimportant" and one was anchored by "a bad thing" and "a good thing". Participants were asked to rate their attitudes to having the hearing test on these items which together formed a scale with an alpha in this sample of .80.

4.5.2.3 Participants

IDT: A total of 65 mothers whose babies had had their hearing tested using the IDT returned the first questionnaire. Of these 35 were mothers whose babies had received a satisfactory result and 30 were mothers whose babies received a referral for follow-up. However, only mothers who had returned questionnaires at both measurement points were included in the final sample, giving a total sample of 48, 27 mothers of babies who had received a satisfactory result and 21 who had received a referral for follow-up testing.

NHSP: Data were drawn from those used in a larger study of the emotional effects on mothers of receiving different hearing tests results following newborn hearing screening. The comparison groups comprised the first 35 mothers recruited whose babies had received a satisfactory result at the first stage of the screen (first or second AOAE test) and the first 30 mothers whose babies had not received a satisfactory result in either ear on the hearing screen, and were referred for follow-up testing. However, only mothers of babies who had returned a questionnaire at both measurement points were included in the final analysis giving a sample of 26 mother of babies who received a satisfactory result and 16 mothers of babies who received a refer result.

4.5.2.4 Procedure

Infant Distraction Test: Mothers were sampled from six of the first phase NHSP sites (where the IDT was still in place for that cohort of babies already born as NHSP was introduced, but who were less than 8 months of age and had therefore not yet had the IDT). A total of 49 Health Visitors were either randomly selected by the study team or nominated from five of the sites to recruit mothers to the study. At the sixth site all Health Visitors participated in recruitment as the IDT was imminently being ended. Protocols for the IDT varied between areas. However, the test was usually conducted in a Health Visitor clinic or general practice surgery on infants aged between 6 and 8 months. If, following the first test, the infant had not passed the test then an appointment was made for the baby to have a further IDT (Davis *et al* 1997). If clear responses were still not recorded then a referral for follow-up assessment was made. Health Visitors invited the mothers of all the babies they tested who required a referral to Audiology after the IDT screen to take part in the research. The mother of the next baby they tested who passed the IDT screen was also invited to take part, to form the comparison group. Health visitors described the study to each mother eligible to take part. Mothers gave their consent to participate by signing a consent form. This was returned to the researcher, who sent a questionnaire to the mother within 3 weeks of, and 6 months after, the baby had had the IDT screen.

NHSP: The screening process was begun prior to discharge from the Maternity Unit. On the morning of their baby's hearing test, screeners gave mothers a leaflet to read entitled 'Your Baby's Hearing Screen'. This leaflet included information on (i) reasons for screening; (ii) details of the screening test; (iii) when screening is undertaken; (iv) the meaning of screening test results and (v) who to contact for further information. Immediately before screening, screeners gave mothers a brief verbal explanation of the screen. Women were informed about the study and the possibility of being asked to participate in the questionnaire based evaluation when consenting for their babies to undergo the screen.

Data concerning all babies who had received newborn hearing screening at the hospital sites was sent to the research team electronically. These data were entered into a Microsoft Access database.

4.5.2.5 Data analysis

Data were examined to ascertain whether they were parametric using Levenes and KS-Lilliefors tests. As these indicated that the data were non-parametric, between group tests comparing mothers of babies who i) had the same test but received different results and ii) mothers of babies receiving the same result but having different tests were conducted using Mann-Whitney U tests. Effect sizes indicated by Cohen's *d*, were calculated using the programme Gpower. An effect size of 0.2 is regarded as a small, but probably meaningful effect, one of 0.5 is regarded as a medium effect and one greater than 0.8 is regarded as a large effect (Howell 2002).

4.5.3 Results

Only mothers who returned a questionnaire at both time points were included in the analysis. Among mothers whose babies underwent the IDT 48% (48/99) returned questionnaires at both 3 weeks and 6 months of the completion of the screen, comprising 49% of mothers of babies receiving a satisfactory result and 48% of mothers of babies referred for further testing. Among the whole data set from which the NHSP comparison data were drawn, 35% of mothers returned a questionnaire both 3 weeks and 6 months following completion of the screen. Among mothers of babies

	IDT (n=48)	**Newborn Hearing Screening (n=42)**
Mean Age	29.27	31.38
Ethnic Background*		
White	100 (46)	98 (40)
Other	0	2 (1)
Highest Qualification**		
No qualification	10 (5)	5 (2)
GCSE or similar	28 (13)	36 (15)
GCE A level	13 (3)	7 (3)
Further education	17 (8)	19 (8)
Degree or similar	28 (13)	33 (14)
Other	4 (2)	0

* 3 cases missing

**1 case missing

Table 4.9. Demographic characteristics of respondents (%(n)).

receiving a satisfactory result, 48% returned questionnaires at both time points, while 24% of mothers of babies receiving a refer result returned questionnaires at both time points. The demographic characteristics of the mothers participating in the study are shown in table 4.9.

The means and standard deviations of the outcome variables are shown in table 4.10. These indicate that overall state anxiety levels, which were between 29.0 and 36.9, were within the normal range. Worry about the baby's hearing was low ranging from 1.1 to 3.3 out of 7, and certainty about the baby's hearing was high ranging from 5.4 to 6.8 out of 7. There were also

positive attitudes towards the hearing test that the baby received with mean scores ranging from 5.6 to 6.8 out of 7. Overall satisfaction varied from 4.3 to 6.2 out of 7

(a)	Time 1:3 weeks post test		Time 2: 6 months post test	
	IDT (n=27)	**NHSP (n=26)**	**IDT**	**NHSP**
State anxiety	28.97 (11.11)	31.79 (11.08)	32.84 (12.39)	32.64 (8.90)
Worry about baby's hearing	1.11 (.32)	1.08 (0.27)	1.11 (0.32)	1.12 (0.33)
Certainty about baby's hearing	6.80 (0.49)	6.58 (0.86)	6.67 (1.18)	6.62 (1.02)
Total satisfaction	4.89 (1.61)	6.12 (0.81)	4.90 (1.64)	6.17 (0.80)
Total positivity of attitudes	6.36 (1.05)	6.80 (0.47)	6.14 (1.08)	6.75 (0.60)

(b)	Time 1:3 weeks post test		Time 2: 6 months post test	
	IDT (n=21)	**NHSP (n=16)**	**IDT**	**NHSP**
State anxiety	33.17 (10.06)	36.89 (11.85)	35.40 (12.76)	30.42 (8.15)
Worry about baby's hearing	3.29 (2.00)	2.63 (2.06)	2.05 (1.62)	1.59 (1.18)
Certainty about baby's hearing	5.38 (1.80)	5.69 (1.77)	6.00 (1.49)	6.29 (1.57)
Total satisfaction	4.30 (1.73)	5.48 (1.27)	4.89 (1.23)	5.54 (1.25)
Total positivity of attitudes	5.65 (1.25)	6.06 (1.52)	5.60 (1.34)	6.06 (1.56)

Table 4.10. Maternal anxiety and satisfaction following the IDT and newborn hearing screening among mothers of babies receiving (a) a satisfactory result and (b) a refer result.

4.5.3.1 Comparisons between mothers of babies having different hearing tests and receiving a satisfactory result

There were significant differences between these groups in relation to total satisfaction and positivity of attitudes. At three weeks mothers of babies receiving the newborn hearing test were more satisfied (U=177.000, N1275, N2=26, $p<0.01$) and remained so at follow-up (U=158.000,N1=27, N2=23, $p<0.01$). There was a large effect of the test type on satisfaction three weeks following the screen of indicated by a d of 0.87, and at six months this had increased to 1.31. There was a non-significant trend for mothers whose babies had the newborn hearing test to have more positive attitudes towards the test their baby had at three weeks (U=253.500, N1=26, N2=26, $p=0.058$) with a small effect size of 0.32. However at six months this difference reached significance (U=211.500, N1=27, N2=25, $p<0.01$) and a d of 0.66 indicated a medium effect size.

4.5.3.2 Comparisons between mothers of babies having different hearing tests and being referred for further testing

There was one significant difference between these groups. At three weeks mothers of babies screened by NHSP had significantly higher satisfaction (U=84.000, N1=21, N2=14, $p<0.05$) and there was a substantial effect of the type of test on satisfaction (d=0.72).

4.5.3.3 Comparisons among mothers of babies having IDT and receiving different results

Three weeks after the screening test, mothers of babies who were referred for further tests following the IDT were more worried about their babies hearing (U=69, N1=27, N2=21, $p<0.001$) and less certain (U=123.000, N1=26, N2=21, $p<0.001$) compared with mothers of babies who had received a satisfactory result on this test. There was a considerable effect of test result on worry (d =1.27) and certainty (d=1.00). Although the effect diminished somewhat, these trends continued six months after the screening tests with higher worry (U=211.500, N1=27, N2=21, $p<0.05$; d=0.80) and lower certainty (U=197.000, N1=27, N2=20, $p<0.05$; d=0.50). There was a non-significant trend for increased anxiety among mothers of babies who were referred (U=180.000, N1=27, N2=20, $p=0.072$; d=0.39). Those referred, compared with those who had a satisfactory result, also had less positive attitudes at three weeks (U=173.500, N1=26, N2=21, $p<0.05$; d=0.6) but this difference did not persist six months after the hearing tests.

4.5.3.4 Comparisons among mothers of babies having NHSP and receiving different results

Mothers of babies receiving a refer result were more worried than those who received a satisfactory result following the newborn hearing test (U=114.000, N1=26, N2=16, $p<0.001$). There was a large effect of the test result on worry (d=1.04) although this worry was not evident six months later (U=164.000,N1=26, N2=16, $p=0.095$).

4.5.4 Discussion

The pattern of results was broadly similar across both tests with those who received the newborn hearing test having being more satisfied and, if a satisfactory result was received on this test, having more positive attitudes to the test. Being referred for further tests was associated with emotional distress, particularly worry about the baby's hearing, regardless of the type of test. Likewise receipt of a normal result was associated with greater levels of satisfaction with whichever hearing test that the baby had had.

Comparison groups differed in two respects: first the type of test; and second the age of the baby. It is therefore not possible to know the extent to which the more positive attitudes and experience of the newborn hearing test are due to the type of test or to testing in the newborn period. Nevertheless, these results suggest that newborn hearing screening does not cause emotional distress in addition to that caused by the IDT. In addition newborn hearing screening was associated with higher levels of satisfaction with the screening test. Greater satisfaction may facilitate attendance at follow-up tests.

The study had two main limitations. Firstly the numbers participating were small and the response rate low, particularly among those referred. There were, though, substantial effect sizes of the type of screening test and screening test result on the outcome variables suggesting that the findings are robust. In addition, our previous unpublished research, in which a higher response rate of 41% was obtained among mothers of babies referred for further tests following the newborn hearing test, found equivalent levels of emotional distress in this group. A second limitation of the study is that because the two groups being compared comprised the mothers of children who had been screened at different ages there would have

been different influences on the mothers' emotional state at these different stages in the development of the child. However, concerns that the screening infants in the neonatal period might have adverse effects on the mother infant relationship (Young & Andrews 2001) mean that it is important to evaluate the emotional distress this screening programme generates in comparison with the existing screening programme.

In conclusion the results of this study do not suggest that testing in the newborn period causes greater emotional distress than later testing.

4.6 Job satisfaction in newborn hearing screeners: a comparison of hospital-based screeners and community-based Health Visitors

Abstract

Background: Newborn hearing screening is being implemented in England in two different ways: hospital-based dedicated screeners who are recruited and trained specifically to test babies' hearing prior to discharge from the maternity unit; and community-based Health Visitors who conduct the screening at their routine postnatal home visit. While community-based Health Visitor screeners are relatively well paid, reflecting their professional status, hospital-based dedicated screeners have, as yet limited, professional status and relatively low salaries.

Aim: This study compares the job satisfaction of the two types of screener.

Methods: All 124 hospital-based dedicated screeners and a random sample of 124 community-based Health Visitor screeners were sent postal questionnaires. The job satisfaction sub-scale of *The Nurse Stress Index* formed the main outcome measure of the questionnaire.

Results: A response rate of 94% (116/124) was achieved among dedicated hospital-based screeners and 81% (101/124) among the community-based Health Visitor screeners. Total job satisfaction was significantly higher among the hospital-based dedicated screeners than the community-based Health Visitor screeners. For both groups satisfaction was predicted by the extent to which they felt people listened to them at work, their job met career aspirations and they were satisfied with their salaries. In addition, feeling part of the team at work predicted satisfaction among hospital screeners.

Conclusions: Despite relatively poor pay and limited career opportunities, dedicated hospital screeners reported greater job satisfaction than community-based Health Visitors conducting newborn hearing screening.

4.6.1 Background

4.6.1.1 Job satisfaction in healthcare professionals

Job satisfaction among health care professionals is now recognised as important to the delivery of effective health services. Job satisfaction and stress in healthcare professionals are negatively correlated: low levels of job satisfaction are associated with higher stress levels (Healy & Mckay 2000, Tyson *et al* 2002). In turn, stressed healthcare professionals give poorer care (Firth-Cozens & Greenhalgh 1997; Firth-Cozens 1999) and have less satisfied patients (Linn *et al* 1985, Baker *et al* 2000, Haas *et al* 2000). Job satisfaction, however, appears to form a buffer between job stressors and psychological strain (Kalliath & Morris 2002) enabling health professionals to provide effective care even in challenging and demanding environments.

4.6.1.2 The NHSP

One of the areas of interest for the evaluation of the first phase of the NHSP is a comparison of the hospital and community modes of delivering newborn hearing screening. Screening in the two settings differs in a number of important ways including the staff who conduct the screening. In hospital sites screening is conducted by health care workers whose only role is that of newborn hearing screening. These dedicated screeners have as yet limited professional status or career structure and are paid less than Health Visitors. The average salary for dedicated hospital screeners is in the range £9,729 to £10,803 (year 2003 figures). In contrast, the Health Visitors who implement the screening at community sites are provided with a

career structure and also status as professionals within the health care team. Health visitors are senior nurses with specialist training in community health who work as autonomous practitioners, with particular concern for the health of children and families. They have a varied workload of which newborn hearing screening is only a part, and earn two and a half to five times as much as the dedicated hospital-based screeners. The contrasting employment conditions of hospital-based dedicated screeners and community-based Health Visitor screeners would be expected to contribute to differing levels of job satisfaction given that structural characteristics of work, including pay have been found to predict job satisfaction (Seo *et al* 2004). Newborn hearing screening, particularly in those recalled, can cause emotional distress to families at what is already a demanding time. Satisfaction with care, facilitated by the satisfaction of the screener, may help to ameliorate any stress parents might experience as a result of screening.

The aim of the current study is to describe and compare levels of job satisfaction in hospital and community-based screeners and to identify the factors associated with it.

4.6.2 Method

4.6.2.1 Study Design

A descriptive study was used to compare the job satisfaction of the two types of screener.

4.6.2.2 Measures

The Nurse Stress Index. The Nurse Stress Index (NSI) was developed to measure the stress that nurses experience (Harris 1989; Williams & Cooper 1997). It includes a factor, comprising six items, that measures job satisfaction. Minor modifications to the wording were made for the current study to ensure that the items were appropriate for non-nursing staff. The Cronbach's alpha for the scale in the current sample was 0.84.

Potential Predictors of Satisfaction. Potential predictors were identified from a consideration of the existing research on satisfaction in health care professionals and are listed in table 4.11. Items 1 to 5 were assessed using Likert response options ranging from strongly agree to strongly disagree. Items 5 to 7 were assessed using 7 point rating scales anchored at either end with "not at all" and "extremely highly". The questions "what do you find most satisfying about your job" and "what do you find least satisfying about your job" were asked with space provided for open-ended responses.

Community-based Health Visitor screeners were specifically asked to answer job satisfaction items in relation to their whole job, not just in relation to their role as newborn hearing screeners.

Demographic information. Age, educational and employment details were collected as shown in table 4.12.

4.6.2.3 The Sample

124 dedicated screeners, the total number of hospital-based dedicated screeners employed at the first phase hospital sites, were invited to participate in the study. From the total number of

375 Health Visitors who had been trained to conduct newborn hearing screening at the community sites included in the first phase, a random sample of 124 was invited to participate. Randomisation was achieved by giving all these community-based Health Visitor screeners a number, entering the numbers into SPSS and using SPSS to generate a random sample. Because hearing screening comprises only a part of the workload of community-based Health Visitor screeners, those Health Visitors who, although trained to conduct the screening had not conducted a screen as part of their work at the time of the study, were excluded from the study.

4.6.2.4 Procedure

Each eligible participant was sent a pack containing a letter inviting them to participate in the study, a copy of the questionnaire, a freepost return envelope and a freepost reply card. Questionnaire packs were sent directly to the work addresses of community screeners. The packs for hospital screeners were distributed via the newborn hearing screening co-ordinators at the hospital sites. In order to maintain anonymity while maximising response rates, participants were asked to return the questionnaires anonymously in the freepost envelope and at the same time to return, separately, their named reply card. Two weeks after sending out the questionnaires, those who had not returned reply cards were sent a reminder to complete the questionnaire.

Job satisfaction item	**Hospital-based dedicated screeners (n=116)**	**Community-based Health Visitor screeners (n=96)**	***P*-value**
Item 1: People listen to and value my views at work. (5-pt scale)	3.89 (1.01)	3.49 (0.99)	.005
Item 2: I feel part of the team at work. (5-pt scale)	4.28(1.06)	3.98(0.94)	.036
Item 3: My job meets my career aspirations. (5-pt scale)	3.19(1.22)	3.29(1.08)	.543
Item 4: I am satisfied with my current salary. (5-pt scale)	1.84(1.08)	2.85(1.24)	.001
Item 5: To what extent do you value your work as an NHSP screener. (7-pt scale)	6.38(0.97)	5.56(1.51)	.001
Item 6: To what extent do parents value your work as an NHSP screener. (7-pt scale)	5.71(0.97)	5.70(1.56)	.961
Item 7: To what extent do colleagues value your work as an NHSP screener? (7-pt scale)	4.53(1.45)	4.26(1.56)	.207
I am satisfied with my current situation at work. (5-pt scale)	3.76(1.04)	3.45(0.86)	.022
I am satisfied with my involvement in decision-making at work. (5-pt scale)	3.67(1.16)	3.27(0.96)	.009
I am satisfied with the degree of support I receive in my job. (5-pt scale)	4.00(1.13)	3.31(.095)	.001
I seldom think about finding another job within healthcare. (5-pt scale)	3.53(1.19)	3.21(1.33)	.07
I seldom think about finding another occupation. (5-pt scale)	3.61(1.22)	3.22(1.51)	.013
Total job satisfaction subscale.	3.71(0.93)	3.28(0.81)	.001

Table 4.11. Job Satisfaction in Hospital And Community Screeners (M(SD)).

4.6.2.5 Data analysis

Job satisfaction was compared between the two types of newborn hearing screener, using t-tests. Hierarchical multiple regression was used to explore the predictors of job satisfaction, the job satisfaction measure from the Nurse Stress Index being used as the criterion variable and job satisfaction items as predictor variables. In the first step of this analysis, demographic variables were entered into the model; on the second step, the predictor variables were entered; on the third step the mode of implementation was added; and, on the fourth step, an interaction variable of study group by each of the predictor variables was added.

4.6.3 Results

A response rate of 88% was achieved (217/248), 94% among dedicated hospital-based screeners (116/124) and 81% among community-based HV screeners (101/124).

Screener Characteristic	Hospital-based dedicated screeners (n=116)	Community-based Health Visitor screeners (n=100)
Age (Mean/range)	37.9 (20.0-59.0)	46.1 (30-65)
Education (%(n))		
No qualifications	2(2)	0
GCSE level	35(40)	1(1)
GCE A'level	22(25)	2(2)
Further/higher education	30(34)	27(27)
Degree	11(13)	70(70)
Most recent previous job (%(n))		
NHS professional	15(16)	84(81)
NHS care worker	12(12)	0
NHS administration	11(12)	1(1)
Non-NHS professional	9(9)	3(3)
Non-NHS care worker	7(7)	1(1)
Retail/customer services	11(11)	0
Office work	7(7)	0
Domestic services	1(1)	0
In education	11(11)	2(2)
Raising a family	13(13)	8(8)
Other	5(5)	1(1)

Table 4.12. Demographic characteristics and previous types of work of participants.

4.6.3.1 Demographic characteristics

Hospital-based dedicated screeners and community-based Health Visitor screeners differed in their demographic and previous job experiences (table 4.11). Community-based Health Visitor screeners were somewhat older than hospital-based dedicated screeners and also had a higher level of education. While hospital-based dedicated screeners were drawn from a wide

variety of previous occupations, most community-based Health Visitor screeners had been most recently previously employed as an NHS health professional.

4.6.3.2 Job satisfaction of newborn hearing screeners

Hospital-based dedicated screeners expressed a significantly higher level of job satisfaction than did community-based Health Visitor screeners ($t(205)=3.547$, $p=0.001$ see table 4.13). Reflecting this, hospital-based dedicated screeners felt significantly more strongly than community-based Health Visitor screeners that people listened to them at work ($t(205)= 2.847$, $p=0.005$), that they were part of the team at work ($t(206)=2.116$, $p=0.036$) and that they valued their work as a screener ($t(209)=4.585$, $p=0.001$). Community-based Health Visitor screeners expressed a significantly higher degree of satisfaction with only one potential predictor of job satisfaction, namely satisfaction with their salary ($t(204)=-6.226$, $p=0.001$.)

Hierarchical multiple regression explained total job satisfaction well, (R_2 =.666, $p=0.002$). The demographic variables entered in the first step did not provide a significant model (R_2 =.014 $p=0.095$). The job satisfaction variables entered into the model on the second step added 62% to the variance explained. Significant predictors in this model of total job satisfaction were that people listen at work, feeling part of the work team and that the job meets career aspirations. When the variable group, that is, whether the screeners worked as hospital-based dedicated or community-based Health Visitor screeners, was added in the third step, the variance explained by the model increased by 1% (R_2 = .636, $p<0.001$).

In the final step new interaction variables of "group by job satisfaction" variables were entered into the regression equation. The variables "feeling part of the work" team, "satisfaction with salary" and "job meets career aspirations" were all significant, indicating that these variables differed between the two groups in the way that they predicted job satisfaction.

Step	Variable	**Partial Correlation**	**Beta (Step 1)**	**Beta (Step2)**	**Beta (Step 3)**	**Beta (Step 4)**	**Increase R2**	**Overall AdjustedR2**
1	Education	-.057	-.146*	-.090*	-.037	-.036		
	Age	.108	-.044	.007	.052	.072	.014	.014
2	People listen at work	.252		.282***	.263***	.260***		
	Feel part of the work team	.395		.270***	.269***	.390***		
	Satisfied with salary	.158		-.036	.007	.193**		
	Value work as a screener	.248		.106	.061	.136		
	Parents value work	-.069		-.101	-.075	-.081		
	Colleagues value work	.080		.048	.048	.072		
	Job meets career aspirations	.303		.431***	.428***	.284***	.618***	.625***
3	Group	.122			-.155*	.499	.012*	.636***
4	Group x people listen at work	-.009				-.023		
	Group x feel part of the work team	-.187				-.570*		
	Group x satisfied with salary	-.249				-.458***		
	Group x value work as a screener	-.068				-.283		
	Group x parents value work	.018				.078		
	Group x colleagues value work	-.017				-.035		
	Group x job meets career aspirations	.237				.542***	.040**	.666***

*=p<0.05, ** p<0.01, ***p<0.001

Table 4.13. Hierarchical Multiple Regression of Job Satisfaction. N=208, adjusted R2 = 0.67

4.6.3.3 Screeners descriptions of the most and least satisfying aspects of their work

The way in which predictor variables affected job satisfaction is illustrated by the comments that the screeners made about their work. A selection of these comments is shown in Boxes 4.1 and 4.2.

Box 4.1. Screeners comments on the predictors of job satisfaction in relation to most satisfying aspects of their job.

Feeling part of a team:
"I also enjoy working with my fellow screeners-we have developed an excellent team spirit." (Hospital-based dedicated screener 195)
"Working with an excellent team and colleagues." (Community-based Health Visitor screener 130)
Job meets career aspirations:
"Feeling it is a rewarding useful job." (Hospital-based dedicated screener 64)
"Doing something valuable – not just making money for someone else." (Hospital-based dedicated screener 74)
"Health visiting-all aspects." (Community-based Health Visitor screener 171)
Value placed on role as screener by the screener:
"The feeling that you are doing something beneficial for such a young child that can affect the rest of his/her life." (Hospital-based dedicated screener 66)
"As a Health Visitor I value the OAE screening and am pleased to be able to offer this service to parents." (Community-based Health Visitor screener 136)
Value placed on role as screener by parents:
"Parents appreciate the test-reassures in most cases." (Community-based Health Visitor screener105)
Value placed on role as screener by non screening colleagues:
"Meeting grateful parents and SCBU nurses too." (Hospital-based dedicated screener 9)

Box 4.2. Screeners comments on the predictors of job satisfaction in relation to the least satisfying aspects of their job.

People listen to and value my views at work:
"Work 'inflicted' on my profession without proper (or sometimes no) consultation/debate or discussion." (Community-based Health Visitor screener 117)
"Not being listened to by management who are only interested in putting nice sounding phrases on paper reports." (Community-based Health Visitor screener 132)
Feeling part of the team at work:
"Working on my own." (Hospital-based dedicated screener5)
"Lack of support and communication." (Community-based Health Visitor screener 128)
Job meets career aspirations:
"One of the most difficult aspects is that there appears to be no career progression for screeners. It is easy to become bored with no involvement with parents or babies who are referred." (Hospital-based dedicated screener 13)
"Often work is brain numbing, too routine and not being able to utilise professional skills for public health practice approach to job." (Community-based Health Visitor screener 116)
Satisfied with current salary:
"Having now being doing my new role for months now, I can clearly see the pay doesn't match what we actually do and are responsible for on a daily basis. If it isn't addressed, screeners will become dissatisfied and you will loose them." (Hospital-based dedicated screener 95)
"I think the pay is an insult." (Hospital-based dedicated screener 56)
Value placed on role as screener by the screener:
"It should be conducted in hospital by other personnel. It provokes anxiety in both client and screener and I believe it is not conducive to initiating a working relationship with my clients." (Community-based Health Visitor screener 117)
Value placed on role as screener by non screening colleagues:
"Sometimes I feel that the service that we offer to parents is not looked upon seriously or valued by other health care professionals." (Hospital-based dedicated screener 25)

4.6.4 Discussion

Both hospital and community screeners expressed a relatively high degree of satisfaction with their jobs, with small standard deviations suggesting low variability. The mean scores on the job satisfaction sub-scale of the NSI are comparable to those found in samples of nurses (Healy & Mckay 2000, McGowan 2001) and dental hygenists (Gibbons *et al* 2000, 2001). Overall, hospital-based dedicated screeners indicated higher levels of job satisfaction than did community-based Health Visitor screeners. For all screeners, the variables "people listen at work", "feeling part of the work team" and "job meets career aspirations" were all significant predictors of job satisfaction. In addition the variables "feeling part of the work team", "satisfaction with salary" and "job meets career aspirations" differed significantly between dedicated and Health Visitor screeners.

Adams and Bond (Adams & Bond 2000) found that job satisfaction increased from lower to higher grades of nurses. This is contrary to the findings of the current study, in which the hospital-based dedicated screeners, who occupy a more junior position within the healthcare hierarchy than the community-based Health Visitor screeners, had higher levels of job satisfaction. However, the comments that the community-based Health Visitor screeners made, (see Box 1), suggest high levels of frustration with their management structures which may have led to their greater job dissatisfaction. By contrast the senior nurses in the study of Adams and Bond worked in hospitals and would therefore have worked within a wider team. Health visitors work away from the structures that an institutional setting provides which may heighten the need for sensitive management and a supportive interpersonal environment. Evidence to support this in the current study comes from the observation that community-based Health Visitor screeners felt less strongly than hospital-based dedicated screeners that their views were listened to and that they were included as part of a team, both variables that predicted job satisfaction (Adams & Bond 2000).

Community-based Health Visitor screeners indicated significantly higher satisfaction with their salary than did hospital-based dedicated screeners. However it should be noted that, for both types of screeners, mean satisfaction with salary was lower than for any other variable. In addition, many of the hospital-based dedicated screeners expressed considerable dissatisfaction with their salary, to the extent that their dissatisfaction led some of them to consider leaving their jobs. None of the variables in relation to the value placed on the work as an NHSP screener was a significant predictor of satisfaction. Although hospital-based dedicated screeners did value their work as an NHSP screener significantly more highly than did community-based Health Visitor screeners, this may well reflect the fact that hearing screening is only a small part of the workload of community-based Health Visitor screeners.

4.6.4.1 Strengths and limitations of the study

The study achieved an excellent response rate and therefore these findings are likely to be representative of the views screeners held about their jobs. One possible limitation of this study was the use of the NSI job satisfaction measure. The NSI was developed specifically for use among nurses. Although the issues facing hospital-based dedicated screeners might be similar to those faced by hospital nurses, an instrument that was not specific to nurses might have been more sensitive to detecting differences between the two types of screener not tapped by the NSI, for example satisfaction with the variety of tasks in their jobs. However, such instruments, such as those identified in a recent systematic review (van Saane *et al* 2003) are typically multidimensional consisting of several scales which would have made the

current questionnaire too long. A further limitation of the current study comes from trying to compare two types of healthcare worker with very different roles and work environments. However, as policy decisions about the future implementation of NHSP involve decisions about the extent to which the community-based model of screening should be incorporated in NHSP, direct comparison of aspects of these different modes of implementation were felt to be appropriate.

4.6.5 Implications and conclusions

Hospital-based dedicated screeners reported higher levels of job satisfaction than community-based Health Visitor screeners. Although the two groups differed in their levels of job satisfaction, their satisfaction was influenced by similar factors. Thus their different levels of overall satisfaction reflect differences in levels of the same predictors. These results have implications for policy formation as new screening programmes are developed. They suggest that hospital-based dedicated screeners can have high levels of satisfaction which can contribute to providing a service with which patients are also satisfied (Linn *et al* 1985, Haas *et al* 2000) and are therefore an appropriate addition to the healthcare workforce. However, while the community-based Health Visitor screeners had many years of experience, hospital-based dedicated screeners at the first phase stage would have had relatively little experience of this work, decreasing the time over which dissatisfaction with the job could build up. Over time, the impact of dissatisfaction with pay might impact on the wider job satisfaction of these screeners, particularly if the support that has been available in the early stages of the programme were to decrease. Further evaluation of the long-term job satisfaction of these screeners is needed before the introduction of dedicated screeners is advocated in other screening programmes.

5. THE TRUE CASE STUDY – THE EXPERIENCE OF PARENTS[8] WHOSE CHILDREN HAVE BEEN CORRECTLY IDENTIFIED AS DEAF THROUGH THE SCREEN

5.1 Introduction and methodological approach

Parents are uniquely placed to inform the evaluation of the introduction of newborn hearing screening. Their inclusion in the research is not simply about including their perspective, or ensuring they have a 'voice'. It is also about exploiting their epistemological privilege; that is to say, providing the opportunity for *their* definition of what should be known about this event and the terms in which it should be known. In designing the study we were, therefore, concerned to choose a methodological approach that would uncover rather than predefine what was important in parents' experiences, enable them to set the conceptual and experiential criteria against which the success of the experience should be judged, and promote both confirmation of and challenge to professional assumptions and practices.

The methodological approach is a qualitative one, based on narrative. Parents are invited to tell their own stories, in their own words, within the broad framework of covering: the experience of the screening from first screening test; through referral and diagnostic assessment to confirmation; the experience of early intervention and professional support; and their advice to other parents and professionals engaged in the same process. The interviewer's job is to clarify points in the narrative as it progresses to ensure information is collected about comparable events across all interviews undertaken; to support the narrative-telling through empathic engagement with the teller; and to record the interview for later analysis. In this way, parents do not respond to a set of pre-defined questions in which to fit their experience, but rather are given the scope to make decisions themselves about what is meaningful and important in their experiences, and to set the criteria by which they would want their experience to be understood and evaluated. Further details of methodological approach can be found in Young *et al* (2004).

[8] The term 'parent' is used throughout to include also principal caregivers.

5.1.1 Details of method, sample and analysis

5.1.1.1 Aims

- to evaluate the impact of the screening process and its consequences for intervention from the perspective of parents of true cases
- to explore socio-demographic influences on parents' experience
- to enable parents to contribute to the identification of what is good practice

5.1.1.2 Sampling and recruitment

The sample was a purposive one since only those parents whose children fulfilled the definition of a true case identified by NHSP could be invited to participate. To be classed as a true case, the child had to meet the criteria of having 'a permanent bilateral hearing loss with hearing threshold ≥ 40 dB HL based on the average in the better hearing ear at 0.5, 1, 2 and 4 kHz.'

Between the period 1st December 2002 and 31st December 2003, the evaluation team were notified of a total of 108 true cases by the appropriate audiology staff located in each first phase NHSP site. After a six week period the researcher requested the responsible clinician to send the parent letter and information sheet to the parents/family. At this point the name and address of the family was unknown to the researcher. If the family wished to be involved in the study they completed the response sheet with their name and address and sent it directly to the researcher; an interview would then be arranged. If no response was received from the family after three months the host service was asked to send a reminder letter. There were no further reminders.

The invitation to participate as well as the information and consent materials were available in a variety of community languages including in British Sign Language in the format of a video letter. For full details of the recruitment methods used, the creation of parent information materials, a discussion of the ethical issues involved and the challenges of sampling see Young *et al* (2003).

Of the 108 notified true cases, 91 families were invited to participate in the study. In five cases the child had died, ten cases were thought to be auditory neuropathy[9], and in two cases it was unclear whether the child fulfilled the true case criteria. Of the remaining 91 families, 28 responded positively and 27 were interviewed. These 27 interviews involved participation from 45 parents/carers/extended families.

[9] Babies thought to have auditory neuropathy were excluded from this study. The degree of uncertainty relating to this condition would mean that these families' experiences of screening would be significantly different from the experiences of the majority of parents of true cases.

Characteristics of the sample

- 25 from sites with hospital-based screening, 2 from sites with community-based screening (but of these one baby was screened in NICU and therefore the experience was more similar to hospital-based screening)
- 6 babies from the NICU population
- 2 families had other deaf children
- 22 per cent of infants had disabilities/illnesses
- In 11 of the 27 families the deaf child was their first child
- Five families from black/minority ethnic backgrounds (+2 other cross cultural families)
- All parents/carers 'hearing' (but 2 with unilateral losses)
- Bias towards high-income families (12 out of 27 had family incomes of £35,000 or over)
- In three cases languages other than English used in the interviews [one more family used a language in addition to English at home but not in the interview] [10]
- Degree of deafness identified in the babies: 44% moderate; 19% severe; 37% profound.

5.1.1.3 Data collection

Parents completed a narrative-based interview lasting on average one and a half hours. All parents chose to be interviewed at home. They chose who should participate e.g. in some cases both parents were present, other interviews were with one parent alone, others included extended family members where they had a significant care-giving role. In addition parents completed a simple questionnaire to collect socio-demographic information.

[10] For a discussion of qualitative data handling and analysis where data are collected in more than one language/modality see: Temple & Young (2004)

5.1.1.4 Analysis

Data were audio recorded and transcribed in full. A thematic content analysis was carried out with the assistance of the sort and retrive programme QSR NUD*IST 4. This analysis used cross-sectional techniques from both within case and cross case perspectives.

5.1.2 Selection of findings to be discussed

We will confine the discussion to findings from three particular stages in parental experience: (i) the period of time from the start of screening to the referral for diagnostic assessment; (ii) the waiting time between the end of the screen and the start of diagnostic assessment; (iii) the experience of diagnostic assessment and confirmation of deafness. In presenting these data, attention will be paid to the variation in parents' experience as much as the similarity. In what follows the code numbers used relate to specific parents/families and so it is possible to see the development of experience of the same families across different segments of data presentation. All names have been changed and identifying features removed. Quotations used are indicative.

5.2 The period of time from the start of screening to the referral for diagnostic assessment

5.2.1 Introduction

This period of time from first screening test to referral is particularly interesting because it represents a new condition for parents of deaf children. In the past much research attention has been focussed on parents' recollection of the usually protracted process of discovering their child was deaf, its attendant frustrations, and the extent to which parents 'knew' long before it was ever confirmed. Now, not only is that process condensed in terms of timescale, but the discovery emphasis has changed. Instead of deafness being something that in many cases emerges over time and with the experience of the developing child, it becomes instead something almost immediately identifiable. Instead of parents' suspicions often being instrumental in that discovery, it is technology and postnatal procedures that take over that process. This latter shift, from experiential and developmental discovery to routine investigation, is particularly powerful because of the unseen nature of deafness. Unlike many postnatal conditions deafness is usually not one with visible markers to indicate its presence, so the idea that the invisible can be detected, and in a way that is not dependent on parental experience or milestones of child development, is especially striking.

For the parents in our sample, the screening experience in this period of time from birth to referral is marked by one overwhelming theme: how to interpret the *inconclusive* message that each stage of the screening delivers. That is to say, with each screening event comes the message that there is no clear response and a further screening event is required. This is a message that is neither positive nor negative, certain nor uncertain – it cannot be. In what follows we will focus on:

- what parents understood by their child requiring further screening tests
- the impact of that knowledge and how they handled what happened next
- what influenced variations in parents' experiences of that inconclusive outcome
- what makes for a 'good' screening experience

In addressing these issues, parents fell into three broad groups:

- The inconclusive message gave little or no cause for concern
- The inconclusive message did give cause for concern but that was linked by parents to other factors in their lives/contexts rather than to anything about the process of the screen itself.

- The inconclusive message did give cause for concern and was linked by parents to issues in the process/delivery of the screening[11].

5.2.2 Parents for whom the inconclusive message gave little or no cause for concern

For around a half of parents the idea that the screen had produced an inconclusive result and their baby needed to be screened again was not a particular cause for concern. The following were typical:

> *"they did it a few times, obviously with the consent, but I wasn't too bothered about it." [12]*[12]

> *"he was tested the day after he was born in hospital and failed that which wasn't a worry 'cos she said many babies fail that one…and it's kind of not alarming or worrying" [23]*

It was only as the process progressed further following referral that for some, (not all), concern did set in. Compare:

> *"I don't think we were worried at that point, it was only when he failed the second one the next day, that we started to really worry about it…" [03]*

with:

> *"I got a leaflet saying a lot of babies are referred, it could be this, that or the other, so I wasn't unduly worried" [06]*

From parents' perspective there were two main factors that ensured they did not interpret the inconclusive result and the need to test again too anxiously:

- the manner in which the screener went about his/her job and

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[11] Each group is considered in turn, however, contrasting examples from parents outside the group being considered are also sometimes used to illustrate a point by way of exceptions to the experience casting light on their opposite.

[12] Numbers in square brackets after quotations are codes for each respondent. Their inclusion enables the reader to see the range of parents from whom we have drawn illustrative examples and also to track particular parental experiences through the different stages of data presentation.

- the content of the explanation they were given.

5.2.2.1 The importance of a reassuring screener manner

Many parents commented on how they valued how "kind", "patient", "nice", and "understanding" screeners were and how their confidence and reassurance was key to parents not being particularly worried by the fact that their baby needed to be screened again. Their patience and sensitivity in handling a new baby was also commented on. For example:

> *"Certainly the lady, the day after she was born, was very reassuring and it was reassuring that she came back when she said she would." [01]*

> *"…she was extremely nice. We both went in and she was very understanding 'cos it must be difficult doing tests on new babies 'cos they don't do what you want them to do…she explained every test as it went and she was very patient and I think Lucy might have even wanted to be fed half way though and she was very understanding…" [24]*

What is interesting in parents' reflections on screener reassurance is that they are not just about what screeners say, but how they seem as people. The descriptions used are often about the screeners' personality and character, not just their professional communication.

5.2.2.2 Looking back, was the reassuring approach the right one to have taken?

Even looking back now with the knowledge parents had that they had a deaf child, the vast majority who appreciated the reassurance that accompanied the inconclusive message from the screen, still appreciated it now. Most of this group of parents still thought, with hindsight, that playing down the possibility that the screen result might indicate deafness was exactly the right thing to do, otherwise they would have become alarmed unnecessarily.

This mother contrasts the reassuring approach of the screen (even though the tests were inconclusive) with that of the paediatrician who was equally unsure about a possible heart murmur the baby might have:

> *"I think it was better that she was reassuring in hospital 'cos when we went to see the paediatrician he said…'I'm not sure if I can hear a murmur or not' and that was it, I was in floods of tears…a heart murmur, I thought oh my god…[but] there wasn't nothing wrong, he got a second opinion and there was nothing at all…I don't think it [the screen] could have been done differently because I think we needed reassurance, because when you've just had a baby you're sort of all over the place…" [09]*

5.2.2.2.1 The content of the screener message – when and how does an explanation work?

Clearly what a screener says and how they say it are not easily separable – both are inextricably linked to an outcome of reassurance (or not). However, in this analysis manner and content are, to some extent, being artificially separated, because parents demonstrated some unexpected interpretations of the content of what they were being told and had some good ideas about what kinds of information it was best to give and what it was best to withhold.

Parents expressed a clear preference for being given a reason why the screen or test result might be inconclusive. Having a likely explanation made the fact that the results were not straightforward much easier to cope with.

> *"…they had reassured me 'don't worry too much I'm sure everything will be OK' and there were lots of reasons why she hadn't passed it such as fluid in the birth canal, the ears after being born, it could be a few other reasons I can't remember. And it's not you know, they didn't say she wasn't deaf, but they'd tried to say there's lots of reasons why you've got this results so not too worry too much…" [13]*

> *"I reckon she did a good job to be honest with you…she explained everything, what she was doing and when it turned out she didn't get any responses …why she ain't getting a response and what she'd do then…" [16]*

One of the interesting effects of parents being given reasons for why a test might be inconclusive is that for many it shifted attention away from the idea that there might be something wrong with their baby's hearing, to the idea that there might be something wrong with the test itself. What was not working was the test, rather than their baby's hearing. So, for example, the common message that the test was probably not definitive because the baby's ears were congested, was not usually interpreted to indicate that congestion meant that at least temporarily the child's hearing was not fully functioning. Rather it was very commonly interpreted as meaning the screening system/technology itself was not very good because it could not work if the baby's ears were congested. In retrospect, many parents put down their lack of undue anxiety to their understanding that the screening tests themselves were not particularly "good" or "sensitive" or "proper". As one parent pointed out, being worried about a baby not passing the screen is entirely different from being worried about a baby being deaf.

> *"… we weren't expecting there to be any problem, obviously, so we didn't really think about it. She said the test might not work. We weren't concerned when it didn't…we just thought it was one of those things that he was too young to test and problems doing the actual test itself with them having to be completely still." [17]*

> *"I just thought it was one of those things, perhaps it's down to their equipment, you know and didn't think any more about it." [22]*

Parents reported far less satisfaction with the screening process and more anxiety about why their baby had not passed in those situations where they felt the message they had been given was 'vague', without a context, or where the explanations offered did not strike them as credible. These experiences are discussed in the following sections (note also that the issue of anxiety levels and knowledge in mothers of screened babies who were not true cases is discussed in Chapter 4).

5.2.3 The inconclusive message did give cause for concern but that was linked by parents to other factors in their lives/contexts rather than to anything about the process of the screen itself

As parents themselves reflected, even though the manner of the screen is reassuring and the explanation good, you cannot legislate for other influences on how parents might experience it. There was a smaller group of parents who for a variety of reasons had other influences in their lives that meant that the inconclusive message that came with the screening experience did ring alarm bells for them and create anxiety.

5.2.3.1 Characteristic appraisal styles

As one parent pointed out, screeners cannot control for how their well-meaning explanations might be interpreted. The facts they offer inevitably interact with the kind of person you are as a parent and the kinds of appraisal you have a tendency to make of situations. In her case, the idea that very few babies that are referred turn out to be deaf was not necessarily comforting. As the screener explained to her that since the start of screening only 2 or 3 babies had been picked up in that area, she was thinking that did not mean hers would not be the next! She was just that sort of person. It should be said, however, that other parents in our sample found specific information from screeners about how few babies were identified especially comforting because it gave them a more realistic context for what they might be fearing. One message will never fit all.

5.2.3.2 Deafness in the family

In the case of two families, they discussed the fact that there was deafness already in the family made them think that it was more likely that their children were not passing the screen because they did have a hearing loss despite the reassurance they were receiving. They described themselves, therefore, as worried by the outcome of the screen but linked that with the family knowledge rather than concern over the outcome of the screen being inconclusive per se.

5.2.3.3 Other sorts of pre-existing knowledge

One father reflected that he was perhaps more suspicious than most parents because he was himself a medical professional, although his wife had not had any doubts about the reassurance she had been given. He took the inconclusive outcome of the screening as his cue to start testing his baby at home with loud noises etc whilst waiting for his follow-up appointment with audiology. By the time that came he was fairly sure his child had a hearing loss, but his wife was still reassured that nothing was certain because of what she had been told about the screen.

5.2.3.4 Instinct

Another mother noted her tendency to doubt the reassurance she had received following the end of screen:

"They came and like just did a bit and said they weren't getting no response from her and they said it could have been 'cos she could still like have fluid and that in her ears…so I got an appointment…but like I said to my cousin, the day I were going, I said I'm gonna have to go to this appointment because I've just got this funny feeling she's gonna be deaf…anyway I took her and like they did that same test as what they did first time and like weren't getting no response still and I knew then, I thought I know I were right." [25]

Once again this mother had no criticism of the screen, it was just that she felt she knew better. [This experience stands in contrast to other parents (see next section) who also had a 'feeling' their baby was deaf but who blamed the screen directly for not being able to deliver a definitive result that would have supported that feeling].

5.2.3.5 Physical signs

For another parent, whose child had additional needs, the inconclusive screening result did make her think it was more likely that her child had a hearing loss, but she linked this firmly to the fact that her child had other obvious problems, so it might be expected. In these circumstances that fact the screening was being done quickly was seen as supportive in helping her to prepare for the possible outcome of deafness.

"they were within 24 hours of him being born and both tests showed kind of basically no hearing, nothing that was conclusive…at that point we were thinking you know he could be profoundly deaf…we had to look at the worst case scenario, so you know, he had the ear problems, certainly no [ear] canal on one side, could be profoundly deaf, so that was quite a lot to take in…

…it's not always conclusive [the hearing screen] and it can be [because of] a number of conditions…that can mean you don't get the right response, but at least it gives you the opportunity, if there isn't a right response, to make sure there is early intervention and to assimilate what it is." [05]

However, it should be noted that there were other parents in the sample with children with additional needs who did not respond as positively as this mother had done. As we will see (below) for these parents the difficulty lay in a seeming failure to acknowledge the possibility of a connection between the screening outcome and physical problems they could actually see the child had.

5.2.4 The inconclusive message did give cause for concern and was linked by parents to issues in the process/delivery of the screening

5.2.4.1 Introduction

In looking in fine detail at the following parents' experiences it is important to bear in mind the overall context i.e. that for the vast majority of parents in our sample screening was satisfactory and highly valued (although parents may have differed on how reassured/anxious they felt). For the seven parents in this group, the screening process caused significant concern and they linked that directly to issues in how the screen was carried out. Whilst it might be easy to dismiss some of the points they make as 'their' misunderstanding, rather than unsatisfactory practice, the fact remains that their interpretation of what happened was their reality and the one they emotionally and psychologically reacted to. They also do raise important questions about practice. Becoming sensitive to the full range of parental experiences and examining whether some of the more distressing can be predicted and/or avoided is at the heart of the challenge thrown up by these stories.

5.2.4.2 Wanting the possibility of deafness acknowledged

Three couples in particular discussed that it would have been important to them, looking back, if someone had acknowledged that one of the possible reasons for not passing the screen was in fact because their child might have a hearing loss.

One couple had a baby with a range of additional needs and was in NICU at the time of the screening. Their son had very obviously small ears (through a chromosome abnormality) and so from their perception, common sense suggested this might affect his hearing. The problem was that the reassurances and explanations they had from the screener never acknowledged this possibility and diverted attention instead to the difficulties of the test if the child had small ears. Looking back they were very annoyed that reassurances continued despite what they regarded as the evidence of their own eyes.

> *"that newborn hearing test, I think is absolutely appalling…they shouldn't have been so reassuring that test was not a good test [because] obviously it was a good test and it did work because it came back as inconclusive both times and then a referral*

> *…the lady that was doing the test could have said, like I said to you before, could have said 'it could be that he's got a hearing loss or it could be that the machine can't work with his little ears' rather than like they just said the machine can't work with his ears, you know, it's obviously his ears that are affecting his ears as opposed to the machine…" [10]*

In part, their difficulties with the reassurances they received were also based on the fact that they had made observations themselves about their child's hearing over the period of time he was in NICU and these had led the mother to admit the possibility that their son had a hearing

loss. But once again because this was never explicitly considered as a possibility within the screening then it made the parents' responses to the screening experience more negative.

> *"I think it's quite hard because we, I knew, he couldn't hear. Babies, they jump to noises. Billy, didn't jump. One of the nurses knocked a metal tray right next to him and he didn't flinch and you could call him and he didn't look. At about six weeks babies start to look for your voice and smile and interact. Billy didn't, but they were still reassuring us that it was OK." [10]*

From this mother's perspective the cumulative effect of too much reassurance without mentioning the possibility of deafness was that whilst she was prepared for the eventual diagnosis, her husband was not. He had followed what the screener had told him and invested authority in the testing procedures, rather than believe what seemed little more than his wife's personal and unfounded suspicions.

Another mother was very concerned that nobody had acknowledged in their reassuring explanations after the first screening test, that her baby might actually fail the second screening test. This meant, from her point of view, that she had not prepared herself for that eventuality and had not had the opportunity to plan appropriate support for herself during the screening process. Consequently she experienced more distress than she might otherwise have done. She said she was so upset that she requested to be discharged early, after only two days, despite having had a caesarean section.

> *"I don't think it was very clear to us…and this is probably us as well as the hospital…it wasn't really explained or we hadn't really thought about what happens if he had failed. When they did the first test it was just me on my own with the baby so there was no-one there at all who could support that, the fact he didn't get through it, so maybe a point would have been if someone had said at that point, you know, 'have you thought about how you would feel if he doesn't get through this test?' and then maybe I would have thought 'oh maybe I should have someone here just in case' 'cos obviously having just had an operation and not slept for two days it's quite a distressing time anyway so to have that on top of that and not have any support…" [03]*

This mother was also very keen for it to be recorded that the timing of the second screen was not helpful because it was just before the one and a half hour period when fathers are not allowed on the ward (because it is mothers' sleep time) so when she was told that her son had not passed the screen for a second time she could not even turn to her partner for support because he was not allowed on the ward.

This mother's situation was also not helped because she never received the standard information given to parents on point of referral and so did not go home with a clear enough explanation about what a referral at the end of the screening process meant, and what the wider context was i.e. that very few babies who are referred are actually deaf.

"I knew what the screening was for, it wasn't that I didn't have enough information in that respect, it was the after care really that was the problem…having had no idea at all that he was going to fail at all, there was nothing after that to be able to give out, you know, to give you answers and after we did the second lot of testing…we were talking about it and asked her, you know, 'does this happen often, you know, should we be worried about it?' and she didn't give us any statistics that made us feel better. That was something I realised with our teacher of the deaf and I think she's taken it up because she told us after the event that of the babies that are referred to the hospital about nine out of ten are fine…now if I'd been told that at the time, although it would not have made a difference, 'cos John had the problem, it would have made me feel better in the interim…" [03]

For another couple, looking back, failure to mention that the baby might not be passing the screen because she might have a hearing loss was now considered unacceptable, because it had robbed them of the possibility of being able to prepare in advance for the eventual diagnosis. The reassuring but inconclusive message they had received was not considered appropriate. For them the problem was that the message stayed the same whilst the odds of the baby being deaf narrowed and concerns were raised. They would have preferred communication that reflected these changing circumstances.

"I mean I can go with the first one you know, he could have had fluid in his ear, but the second one when he failed that you know, they could have said we have concerns, we need another test. Yes you are going to be worried, but you can prepare yourself, because when they are re-testing you are going to be worried whatever, even if the outcome had been you know positive…if they have got concerns, they have not got concerns for nothing and especially with the level of testing now. You know they're doing brainwave patterns…" [15]

4.2.4.3 Believing that failing screening meant their child was definitely deaf

Of considerable concern were the two couples in our sample who believed that their baby being referred from the screen meant that their baby was definitely deaf. In one case, the family already had a deaf child. They just presumed when the baby was referred that the only issue to be resolved was how deaf their new baby was, not whether or not she was deaf. This response is perhaps understandable from a family who must have been veterans of hearing assessments and might have found it hard to distinguish a screen from a hearing test.

Another mother had totally misunderstood what referral from the screen implied. She and her partner had believed that the AABR test was diagnostic and that at two days old they had been told their baby was deaf. With this in mind, the trauma and distress she recounts is entirely understandable as she thought despite the devastating news, she was simply being left alone and nobody was providing her with support and information.

"well I was just desperate, to be honest. I was left, I was in my own room because I'd had a section, I was in my own room. All the equipment and people had gone and I was there left thinking and it was all dawning on me that there was a problem and basically it was very difficult" [13]

Both mother and father were still very angry at what they saw as a failure of support at the end of the screening process, a failure made all the more acute by their false understanding that referral meant deafness.

> *"It's emotive. I think they should have people there who are more knowledgeable and they could say 'well there's this hearing loss, we don't know why it's there, but she's got this. We know that because of the technology that we've used and the sophistication of the equipment' and then there's people like social workers or counsellors or whomever, but someone you can ask a question to get a straight answer. The people who we were dealing with us first were just technicians and they were just giving us instructions on how to operate this piece of equipment with no understanding of why it was doing what it was doing…that's where I think it breaks down. It's like a pyramid, an inverse pyramid, where at the bottom you've got nobody who knows nothing but they're the first people of contact…" [13]*

A few days after the AABR screen a ward doctor picked up the fact that the mother had misunderstood the implications of not passing the screen and a screener came back to talk to her in detail. However whilst the mother, looking back, found this extra information helpful, it only served to reinforce for her the idea that something must be wrong, the only question was what exactly.

It might be easy to dismiss these parents' experiences as exceptional and simply accountable for because of a failure on their part to understand what screening actually was. Certainly a lack of understanding that screening did not equate to diagnosis contributed to their negative response to the experience. But regardless of cause, these parents' experience was deeply upsetting and clearly continued to be. They reinforce how important it is to be alert to those parents who might confuse screening with diagnostic testing in order to avoid unnecessary distress.

4.2.4.4 The effect of not understanding why the screen could not be definitive

As previously discussed, one of the unexpected issues parents brought up concerned their interpretations of the technology that was used in screening. For many, the idea that the technology or the test was not quite good enough was a helpful way to make sense of why their baby had been referred. However, for two parents in our sample, queries about what the technology could or could not do, had precisely the opposite effect.

One couple could not understand why the test could not be definitive (instead of simply pointing to the need for another test). The fact that they were being repeatedly told that the child was not responding only made them believe there must be something really wrong, because tests should work.

> *"…so she just explained that she took Joseph for this screening test and that he'd not responded to it, so I were like 'so what you trying to tell me, that he can't hear anything, that he's deaf?' And she said 'no I'm not telling you that' she says 'I can't tell you 'cos I don't know for definite' so I says to her ' well he either heard it or he didn't so you've got two choices' she said 'well he didn't hear anything'…and then like my heart dropped and so she says 'it could be that he's*

just too premature like to be developed, his ears aren't developed enough so we'll have to do another test'…

…well we wanted to know why he weren't responding, we didn't have no idea…" [26]

Similarly another couple wondered why a hospital simply could not tell them what was wrong with their daughter if clearly she was not passing the tests.

"They came round the hospital, yes. They checked the one ear and one is OK and the other ear they go that they can't tell nothing. It got me worried, yes. I go, why is that for? You are a hospital – what is wrong with her? I was getting really worried." [18]

5.2.4.5 Finding the inconclusive message deliberately misleading

One couple recounted a different kind of inconclusive message than others. The problem was not lack of specificity but rather too much specificity. Instead of simply being told their baby was not responding or the test was inconclusive, they had been told that "there was a little response, but it wasn't an effective response" which they had interpreted as meaning there was problem but the screener was trying to make them feel better about it by suggesting that the problem was not as big as it might be:

"MOTHER: It was a bit misleading I think

FATHER: It was a bit whether she sort of wanted to make us feel better…at least there was something there just to make us feel a bit happier. She didn't want to tell us ' I couldn't get no response at all' at the time, which you know, if we'd just had the baby then that would probably have made us feel really bad sort of thing…

…MOTHER: You shouldn't really say a partial response if there isn't one

FATHER: …'cos you know it's not being truthful with people. " [27]

The longer-term consequence of this experience was this couple found it difficult to have trust in or confidence in the audiology services. The father was beginning to question whether in fact it was the 'tests' that had made their child deaf. [This also was a family who never received the explanatory leaflet at point of referral that might have put the screening outcome into a wider context].

With the benefit of hindsight, these parents would have preferred a more clear explanation from the screener that they simply were not sure why the baby was not responding and so needed to do more tests.

"If she didn't' tell us there was a partial response, if…she said 'you need to go to the hospital because I can't really sort of fathom this out 'cos the thing keeps slipping out and I can't quite get a good response' then we'd have sort of like been a bit more aware you know, could be problems here sort of thing…" [27]

5.2.5 Conclusions: screen up to point of referral

The interviews have successfully captured a wide range of experiences of screening (up until the point of referral) and with them the fine detail of similarities and differences in parents' experiences. These provide us with some important indicators of what works well and cautions for where things might go wrong for some parents. In summary, the main messages are:

- For parents, the defining experience of screening is how to interpret and how to respond to the inconclusive message that each stage of the process delivers.
- For about half of the parents in the sample, the inconclusive message gives little or no concern.
- This lack of concern is assisted by two main factors: the totally reassuring manner of the screener and the content of the explanation offered.
- Positive appraisal of screener manner was not just made on grounds of what they said, but also how they seemed as people – their character and their sensitivity.
- The offering of an explanation why the baby had not passed the screen was important in reducing anxiety. Where explanations were vague parents were more worried.
- For some parents, an important element in that explanation must be an acknowledgement that deafness might be one of the range of explanations why the baby was not passing. This was of particular importance in situations where there were potentially other signs that the baby may be at higher risk (e.g. in NICU, the presence of disabilities, a history of deafness in the family). In these circumstances, to persist with explanations such as the ears might be congested or it may be test that is not good enough, could be infuriating and raise rather than lessen parental concern.
- An explanation that set the screen outcome in a wider context was considered vital i.e. one that showed that few babies that were referred actually had a hearing loss. Where parents were told this, it was very helpful, where parents were not, it added to their growing concerns.
- Caution should be exercised in how specific to make the reassuring message. In a situation where the screener had offered additional information indicating a partial response rather than an inconclusive response, parents interpreted this as an indication of deafness being present, rather than an indication of something positive.
- Many parents readily believed that the reason for the inconclusive message was a problem with the testing/test equipment, rather than potentially a 'problem' with their baby. On the whole, this interpretation was helpful in making them less anxious. It does raise ethical issues about whether and how such false belief should be challenged.

- Those parents who did experience the inconclusive message of the screen as a cause for concern fell into two groups: those who linked their concern with other factors in their lives that made them more likely to interpret the screen result as indicating possible deafness; those who blamed in some way the screening process for making them interpret the screening result as possibly or definitely indicating deafness.
- Reasons parents recognised from their own experiences influencing their interpretation of the screening outcome included: their character in how the tended to appraise ambivalent situations; professional identity; family history of deafness; 'instinct'; other physical signs.
- It is of cause for concern that there were two couples in the sample who believed that the AABR screen was actually diagnostic and that they were being sent home with a deaf child, and with no support until their audiology appointment. These two cases point to the importance of checking that parents really have understood what the screen result implies rather than simply assuming that the reassuring message will of itself be adequate explanation.

5.3 The waiting time between the end of the screen and the start of diagnostic assessment

5.3.1 Introduction

This section concerns the period of time between the end of the screening (which results in a 'refer') and parents first appointment with audiology for diagnostic assessment. We know that over 90% of babies that are referred do not have a hearing loss. By contrast, these families' represent those whose experiences of 'refer' in retrospect turn out to be the next step on the road to discovering their child is deaf.

In what follows we will look at: the variations in the amount of time between the referral and the actual appointment; what parents views are about whether the time they waited was acceptable or not; what underlies parents' different appraisals and experiences of this time; and what parents actually did whilst they were waiting.

5.3.2 Variations in the time between referral and first appointment

There was considerable variation in the amount of time between refer and first appointment amongst our group of parents. Over half of the sample (n = 20) did begin diagnostic assessment within the target period of 4 weeks from referral, with one family beginning that process on the same day of the referral.

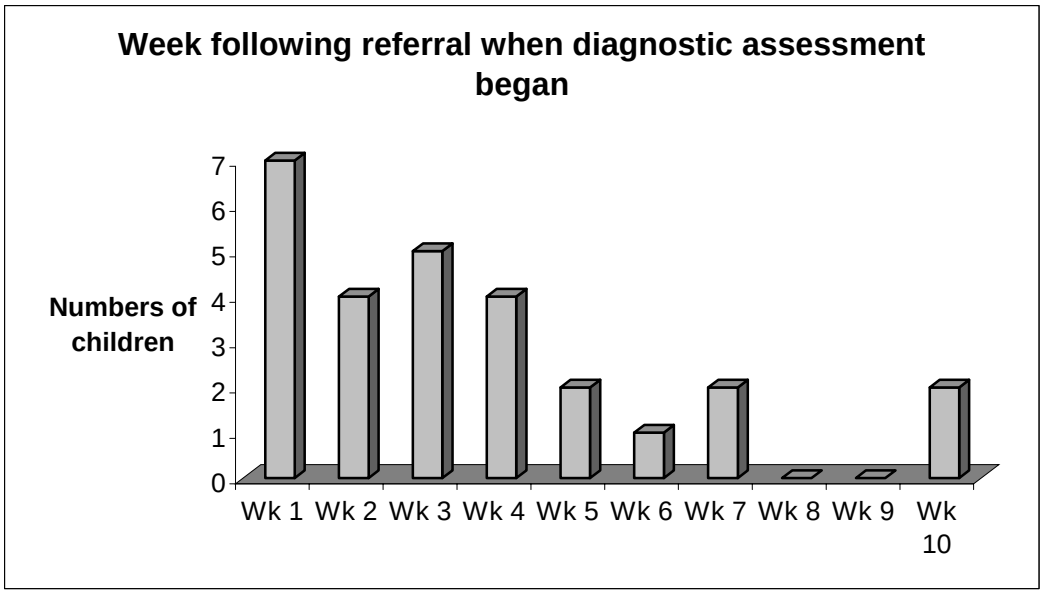


Figure 5.1. Distribution of children by the delay between

In many respects the actual amount of time between referral and the beginning of diagnostic assessment is of less importance than whether for parents this amount of time was considered acceptable or not, and what they felt during it.

Of the 27 interviews in our sample, in 15 cases parents felt the period of time they had to wait was perfectly all right, 8 were unhappy about it, and in 4 cases the notion of time lag did not really apply (or parents' focus was firmly elsewhere).

Each of these groups will be considered in turn.

5.3.3 An acceptable time lag

For 15 of the families the amount of time they waited between referral and first audiology appointment was considered perfectly acceptable.

5.3.3.1 Perceived to be a quick process

For the majority of parents satisfied with the time lag, the main reason was that they perceived this part of the process to be very quick. For some, objectively in terms of days and weeks this was indeed the case. For others the perception of quick and, therefore, of acceptable was also to do with having begun with expectations of a drawn out process and being surprised by the opposite.

> *"When I got the appointment through…you know I was surprised it was so quick because you know often you can end up waiting…especially then as there was something wrong and as I say, to be honest, I didn't think there was going to be anything wrong so I wasn't sort of really concerned about it, but yes, it was nice to have had the appointment quickly rather than you know, 'yes that's going to be in six months time or something'." [02]*

Some parents explicitly linked the short time between the referral and the beginning of diagnostic assessment with feeling "reassured". Professionals were perceived to be "getting on with it". However, interestingly for these parents it was not just the quickness of the process that was important, but rather the fact that many of them had left the last screening stage with an actual appointment for audiology. It had been made there and then. In other words, it is not just the short time scale, but the feeling that the elements are connected up, even if some waiting is involved, that these parents identify as contributing to reassurance.

> *"I was given the appointment that same day, she actually rang up there and then, she rang the hospital and got me an appointment and she was really helpful actually…" [19]*

One parent also linked the process that she was now part of with the video on NHSP she had seen at the antenatal class and so felt she also knew what to expect and that things were progressing in predictable stages. This predictability was also reassuring.

The fact that the process was, as one father put it, "handled quickly" also meant that as several remarked they did not actually give it much thought. It appeared routine and quick and, therefore, in many respects just the next stage of the same screening process.

This perception of continuity is interesting because for these parents there was not a perception that anything different or more critical might be happening if their baby had been referred.

5.3.3.2 View that there must be understandable reasons for the wait

Another reason why the time lag was thought to be acceptable was that parents felt that there were good reasons why it was necessary to wait. For one this was based on the simple presumption that professionals must know what they are doing and there must a good reason for the wait:

> *"[Waiting]. No, we didn't mind much at all, no. We just felt that however long they leave it, they're the professionals in this area and they know, you know, how long to give things…" [14]*

For another family they too took some comfort in having a reason for why there was a delay in beginning diagnostic assessment. They were very worried during this time, but the wait was acceptable because it was perceived to have a necessary purpose. Also knowing why it was not possible to proceed straight away helped.

> *"I think the two weeks in between were very hard, but I don't see how you can get round that because I understand why the two weeks were there because obviously they were waiting to see if the fluid, if there was fluid, if it would clear, so I understand that they couldn't give an appointment the next day 'cause it would give the same result, if that was the problem. So I don't see how you would get round it." [03]*

This mother did go on to say, however, that she would have appreciated some support, someone she could have talked to during this wait even though she understood why the wait was necessary.

In the case of another couple the wait was acceptable because they perceived the test they were going on to have to be a "better" one in better conditions and, therefore, one that could sort out exactly what might be wrong with their baby (or not). They were happy to wait to get to this "proper" test. It is interesting in this case, that this is a baby who was screened at home. The mother was very uneasy about whether such a seemingly special test could actually be done well in the noisy conditions of the home anyway. She was more likely to put her faith in a result that came from testing in obviously clinical (hospital) conditions rather than in her own home.

> *"I think it was more part of the same process in that they at [the hospital] they had a room that could be sound proofed and that there wasn't a problem with picking up background noise. So it was more that the conditions for doing the test were more favourable really. You were more likely to get a proper test result from it rather than doing the home testing when obviously there are background noises, there is traffic outside…" [17]*

5.3.3.3 Too busy to give it a second thought

Finally, for one mother the time lag was perfectly acceptable because she was too busy to give it a second thought. Also from her point of view if she the timing had been any quicker she simply would not have been able to fit it in with everything else she had to do with her other children.

5.3.4 An unacceptable time lag

In eight cases the time lag between the end of referral and the beginning of diagnostic assessment was clearly felt to be unacceptable although actually only two waited more than four weeks (family 10 who waited 10.4 weeks, and family 13 who waited 6.4 weeks). Also, two of the parents with this view actually waited less than two weeks. There were three main reasons for this perception that the time lag was unacceptable: parents for whom the wait was a continuation of what had been regarded as a dissatisfactory and unhappy screening process; parents who thought that audiology should simply be able to provide appointments more quickly; and parents who simply found the wait very distressing did not feel they had been given a reason for it nor acceptable support during that time.

5.3.4.1 Residual dissatisfaction carried forward from the screening process

It is interesting that five of the families who were most articulate about finding the wait between referral and the start of diagnostic assessment unacceptable were also amongst those who had described a very unsatisfactory screening experience. Couple 13, for example, were the ones who had believed the screen to be diagnostic and the mother had described considerable trauma when she was left alone after her baby did not pass the first screen and thought her baby was deaf. Although this misapprehension had been picked up and the mother given considerable reassurance and information, she clearly carried through that initial distress into the period of time whilst she was waiting for the first diagnostic assessment appointment:

> *"You're basically just left with the worry, then we came home and rather than having the joy of bringing a new baby home all we had in our head was worry… and [despite] all the information, you've still got to deal with it, you've still got to live with it. It's not going to go away. But the not knowing, the months of not knowing…" [13]*

In actuality this mother waited 6.4 weeks for her baby's first diagnostic assessment.

Couple 10 were amongst those parents who, having a baby in NICU, were also dissatisfied that during screening nobody had acknowledged to them the possibility that their child might be deaf, particularly given the visually obvious additional risk factors. They had felt nobody had acknowledged the evidence of their own eyes and had become very unhappy about the same message every time (no clear response) instead of a more elaborated conversation that would have acknowledged that there might be additional reasons to be concerned. For this couple, the period of time between the referral and the beginning of diagnostic assessment was seen as just a continuation of the same experiences of frustration and confusion and to some extent, distress. In actuality, this couple did wait the longest period of time (10.4

weeks) largely because of other considerations associated with the child's medical condition and continued stay in NICU.

For family 15 their experience of screening had been one of suspicion. They could not understand why screeners could not be clearer about whether something was wrong. In retrospect they had felt that if the baby had been referred then someone must have suspected that the baby was deaf and were angry that nobody shared those suspicions with them. This feeling that professionals were not being as honest with them as they might have or that information was being withheld from them was one that they also felt influenced the period of time when they were waiting for the first audiology appointment:

> *"They must have had concerns and that, they must have known, suspected that he was going to he deaf. I think that they should have prepared her from then [point of referral]…I think that is what they should do, I really do. Instead of like you can come home. My mum is phoning, [the father's] mum is phoning [and you say] 'oh it's probably wax' and so you convince yourself then, that this is probably what it is…" [15]*

For family 27 dissatisfaction with the amount of time they waited was compounded by the very fundamental experience of just trying to get the correct information about what exactly they were waiting for. This is the family who at point of referral had been given the wrong information leaflet then when they had requested the correct one had again been sent the wrong leaflet. Dissatisfaction with the time they were waiting merged into dissatisfaction with the whole process and thus not feeling they were in any way prepared for what was to happen next. In reality this family waited just under 4 weeks.

Family [26] expressed dissatisfaction because the waiting time caused them distress.

> *"…them months seemed to be like weeks, seemed to be like years, if you know what I mean. It were a right long time, even though it weren't, it felt like a long time…Once it were here, you were like nervous and really stressed about it, not knowing what you were going to find…it were like hard, weren't it…It seemed to be like never ending, but once it come, you were glad but then…it were very strainful (sic). " [26]*

In reality they waited about a month largely as a result of complications with their baby's prematurity. It should be noted that this is a family who again had been very unhappy with the screening process questioning why exactly technology could not be definitive and who found the inconclusive 'no clear response' outcome to screening deeply unsatisfactory. Their unhappiness at the process and outcome of the screen continued into their unhappiness during the waiting time.

5.3.4.2 Audiology waiting time is too long

Two families queried why exactly audiology could not provide a quicker appointments system following screening. One family, who already had a deaf child, were familiar with things taking time. But their experience of screening had been so very different from the

protracted experience of learning that their other child was deaf, that they could not understand why if screening could happen so quickly, audiology appointments could not follow close on behind:

> *"You don't mind waiting if you know what the reason for the wait is…but if you are expecting it soon afterwards and there's delay , you know, it just adds to the uncertainty really." [04, father]*

This couple felt that they were waiting because of a bureaucratic problem with the audiology clinic (they could not be fitted in sooner) rather than for any clinical reason. They suggested that perhaps their local audiology service should operate an appointment system whereby some priority was given to babies being referred so families were not left waiting and worried unnecessarily:

> *"INTERVIEWER: and then did you actually leave with a date for the next appointment?*
>
> *MOTHER: No, no…and that as I said was the worst period. Thinking about it before you were coming, I was thinking, I know audiology are really busy and got lots and lots of things to do, appointment and al the rest of it, but whether... audiology could have like a slot sort of every Monday afternoon, they knew they would get the babies that were tested in the last 6 days and they could go in like straight away… rather than having booked up the clinic…they could always sort of like keep the slot open and offer it to someone else on the Friday afternoon or the Monday morning if it hadn't been taken. I know it's about organisation and so on and everything but it certainly was the period of waiting, that was the worst." [04]*

In reality, this family waited just over 3 weeks.

Another family simply said they were "totally horrified" to be told they would have to wait four weeks following referral for an audiology appointment.

> *"She actually said, 'we can't do anything further now, you'll have to go up to audiology'. I said, 'can I go today'? She said 'no because they haven't got an appointment, we'll have to send you an appointment.' So I said' when's that going to be?' And she said 'if you're a bit worried I'll phone through now and I will try and get you an appointment as soon as possible.' So I said 'OK' and the next appointment was about 4 weeks later and I said 'you're kidding you can't make me wait that long!' And she said ' well if I hear of a cancellation come up I will phone and let you know, but unless that happens you will be waiting that amount of time.'" [21]*

Once again the problem was that the wait seemed to be a result of the audiology clinic being too busy, rather than for any reason associated with their baby and so was not seen to be an

acceptable reason to wait – particularly given the fact that screening had happened so quickly.

5.3.4.3 The wait is too long because it causes distress

Some of the families who had been satisfied with the waiting time had described worry and distress whilst they waited. A quick appointment did not necessarily take that away. But there were two families in our sample who had received a quick appointment (both saw someone under two weeks following referral) and who was nonetheless dissatisfied with the wait because they were very distressed.

> *"I think they should do it after a week then don't prolong it, it's not that good…I definitely think they should…have given me more support during them two weeks, to build you up…they could have said to us 'look it could be this, if it is this, do this, this and this…" [16]*

In other words, a quick appointment does not necessarily lead to satisfaction with waiting times and although this family linked distress with having to wait, in objective terms they waited for a far shorter period of time than others who were more satisfied.

5.3.5 Notion of time lag or time delay not considered relevant to the experience

There were four families who, for different reasons, really did not have an opinion on the acceptability or not of the time they waited between referral and first audiology appointment.

For one family, their baby was so long in NICU and had so many additional needs that for them concept of time between one part of a process and another does not seem very relevant because there was just so many complex things occurring at the same time. For another their focus was firmly on the whole experience of having a new baby at home and so the idea that they had to wait for an audiology appointment was just subsumed into the neonatal experience and not perceived to be anything special or different requiring any more or less attention than anything else. Consequently enquiries about whether the time they waited was acceptable or not did not feel relevant to them.

For another mother an evaluation of the amount of time she waited was also considered an irrelevant issue because for her there was a far bigger issue – namely what to do during that time. This family were very concerned about whether they should try to communicate with their baby until they knew whether she was deaf or not. Their focus was firmly on that question rather than having an opinion about the amount of time they waited.

> *"It was awful, it was, you know, you didn't know whether to talk to your child or not, you know that you should but you're feeling like 'am I… is he really hearing me?' you know, so every time that you wanted to talk to him you were , you could, you were reminded and you felt that kind of 'oh' you know, 'is he gonna be all right?" [05]*

It is perhaps of some concern that although this family only waited ten days, during this period of time they could feel so dislocated from their newborn baby.

Finally, one family, despite waiting amongst the longest of any of our sample, had no opinion on whether it was an acceptable wait or not for the simple reason that they had no idea what to expect. Consequently they just waited patiently until someone got round to them:

> *"We wanted to know what the doctor's opinion is and so we just waited three months" [08]*

It is perhaps relevant to note, in terms of this family's not knowing what to expect, that this is a family for whom English is not preferred language. In this respect they also commented on never having received information leaflets in a language they could understand and the audiology appointment letter being in English only.

5.3.6 Conclusions: time between referral and follow-up

- A short waiting time between end of screening and first appointment with audiology was helpful for many families. In addition the possibility of receiving the appointment date immediately at the end of screening was especially reassuring.
- A quick appointment did not necessarily take away worry or distress but for the majority of parents it did help
- Also knowing exactly why they were required to wait (e.g. giving time for fluid to clear from baby's ears) was also helpful.
- For some parents the quickness of the appointment was less to do with the objective fact of how long they had to wait and more to do with the fact that it exceeded their previously low expectations of how long they would have had to wait.
- When the appointment followed on quickly it tended to be positively perceived as being part of the same process that was being handled efficiently by professionals who knew what they were doing. This routineness was linked by parents to helping to reduce stress/worry.
- There is evidence to suggest that in cases where parents have had a particularly dissatisfying experience of the screening process then they are more likely to experience the waiting time between referral and first appointment as unacceptable and particularly distressing.
- Parents who received an explanation for why they had to wait in terms of how busy the audiology clinic was, did not find this acceptable, questioning why if early screening is possible then why is a more flexible approach to seeing referred families not possible also.

- Two cases raise particular concern: (i) the family who during the waiting time felt unsure whether they should communicate with their baby and if so how; (ii) the family who had received no information in their preferred language, an appointment letter in English that they could not understand and who waited 3 months for an audiology appointment without being sure if that was a usual period of time to wait or not.
- Families made good suggestions about how to improve the transition to audiology services by e.g. setting aside slots of time on a regular basis for those who had been referred so that there were no unnecessary service-linked barriers to their progression through the system.

5.3.7 Parents' attitudes during the waiting time and what influences these

The above discussion has concerned whether the amount of time parents had to wait was considered acceptable and what influenced that appraisal. Quite separately from those considerations is also the issue of what parents actually did during that time including what their attitude was to the possibility of their child's deafness.

As previously noted there was a group of families where issues about what they did or thought during this waiting time was rather irrelevant because of other concerns. There were also three families where no data were offered relevant to their attitudes/feelings/actions during this time. This leaves twenty families from which we are able to derive some idea of what occurs during the waiting time and what influences that.

The experience of these twenty families is best described through envisaging a continuum. The mid point on such a continuum would be termed *"I put it at the back of my mind"* a phrase used by many of our families. Then either side of that mid point there were families veering towards not being worried at all or veering towards being definitely worried but not wishing to dwell on it. In fact both kinds of feelings could underlie the expression '*put it at the back of my mind*'.

For example, these parents all described a back of the mind attitude but varied in the extent to which they presumed their child might be deaf or not and the extent to which they were worried or not:

> *"Yes, I suppose at the back of your mind you think oh maybe there is a problem, but no at that stage we were still thinking it was the equipment" [17]*

> *"I was more worried because I was aware now that he had failed it twice, it was like ooh, perhaps there is something…So it was a few weeks before we went. It was really worrying – I mean you try to put it to the back of your mind…" [19]*

Similarly others described a process of rationalisation of what was happening where again there was a recognition of the possibility of deafness but a desire to put it away somewhere and not to focus too firmly on it:

"I remember I kept thinking 'oh she might be deaf' and then getting tearful about it and the thinking 'oh that' ridiculous' and all my friends and family were saying 'oh of course it's just a bit of wax'. So I was definitely feeling quite anxious at that stage but sort of trying to rationalise it. It was probably nothing. My husband quietly sort of, thought there was something more serious going on." [24]

There were also, as we have seen, families who definitely sat at either end of the continuum because of the experiences of screening they were carrying forward (these had been negative so the ongoing experience was experienced as such) or just the kind of family they were (e.g. so busy never gave it a thought). The previous section on attitudes to the waiting time has provided examples of those at the worried end. Those at the not concerned end included, for example:

"We just dismissed it and thinking 'oh not, it'll be fine'. It happens to everybody else" [20]

"I wasn't unduly worried because I had been given a leaflet and I thought well there could be a lot of explanations for what is going on." [06, mother]

"FATHER: Didn't even give it a second thought

MOTHER: No we just thought oh, he's got glue ear, it's fine.

FATHER: I didn't give it a thought. Full stop." [09]

5.3.7.1 'Testing out' behaviours

It has been well documented in the past that it is often been parents who have suspected their infants' deafness long before any professional confirmed it and that during the period of growing suspicion before the deafness is confirmed, parents often tested out whether their child could hear or not (see Gregory 1995). For many the priority was to check the child's deafness and to provide some confirmatory evidence of that in the face of not being believed. NHSP brings with it potentially the promise of an end to such periods of protracted suspicion (which had often caused so much distress). However, as our interviews demonstrated, the wait between the referral and first appointment with audiology still prompted testing out behaviours for many parents – either overt tests like banging trays or more implicit tests like watchful observations. What is interesting is that the emphasis in these testing out behaviours was rather different than has been recorded in the past. Whilst for some parents who did suspect deafness these testing out behaviours were about demonstrating the child did have a hearing loss, for many others they were about just the opposite – testing out the baby could hear. In other words testing for confirmation of hearing (and the referral was nothing to worry about) rather than testing for confirmation of deafness (a suspicion they have had).

"We still didn't' really think there was a problem then because we kept testing his hearing by slamming doors and shouting behind him and stuff like that. Loud noises he will respond to. My mum was convinced her could hear because she said he does respond when he could hear my voice and things like that." [17, mother]

Within the context of screening where the vast majority of these referred infants will turn out to be hearing and parents know that, this shift to seeking assurance of hearing, rather than evidence of deafness makes perfect sense.

5.3.8 Other issues from the waiting time

5.3.8.1 Knowing what to tell people

A few parents discussed the difficulty of not knowing what to tell friends and family during this period of time whilst waiting for the first audiology appointment. The problem was not necessarily not knowing what to say, but rather not wanting to deal with other people's presumptions that the referral meant there must be a problem if parents themselves did not necessarily perceive its significance that way:

"We just kept it between ourselves, we weren't concerned, so we just kept it between ourselves, didn't' we? Everyone was gutted after the third test weren't they?" [22]

In this context, another family talked about how useful it was to have had the third information leaflet, because that was something they could give to curious friends and worried extended family.

For the family who already had a deaf child and who were almost certain their new baby was deaf, (the only issue was how deaf), not telling extended family and friends was part of a strategy of not wishing to worry then unduly until the situation was more certain.

5.3.8.2 Wanting support

Whilst the overwhelming majority of parents did not express a desire for professional intervention and support during the time between the end of screening and the beginning of diagnostic assessment, there were three families who did. In one of these cases the issue was simply information. This family did not believe they had had enough information about what was going on and said they would have found this helpful in preparing themselves for the appointment with audiology – both emotionally and in terms of what questions to ask. It should be noted that this is a family who only remembered having received leaflet one and no others. In particular they had no recall of leaflet three.

In the case of two other families, both would have welcomed a person to actually support them during the period of waiting time. For one mother this reaction is perhaps explicable again in terms of their experience of the screening process. The couple had initially believed the screen to be diagnostic and had described trauma as a result. They also had expressed dissatisfaction with the amount of time they had to wait for their first diagnostic assessment

appointment. In the case of another family they linked the requirement for more information with querying who should provide them with emotional support following a referral outcome if, as it seemed to them, that was not the screener's job:

> *"More information at the time would have been better. If I'd known the level of problems you know, if most people are actually ok, I might have felt a lot better about the wait…once that health screener had walked away that was, that was it…'sorry we're referring you' that was it. I never saw her again. I'm not saying that she should be there to support me, but someone need to be there to support people when they have had results." [03]*

5.3.9 Further conclusions about the time between screen and follow-up

- The vast majority of parents during the waiting time adopt a '*back of the mind strategy*'.
- Amongst those who adopt a back of the mind approach there is a continuum from those who veer towards not being concerned and those who are underneath more worried.
- Only a few parents are consciously and overtly worried during this period of time.
- Experiences of screening appraised as unsatisfactory appear to influence the waiting time experiences of this more worried group.
- Similarly the attitudes and experiences of the groups of parents who never gave screening and its outcome a second thought appear to be continuous with their attitude to the waiting time period.
- Testing out behaviours (including watchful observation as well as overt testing) are very common during this waiting time. However in many cases the issue for parents is testing to confirm they have nothing to worry about, rather than testing to confirm their suspicions that their child is deaf.
- For some parents the waiting time can create the difficulty of not being sure what to tell friends and extended family. This difficulty is principally related to not wanting to deal with others' presumptions of the significance of referral.
- A minority of families would have appreciated active support during this waiting time. These families were also ones who had inadequate information about the screening process including the meaning of refer and/or negatively appraise the screening process. But there are other families who also meet these two conditions and who did not express a wish for more support during the waiting time other than for the process to be speeded up.

5.4 The experience of diagnostic assessment and confirmation of deafness

5.4.1 Introduction

This section concerns parents' experiences of the 'diagnostic process'. Although the word 'diagnostic' is properly associated with aetiological investigations, the term 'diagnostic process' will be used here to differentiate from the screening process and to define it as the period of time that begins with parents' first visit to audiology for audiological assessment after being referred from the screen. Aside from the clinical perspective of the diagnostic period, it is important that we understand how parents perceived this period of time. There were in fact no parents that reflected on the "diagnostic process", using these exact words. Most parents saw each audiology appointment as an event in itself; some saw the appointments as stand-alone events whilst others made connections between them. However, for the majority of parents in our sample, the defining moment for them during this period of time was when they were told (with some degree of certainty) that their child was deaf – for some this was the end of the process, for others it was just another step in their journey.

The following discussion will not be based simply on and limited to the clinical definitions and parameters of the 'diagnostic process'; instead we will attempt to follow parents' own definitions and meanings (the first visit to audiology will be used as the starting point for this process, since this event is clearly identifiable in the majority of parents' accounts).

5.4.2 What was the key predictor of parents' experiences?

The experience of the diagnostic process was found to be hugely variable for the parents in our sample, with considerable differences in relation to time span of the process and the number of appointments attended. It would be reasonable to assume that these factors would influence how parents experienced the diagnostic process, however they were found to be of less importance compared with the communication style and manner of the professionals encountered during this period. Since parents often encountered several different professionals during the diagnostic process, they too may have experienced an array of communication styles and personalities/ characters of professionals. Therefore, parents' experiences tended not to be either good or bad, rather a combination of both, depending on who they came into contact with and on their appraisals and perceptions of the same.

5.4.3 What do parents perceive as good professional communication?

5.4.3.1 "Good" explanations

Good explanations were seen as key to good communication by the majority of parents. Parents had their own views as to what made a good explanation. Three different types of explanation were identified: explanations using appropriate register, thorough explanations, and explanations using examples in context. To further explain the latter approach, parents

appreciated explanations that used terms and examples that were meaningful to them. For example, the following family specifically highlighted what was lacking in the explanations given them by audiology.

> *"it would have been helpful if they'd actually written it down or told us in plain words, this is what it means, this is what we think he could possibly hear. And I mean like we didn't really know what it definitely meant until [the Teacher of the Deaf] wrote that thing for Disability Living Allowance. And she explained it in words saying like, 'if a jet plane comes over the house all he'll hear is a little buzz." [09]*

Another family described how professionals had explained their daughter's diagnosis using terms they did not understand. Subsequently, the mother attempted to elicit a more meaningful explanation but felt that the one she got was still insufficient.

> *"Well I said 'if mine's on a level of 1 – 10, if my hearing's 10 what's Alicia's?' And he said 'oh it's barely a 1.' And that was as much as we got. That was as much as we got then. I mean I'm sure it's very difficult for them to give that sort of news to parents but they do have to give that sort of news to parents, they should have training to do it better." [13]*

5.4.3.2 Sensitive

Unsurprisingly, many parents in our sample valued a sensitive approach by professionals during the period of time in question. By sensitive, parents meant more than simply being responsive to their needs, (e.g. answering parents' queries and questions or providing for parents' needs in a practical way). When parents talked about professionals being sensitive, they were referring to the fact that their needs were being met at an emotional level and that the professionals appeared to be aware of their feelings. Several parents used the word "gentle" to describe this kind of sensitive approach:

> *"…(they) said oh, they had detected there was a loss there and sort of explained roughly about it, but I think she was trying to break it to us gently and let it, sort of sink in…(…)…I think it's nicer the way we…they did it they said 'oh yes, um there is a problem there you know, come back for more tests' and at least that gives you a week or so for it to sink in or to think about it rather than just to suddenly be told, 'oh yes, your child is profoundly deaf,' I mean that would be you know, your world's ended kind of thing." [02]*

> *"…even before we had the diagnosis, you know, she was very gentle and explained things over again and didn't make us feel stupid for asking silly questions and things." [24]*

Another mother described the sensitive approach the head of education support services had taken on the day her child's hearing loss was confirmed – the example she gave was in fact in stark contrast to her account of the approach taken by the personnel within audiology.

> *"…he came there and he was quite sensitive I must admit, he, he was you know, quite, I felt he was quite aware that you know, something like that being told to you is awful and he came and gave me a card and 'ring, you know, we'll discuss things more.'" [06]*

Unfortunately, not all families experienced such a gentle approach from professionals. For example, one family had arrived for their first diagnostic appointment at audiology and since they had received no leaflet prior to the visit, they had little preparation for the news they would receive that day and found the professional's approach unacceptable:

> *"FATHER: She told us the bad news basically as we were leaving the room….*
>
> *MOTHER: …she just, she said, you know like Jeff said almost as we were going out, 'he's got permanent damage to his inner ear' and that was it kind of thing." [27]*

What is particularly interesting to note is that the sensitive/gentle approach was not appreciated by all parents. The following family described a 'cautious' approach by professionals and although understanding why this approach was taken, explained that they preferred to be 'told straight.'

> *"FATHER: …they were quite cautious as to how we were going to react.*
>
> *INTERVIEWER: Was that helpful or not?*
>
> *FATHER: I can see it being…I mean, to us, it's not a problem, we…the way we react, we wouldn't have minded, you get on with it…(…)…Yeah, I think they were a bit unsure as to maybe how to tell you. As I say from our point of view, we say, 'well it's fantastic you've picked it up, what's the next step?' But I find that all the way along the line, Judith I think you said was the same, Colin was certainly the same. They're very very cautious how they tell you what's happened… We prefer to be told straight, rather than be tiptoed round." [01]*

5.4.3.3 Inclusive

Attending diagnostic appointments meant for most parents that they were in a potentially confusing and perhaps anxiety-inducing situation. Just under half of the families in our sample commented that one thing they had (or would have) appreciated during the diagnostic process, was professionals sharing available information with them and including them in

their conversations or correspondence. The kind of inclusiveness that parents were looking for was more than just receiving good explanations, it was professionals including them as equals and it was something that made parents feel like partners in the whole process.

One of the ways that parents were made to feel partners during the diagnostic process was when professionals engaged them in the testing process, i.e. professionals would include parents in the process by explaining the testing procedure, how the equipment worked, and what the results should look like. Four families experienced this kind of inclusive approach from professionals. For example, this father felt that he knew exactly what was going on at the first audiology appointment and he even went as far to say that he felt he could see for himself what his son's results meant. He valued being included.

> *"…[the audiologist] was there and explained thoroughly what was…I mean I asked…I mean she explained thoroughly what we were trying to do, what the results should look like and then you could, I could sort of tell for myself that the result wasn't anything like it should be looking like, which is why I probably knew before I was told, about his hearing loss erm…I think the whole thing was very professional and…you know, I wouldn't want to change it." [07]*

For another family, even though they already had a child that was deaf, there were still things they did not understand about the testing procedures and so they too valued being informed of and included in the process second time round.

> *"…explaining what she was looking for and what was a good result, what was a bad result or whatever was quite useful 'cos I never actually realised or seen them do that before…I mean I know we've seen them do it, but not really understood what was going on…we were looking at these things on the screen and thinking 'what does that mean?' So yes, she was explaining that, that matched and that one didn't, that sort of thing was helpful." [04]*

Contrary to the positive experiences above, some families felt excluded because professionals were not engaging them in the testing procedures and they were left wondering what was going on. The following family contrasted their first visit to audiology with their experience of screening. During the screening the family had felt included because even though they did not fully understand what was going on, the screener had attempted to explain the process to them. At their first visit to audiology, however, the professional had failed to engage them in the process and so they felt excluded.

> *"MOTHER: …whereas the previous test (the screen) she had shown us the graph, even though it meant nothing to us in a way, she'd shown us the stuff, I didn't think [the audiologist] showed us what the equipment was particularly doing and what she was looking and checking for…*
>
> *INTERVIEWER: So you didn't really feel part of what she was doing?*

MOTHER: No.

FATHER: No." [23]

5.4.3.4 Honest/open

Not dissimilar to the idea of professionals being inclusive of parents was the notion of honesty and openness between professionals and parents. Just under a third of the families specified that "honest" communication from professionals was something they valued. The quote from the following mother best illustrates this idea.

> *"Well I mean like we've appreciated the openness and the honesty…the openness that the consultant has been with us like all right…when you're there you don't want to hear what they've got to say to you sometimes, but you come away and you think about it and say well…I'd rather him be open with me because then I know where I stand and where Joseph stands whereas if they don't tell you everything, you don't build that picture up whereas we know everything what's going on with Joseph…" [26]*

The experience of one mother perhaps highlights how much honesty was valued in professionals' communication. At the time of interview, the diagnostic process was ongoing for this mother and she still felt that she was receiving conflicting information and messages from the audiology professionals every time she attended the clinic. It is interesting to note that she makes the link between professionals' honesty/openness and trust.

> *"But it is just really, really difficult and we were really keen for their help and every thing at first but now we're a bit erm…well, we're not sure who to believe really, we're not sure who to trust and I suppose we're a bit dissatisfied with the whole service really. Just because we've been told so many different things… and I have noticed the last couple of times we have been they have 15 minutes together first… and I almost feel they are getting their stories straight before they speak to me." [19]*

When asked to describe an incidence of good practice from her whole experience of screening and follow-up support she specified that:

> *"But I suppose the main thing is when we have been told information really that has been the best thing and when they have been honest, you know although it was very kind of them to try and spare my feelings, I don't want that, I just want clear information to be told what is what." [19]*

5.4.4 What do parents perceive as a good professional manner?

5.4.4.1 Approachable

Five parents stated the importance of professionals being "approachable". Interestingly, the professionals that were often described by parents as unapproachable were those seen at the first audiology appointment. Parents described them in terms such as lacking "people skills" and not being a "people person." The following family found their first audiology appointment incredibly difficult because they felt that they had not been given "permission to cry". They described the professional involved as being unapproachable and unable to acknowledge and manage their emotions.

> *"…she kept going out of the room and then we'd have like a few tears and then hold them back when she came back in, 'cos…I don't think she could manage that…I feel that she wasn't very good at managing that side of things…And I think that's difficult because we were then not able to comfortably express how we were dealing with it, well, you know, we didn't feel comfortable to just cry so we were sort of holding back these…and obviously she found that difficult I think…" [23]*

Another family identified that the professional that gave them the 'news' at the first diagnostic appointment perhaps did not have the right personality. The parents' reasoning was that in fact this professional was a scientist and so perhaps could not be expected to be skilled in the area of interacting with parents.

> *"MOTHER: The other thing is whether people have enough training in actually being able to deliver that initial bad news erm…'cos that's not what they're doing as a job, but…*
>
> *FATHER: Yeah, I mean she's a scientist at the end of the day and…*
>
> *MOTHER: Yeah, she's plugging up the baby…*
>
> *FATHER: Yeah, plugging the baby up and testing them out and basically maybe it should be someone else that has that persona really…that personality in being able to give the parent the bad news…" [27]*

This family later went on to recommend that if such professionals were to be present at the first audiology appointment, they should at least be able to recognise when to bring in another professional more skilled at communicating with parents.

Similarly the following mother felt that the professional she saw at audiology appointments was not approachable and in addition she highlighted the fact that this same person was unable to relate to her child; she found this was also unacceptable.

"I mean her manner's not…she's not awful, but erm…she's not easy to talk to and you know it's…it's just like it's a job and she's got a job to do and she's gonna do it…You know, she doesn't take that much notice of Paul, I mean, 'oh, you're getting big aren't you?' And that's about the only comment she'll make, but when you are dealing with…you've got to try and you know…I mean she's not horrible, don't get me wrong, but I can't talk to her…" [06]

5.4.4.2 Patient

Seven families saw the patience of professionals during the diagnostic process as something to be valued. Three parents praised the efforts of the audiology professionals involved at the diagnostic appointments – these professionals had persevered for a significant amount of time in order to complete the tests. Although the testing sessions had been lengthy, parents perceived this as admirable – it illustrated the patient and persevering nature of the professional involved.

"I think the helpful part would be the sheer patience of the professionals in trying to test him. When we were getting bored and impatient, they would just stick with it." [12]

Other families commented that the professionals had taken their time at the appointments and this was commended.

"And he didn't let us leave until we understood, you know, her condition and you know, what was going to happen…He said to us, 'I appreciate…it's going to take a time to sink in,' but… he allowed us plenty of time to ask questions." [01]

5.4.4.3 Accommodating

About one third of parents recounted that professionals had been accommodating during the diagnostic period; this was something that was particularly appreciated since it could potentially be quite challenging attending a series of appointments with a young infant. Examples of accommodation included advanced warning of the likely duration of appointments, flexibility over appointment times, and understanding about how a busy lifestyle, work, and other children might mean that appointments could be stressful to organise from the perspective of the parent.

One mother contrasted the accommodating approach of the audiologist in providing her with helpful information prior to attendance, with her experience of ENT:

"…and audiology were very good, so, 'be prepared to, you know, try and make sure your child's asleep,' and we really made sure we did that, we would turn up to the hospital two hours early to do a feed, get him down and we had him in a deep slumber so that, you know, you could fiddle round with his ears, the trouble with the ENT guy was he was very busy, he was over an hour late for the appointment, by which point Joe was waking up again…so he couldn't stick things in his ears, so

that was frustrating, he did apologise to us, but it's like, we have a tiny baby here and…you know, what he said is in future he would always make sure that when a baby comes in, they're seen on time, you know, especially if a parent's made that effort." [05]

5.4.5 Conclusions in relation to professional manner and communication

- Good explanations were a key component of what parents perceived to be good professional communication. In order for parents to positively appraise an explanation, it had to be thorough, using appropriate register or using examples that were connected to a reality with which they were familiar.
- When professionals were aware of parents' feelings and attempted to meet their needs at an emotional level, this was generally appreciated. However, it should be recognised that this approach did not work for all parents.
- Parents identified that being made a partner in the process was a key feature of good communication. One way of achieving partnership with parents is by engaging them in the testing procedures.
- Honesty and openness from professionals was valued. A point to be particularly noted is that one parent made the link between honesty and trust.
- Being approachable was identified as an essential component of professional manner. Interestingly, those professionals described as unapproachable were generally those seen at the first audiological assessment. Families noted that audiological assessments took a significant amount of time to complete and so when professionals' demonstrated patience under these circumstances it was greatly appreciated.
- The practicalities of the diagnostic process could be challenging for many families. However, having a professional that was accommodating helped to counter this. One way that professionals could be accommodating was by notifying parents of the duration of appointments so that they could prepare themselves and the baby appropriately.

5.5 Conclusions

Newborn hearing screening is not an event, it is a process. The qualitative, narrative approach that we took has enabled us to capture in fine detail that process and the diversity of parent experiences associated with it. Parents have confirmed the value and importance of newborn hearing screening at the same time as raising subtle and at times unexpected questions about professional practice. Such accounts have taken us inside the earliest experiences of those for whom screening has the most significant consequence – those who discover they are parents of deaf children. We are very grateful for the time and care parents have taken to tell their stories and help influence further the future of NHSP.

6. IMPACT OF NHSP ON SERVICES

6.1 Introduction

The brief for the NHSP implementation included 'the development of audiology services to meet the needs of the very young, and furthermore the development of the involvement of Education and Social Services in the care of deaf children and families'. The latter derives from an existing service model in the UK, where specialist LEA services are informed by Audiology services about any new child with a significant hearing loss, and a Teacher of the Deaf (ToD) is allocated by the service to support the family and help the development of the child's communication skills. The involvement of ToDs continues through preschool and school age, and has been strengthened by the protocols recommended by the recent DfES Early Support Programme (ESP) for children with disabilities. While the model of Education services involvement is widespread and well-established in the UK, the involvement of Social Services with families of deaf children has been more varied and uncertain.

This strand of the NHSP evaluation is concerned with identifying the changes to practice brought about by NHSP, using survey and interview techniques with professionals in first phase NHSP sites. In some cases, a before-and-after approach was used (i.e. before and after the introduction of NHSP) while in others surveys were done only after NHSP had been introduced.

Three studies addressed the possible impact of NHSP on:

- Audiology, Education and Social Services
- Health Services including Hearing Screeners, Health Visitors, Health Visitor Managers and General Practitioners
- D/deaf professionals (the term Deaf with a capital 'D' will be used when referring to those individuals who identify themselves as being culturally and linguistically deaf)

6.2 Method

6.2.1 A two-stage data collection process with the main service providers in Audiology, Education and Social Services

An initial structured postal questionnaire was used to collect baseline service information pre-implementation. This included both closed questions using scaled responses, and open

questions. Three separate questionnaires (one for each service) were designed to probe similar areas but also to cover service-specific areas. The initial questionnaires focussed on expectations of changes, preparation and planning for service response/development and perceptions of opportunities and challenges (see appendix for questionnaires.).

Questionnaires were followed by a semi-structured telephone interview lasting about 30 minutes. The aim of the telephone interviews was to clarify any ambiguity in the questionnaires and to elicit more general attitudinal information about the perceived impact, and how opportunities and concerns were being defined.

One year after the start of NHSP at the site, a second telephone interview was undertaken. In all cases the interview was either with the Head of Service or with a person designated by the Head of Service. Both 'before' and 'after' interviews were, wherever possible, carried out with the same respondent. This was in some cases not possible because of staff changes. In the case of Audiology Services (but not Education or Social services) a second structured postal questionnaire was also used one year after the start of screening. This questionnaire was identical to the first questionnaire except for a few modifications reflecting its 'after' status (see appendix).

Health, Education and Social services are not co-terminus; this presented a considerable challenge initially. In the 23 first phase NHSP sites there were 23 paediatric Audiology Services, 27 Education Services and 34 Social Services. In the case of audiology and education services contacts were known or easily identified through professional lists and networks. In the case of Social Services locating the ‘appropriate person’ to fill in the questionnaire was problematic as services for deaf children could be located in one of a number of teams: e.g. Children and families, Disabled Children’s team, Sensory team. There was often no clear list of contacts. Such a list was generated by contacting Social Services representatives who had attended an NHSP information day, searching appropriate websites, and seeking names from the Education service contacts at each site. Table 6.1 summarises the response rates for the 'before' questionnaires and interviews for study one.

Service	Number of services within phase 1 NHSP sites	Number of services returning 'before' questionnaire	Number of services completing 'before' interview
Audiology	23	19	20
Education	27	26	27
Social Services	34	20	15

Table 6.1. Summary of response to questionnaire and interviews across services.

6.2.2 Study two: A one-stage postal questionnaires was used one year after the start of newborn screening in each first phase site, directed at Health Visitors, Midwives, and General Practitioners

Questionnaire responses from Health Visitors, Midwives and GPs were collected from NHSP first phase sites one year after screening had started in each.

The aims of the questionnaires (see appendix) were to identify knowledge of the newborn hearing screening programme and to explore the views of these health professionals in relation to the impact of NHSP on their work.

For Health Visitors, GPs and Midwives, a 20 per cent sample from each screening site was taken; three sites with prior experience of universal newborn hearing screening were omitted and the Midwives from those sites using the community-based screening model were not included. Table 6.2 gives the numbers sent and the response rates

	Health Visitors	GPs	Midwives
Number sent:	297	272	352
Response rate (%):	36%	25%	29%

Table 6.2. Response rates from Health Visitors, GPs and Midwives.

6.2.3 Study three: Two half-day focus groups were run for D/deaf professionals who had service support roles with the families of newly-identified deaf babies

The groups were held one year after the start of NHSP in the first site. The focus groups, one in the South of England and one in the North, were carried out in BSL via interpreters where necessary. The broad areas for discussion were similar to those of the main service providers and covered information and preparation for NHSP, resultant changes in working practice, training, challenges and opportunities of NHSP for D/deaf professionals, values and perceived benefits of NHSP for deaf children and their families, and the challenges and opportunities of the screening programme for D/deaf professionals themselves.

Respondent identifiers	Service/organisation within which individual is located	Role of individual within service/organisation	Sign language user
DP01	NHS Trust	Home sign tutor for parents of deaf babies/children	Yes
DP02	Education	Communicator	No
DP03	Education	Deaf support worker	Yes
DP04	Education	Parent support worker and teaching assistant at school for deaf children	No
DP05	Education	Deaf instructor	Yes
DP06	Charitable organisation	Team leader of youth services	Yes
DP07	Education	Deaf role model and support worker	No
DP08	University	Researcher	Yes
DP09	Education	Family sign language worker	Yes
DP10	Education	Family sign language worker	Yes
DP11	Education	Family sign language worker	Yes
DP12	Education	Family sign language worker	Yes
DP13	Education	Coordinator of family sign language workers	Yes
DP14	Education	Family sign language worker	Yes
DP15	University	Researcher	Yes
DP16	Education	Bilingual instructor	Yes

Table 6.3. Details of participants in the D/deaf professionals Focus Groups.

Letters were sent to the Health, Education, and Social Services contacts for each of the twenty-three first phase NHSP sites, asking them to forward the information about the study

to the D/deaf professionals within their service with role responsibilities for family support of deaf children.

Sixteen D/deaf professionals were identified, representing nine NHSP areas. In the event, 14 of these attended the Focus Groups. Their employing agencies included five Education services, one Social services department, one charitable organisation that provided social services for the area, and one NHS Trust. Interest in participating in the Focus Groups was also shown by two deaf researchers involved in work relating to deaf children and families; they later attended one of the Focus Groups. Table 6.3 summarises the details of participants. In preparation for the Focus Groups, each participant was sent a list of topics to be discussed (a written English version together with a BSL video version). The two professionals that were unable to attend were sent the list of discussion topics (in English and BSL) and invited to respond with their contributions. For the purposes of recording the data, the interpreters simultaneously translated the discussions into English and this was recorded onto minidisk. The timing and procedure for data collection are summarised in table 6.4.

Service	Before Implementation	After Implementation
Audiology services	Questionnaire Telephone semi-structured interview	Questionnaire Telephone semi-structured interview
Educational Services	Questionnaire Telephone semi structured interview	Telephone semi structured interview
Social Services	Questionnaire Telephone semi structured interview	Telephone semi structured interview
HVs, Midwives, GPs		Questionnaires
Deaf professionals		Focus groups

Table 6.4. Summary of data collection for the three impact studies.

6.3 Data analyses

6.3.1 Quantitative analysis

Quantitative data generated through the 'before' questionnaire, and, in the case of Audiology services, in the 'after' questionnaire, were analysed using SPSS 10.1 for Windows. Two-tailed Kendall's tau-b was carried out to test relationships between two ordinal variables, as well as between ordinal and interval variables. To assess a paired relationship of ordinal variables a non-parametric Sign test, a generalisation of McNemar's test was used. The non-parametric Mann-Whitney test was used to test possible relationships between ordinal and nominal variables. To assess a paired relationship between interval variables Two-tailed paired samples t-test was used.

6.3.2 Qualitative analysis

Qualitative data generated through telephone interviews or via focus groups was analysed inductively using a thematic content analysis (Wolcott, 1994) using QSR NUD*IST 4 (a

search and retrieve computer software programme). Each interview of focus group was fully transcribed prior to review. Members of the research team reviewed the transcripts of the interviews in order to establish themes. These themes are classified as ‘codes’ to be applied to the data; for example, ‘descriptions of good working relationships', 'training needs', 'the definitions of opportunity' and so on. The validity of such themes was increased by the analyses being undertaken by a multi-professional team which included researchers who also held professional qualifications in audiology, education, social care and medicine. In this way professional bias and perspective could be recognised and minimised.

6.4 Results

The results of the studies are considered in the following sections; sections 1-5 and section 8 merge the data from studies 1 and 2; the results from the questionnaires to other professionals are reported in section 6, and study 3 results are reported in section 7:

- Links between services
- Inter-agency working
- Changes in working practice
- Training
- Funding and resource implications
- Health professionals' perspectives
- Deaf professionals' perspectives
- The perceived opportunities offered by NHSP

6.4.1 Links between services

6.4.1.1 Links between Audiology and Education Services

There was evidence of open and regular dialogue between Education and Audiology services through both formal meetings as part of audiology reviews, case reviews and through Children’s Hearing Services Working Groups (CHSWG).

The pre-existing relationship between these two agencies produced a strong foundation upon which joint working, an understanding of roles and remit, and joint working practice could be developed or further extended following the introduction of NHSP. In all cases the satisfaction with the quality of links between Education and Audiology was described as ‘strong’, or ‘well established’ and ‘even better’. Such positive joint working was presented in

a range of ways that demonstrates mutual respect, recognition of professional expertise, an appreciation of the value of joint working and an expectation that such working would benefit both the deaf child and the child's family.

When Education professionals talked about their links with Audiology services the emphasis was on the exchange of information rather than the opportunity to meet.

"We share everything together, we really do. There's nothing we don't share. There'd be a huge list (if itemised)." [T3]. From the perspective of Education services, the speed of communication was the overriding issue in relation to good links with audiology around identification of a child's deafness. The rapid communication between audiology and education was most effectively achieved through immediate and to some extent informal communication- a phone call or a fax. The effectiveness of the system relied heavily on professionals' pre-existing relationships with each other. This feature is seen most clearly in the comments from respondents that described in some way a culture of sharing in which audiology colleagues took into consideration the different kind of knowledge that their education colleagues may have knowledge often rooted in the context of the home and family.

> *"There's a good dialogue about you know, the most appropriate hearing aid. If the audiologist fits an aid and we think it's not suitable, then he's very willing to listen to that and try different things, so its very much two way." [T18]*

The open style of information exchange was one that allowed for constructive and critical dialogue between services.

Opportunities to meet jointly to discuss, review and plan for both individual children and for the service as a whole was regarded as a vital feature by both Audiology and Education services. Several respondents emphasised the importance not just of meetings but also of having joint meetings in a predictable, planned cycle that could be relied upon to happen. This boosted confidence that working relationships between agencies could be effective, that matters of mutual concern would be addressed, and that decisions would be taken. For example:

> *"(The consultant) from audiology, myself and the nursery staff if they're involved with the child, the Health Visitor and anyone else, Speech and Language Therapist is always there and we are all clear about why we are meeting before it happened and the standard format is it's an exchange of information and planning for, you know, the future and because we've got all of use then we can make decisions on the spot." [T14]*

After NHSP implementation the Education services all commented that links with Audiology services had improved; two services used the phrase (relationships) 'being cemented' by NHSP.

"I think it was very separate, they did their job well, we did our job well and we sort of interfaced in the middle a bit, but I think we work together much closer now " [T22]

Improvement in links between Education and Audiology services was characterised by a number of features of service provision:

- Increased frequency of contact
- Use of technology to enable fast referral by use of email
- The joint development of protocols to redefine roles and responsibilities
- The inclusion of education staff at the point of disclosure and in two cases (T24,T6) during the period of audiological uncertainty between screen and diagnostic audiology
- Other national initiatives relating to young deaf children in addition to NHSP—MCHAS (Modernising Children's Hearing Aid Services) and ESP (Early Support Programme) were noted as stressing the imperative of joint working
- The establishment of joint care pathways
- The joint development of web- based resources aimed to improve knowledge and understanding of both teams

6.4.1.2 Links with Social Services

In sharp contrast links with Social Service departments before and after implementation were less well established. In the few cases where links between Social and Education services were good they were highly valued and characterised by mutual respect, an understanding of complimentary roles, positive exchange of information, and what was perceived to be flexibility of approach:

" because she (the family support worker for education) only does early years work she can be much more flexible and she's doing much more sort of evening visits and things like that, so she can talk with the family, with dad and whoever's out at work." [S2]

Characteristics of good links between Social and Education services:

- Inter professional contact and dialogue
- Individual personal links
- Openness of professional exchange

However, both Audiology and Education services predominantly characterised their links with Social Services as poor; furthermore there was little evidence that NHSP had led to improvements. Of the 27 Education services, 24 reported no change in their relationship, 2

reported that the relationship had deteriorated and only one reported a positive change. A number of barriers were identified as militating against improving links with Social services:

Social Service criteria for referral not being met:

> *" the Disabled Children's Team take them on board if…they have to met their criteria. There has been no change in terms of criteria so a child with a moderate loss or a mild loss or just a severe loss, its unlikely they will take them on board. Very, very few of our children are under the DCT." [T1]*

Specific contact and referral routes being either undefined or poorly defined:

> *" I don't think we really do have a contact, we certainly don't have someone who is aware of the screen who could say something useful to families. [T11]*

> *" I refer them through (to Social Services). I don't think Social Services know what their role is. [T14]*

Staff shortages and unfilled vacancies:

In five of the 27 Education services respondents noted that positions had fallen vacant and not been filled, that pressure on staff working within Social services compromised any involvement with deaf children and that in two cases Social Service staff who were keen to make links were prevented from doing so by their job descriptions.

> *"What it did (preparation for NHSP) was actually highlight the fact that her job was with adults and although she would have so much liked to do more with children that wasn't within the remit of her job and she subsequently moved.." [T24]*

Structural and procedural barriers:

> *" it seemed that at every turn we made, we came across guidelines and constraints within Social Services which impeded movement really so we...as a multi-agency group sort of turned things around and put it in Social Services jargon but basically it needs a vast input of awareness and money to allow social workers to get out and see these children.." [T17]*

- Social Services may rate their relationship with audiology to be good (65 per cent of services interviewed stated they were extremely satisfied with their links) –yet often this is linked to their work with older deaf children, young people or adults, as opposed to deaf children 0-2.
- There is a minority of Social services that have no links with audiology

- The type of team and worker cannot be used to predict whether a service has good/poor links with audiology e.g. having a specialist social worker within a team does not necessarily mean that the service will have good links with audiology
- Workload and lack of resources appear to be some of the main barriers to services being able to improve their links with audiology or education

Characteristics of poor links between Audiology and Education services on the one hand and Social Services on the other:

- Difficultly in establishing a specific contact point or person
- Lack of contact or referral route
- Lack of clarity about roles
- Strategic level barriers

6.4.2 Inter-agency working

Amongst NHSP first phase services there was universal agreement that partnership working needed to be improved. NHSP was viewed as a driver in promoting joint working and providing an impetus for tackling areas that were perceived to be more difficult. The effectiveness of inter-agency working was perceived to have changed markedly after NHSP implementation and demonstrated that even within a restricted timeframe considerable positive change could be achieved.

Prior to the introduction of NHSP, education services were aware of the partnership imperative but cast achievement of such working within a framework of structural and attitudinal barriers. In several cases achievement of joint working was seen to be a medium or long-term goal rather than short-term option.

> *"It's a bit like hitting your head against a brick wall, I think we'll use UNHS (NHSP) guidelines to push it on really, try to get people to meet on a regular basis." [T 10]*

Four audiology services explained in some detail that from their perspective interagency working would only be achieved if links were developed at a structural level. This was echoed by education services, for example

> *" I think where we fall down is links with other services, particularly Social Services, you know we can be as ready as you like, but if other services…..[T22]*

Joint clinics between audiology and education services underpin common working practice within the care programme for deaf children and their families. The opportunities offered by this are considerable and provide a foundation upon which to develop inter-agency working at practitioner, organisational and structural levels. Of the 20 interviewees from audiology

services 14 had such joint clinics. These audiology services expressed confidence that where a child was identified with permanent childhood deafness a fast and appropriate response would be provided by education. On the other hand, despite the statutory requirement (HMSO, 1989) for all deaf children to be referred to Social Services there was scant evidence of routine referral.

Joint working with Social Services was generally reported to be poor by both audiology and education services. Joint working was achieved in a small number of services where an organisational decision had been made to promote and develop such practice. For example one respondent reported the opportunistic approach taken during re-organisation of local educational services, pre implementation:

> *"…we have been included from the beginning in a multidisciplinary working group re implementation of NHSP and as a result of this expect links to improve especially with health." [S5]*

In the case of another respondent reporting very positive inter-agency working this was linked to individual personalities rather than to any organisational changes. Additionally links with positive inter-agency working are ascribed to professional role release and a sense of trust. Another Social Service provider anticipated that with NHSP would come a change in practice for audiology, which would in turn influence their joint working with Social Services. This service saw that NHSP would highlight the need for audiology to review their practices with regard to referring newly identified deaf children.

"…we're constantly debating that now, well where is the role of the teacher of the deaf in, say a twelve day old baby, what would they actively be doing erm…and it's about audiology kind of realising that and kind of coming away from that safety net, in a sense of where before it was make a referral and they'll kind of pick it up and do whatever, but actually you've got to look at new strategies…" [S16]Interestingly responses from Social Services were less optimistic about positive change, either in perceiving it to be necessary or achievable. There were some services that expected NHSP to have little effect on their links with audiology, for example:

> *"I mean I think our links are pretty reasonable now for what contact we need to have but you know, if they improve then all the better really." [S11]*

For another service who had expressed that they were 'extremely satisfied' with their links in this area, the advent of NHSP was expected to have little impact because (i) they already had good links; (ii) there was 'no room' for improvement due to lack of resources.

For one fifth of the Social Service providers interviewed it seemed that NHSP was a welcome impetus that would improve the joint working between audiology and Social Services. There was strong evidence across all service groups that opportunities to develop multi-agency working was enabled through development of local Children's Hearing Services Working Groups (CHSWG). These were seen as a focus for inter-agency exchange, and for the development of joint protocols. The organisation and strategic empowerment of CHSWG

varies across England. Many have no budget and no statutory power to enforce change but are reported to act as an important inter-agency forum.

Across all services CHSWGs were identified as the organisational grouping that positively encourages inter-agency work. CHSWGs did not have the same constituent members across all first phase sites. In six cases Social Services were reported to be actively involved in CHSWG meetings. In thirteen cases no representative from Social Services attended despite repeated invitations for such attendance. Before implementation nine areas reported having established CHSWGs; after NHSP implementation this rose to 14 areas, with NHSP being cited as the prompt for their development. In one area the CHSWG was reported to have become "unwieldy" and had been replaced by a sub group—Education and Tertiary Services subgroup—made up of health and education professionals only.

Two areas had established parallel groups, one pre-screen, a Deaf Interagency Group (T3) and one post-screen group for joint training:

> *"Audiology, specialist Health Visitor, Speech and Language therapist, Social Worker for the Deaf, a generic Social Worker, Deaf Tutors, parents of deaf children and representatives from the Deaf community." [T22]*

- In summary, all service providers viewed inter-agency working as an important goal in the context of NHSP. Achievement of this goal was viewed as relatively easy to achieve between Audiology and Education services but less easy to achieve with Social services. Traditionally audiology and education have a long established working relationship relating to deaf children, which are reported to have been strengthened through the introduction of NHSP and a consequence of this has been closer joint working

6.4.3 Changes in working practice

6.4.3.1 Changes in working practice within Audiology services

For each agency, specific challenges are presented when services are required for very young deaf children and their parents. Within audiology services changes were primarily related to audiological assessment techniques: the focus of Audiology services was on the importance of being able to gather:

- Frequency-specific information across the speech frequencies, by using tone pip ABR in conjunction with other tests
- Identification of any conductive element of the hearing loss, by using high frequency middle ear measurements and bone conduction auditory brainstem response measures
- Use of a test battery approach to increase the accuracy of assessments

- Use of an appropriate paediatric hearing aid fitting protocol

Pre-screen it was evident that services had to develop quickly to achieve these goals and to ensure that sensitive and efficient assessment and hearing aid fitting procedures were in place for when screening started. The 'before' and 'after' use of key test procedures at first phase sites is detailed in figure 6.1 There is clear pattern of improvement in services with over 90 per cent reporting using bone conduction click evoked ABR and tone pip ABR as a result of NHSP implementation. Additionally, there was an increase in the use of high frequency tympanometry, well suited to the needs of infants (Sutton, 2002). Just over 50 per cent of services were undertaking probe tone measures for accurate hearing aid fitting. It is also notable that whilst just over 80 per cent of the services were employing paediatric hearing aid fitting protocols this does mean that just under 20 per cent are still failing to apply appropriate fitting and verification techniques despite very considerable information and training efforts.

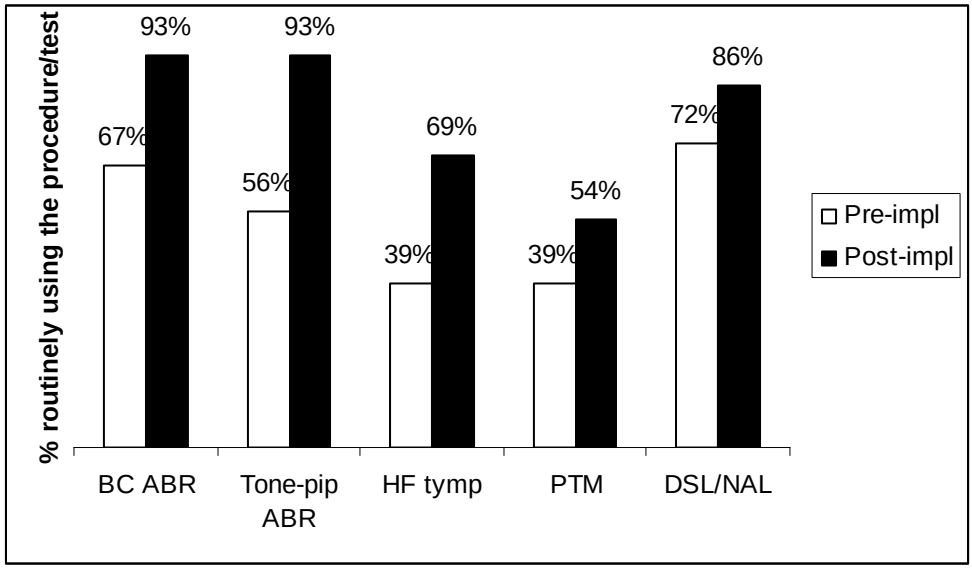


Figure 6.1 Proportion of services routinely using the tests in infants younger than 6 months before (pre-impl) and approximately 1 year after the implementation of NHSP (BC ABR: bone-conduction ABR; HF tymp: high-frequency tympanometry; PTM: probe tube microphone; DSL/NAL published hearing aid fitting prescription).

When audiology services were asked before implementation to rate their state of readiness for the introduction of NHSP on a five point scale, no correlation between self-perceived readiness and the use of the necessary tests/procedures was found. However, despite this failure to relate readiness with availability of paediatric-focussed audiological procedures there have been significant improvements in the availability and use of such equipment and procedures since NHSP implementation.

6.4.3.2 Changes in working practice within Education services

Education services in the UK provide support services to families with pre-school (and school age) deaf children. Such personnel work within the family setting and thus deal with a range of factors some predictable and some child or family specific. Year-round cover was recognised as a key feature of family friendly provision by the majority of services. Whilst many saw this as a national issue, recognising that reliance on ad hoc agreements and

goodwill was no longer appropriate a number had taken this issue on locally. In many cases organisational efforts were being made to achieve cover, for example:

"We are trying to arrange staffing so that cover is available and not left exposed if someone is ill" [T2]. One service had approached this at a strategic level and had agreed pay and conditions that allowed a more flexible and reliable basis for year round provision.

> *" We've reorganised our service, one member of staff who does pre-school early years work-we had a meeting with the unions about flexible working conditions, so we can work throughout the school year" [T22]*

A number of services expressed concern that changes in working practice and the need for forward planning was not being reflected by priorities set at a strategic level. Staffing levels attached to increasing caseloads and current understaffing was seen as a problematic area that in some cases was not being acknowledged.

> *" I think my pre-school caseload is 14, we need to make sure the LEA is aware this is not a needs-led service. We're just inundated and we've (education service for deaf children) got thinner everywhere, which is not good" [T1]*

In other areas such needs were recognised and agreed at a strategic level, for example:

> *" Making sure the LEA is aware of activities we're undertaking and the fact that it is going to be an issue for future support. (Children with) Mild and moderates (hearing loss) arriving earlier to our service so services will be stretched. An application for additional staffing has been approved." [T2])*

The pattern of funding for extra staff in first phase sites was patchy and included sites which had been funded for an extra member of staff and sites where funding requests and bids had been turned down. In one service an innovative approach had been taken by the Audiology service in conjunction with the education service to cover a perceived new area of need.

> *" So what's happened is that um health have actually funded point 4 of a teacher of the deaf time for post called the early language facilitator…that role is to monitor children um who have got a very slight hearing loss. [T6]*
>
> *Interviewer "A mild hearing loss?"*
>
> *"A mild hearing loss, picked up through the screen, um to monitor, I see them monthly, so monthly for the first 8 months and we will monitor their communication development " [T6]*

6.4.3.3 Changes in working practice within Social Services

Only 4 respondents (20 per cent) said that they were 'always' notified of newly identified deaf children. Analysis of data from social workers in phase one NHSP sites revealed great diversity in both attitude and practice concerning their role with newly identified deaf children and their families. In broad terms responses fell into three groups:

- respondents who were clear that they had a significant role to play with deaf children and their families post-diagnosis, were pro-active in seeking to expand and evolve further that role in the light of NHSP and who were in many respects frustrated that they could not fulfil all aspects of the role they identified.

- respondents who were generally satisfied with their role currently but saw it as minimal and circumscribed and who thought that the role was perfectly acceptable given the role of other agencies, in particular education. They did not perceive a great deal changing in the light of NHSP.

- one respondent who found it difficult to explain what the social work role might be or should be with newly identified deaf children, who honestly admitted he had very little knowledge about the area of work (or of NHSP) and for whom it was a genuine puzzle whether there was a role Social Services should be considering that they were currently not fulfilling.

Some services saw the opportunity for changes in working practice:

> *" Well I hope newborn screening would actually give us, like the Teachers of the deaf, a sort of right to erm…be informed of all children diagnoses and…they don't have to get parental consent to refer to teachers so I think they shouldn't have…need to get parental request to refer to us… At the point of diagnosis, if there's an automatic referral to education, there should be no reason why there isn't an automatic referral to Social Services…that's not saying the parents have to take it on board…" [S8]*

Others were satisfied, as a first step, with the idea that Social Services would at least get mentioned as a contact on any information given to parents, whereas this had usual not been the case before. Three services had been proactive in seeking contacts with audiology services both to better understand the process of screening but also to explain the role and remit of Social Services in respect of newly identified deaf infants; for example:

> *" We've been up to see the audiologists and talked with them on an informal basis , and we have explained if there is anybody whose child has just been diagnosed and they can refer to us, but they can at least give the families information about us,*

give the family the choice to get in touch and they've taken that on, that's not a problem……..People are phoning us and asking about stuff now, whereas before they wouldn't" [S1]

Respondents offered very detailed analyses of why they thought it was or would be difficult to respond to referral and to provide a service even if they were informed. Their comments were overwhelmingly to do with the structure of Services, where responsibility for deaf children lay, and the difficulties that arose because of confusions about this.

It is not unusual for Social services to be organised so that, on the one hand, there is a specialist sensory team with expertise in deafness but which only dealt with adult services. There was a children with disabilities team (or children and families team) which dealt with referrals across disabilities, including deaf children but which lacked specialist deaf child knowledge and generally was poorly connected with multi-professional networks that were deaf child specific. This organisational structure leads to several problems. In some cases, basic information about NHSP and related developments did not actually get to the services that had responsibility. This could lead to children's teams receiving information too late to attend events and appearing to be not interested in what was happening.

" I mean I don't have any direct links with the audiology department, I don't know any names, I don't know any people, that's what I'm meaning here. And that's partly down to the fact that whilst the pilot was being conducted, although I've got names on paper, a lot of the time I got notification well after the event, so we weren't actively involved in any of it. " [S12]

Complex arrangements for joint working require resolution if, as in this example, the children's team are responsible for receiving referrals and holding the case, but require expertise of the adult team to carry out joint assessments to decide what work is actually needed.

" There are no designated social workers to work with deaf children in (name of city) because there is a child centred team erm…I don't know what you call it, but the child centred team….What has happened when we were working with children is that we have co-worked because we feel that its difficult to work both with adults and children because it's two different roles and I have made representation about that so that's already known, but we have limited resources." [S7]

However, in other cases where there was a split between adult sensory teams and children's disabilities teams, joint working with deaf children was actually characterised as very unlikely because the children's team would not routinely take a referral for a child who was 'simply deaf' as this did not meet their thresholds for responding to a referral. It was left with the adult sensory team to try to provide a service, even though they were not trained to work with deaf children.

Interviewer: "…the role of Social Services under two in your area, how would you characterise that role at the moment?"

Respondent: " It's a nightmare! It's a nightmare because what happens is…we've got a duty assessment team who primarily deal with child protection issues, then you've got a children's physical disability team who won't pick children up that are only deaf…because they don't have additional needs on top of that. Their needs aren't being met, so actually comes back to me in actually trying to identify, OK.. theoretically they come out as a child in need automatically because they have a disability so…its about going out., seeing what we can do…" [S16]

For other respondents the issue was not about how to develop protocols of joint working or how to respond as an adult service, but more simply, that what was missing was a designated specialist social worker for deaf children. If one existed then problems both of under capacity to be able to respond in adult services, and the lack of skills and experience of being able to respond in children's services could be overcome.

6.4.4 Training

"(However), for the potential benefits to be realised (of NHSP), it is vital that all the professionals involved have access to high quality training and acquire the knowledge, skills and understanding necessary to work with deaf babies and their families." (Garner, 2000).

6.4.4.1 Audiology

All responding heads of Paediatric Audiology services expressed a pressing need for more training. There was one clear trend — the better the service, the more prioritised was the issue of training for working with families ('sharing the news' and 'counselling').

'We have always done it [counselled the families]...with proper training we would do it better'[A1]]

It became apparent that some professionals found it uncomfortable to admit to their lack of expertise and ask for assistance. This reticence was an obstacle to development

'When you've been a professional running service for some time … it is very difficult to phone up somebody and say look I am not very experienced at this, would you mind showing me how you are doing it...' [A4]

When asked how or by whom would these training needs be met, a number of interviewees, often from the more well developed services, deemed that in-service training and peer training were valuable, as was general networking with other colleagues. Others felt that visits to the clinical centres of excellence could be helpful. There was a feeling that Universities ought to become more responsive to the training needs of paediatric audiology staff. The interviewees also expressed hope that voluntary organisations (e.g. in the UK the National Deaf Children's Society) would continue contributing to the training opportunities.

As a whole, the heads of services emphasised a need for centrally funded, high-quality, hands-on training.

6.4.4.2 Education

Pre-implementation all education services recognised the need for training, with professional recognition of missing or incomplete skill sets. Ability to take advantage of training opportunities and to meet the demands of courses varied across sites. The importance of cascading training within individual services, and of a co-ordinated approach to meeting training needs was highlighted by services as central to achieving a service that was "ready".

Education services had mixed views about who were the appropriate individuals to undertake training. In some areas it was feasible to have dedicated pre-school staff, in others all teachers of the deaf had to have at least a basic training in very early years, although this was perceived to raise some practical concerns.

> *" We need to be well prepared and we area geographically spread out LEA and its not like perhaps in, …in the city where you can have a specialist in an area, you can't, you have to be all things to all people really… Some of our staff are not au fait with baby handling –if you haven't had your own child…. you know you haven't got an extended family where do you get access to very young children the prospect of holding a couple of weeks old baby, it's a bit daunting " [T18]*

For small unitary authorities where the team consists of a single or two Teachers of the Deaf within the team catering for all deaf children from identification to school leaving a more pragmatic approach was necessary. Training could not be accommodated, simply providing a service was compromised, with caseloads being reshuffled and support being minimised.

> *" This service is ridiculously stretched, thread thin, I don't alter the length of the visit but what I am finding is that the ability…for me to be able to regular visits in really quite tough…I asked what their preferred support form me would be in terms of visits and their response was a weekly visit from a Teacher of the Deaf to which I had to pass and say unfortunately you know that just isn't possible. In a hypothetical world that would be marvellous" T1*

For those in urban areas coverage could be considered in a more detailed way to include both professional and personal qualities where a good match to the needs of families with very early-identified deaf children.

Specific funding for training was earmarked and linked to NHSP by the DfES via a ring-fenced resource, Standards Funding (SF). This potentially set up a funding route for training and other small scale spending by education for activities related to NHSP. This resource required a 50% contribution by the Local Education Authority and was allocated locally according to the perceived needs within each local area. These did not necessarily include NHSP related services from an LEA perspective. Of 27 education services within first phase sites 8 were successful in achieving SF. This funding was used in a variety of ways:

- to support staff training (n=4)
- to employ additional staff (n=2)
- to purchase assessment tools (n=1)
- a mixture of the above (n=1)

A number of additional routes for funding were identified by education services and wherever possible were accessed to help support and develop early years services. These funding sources are summarised below:

- The Early Support Programme (ESP): (n=8)
- LEA early years budgets: (n=3)
- Early Excellence funding as part of a bid by the Institute of Hearing Research (IHR) to the DfES: (n=2)

The Early Excellence funding obtained via IHR was used to fund the development of centres which, whilst having shared goals and aspirations, were different in their approach to service delivery. One centre is based in a new purpose built resource attached to an established Early Excellence Centre and has brought with it new audiology facilities an opportunity for families to attend the nursery and to have a range of professionals and other parents readily available. The other centre is a virtual centre that covers 17 LEAs and aims to promote cross boundary use of resources and training opportunities, to enrich but not replace local service provision (evaluation of these resources is the subject of another publication).

6.4.4.3 Social Services

For social workers in first phase sites the implementation of NHSP was in many ways viewed as a rather distant health initiative that represented a very small demand in relation to the many other responsibilities that had to be met. Despite the statutory requirement for automatic referral of all deaf children to this service, social workers were sceptical of their ability to offer appropriate services. This scepticism was linked to three main approaches:

- Those who worked within Children with Disabilities teams who had knowledge, understanding and skills in child development, family dynamics and child protection in addition to a generic social work skills
- Those who worked in Adult Sensory Teams who either recognised an inappropriate skill set but who had an appropriate understanding of deafness
- Those services that abrogated responsibility to education or health services

There was a clear recognition of the need to graft deaf specific training onto the skills base that 'Children with Disabilities' teams already possessed well summarised by one respondent:

> *" I mean we are not lacking in skills, but they are broad based skills, they're not specific to the issues parents are going to face (post NHSP)…[S12]*

This represented a sharp contrast to adult-based sensory teams who felt secure in their disability specific knowledge but insecure in meeting the needs of deaf children and their families. For example:

> *"We both feel its difficult working with children because most of our work is with adults and its completely different" [S5]*

In four cases social workers whist recognising the need for services were confident that to this role could and indeed should be met elsewhere. One service noted this might be either via Health or Education whilst the other three firmly placed the responsibility for social services within the remit of education.

> *"We have no social worker trained in any way to work with deaf children within the Children with a Disability team. All work is undertaken by the Sensory Inclusion team within the Education department." [S23]*

This stance was justified as a pragmatic approach to providing a service when social workers were over-stretched and being asked to work out of their area of expertise.

> *"We really see children with deafness so rarely that we don't have the expertise." [S10]*

In one case there was an open admission of a basic lack of understanding of the remit to be undertaken with deaf children.

> *"It would be good to know what our role is, the expectations of our role in it, you know what the process is in a simplified way." [S11]*

It became clear from the analysis of the data that where a responsibility for service delivery to deaf children was recognised that social workers recognised the need for training. There was agreement across all services interviewed that training routes were either unknown/unidentified or very restricted, a typical comment being quite simply:

> *"Very few opportunities specifically in this field to my knowledge." [S6]*

Beyond simple observation of the screening process locally only two sources of training were identified: the National Deaf Children's Society and the Handsel trust. It should be noted that services able to identify a possible training route were in the minority. Even when a specific route was mentioned this was, in a number of services linked to broader issues rather than to NSHP.

Education	Social Services	Audiology
Counselling	Child development	Working with very young babies
Multi agency working	Audiology	Advanced audiological techniques
	Impact of deafness on a child	Habilitation
	Impact of deafness on the family	
	Counselling	
	Language development	
	Care co-ordination	
	Goal setting with families and other professionals	

Table 6.5. Perceived training needs by service and ranked in order of importance.

There was agreement across all groups that counselling is an important missing skill that needed to be developed by all services.

All service providers recognised the importance of multi-agency working within the context of NHSP and saw a natural extension of this to be joint training opportunities. In a small number of cases such opportunities had already been taken, notably through ESP, the North West Regional Early Excellence Programme, and in two areas through local initiatives to share information not only about individual cases but also about roles. Within the data there were three examples of social workers taking a proactive lead in seeking information from colleagues in audiology about the screen. In one case the audiology service had taken a lead in providing a training opportunity about the diagnostic process for local social workers and midwives. In another an audiology service had shared 'breaking the news' training with staff form the education service. There were no examples within first phase sites of social workers and education sharing any training even when opportunities for such sharing had been provided (for example through NW REEP).

In summary, a number of themes emerged with regard to training needs:

- All services linked appropriate training with their ability to provide a high quality service for very early identified deaf children and their families.
- While there is guidance regarding the professional competencies required by Teachers of the deaf working with children 0-2 years and their families, no such competencies exist for the other core professionals (paediatric audiologists and specialist social workers).
- There is a range of training opportunities available for Education services staff; some of these are also accessed by doctors and speech/language therapists
- Audiology services are able to identify specific training needs and potential training routes that could meet such needs.

- There is no evidence of any specific training courses that have been developed to meet the needs of social workers working within the context of NHSP.
- Social workers within first phase of NHSP are aware that they do not have deaf child specific skills and are able to identify training areas that would, in their opinion allow them to offer a more appropriate service.
- Some Social services do not prioritise this area with their package of service delivery.

6.4.5 Financial and resources implications of NHSP

Although respondents across all service providers were in general enthusiastic about the benefits and opportunities of the introduction of NHSP, they were also realistic about the problems they faced. Top of the list for all services was "staff shortages" but this meant different things in different contexts. For a minority of respondents they had managed to secure resources to employ more staff, but were having problems finding suitably qualified and experienced ones to recruit. For others the issue was opposite. They had identified a need for more staff but were unable to employ any, either for financial or organisational reasons:

> *"We cannot do NHSP without extra staff…. We were late to advertise as we were waiting for the confirmation to get central funding. If we do get somebody it will be at the expense of another audiology department in [the area]." [A19]*

> *"We have a chronic shortage of teachers of the deaf nationally, we're having recruitment problems anyway, so I don't know what we can do about that.." [T10]I'm also very conscious of erm…sort of financial restrictions within Social Services, which actually limit what we can do at the minute . We're very ,very limited" [S6]*

Other respondents focussed on the way in which potential differences in caseload tasks could require more staff time and thus lead to staffing shortages. For example, within audiology more time might be needed than in the past to:

- assess (the now very young) referrals,
- there would be a number of false positives,
- more time and more frequent appointments for young deaf babies would be needed with ear moulds, hearing aid fitting or just in general with parents.

> *"The only unknown factor is the time that we will need. Patients' expectations have gone up and we need to spend more time with the parents." [A21]*

In one case where there was an all-age audiology service the very considerable demands of other client groups were ranked as more important than those resulting from NHSP. It should be noted that this was not a typical response.

> *"For my service there aren't any [opportunities]!...long waiting lists for elderly adults are a far bigger issue for me than 2 or 3 babies who might have to wait an extra year before we find their hearing loss. You have to put it in context…Even as paediatric audiology is our priority, we cannot switch our staff doing paediatric audiology from adult work just because of NHSP. If we do not have the screening system in place that we feel is appropriate, we will go on with our targeted screen. It is a cost-benefit analysis. We are doing a good job anyway. The incremental cost of introducing NHSP may well outweigh the benefits to the parents overall. We don't want to harm the adult work." [A19]*

Similarly one Social Service respondent stated that their desire was to be proactive in relation to NHSP, however the social worker's lack of time meant that they were forced into taking more of a passive role.

> *"…if we had time, this would be something where I would very much like X (the specialist social worker), for example, to be more proactive in going to find out about this (NHSP). At the moment because of the demands on our service erm…for example, mental health clients and so on, we have to prioritise and at the minute that probably…I mean, our priorities at the minute are only to deal with clients in high risk…" [S6]*

Within education increased demand on staff time was linked to:

- more families with babies requiring support,
- the caseload would include new groups of children, those with moderate degrees of hearing loss and those children who had been identified with auditory neuropathy,
- the need for increased access to services outside normal working hours,
- an increase in multi-agency working.

Eight of the education services linked NHSP with the need to re-organise service provision, to reassess how resources of both time and staffing could be best employed to meet the demands of NHSP. This was in all cases linked to a potential decrease in support elsewhere within the service, which was in itself viewed as problematic.

> *" So I think we, like a lot of authorities are looking at the other end and maybe reducing support for milds or monaurals or…or what have you, but as*

professionals you don't want to reduce the service to anybody. But …you know economically something has to go." [T17]

Time and staffing are inextricably linked but there were other time related issues that were highlighted by respondents across all services. Two social workers were specific as to the resource they perceived would be impacted most by NHSP: time. The general feeling of these services was that they did not have sufficient time to devote to NHSP. One service, on commenting on their relationship with audiology, maintained that it would be difficult to improve their relationship with audiology since the workers simply did not have enough time to devote to this.

"[So your links with audiology haven't been particularly strong but are you hoping that they'll increase?] Yes, that's more about worker time really, not because of anybody reluctant to do anything, it's just with one worker we haven't had enough resources really." [S2]

Another key concern was a skills shortage. Inability to access training was not linked to lack of training opportunities but rather to lack of time resulting from staff shortages

"I don't have the chance to keep up to date as we're so understaffed." [A4]

Additionally funding had to be identified to cover course fees and in some cases to pay for cover. As one respondent noted:

" I think the challenges of getting cover to release staff for all this training has been horrendous." [T8]

Physical resources were also identified as a key cause for concern across all services. In the case of Social services it was possible to ascertain from the questionnaire data that 70 per cent of services perceived lack of resources to be one of the main problems in the implementation of NHSP.

For audiology and education services physical resource issues were dominant, primarily equipment and space. For some, the issue was that they did not have the equipment deemed essential to meet the needs of very young children; for example, high-frequency tympanometers in the case of audiology and video cameras and editing facilities in the case of education services. In other cases, particularly within audiology services, the equipment was old and in need of replacement. The importance of appropriate working space to meet new client needs was also emphasised by both audiology and education. In the case of audiology this was linked to appropriate test and assessment facilities, for example, accommodation for screeners, or additional soundproofed rooms. In the case of education services, four respondents mentioned the need for appropriate space to run pre-school parent groups.

Four respondents in audiology services and seven within educational services specifically mentioned difficulties at a management or organisational level in arguing for additional resources and the legitimacy of these needs being recognised.

> *"The education side of it really at the um…well it's left up to the LEA's and they can embrace it or ignore it or whatever……I'm sure they are under lots of pressure with other areas of special needs, but I, you know pretty much directly I've been told that there is no additional funding for HI service…" [T1]*

6.4.6 Health professionals' perspectives

6.4.6.1 Knowledge about NHSP

Health professionals (HVs, midwives and GPs) were asked 4 questions about NHSP and awarded points from 0 (wrong) to 3 (right) for each answer. The questions were about issues that parents were reported to be likely to misunderstand (Baker *et al* 2004).

Questions were the following:

1: What are the 3 main results a baby could get after having the NHSP?[13]
2: What does it mean when a baby has recorded a clear response on the first test (OAE)?
3: What does it mean when a baby has not recorded a clear response on the first test (OAE)?
4: What happens if the baby has not recorded a clear response on the second test (AABR)?
5: Of all the babies who are referred by the screen, what percentage will be found to have a hearing loss?

To all questions, HVs gave the most correct answers and GPs gave the most incorrect answers (and often no answer at all). For question 2 there was a significant difference between the knowledge in the professional groups $F(2,273)=17.2$, $p<0.001$ and they all belonged to different subsets for $\alpha=0.05$. For questions 3 and 4 there GPs showed significantly inferior knowledge $F(2,273)=30.6.2$, $p<0.001$ and $F(2,273)=7.7$, $p=0.001$. However, knowledge about the positive predictive value at screen referral (question 5) was similarly low in all health professionals $F(2,273)=1.8$, $p=0.175$.

6.4.6.2 Attitude towards changes

NHSP has brought about changes for HVs and midwives and worry has been expressed in how satisfied these two groups of health professionals are with these changes.

A massive 93% of the HVs who responded stated some degree of satisfaction with the changes reporting to be either very satisfied (N=49/107), quite satisfied (N=34/107) or somewhat satisfied (N=12/107). Only 7% expressed dissatisfaction with the changes. There was no statistically significant difference in the level of satisfaction in hospital-based sites where the HVs had given up screening and in the community-based sites where screening was preformed by HVs on the 10-day home visit ($p=0.360$).

[13] Question 1 was excluded from the analysis.

The main reasons for high level of satisfaction was the recognition of superior effectiveness and efficiency of NHSP as well the ability to identify hearing loss at an earlier age. Positive parental response was also reported as a source of satisfaction.

The causes of concern were mainly to do with the lack of clarity about the surveillance for acquired and progressive hearing loss together with the worry about babies who were missed by NHSP. It also became apparent that HVs were not clear about whether or not IDT screen was to be phased out completely.

In midwives, 90% expressed satisfaction (N=68/102 very satisfied; N=13/102 quite satisfied; N=11/102 somewhat satisfied). Midwives appear to be happy with the role that has been given to them: although they do not seek to get too heavily involved with the screening (mainly because of their busy schedules), they are happy to support the screening team and see the following as their responsibilities in NHSP: (i) giving information to parents; (ii) ensuring test is performed; (iii) ensuring mother is referred either for completing the screen or follow-up assessment.

The majority of the midwives reported good professional relationships with the screening team whom they hold in high esteem.

Of the ones who expressed concern, it is mainly to with the business of the maternity ward and the comments were along the lines of "*There are too many people around one patient*." [MW23], "*…too many people in the busy ward…*" [MW4] and "*Everybody wants the baby's medical notes at the same time…*" [MW87]

As for GPs, a substantial 60% feel NHSP as no impact on them. Of those who thought that they would feel the impact believed that NHSP would reduce their workload. They were also expecting NHSP to give greater assurance about child's hearing both for parents as well as for GPs themselves. 56% of GPs felt there is a need for ongoing surveillance. They are mostly concerned with glue ear and meningitis, but also feel surveillance is needed for babies missed by NHSP as well as false negatives. When asked about GPs' role in NHSP, 55% felt that their major role is in listening and responding to parents' concerns.

6.4.6.3 Training needs

HVs point out various surveillance issues as an area with most pronounced training needs. They also want more training in supporting parents at various stages of screening and diagnostic process the management of screen referrals and encouragement of attending appointments. In general HVs articulate need for more information on NHSP.

Midwives also communicate the requirement for more information on NHSP and hearing loss in general. They also feel they need a better understanding about the routes of referral.

6.4.6.4 Summary

- Out of the three groups of health professionals (HVs, midwives and GPs), HVs are the best informed and GPs are the least informed about NHSP
- 93% of the HVs and 90% of the midwives are satisfied with the changes brought upon by NHSP

- HVs want more training on surveillance issues
- 60% of GPs don't feel NHSP has any impact on them, nevertheless 55% see their role in listening and responding to parents' concerns. With low level of knowledge about NHSP that could prove challenging.

6.4.7 Deaf professionals' perspectives

6.4.7.1 Information and preparation for NHSP

The majority of D/deaf professionals involved in the focus groups reported having either little or no preparation for Newborn Hearing Screening and in addition little or no information about the programme subsequent to its implementation. Those that had received information, reported fairly informal and unstructured means of obtaining the information, e.g. reading a magazine article, speaking to delegates at a conference and one professional commented:

> *"Some people at my school have some of the information, they have information sometimes, but not very often." [DP3]*

In general, there did not appear to be any formal communication (i.e. through established structures) at a local, regional or national level to D/deaf professionals regarding NHSP. Of the sixteen professionals involved in the focus groups, only one professional reported having had preparation for NHSP. This particular professional described how she received information through her workplace, e.g. magazines, newsletters and she had attended a conference in relation to NHSP and had also watched the NHSP video.

6.4.7.2 NHSP and changes in working practice

One of the aims of the focus groups was to assess the impact that NHSP had made on the working practice of D/deaf professionals. Although the majority of professionals were working with pre-school infants located within first phase sites, the impact of the screening on their working practice was in fact minimal. Of those who maintained that the screening *would* make a difference to their working practice, one professional stated that although there had been no change as yet, they were expecting an increase in the number of children that would be referred to their service. One professional expected that NHSP would mean a quicker referral process for families resulting in his earlier involvement with families as a D/deaf professional.

6.4.7.3 Timing of involvement

An area in which the deaf professionals argued for change was in relation to the timing of their involvement with newly diagnosed infants and their families. One suggestion was that D/deaf professionals be available to the family from birth, to act as a role model. Others suggested that D/deaf professionals should be involved as soon as the child's deafness is identified:

"…and obviously there's teachers of the deaf involved in the programme, but really if you're thinking about when parents are ready for us (deaf professionals) to be involved, we're following their advice, really we come into play once the teachers of the deaf say, 'ok, yes, the families are ready.'" [DP1

6.4.7.4 Working partnerships

It was proposed that joint working would help in promoting equal access and standing with the family:

"…we could go in with a co-approach to families…for a period of time you could co-work and then one or the other could remove themselves from the equation or whatever, you know, it just depends on what the parents decide they want." [DP13]

In agreement with this idea, another professional commented:

"…I think I would like deaf person in the equation, alongside a hearing person, obviously probably need an interpreter if it's a sign language user as the deaf person, delivering information, giving that information…" [DP09]

Another way to establish partnership working between teacher of the deaf and D/deaf professional was by alternate visits. As well as giving both professionals equal access to the families, it was proposed that this would have the added benefit of giving parents the experience of both D/deaf and hearing professionals.

6.4.7.5 Training

Although all D/deaf professionals involved had received training for their role within the workplace, in response to the issue of training regarding NHSP only one professional reported receiving any. This particular professional's training had consisted of attendance at a conference, general training through her workplace (e.g. meetings and support from colleagues) and observation of the screening of a baby. For the other D/deaf professionals, no formal training regarding NHSP had been received. A point worth noting is that the team implementing NHSP had organised an NHSP information day in September 2001 specifically targeting D/deaf professionals. However, none of those D/deaf professionals attending the focus groups had attended this event, in the main because they had not been informed about it; this suggests poor communication at local levels within services. (although some had not been in post in 2001).

Since the majority of D/deaf professionals had not received any training in relation to NHSP, they were asked what training they would like. Responses were varied. Two key areas of interest emerged one focussed on audiological aspects of the process and one on D/deaf /hearing working partnerships. Standardised work practice in early years' provision for deaf children was proposed, to include both positive communication practice with families, appropriate sharing of information packs and clearly established routes of referral. Other training needs specified were counselling skills; awareness of the roles of other professionals

working with deaf children and their families and training in the skills needed to work with very young children.

6.4.7.6 Routes of referral

Routes of referral for newly diagnosed deaf infants was the main challenge cited by six of the D/deaf professionals involved in this study. In Education services local management decisions about appropriateness of introducing Deaf professionals have yet to be resolved in some cases:

> *"…we're sort of…waiting for the peripatetic teachers to tell us…we're given the green light, we're given the go ahead by them to attend to you know, attend a family, to visit the house, but it's…we really take the lead from them…but my feeling is we really should be moving forward and there should be active change…for the sake of deaf babies and their families, for the sake of them, they need to have more access to deaf adults…" [DP12]*

6.4.7.7 Information for families

D/deaf professionals were keen for families to receive balanced information.

> *"…the procedure of giving information doesn't seem to be adequate, there isn't enough information on deaf issues, there's so much coming from the medical profession, there doesn't seem enough coming from deaf people…" [DP13]*

6.4.7.8 Supporting families

Several D/deaf professionals identified giving support to families as a significant challenge brought about by NHSP. Two professionals recognised that the time just after the diagnosis of their child would be an emotional time for most families and thus the challenge for them as professionals was how to work with such families in a positive and encouraging way, yet also acknowledging that it was a difficult time and being aware of parents' feelings.

> *"…it's quite a difficult situation because if I go to a family they will be upset and that will be a big challenge to reassure that it's not a bad thing…and that will be a big challenge to er…not to upset people more, try to be more positive, trying to encourage them to look to a different view and try to think, it's not all bad and that will be a big challenge for me…" [DP01]*

6.4.7.9 Status of D/deaf professionals within a service

One final challenge of NHSP can be seen in the comments of one D/deaf professional who felt that the current status or position of D/deaf professionals within early intervention services produced a challenge in itself. The first problem this professional identified was that D/deaf professionals were often at the end of a long line in terms of receiving information – she cited the example of cochlear implants and how D/deaf professionals had received the

information and related training approximately five to seven years after other professionals and parents; she felt the same was happening in relation to NHSP.

6.4.7.10 Values and benefits

In general, the D/deaf professionals involved in the study felt that screening was a good thing for deaf children and their families. Earlier diagnosis would bring about a whole range of benefits: one professional felt that it was a positive thing because it would help the family prepare for school earlier and another commented that it would be beneficial to the development of the child's communication skills; two professionals simply said that it would mean parents received earlier help and support; and another professional stated that early diagnosis was good because it meant earlier involvement with education services and earlier hearing aid fitting. One professional, having seen a successful model of early intervention from another country, felt that NHSP should mean that parents accept their child's deafness earlier. Parents having more time was seen as the key benefit by two other professionals; one felt that parents would now have more time to make decisions about communication and education issues.

6.4.7.11 Summary

The data collected from the two D/deaf professional focus groups has given an initial indication of the impact that NHSP has had on this particular group. The main conclusions that can be drawn from these data are set out below.

- The majority of D/deaf professionals (involved in this study) had received limited information on NHSP both prior to its implementation and once screening had commenced
- Where information had been received, in general it was not the result of a strategic approach to information dissemination; routes of communication were not well established. This was true at a local level since there were seemingly poor communication between D/deaf professionals and other professionals groups working within early years settings. It was also true at a national level.
- D/deaf professionals generally reported minimal impact of NHSP on their working practices.
- Only one of the D/deaf professionals reported receiving any formal training regarding NHSP. With the lack of training amongst the group, many reported their perceived training needs; these were varied and included screening rationale, counselling skills and strategies for D/deaf and hearing professionals working together in partnership.
- With a lack of awareness of the screening and lack of any training, D/deaf professionals were keen to point out the challenges they faced as a group in light of the introduction of NHSP.
- There was a general feeling that teachers of the deaf acted as gatekeepers to families and this led to frustration on the part of D/deaf professionals because they felt earlier involvement of a deaf professional would help.
- There was a concern that families would be overwhelmed by the intervention and by the number of professionals involved (after identification of deafness)

- The conclusions drawn from this analysis point clearly towards the fact that to date deaf professionals have had little involvement in NHSP and unsurprisingly it has had little impact on their working practices. In response to this, serious consideration needs to be given as to how to change the situation, and thus affirm deaf professionals as active and valued members of the early years team.

6.4.8 The perceived opportunities offered by NHSP

For the majority of respondents, NHSP was perceived to be a springboard for development. Core services constructed the opportunities offered by the introduction of NHSP in relation to five key domains:

- Improved outcomes for deaf children and their families
- Improved service delivery
- Raised professional profile relating to each group of professionals
- Improved inter-agency working
- Improved level of resource

6.4.8.1 Improved outcomes for deaf children and their families

The opportunity most frequently cited as the principal positive product of NHSP was concerned better outcomes for infants and their families. Within audiology services such improved outcomes for children were linked to earlier diagnosis, earlier amplification, improved early intervention, more sustained and detailed work with families, better interagency working, and better long-term outcomes. In the case of educational services the main focus was on improved educational outcomes and overall long-term outcomes. In some cases specific focus was placed on improved language outcomes and improved success within inclusive settings. Some placed key benefits within specific groups (those with severe and profound degrees of hearing loss), in relation to a specific habilitation approach (oral) or in relation to reduced need for support services at school age.

For Social Services the potential of achieving improved outcomes for deaf children was strongly linked to parents being offered informed choice in respect of communication approach. Social workers saw themselves as representing the values of self-advocacy, empowerment and family directedness and having the skills in how to work with families that ensured these values were fulfilled in practice.

6.4.8.2 Improved service delivery

All services viewed the introduction of NHSP as potentially offering opportunities for service development, both at the level of individual practitioner and at the level of organisational and strategic change. As a low incidence disability childhood deafness is frequently viewed as

having a low profile at strategic level where an overview of generic disability needs is felt to dominate. In the case of education NHSP was seen to be a force that raised the profile of Sensory Educational services, specifically in relation to deafness. Five services reported that NHSP and related activities had raised the profile of the service. This was seen to be important in its own right but also in providing an opportunity to accrue benefits for service development.

> *" I think it is actually highlighting to education services that education is about from nought upwards." [T16]*

A number of services highlighted additional benefits that they felt would be gained from the introduction of NHSP. These included raising deaf awareness amongst co-professionals, positively raising the profile of deaf children at a strategic level and in a number of cases achieving additional staffing to meet new needs. In one case such additional funding was through health service provision:

> *"We've got an additional 0.5 post (as a result of NHSP)" [T3]*

Whilst Social Services did not discern such a range of positive opportunities for service development there were positive service developments in a minority of areas, including, in one area, an opportunity to increase staffing:

> *" We have actually appointed one…. we have actually got a two thirds social worker who I forget to mention before….she was appointed to do post diagnosis work, but not specifically with deaf children (although including this group)" [S12]*

For audiology services developments were linked with the availability of more paediatric focussed service delivery, of achieving early hearing aid fittings and providing a more cost efficient service.

6.4.8.3 Raised professional profile relating to each group of professionals

For both audiology and education services NHSP was seen to be a strong focus of motivation linked to improved perceptions of service worth. At a time of government innovation, review and change, services are under considerable pressure to meet a range of targets. For those authorities where services had the potential to develop and grow in response to NHSP pre-implementation, morale was high and motivation to meet the challenges clearly expressed.

> *"It has involved a considerable amount of training and staff time which people have been prepared to give willingly because they are so excited about the project." [T6]*

Such enthusiasm was echoed within audiology services, for example:

"It has given us something to be proud of…an incredible morale boost" [A3]

All services shared both the belief and hope that organisation and structural barriers to joint working would be weakened and that multi-agency work would be positively encouraged. In some cases this was linked to a conviction that the process itself was so powerful it would drive positive change:

"[NHSP will] bring different services together…political boundaries will be brought down." [T27]

Services placed considerable emphasis on the potential of using NHSP as a driver for change in enabling positive links with other service providers to be forged. In the case of links with health service providers, including audiology, SLT and HV services, education respondents cast NHSP as either a way of strengthening or further developing links. In the many cases such links were already in place at a field worker and operational but required development at a strategic level.

" I think there is a culture of people trying to work together, but often you do that on the ground and it works quite well, but there are no structural issues to really support it and it's a bit personality-driven. Hopefully this will set some mechanisms for things to happen regardless of the people in the places." [T24]

In some cases changes implemented at organisational levels had led to the development of protocols that set parameters for inter-agency working and that actively promoted contact between service providers.

" I think it will just increase (multi-agency working). It has increased at the moment just because of the fact we had to come up with protocols and make decisions about how we are going to respond to various things." [T3]

In addition to developing and improving links with audiology service providers there were some specific examples of improved practice where NHSP had acted as a trigger. In several cases audiology services had invited educational services to be present at confirmation of deafness. In one authority the education service was invited to offer information and support to families who were waiting for diagnostic audiology. Whilst there was no confirmation of deafness families were reported to value the extra information and clarification offered by educational services during this time.

A strong emphasis was placed on the need and imperative to engage with Social services. In many cases the establishment of such links was perceived to be problematic, unsatisfactory and a reflection of a lack of structural organisation facilitating such engagement of Social Services with educational services for deaf children.

" It would be nice to think that it promoted better provision through Social Services and that Social Services felt they could or had something to offer," T7

There were perceived to be very real opportunities for improving and developing links across services. Post implementation education services noted very positive changes in their working practice with heath providers. Considerable enthusiasm was expressed about links with a range of health services including audiology, pre-school teacher counsellors, HVs, paediatric nurses and SLTs. Such improved links were characterised by skill sharing, inclusion in clinical settings and in sharing roles.

No single education or audiology service provider mentioned that improved links with Social Services had been achieved. Social Service respondents did however see that NHSP offered the opportunity of closer links with audiology. For one fifth of the social services interviewed it seemed that NHSP would provide a welcome impetus that would improve the joint working with audiology. However, while education services saw NHSP as a possible route for closer working with Social Services no such reciprocal view was expressed by any Social Service within first phase sites. Whilst practitioners were aware of the need to work across agencies the lack of structural empowerment to help achieve this was identified as problematic.

" I think we work very well at our own level but there needs to be a higher level of management really, which there isn't" [T 13]

Current government initiatives are actively promoting interagency working and seek to address such concerns (e.g. Children's Trusts, Early Support Programme). Whether the provision of structures alone will be sufficient to meet the challenges is less clear.

6.4.8.4 Improved level of resource

Both audiology and education itemised specific resources needed or developed as a result of NSHP. Within Social Services 65% of services expected an increase in resources to be one of the main opportunities of NHSP. However, services varied in their responses, some ranking it as the most significant opportunity, others viewing opportunities such as improved links with other services to be more significant. One social service was hopeful that NHSP would lead to an increased awareness of the need for work with deaf children and their families and this in turn might result in an increase in resources.

"[How about the main opportunities (of NHSP)…?]…well increased resources, if it's highlighted more with this programme, the issues about deaf children, then there might be a link with some resources which I'm sure would be useful…" [S11]

6.4.8.5 Summary

"The opportunities-we've had being a pilot site have been enormous"

This statement is indicative of the positive challenge, excitement and pride that the majority of respondents in audiology and education felt resulted form being a first phase site.

NHSP was perceived to bring with it opportunities across all services.

- All services recognised the opportunity to achieve or improve inter-agency working
- For audiology and education NHSP was seen to offer the very real potential of improved outcomes for deaf children and their families.
- Social Services, where they recognised a responsibility for service provision, saw NHSP as a vehicle for improving links with audiology and linked this to a system of automatic referral from audiology to Social Services
- Audiology and education services saw NHSP as offering an opportunity to both improve service provision and raise their profile at a strategic level.
- All services linked NHSP with the opportunity for an improved level of resource, this was primarily linked to the need for more staff but also included new equipment needs in education.
- All services recognised the need for further training opportunities.

6.5 In conclusion: looking to the future

The implementation of NHSP, whilst demanding changes and new working practice, was welcomed across services. It is perceived to have been an initiative that both requires and supports joint working. This, in itself, requires services to re-evaluate their roles and responsibilities in respect of very early-identified deaf children and their families.

Outstanding concerns remained in a number of key areas:

- how to manage babies identified with mild hearing losses;
- how to remain alert to children with progressive hearing loss;
- how to provide families with informed choices;
- how to address the shortage of specialised trained staff to work in audiology, deaf education and Social Services;
- how better to integrate Social Services within service delivery for families with deaf children;
- how to provide appropriate training for audiology, education, social, and D/deaf workers active with families of young deaf babies;
- how to include D/deaf professionals more centrally within service provision for early identified deaf children and their families.

7. COST AND COST EFFECTIVENESS

7.1 Previous literature

Few data have been published on the costs and cost effectiveness (i.e. studies combining costs and outcomes within a formal economic evaluative framework) of hearing screening[14]. Given the difficulties in directly extrapolating healthcare resource and costs across different countries this review of literature focuses on UK data. The findings of the previous non-UK hearing screening cost effectiveness studies will be picked up at the end of this chapter.

A survey in the mid 1990's of UK centres by Davis and colleagues (Davis *et al* 1997) found that most UK districts provide neonatal or health visitor based screening but did not have any data on costs. Two comparative cost studies of hearing screening in UK have been published.

The costs of the IDT (the 8-month Infant Distraction Test screen usually performed by Health Visitors) screening in London in 1986 were examined by Brown (1992). The study undertook a detailed analysis of referral patterns and the modelling of local data. Linking these costs and a referral rates from their survey of UK practice, Stevens *et al* (1998) estimated that the range of costs of the IDT per 1000 live births ranged from £3,316 to £5,757 at 1994 prices.

More recently, Stevens and colleagues (1998) undertook the only comparative costing of hearing screening in UK. A questionnaire was sent to a number of centres across England and Wales in 1994. Valid data were available from five centres for targeted neonatal hearing screening (TNS, targeted at babies with relevant family history, NICU history, and cranio-facial anomalies), three centres for universal newborn hearing screening (UNHS), and nine centres for the IDT. Costs for both the screen and the follow-up work were estimated directly from the survey of staff time. The costs of TNS, UNHS and IDT at 1994 prices were £5,052, £13,881 and £24,519 for a standardised district of 1,000 live births. The breakdown for the IDT cost was £19,826 for the screen component and £4,693 for follow up. This breakdown was not reported for TNS or UNHS.

Cost effectiveness estimates for UNHS and IDT were reported by Davis *et al* (1997). Table 7.1 shows the estimates obtained by combining the cost data of Stevens *et al* (1998) with screen performance data sought from the same sites.

[14] The search was carried on Medline for relevant articles published in English using key words neonatal or newborn; hearing screening; cost; and cost effectiveness.

	IDT	UNHS
Cost per child screened	£24.50	£13.80
Yield per 10,000 children screened*	2.4 to 3.0	7.0 to 14.0
Cost (£000) per child detected	81.7 to 102.1	14.0 to 19.7

Table 7.1. Cost effectiveness estimates of IDT and UNHS. *Range based on a optimistic to mid-estimate of yield across surveyed sites. Modified from Table 19, Davis *et al* (1997)

Given that the cost of UNHS per case identified was considerably less than the IDT and screen performance is superior it can be concluded that UNHS is both cost saving and more clinically effective. However, the authors expressed caution in over-interpreting these results, the costs not including set-up costs, items of equipment, consumables, or other non-direct staff costs used by each service. Furthermore, the representativeness of the sites included in this study and therefore the generalisability of the results, is unclear.

7.2 Aims

The aims of the health economic component of this evaluation were:

- To assess the relative costs and cost effectiveness of NHSP versus IDT;
- To explore the potential cost differences and cost effectiveness of NHSP when implemented in a community or hospital-based setting.

7.3 Cost and cost effectiveness of NHSP and IDT

7.3.1 Methods

7.3.1.2 Sample

Out of twenty-three first phase NHSP sites, twenty sites were asked to provide information with regard to costs associated with NHSP[15]. Sixteen NHSP sites out of 20 responded (80%). Fourteen NHSP sites were requested to give detailed costs of IDT screen[16]. Response rate was 71% (N=10/14).

[15] 3 sites that had already been involved in UNHS before of the start of NHSP were excluded.

[16] 9 sites were excluded: (a) sites that did not have IDT screen in place; (b) sites that employed a community-based model of NHSP; (c) sites that already been involved in UNHS before the start of NHSP.

7.3.1.3 Screen performance data

Screen performance data (number screened, number of referrals, and number of true cases) were derived from empirical data from the NHSP first wave sites. Details of this are given in Chapter 2 of the Report.

7.3.1.4 Costs

A societal perspective to costs was taken, and both heath care and family costs were considered. Categories of healthcare costs included: staffing; equipment (including IT); overheads; staff training and travel; and audiological follow up costs. Costs have been derived from empirical data from the NHSP first wave sites. Long-term costs were not included such as cost of treatment of detected cases and any potential cost saving from early treatment. Family costs included travel, car parking and lost parental employment. All costs were rounded to the nearest £1.

A proforma was designed to enable a comparable method of resource utilisation for IDT and NHSP models. The proforma 5.1 (see appendix) collected the following data: staff grade and full-time equivalent numbers (screeners, local coordinator, team leader, clerical staff); quantity, make and model of screening equipment; quantity, make and model of computers and printers; quantity and make of consumables; staff travel costs; and any additional costs (e.g. recruitment, refurbishing rooms, stationary). IT costs and training costs were obtained from National Health Service salary scales, the National Health Service Rehabilitation Services Catalogue (screening equipment and consumables), and the Medical Research Council Institute of Hearing Research for calibration costs. Additionally, training costs for screening and IT training costs were obtained using proformas for those attending training. Audiology services reported follow-up costs for 10 consecutive screen referrals and for all true cases.

Staff costs[17]

To calculate salaries, midpoint was taken if not specified otherwise. Health Visitor's time for IDT was estimated at 1%. This estimate was based on a health visitor screening on average 1.3 children per week and spending ca 20 minutes on the screen, which was based on data from the sites and Unit costs from Netten *et al* (2001). National insurance and superannuation were taken at 13%.

Overhead costs

Non-staff related costs refer to the overheads, building capital and equipment costs associated with running hearing screening services. Most NHSP services use a number of different facilities to deliver the different components of the programme and do not have these figures readily available. Hence, to determine these costs, the following steps were taken: allowances

[17] See references and text in the modelling paper.

for indirect overheads (the costs of the support services such as human resources, finance and estates required to carry out the services main functions) – taken as fixed cost of £2216, and building capital (the costs assigned to treatment and non-treatment space) relative to the level of pay scale based on Netten *et al* (2001).

Direct overheads

Costs associated with lighting, heating and cleaning were assumed to be 11% of the sum of staff costs, indirect overheads and building capital. This was based on previous studies carried out in hospital settings where the direct overheads were found to account for 4% to 18% (midpoint 11%) of total costs (Lambert 1994, Bricker 2000, Davies 2002). The same was assumed for community settings (we have no studies in this setting).

Equipment and IT costs

When equipment was totalled over 10 years, a 5% annuity for each year of life was allowed for. VAT was charged at 17.5%.

Consumable costs

The sites provided information of the quantity of consumables they used in November 2002 and prices were obtained from the NHS Purchasing and Supply Agency. VAT was charged at 17.5%.

Calibration costs

Calibration costs were based on the manufacturers' specifications.

Staff travel costs

Only reclaimable staff travel costs directly associated with the screen were included.

Staff training costs

For NHSP, initial training cost calculation was based on the forms that were filled in during each training session: (i) cost of attending; (ii) cost of conducting the training; (iii) venue costs. (i) and (ii) consist of travel and accommodation costs and cost of time spent by participants and deliverers. Refresher training cost calculation was based on an assumption that refresher training will be 0.5 day a year per screener.

In the case of the IDT, calculations were based on the description of the training pattern (number and duration of training sessions, involvement of senior staff etc) provided by the sites.

Cost of time spent was calculated as number of days attending or delivering training divided by number of workdays per year multiplied by annual salary.

Follow-up audiology costs

Audiology follow-up costs were calculated based on the data collected through proformas that the Team Leaders and/or Local Co-ordinators were invited to complete to fill in:

Proforma 5.11 for each of the next 10 consecutive babies referred for audiological follow-up from NHSP (see appendix). Response rate 62% (N= 123/200).

Proforma 5.11A: for every true case (born before 1st January 2004) identified through NHSP (see appendix). Data available for 138 children.

Proforma 5.15: for each of the next 25 consecutive infants referred for audiological follow-up from the IDT (see appendix). Response rate 32% (N= 112/350).

Proforma 1.4: retrospectively for each known true case born between 1st May 2000 and 30th April 2001 that was referred from the IDT (see appendix). Data available for 15 children.

Average costs of audiological follow-up to confirm false positive status were £34.99 for NHSP and £21.33 for the IDT. Average costs of audiological follow-up to confirm hearing loss were £183.64 for NHSP and £168.47 for the IDT. The audiology costs associated with identifying a true case included all the diagnostic and other procedures up to hearing aid fitting (included) and consultations by various professionals (e.g. audiological scientist, audiological physician, ENT specialist etc).

Audiology costs for each site were calculated as follows:

Audiology costsNHSP = 34.99 [(CovNHSP x NNHSP x RefNHSP) - (PPVNHSP x CovNHSP x NNHSP x RefNHSP)] + 183.64 (PPVNHSP x CovNHSP x NNHSP x RefNHSP), where CovNHSP is coverage; NNHSP is number of live births in 2003; RefNHSP referral rate and PPVNHSP positive predictive value for the site.

Audiology costsIDT =21.33 [(CovIDT x NIDT x RefIDT) - (PPVIDT x CovIDT x NIDT x RefIDT)] x + 168.47 (PPVIDT x CovIDT x NIDT x RefIDT) where CovIDT is coverage; NIDT is number of live births between 1st May 2000 and 30th April 2001; RefIDT referral rate and PPVIDT positive predictive value for the site.

Family costs

Family costs were calculated based on the data collected through proformas that the Team Leaders and/or Local Co-ordinators were invited to distribute (or ask an appropriate person, i.e. screener, audiologist to distribute) to the parents/caregivers:

An average family cost for the NHSP screen, when the screen had not been completed in the maternity unit, was £20.10 consisting of £9.58 in direct costs (travel, car parking, child minding arrangements etc) and £10.52 in lost parental wage costs. An average family cost for

NHSP follow-up was £36.11 (£20.11 in direct costs and £16.00 in opportunity costs). For the IDT screen the average family cost was £20.24 made up by £13.76 worth of direct cost and £6.48 of opportunity costs. No data were collected for family costs associated with IDT screen follow-up and average family cost associated with IDT screen (£20.24) was used in the calculations.

Family costsNHSP = 20.10 (CovNHSP x NNHSP x OPNHSP) + 36.11 (CovNHSP x N x RefNHSP), where CovNHSP is coverage; NNHSP is number of live births in 2003; OPNHSP is proportion of babies screened in the outpatient facility; RefNHSP referral rate for the site.

Family costsIDT =20.24 (CovIDT x N) + 20.24 (CovIDT x N x RefIDT), where CovIDT is coverage; NIDT is number of live births between 1st May 2000 and 30th April 2001; RefIDT referral rate for the site.

7.3.2 Results

Costs and screen performance were assessed and are presented at level of each site.

7.3.2.1 Costs of NHSP and IDT

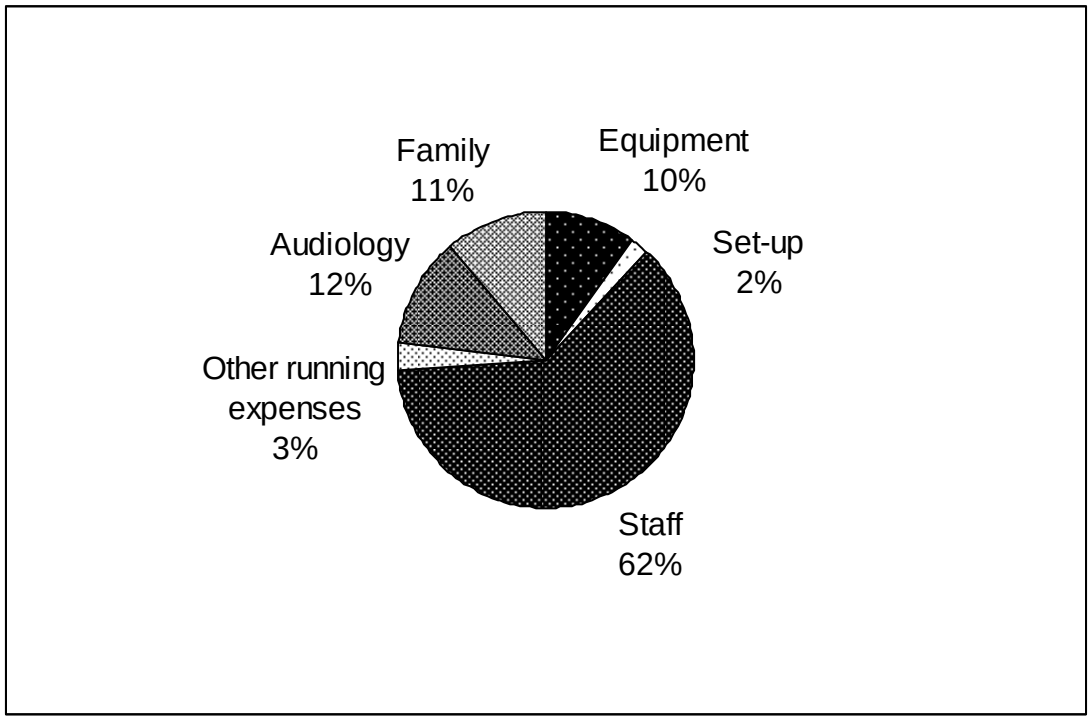
An example of the cost breakdown for IDT and NHSP for one site is outlined in the table 7.2.

		NHSP	**IDT**
SET-UP COSTS			
	Team Leader (2 years)	£9,698	Not known
	Initial training	£12,906	
	Other set-up costs	£42,747	
Set-up total costs		£65,352	
EQUIPMENT AND IT COSTS			
	Equipment	£82,450	£2,137
	VAT @17.5%	£14,429	£374
	Annuity @ 25%	£24,220	£628
	IT	£ 9,634	
	VAT @17.5%	£1,686	
	Annuity @ 25%	£2,830	
10 year equipment and IT costs		£270,497	£6,277
RUNNING COSTS			
	Staff	£218,447	£38,297
	Consumables	£3,459	
	VAT @17.5%	£605	
	Additional costs	£ 17,983.44	£10,706
	Audiology costs	£16,366	£11,972.45
	Family costs	£59,814	£111,751
	YEARLY COST	£ 302,264	£214,553
10 year running costs	10 YEAR COST	£ 3,022,639	£2,145,533
TOTAL 10 year costs	**TOTAL 10 year costs**	**£3,358,487**	**£2,151,810**

Table 7.2. Example of detailed costing for one site for which the number of births in 2002 was 9,655.

The distribution of screening IDT and NHSP costs are summarised in figure7.1.

a. NHSP costs



b. IDT costs

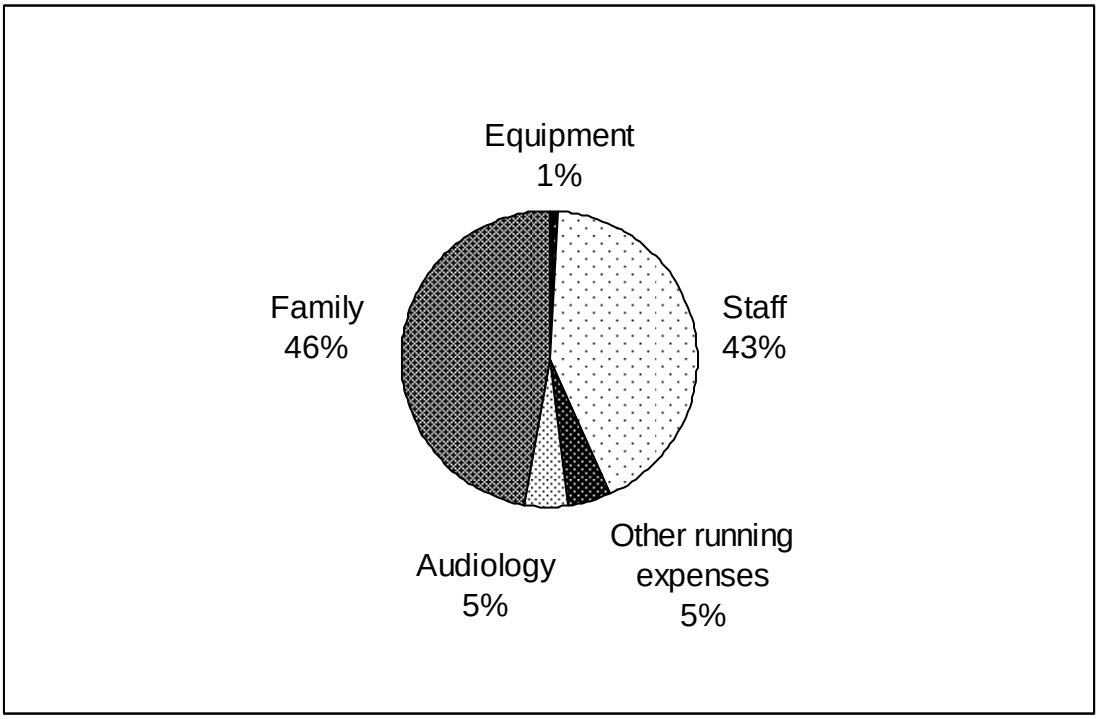


Figure 7.1. Breakdown of hearing screening costs a) NHSP and b) IDT screen.

The screening, audiology follow-up and family costs for each IDT and NHSP site are summarised in table 7.3.

a. NHSP

Cost per 1000 infants screened						
Site	**Set-up**	**Screening**	**Audiology**	**NHS cost**	**Family cost**	**Total cost**
A	£503	£24,623	£1,258	£26,384	£7,568	£33,449
B	£638	£31,271	£1,528	£33,437	£1,846	£34,645
C	£522	£25,576	£1,779	£27,877	£2,047	£29,402
D	£730	£35,747	£1,079	£37,555	£6,542	£43,367
E	£760	£37,218	£806	£38,783	£6,9608	£44,984
F	£789	£38,680	£922	£40,391	£763	£40,364
G	£538	£26,357	£1,695	£28,590	£6,195	£34,247
H	£674	£33,011	£797	£34,482	£2,624	£36,432
I	£1,106	£54,203	£565	£55,874	£6,319	£61,087
J	£429	£21,025	£1,271	£22,725	£8,256	£30,552
K	£510	£24,997	£1,525	£27,032	£1,470	£27,992
L	£635	£31,116	£901	£32,652	£4,593	£36,610
M	£635	£31,099	£2,002	£33,736	£1,408	£34,510
N	£638	£31,284	£496	£32,418	£283	£32,063
O	£867	£42,499	£472	£43,838	£1,071	£44,042
P	£659	£32,308	£306	£33,273	£1,771	£34,384
Mean	£665	£32,563	£1,088	£34,315	£3,732	£37,383
Min	£429	£21,025	£306	£22,725	£283	£27,992
Max	£1,106	£54,203	£2,002	£55,874	£8,256	£61,087

b. IDT

Cost per 1000 infants screened						
	Set-up costs	**Screening**	**Audiology**	**NHS cost**	**Family cost**	**Total cost**
Q	Not known	£19,374	£2,539	£21,913	£20,127	£42,040
R		£45,291	£1,261	£46,552	£11,630	£58,182
S		£12,846	£1,576	£14,422	£17,650	£32,072
T		£8,182	£2,287	£10,468	£15,730	£26,198
U		£34,301	£1,259	£35,560	£17,542	£53,102
V		£20,140	£3,505	£23,645	£20,563	£44,208
W		£17,201	£1,313	£18,513	£16,394	£34,908
X		£8,540	£1,501	£10,042	£16,767	£26,808
Y		£46,263	£1,811	£48,074	£19,855	£67,930
Z		£20,741	£1,778	£22,518	£16,626	£39,144
Mean		£23,288	£1,883	£25,171	£17,288	£42,459
Min		£8,182	£1,259	£10,042	£11,630	£26,198
Max		£46,263	£3,505	£48,074	£20,563	£67,930

Table 7.3. Cost of the screening programme per 1000 screened infants. NHS cost consists of Set-up costs (where known), Screening and Audiology costs. Total cost is a sum of NHS cost and Family cost.

7.3.2.2 Screen performance of IDT and NHSP

A detailed presentation of screen performance results is given in chapter 2 of this report. The yields of true cases across NHSP and IDT sites are shown in table 7.4.

a. NHSP

Site	Identified cases of permanent bilateral moderate or greater HL per 1000 screened
A	1.07
B	1.72
C	1.6
D	0.7
E	0.97
F	1.8
G	0.78
H	1.54
I	0.99
J	0.55
K	1.8
L	0.42
M	1.56
N	0.45
O	0.88
P	0.65
Mean	1.09

b. IDT

Site	Identified cases of permanent bilateral moderate or greater HL per 1000 screened
Q	0.67
R	0.47
S	0.3
T	0
U	0.36
V	0.42
W	0.26
X	0.33
Y	0.55
Z	0.24
Mean	0.36

Table 7.4. Yield per 1000 infants screened.

7.3.2.3 Cost per case of IDT and NHSP

For the purposes of this report, the cost effectiveness of NHSP and IDT is expressed as the cost per child detected. Cost effectiveness results across centres is summarised in the table below.

a) NHSP

Cost per 1 case identified						
	Set-up	**Screening**	**Audiology**	**NHS cost**	**Family cost**	**Total cost**
A	£470	£23,013	£1,176	**£24,658**	**£7,073**	**£31,731**
B	£371	£18,181	£888	**£19,440**	**£1,074**	**£20,513**
C	£326	£15,985	£1,112	**£17,423**	**£1,279**	**£18,703**
D	£1,042	£51,067	£1,541	**£53,650**	**£9,345**	**£62,996**
E	£783	£38,369	£831	**£39,982**	**£7,176**	**£47,158**
F	£439	£21,489	£512	**£22,439**	**£424**	**£22,863**
G	£690	£33,791	£2,173	**£36,654**	**£7,942**	**£44,596**
H	£437	£21,436	£518	**£22,391**	**£1,703**	**£24,095**
I	£1,117	£54,750	£571	**£56,438**	**£6,383**	**£ 62,821**
J	£780	£38,227	£2,310	**£41,317**	**£15,012**	**£56,329**
K	£283	£13,887	£847	**£15,018**	**£817**	**£15,835**
L	£1,512	£74,086	£2,146	**£77,744**	**£10,936**	**£88,680**
M	£407	£19,935	£1,284	**£21,626**	**£903**	**£22,528**
N	£1,419	£69,520	£1,102	**£72,040**	**£629**	**£72,669**
O	£986	£48,294	£537	**£49,816**	**£1,217**	**£51,034**
P	£1,014	£49,705	£470	**£51,189**	**£2,724**	**£53,913**
Mean	£608	£29,806	£996	**£31,410**	**£3,416**	**£34,826**
Min	£283	£13,887	£470	**£15,018**	**£424**	**£15,835**
Max	£1,512	£74,086	£2,310	**£77,744**	**£15,012**	**£88,680**

b) IDT

Cost per 1 case identified						
		Screening	**Audiology**	**NHS Cost**	**Family cost**	**Total cost**
Q	**Not known**	£28,916	£3,790	**£32,706**	**£30,040**	**£62,746**
R		£96,363	£2,683	**£99,046**	**£24,745**	**£123,791**
S		£42,821	£5,254	**£48,075**	**£58,833**	**£106,908**
T	NA (no cases identified)					
U	**Not known**	£95,281	£3,496	**£98,777**	**£48,728**	**£147,505**
V		£47,953	£8,345	**£56,298**	**£48,959**	**£105,257**
W		£66,156	£5,049	**£71,205**	**£63,055**	**£134,260**
X		£25,880	£4,549	**£30,429**	**£50,809**	**£81,238**
Y		£84,114	£3,293	**£87,407**	**£36,101**	**£123,508**
Z		£86,420	£7,407	**£93,827**	**£69,275**	**£163,102**
Mean		£64,689	£5,230	**£69,919**	**£48,023**	**£117,942**
Min		£25,880	£2,683	**£30,429**	**£24,745**	**£62,746**
Max		£96,363	£8,345	**£99,046**	**£69,275**	**£163,102**

Table 7.5. Cost per case permanent bilateral moderate or greater hearing loss. NHS cost consists of Set-up costs (where known), Screening and Audiology costs. Total cost is a sum of NHS cost and Family cost.

7.3.2.4 Cost effectiveness of IDT and NHSP

The incremental cost effectiveness (ICER) of NHSP compared to IDT across NHSP sites can be summarised in the following formula:

ICER (incremental cost per case detected) = (NHSP total costs – IDT total costs)/(NHSP yield – IDT yield)

The average ICER across sites is summarised in table 7.6.

	Mean yield across sites (per 1,000 screened)	**Mean total NHS cost (per 1,000 patients screened)**	**Mean cost per case detected**	**Incremental cost per case detected**
NHSP	1.09	£34,315	£31,410	£12,527
IDT	0.36	£25,170	£69,919	

Table 7.6 Incremental cost effectiveness ratio for NHSP sites compared to IDT sites

In other words, NHSP would on average cost the health service about an additional £12,500 for each additional case detected compared with IDT screening. Ignoring the set up costs for NHSP (which are not taken into account for IDT), the cost effectiveness of NHSP becomes even more attractive at about £11,600 per additional case detected. Taking a societal perspective on costs (i.e. include both health service and family costs), NHSP becomes dominant i.e. both cost saving and more effective in terms of screen yield.

7.3.3 Discussion

The data collected for this part of evaluation provide the largest comparative cost analysis of neonatal and infant hearing screening in England to date. In general, the results underpin the previous evidence of the acceptable cost effectiveness of NHSP i.e. an average additional cost of £12,500 per each additional case detected for NHSP compared with IDT screening. The cost per one identified case of permanent bilateral hearing loss is lower in NHSP. The mean cost of NHSP screening (without the audiology and family costs) to find one bilaterally hearing-impaired child is 86% of the mean cost of IDT screening to discover one such case. Mean audiology costs of following up the referrals from NHSP to diagnose one case are only 25% of the mean costs of following up the referrals from IDT screen. The biggest savings appear to be in the family costs: to find one case with permanent bilateral hearing loss families whose babies are screened through NHSP spend on average just 8% of what the families expend whose infants are screened through IDT screen.

Several studies have evaluated the cost effectiveness of UNHS protocols but only two studies have compared UNHS with other screening strategies, as in this study (Kempler & Downs 2000, Keren *et al* 2002). Encouragingly, their results appear consistent with this study. Kemper and Downs (2000) estimated the additional health care costs of UNHS, compared with selective screening, would be approximately $US24,000 for each additional case detected. Keren and colleagues also compared UNHS with a selective screening strategy, although they modelled costs over the lifetime of the child. They therefore included not only the costs of screening and diagnostic evaluation but also the costs of medical care, education and assistive devices, and lost productivity over the lifetime of the deaf individual. They estimated an additional healthcare cost with UNHS (compared to selective screening) of

approximately $US44,000 (at 2001 prices) per additional infant whose deafness was diagnosed at 6-months. Both these cost effectiveness estimates are similar to those of the present study. The higher cost of Keren *et al* is probably because they modelled the imperfect follow-up rate for diagnostic evaluation in the US. A principle advantage of the present study was the detailed primary collection of cost data across a wide range of different health care sites.

7.3.3.1 Limitations of study

First, the sites entering the NHSP in the first phase may be in the forefront of audiology services and perhaps in other services related to child health (although this is not how they were selected). This could potentially bring in a systematic bias in the costs of services compared with other services in England. Given the learning curve for effective screening, it is plausible that the relative cost effectiveness of the NHSP sites might be seen to further improve over time.

Second, some data were very difficult to collect. Davis *et al* (1997) and Stevens *et al* (1998) also reported that it was difficult to obtain reliable information about some stages of post-neonatal screen. In the case of the present study, data on the costs of the IDT screen were somewhat patchy; costs of NHSP were notably better documented. Also, as data on the IDT screen were collected retrospectively and the IDT screen was a service in the process of being phased out, the motivation to provide the Evaluation Team with data was appreciably inferior to that demonstrated by the NHSP team leaders and co-ordinators, with which the Evaluation Team had established a good working relationship. This has to be kept in mind when looking at the service costs of NHSP and IDT screen.

Stevens *et al* (1998) found that the mean service costs for universal newborn hearing screening were lower than that of the IDT screen, for a standardised district of 1000 live births. The results from the present study are quite the opposite and indicate that the mean cost per 1000 infants screened with IDT screen is lower, just 84% of the mean cost per 1000 babies screened through NHSP. The reasons why the service costs per 1000 live births are higher in NHSP are due in part to the set-up costs that were not calculated in the already long-established IDT screen. There was also more involvement of comparatively more senior staff in co-ordinating the NHSP. Equipment and staff costs are notably higher in NHSP, whereas audiology costs and especially family costs are remarkably higher in IDT screen.

The number of babies screened by an NHSP site has an impact on the cost of the programme. Sites with higher annual birth population have lower costs of screening per 1000 babies. The same trend has been noted in North America (e.g. Gorga & Neely 2003). This has to do with lower set-up and staff costs per baby screened. Costs for each identified case, on the other hand, are not associated with the number of births.

Unacceptably high lost-to-follow-up rates of 40-50% have been reported previously in the US (e.g. McPherson *et al* 1998, Aidan *et al* 1999, Mehl *et al* 2002, Gorga & Neely 2003). Fortunately, only 10% of all referrals are lost to follow up in the first phase NHSP sites (Chapter 2) which is comparatively low. Nevertheless, using the present resources to maintain the low lost-to-follow-up rates and if possible lower them even further has the potential to improve the cost effectiveness of NHSP. Adequate information to parents before, during and after the screen combined with involvement from other professionals (e.g. health visitors) are key to motivating attending appointments.

Finally, the cost effectiveness denominator in this study was the number of cases detected rather than utility, such as quality-adjusted life years (QALYs). Costs per QALY analyses are favoured by policy-makers as they allow resources to be compared and allocated not only across hearing screening programmes but also across other health care interventions. Furthermore, QALYs would capture the utility of earlier successful communication between parents and infants and psychological benefits of improved communication for deaf children and adults, as well as the disutility of false positive tests. Further research should be aimed at measuring the utilities of deaf children and adults given early or late identification and normal or delayed language abilities.

7.4 Cost and cost effectiveness of hospital and community-based NHSP (Acknowledgement: this study was run in collaboration with Eva Grill and the German UNHS Modelling team, and this section co-authored with Eva Grill)

The first implementation phase of the NHSP included four sites where the screening is performed by Health Visitors at a home visit, usually at 10 days of age. This model is called 'community-based screening,' in contrast to the 'hospital-based' model where babies are screened in maternity hospital by a new cadre of screeners prior to discharge (with follow-up of missed cases in a variety of ways). The previous section was based on comparison of the costs and cost effectiveness of the IDT and NHSP solely in hospital-based sites.

A secondary aim of this study was to compare the costs and cost effectiveness of hospital-based and community-based NHSP. The aim of this analysis was to inform policy makers as the extent to which a national screen could encompass the two different models of delivery, and if it could, on what basis areas might be permitted or encouraged to select one or the other model.

7.4.1 Methods

A decision analytic model was used to assess the cost effectiveness of the two screening systems, hospital- and community-based screening using some already-available costs data and screen performance data from the first phase implementation, data from the published literature on newborn hearing screening, and further data collection on costs from the first phase of the NHSP. A modified version of a decision-analytic model which has been developed for a German Health Technology Assessment funded by the German Federal Ministry of Health was used (Anon 2003, Grill *et al* in press).

The absolute and incremental costs and effectiveness of the two newborn hearing screening settings were estimated. The recommendations of the Panel on Cost effectiveness in Health and Medicine were followed (Weinstein *et al* 1996). The target population was all newborn infants. Health effects were presented in terms of the number of quality weighed detected child months (QCM), and true positive and false positive diagnoses at certain developmentally important ages (6 and 12 months). If a hearing impairment was diagnosed within the first month after birth, the baby added six QCM at the age of six months. If the child's hearing loss was diagnosed (strictly, identified) at the age of five months, s/he added

only one detected child month at age six months. QCM, true positives and false positives were reported at the age of 6 and 12 months and with a time horizon of 120 months. Child months which were added until the age of 6 months were weighted with a utility of 1, child months added after the age of 6 months were weighted with decreasing weighting.

7.4.1.1 The model

A state-transition (Markov) model (Sonnenberg *et al* 1993) was developed to characterise the process of screening and diagnosis through all possible stages (see figure 7.2). A child can be in one of the following states:

- Unknown status
- Healthy (hearing) confirmed by diagnostic test or screening – true negative
- Healthy (hearing) not confirmed by diagnostic test
- Hearing impaired confirmed by diagnostic test or screening – true positive
- Thought to be healthy (hearing) but hearing impaired – false negative
- Thought to be hearing impaired but healthy (hearing) – false positive
- Not followed up/not compliant

The model starts with a cohort of newborns being of unknown status and applies transition probabilities recursively to simulate how children progress through different states. In each cycle (lasting one month) children can undergo several possible transitions which accrue costs and utility weights. Ultimately all children from the initial cohort are diagnosed as healthy or as impaired or, if they are healthy, some remain 'undiagnosed' (but with true state healthy).

7.4.4.2 Data and assumptions

A predefined and externally reviewed literature search on newborn hearing screening on all relevant electronic databases has been performed. Search strategy and methods have been reported in detail elsewhere (Anon 2003). Detected publications were scored according to a standardised questionnaire and included or not. All assumptions made and parameters used are shown in table 7.7. Prevalence of congenital hearing disorders has been derived from comprehensive literature searches. The probability of hearing children presenting with falsely suspected hearing disorder has been estimated by a panel of experts. The probability of being detected at a certain age without screening has been estimated from a survey of activity in an area of Germany in 1998 and 1999. Positive predictive values have been calculated from the empirical yield data. In order to account for the heterogeneity of study sites, positive predictive values have been pooled using a random effects model (Laird & Mosteller 1990). Test parameters have then been calculated using the Bayes' formula.

The slope of the weighting function has been estimated by experts making the following assumptions: each month detected before the age of 6 months is weighted with 1, on the general assumption that children detected (and treated) within the first 6 months of life can develop typical speech and language abilities. If not detected within the first 12 months, profoundly and severely impaired children will end up with a utility of 0.85, and moderately impaired children with a utility of 0.90. Assuming that 50% of the children with permanent congenital hearing disorders are moderately impaired gives a utility of 0.875 for every month which is detected after the first birthday. The utilities between 6 and 12 months were calculated by linear extrapolation.

7.4.4.3 Model assumptions

Screening and diagnostic procedures are presented under the assumption of conditional independence, i.e. test parameters are independent of the prevalence of the condition and test results of diagnostic testing are independent of test results of screening procedures. This is plausible because screening and diagnostic testing are based on different testing principles.

Screen Performance Data and Costs

As before, screen performance data and costs for screening and diagnosis were derived from empirical data from the NHSP first wave sites (see Chapter 2). All community-based areas—East Sussex, Shropshire, Wiltshire (Bath) and Wiltshire (Swindon)—and all hospital-based areas that had started NHSP before 1st May 2002—Avon, Barnsley, Bradford, Buckinghamshire, Dewsbury, Manchester, North Staffordshire, Northumberland, and Oxford—were included in the study. Four community-based areas and seven hospital-based areas were able to provide data. Table 7.8 gives the annual birth rates of the included areas.

Within the community model, health visitors' time for NHSP was estimated at 1%. This estimate was based on a Health Visitor screening on average 1.3 children per week and spending ca 20 minutes on the screen, which is based on data from the sites and Netten *et al* (Netten *et al* 2001). National insurance and superannuation was taken as 13%.

Discounting

Future costs were discounted at a rate of 6% per year, future effects at a rate of 1.5% per year. Yearly discount rates have been converted to monthly discount rates.

Sensitivity analysis

One-way and multiple sensitivity analyses were performed on all relevant parameters. Multivariate simulations were used for probabilistic modelling (Monte Carlo). The simulation associates with each of the model variables a probability density function which represents our uncertainty about a fixed but unknown value. The ranges for test parameter estimates derived empirically and from the literature assumed beta distribution based on available ranges of estimates, and ranges for empirical cost data assumed gamma distribution. The model was evaluated for 1,000 trials.

As the number of sites was small the parameter estimates were estimated in a context of uncertainty. We wanted to evaluate the impact of extreme parameter changes on outcome and decision between alternative settings. As described by Felli and Hazen (1998) Monte Carlo simulation was performed on one parameter at a time allowing for the input of extreme values, keeping the other parameters fixed at their baseline level. This analysis was done for prevalence, sensitivity, specificity, coverage and costs. The aim was to show if there is any variation in the input parameter that might result in a change of preference between sites in comparison to the baseline result. One setting can be defined as more cost effective than another if it is (i) less costly and at least as effective, (ii) more effective and no more costly, (iii) more costly and more effective and its additional costs per unit of effectiveness are considered worth paying, (iv) less costly and less effective and the additional costs per extra unit of effectiveness for the alternative setting are not considered worth paying. One unit of effectiveness is defined as one quality weighted detected child month (QCM). The specific goals of the extremes analysis are: to show the probability that one setting (e.g. hospital) is more cost-effective than the other under the assumption that the two sites differ in one parameter, and to indicate which difference in a certain parameter between sites might result in substantial differences in costs.

Data 3.5 (TreeAge Inc.) and Excel (Microsoft Corp.) was used to construct and run the Markov model.

Estimated parameter	**Setting**	**Baseline estimate**	**Range for sensitivity analysis**	**Extremes**	**Source**
Prevalence of newborn hearing impairment %	H C	0.15	0.09-0.3	0.01-0.2 0.01-0.2	Literature (Kennedy *et al* 1998, Watkin *et al* 1998, Aidan *et al* 1999, Parving *et al* 2001, Fortnum *et al* 2001)
Sensitivity of screening %	H C	96	96-100	70-99 70-99	Literature Davis *et al* 1997
Specificity of screening %	H C	99	99	70-99	Data from sites, calculated
Coverage of screening %	H C	97	97	50-99	Data from sites
Follow-up after screening %	H C	95	95		Authors' estimate
Healthy children under suspicion of hearing impairment %	H C	0.1	0.1		Authors' estimate
Discounting factor Costs % Effects %	H C H C	6 per year 6 per year 1.5 per year 1.5 per year			
Probability of "natural" discovery without systematic screening	H C	Distribution, smoothed Weibull curve	Median age at diagnosis 18 months		Empirical data
Costs of screening per child	H C	£ 35.58 (32-40)	£ 31.99 (29-35)	£ 28-59 £ 27-43	Data from sites
Costs of audiological follow-up of referrals	H C	£ 160	£ 160		Estimate from sites

Table 7.7. Data input for the model H = hospital; C = community

This was achieved by the following procedure: The simulation was run twice with all parameters except one held fixed, the first time with the extreme high estimate of the parameter, the second time with the extreme low estimate of the parameter. This resulted in "high" and "low" estimates for costs and QCM for each setting. Differences of costs and QCM were then calculated using the "high" estimate for hospital and the "low" estimate for community and vice versa. This was done for each of the parameters mentioned. If QCM between hospital and community did not vary, only cost differences were calculated. If both costs and QCM varied the resulting distributions in mean differences of costs and QCM were combined using the Net Benefit Approach (Briggs *et al* 1998, Löthgren *et al* 2000). The ICER is defined as the additional average cost of producing one more unit of effectiveness, here the additional cost for one more QCM achieved in one of the settings, e.g. in hospital. Health care planners might decide on a ceiling value for these additional costs so that one setting should replace another setting only if the ICER is below this. From the distributions of cost and effectiveness differences the probability that one setting is cost-effective compared to another is calculated depending on a range of values for the ceiling ratio and presented in the form of a cost effectiveness acceptability curve (Fenwick *et al* 2001, Keren *et al* 2002). The probabilities presented in this curve can be used for formal statistical inference.

Area	Birth rate per 1,000 inhabitants
Avon	11.7
Barnsley	11.7
Bradford	14.5
Bucks	12.7
Calderdale & Huddersfield	13.1
Camden & Islington	14.1
Dewsbury	13.1
East London & City	17.8
East Sussex	9.3
Manchester	13.5
North Cheshire	12.3
North Derbyshire	10.8
North Staffordshire	11.0
Northumberland	11.6
Nottingham	11.5
Redbridge & Waltham Forest	14.8
Oxford	12.6
Sheffield	11.4
Shropshire	11.8
Southampton	11.2
Stockport	11.2
Wiltshire (Bath)	10.6
Wiltshire (Swindon)	12.6

Table 7.8. Annual birth rates of participating areas.

7.4.2 Results

We modelled costs and effectiveness of universal newborn hearing screening in two different settings. As test parameters were held to be constant across hospital and community sites there was no difference in effectiveness, only in costs. Both hospital and community settings yielded 134 true positive cases (89% of all cases) and 794 QCMs at the age of 6 months with

total costs of £3,690,000 per 100,000 screened children in hospital and £3,340,000 in community.

Tables 7.9 and 7.10 show the results of base case and one-way sensitivity analysis. Costs per QCM were higher by £25 in hospital-based sites. Sensitivity analysis showed that prevalence had the most important influence on costs per weighted detected child month. Lower prevalence would result in substantial higher costs for each site and in higher incremental costs. The model was, however, rather insensitive to large variations of the other test parameters. Since incremental effectiveness was set zero for base case and sensitivity analysis, the ICER (incremental cost effectiveness ratio) was not available. Figures 7.4 and 7.5 show the results of the Monte Carlo simulation. Costs would be lower in hospital sites in 48% of the trials.

	Hospital	**Community**	**Incremental**
Effects	**Base case**	**Base case**	
QCM at 6 months	794	794	
QCM at 12 months	1536	1536	
QCM at 120 months	13751	13751	
TP at 6 months	134	134	
TP at 120 months	150	150	
FP after screening and additional diagnostic	12	12	
Costs			
Costs per 100,000 at 120 months	£ 3.690.022	£ 3.343.572	£ 346.450
Cost per detected child	£ 25.813	£ 23.390	£ 2423
Cost per QCM	£ 268	£ 243	£ 25

Table 7.9. Model results base case assumption (discounted) for a hypothetical cohort of 100,000 children. (QCM = quality weighed detected child months; TP = true positives; FP = false positives).

Item	**Hospital site Cost per QCM**	**Community site Cost per QCM**	**Incremental**
Base case	£ 268	£ 243	25
Prevalence (%)			
low (0.09)	£ 437	£ 395	42
high (0.3)	£ 142	£ 129	13
Sensitivity (%)			
low (0.96)	£ 268	£ 243	25
high (100)	£ 263	£ 239	24
Costs (£)			
low (H 32, C 29)	£ 243	£ 222	21
high (H 40, C 35)	£ 299	£ 264	35

Table 7.10. One-way sensitivity analyses (QCM = quality weighed detected child month, detected child months weighted by a utility value indicating the prognosis of further speech development; H = Hospital; C = Community).

Results of extremes analysis

Higher prevalence in hospital resulted in higher costs (figure 7.3) and higher amount of QCM. Figure shows the cost effectiveness acceptability curve for the assumption that prevalence in hospital was higher than in community sites (0.002 in H versus 0.001 in C). If decision makers were willing to pay at least £500 per QCM gained, the probability of hospital being more cost-effective under this assumption would be 95%. If the willingness to pay was

below £30 per QCM, community sites were more cost-effective with a probability of 95%. For the assumption that prevalence in community sites was higher than in hospital sites, community sites were more cost-effective for any ceiling ratio. Any difference in sensitivity predicted differences in costs. Higher sensitivity in any site resulted in higher costs. If decision makers were willing to pay at least £300 per QCM gained, the probability of hospital being more cost-effective under the assumption of higher sensitivity in hospital would be 95%. If willingness to pay were below £150 per QCM, community sites would be more cost-effective with a probability of 95%. For the assumption that screen sensitivity in community sites was higher than in hospital sites, community sites were more cost-effective for any ceiling ratio. Low coverage resulted in low costs. With coverage in hospital being higher than in community, community settings would be more cost-effective with a probability of 95% if willingness to pay were below £350 per QCM. With coverage in hospital being lower, hospital settings would be more cost-effective with a probability of 95% if willingness to pay were below £200. Differences in screen specificity between hospital and community sites resulted in cost differences but not in effectiveness differences. Higher specificity resulted in lower costs. Any differences in costs per screening procedure resulted in output cost differences. With all other parameters held constant in both settings, variance in input costs completely predicted variance in output costs.

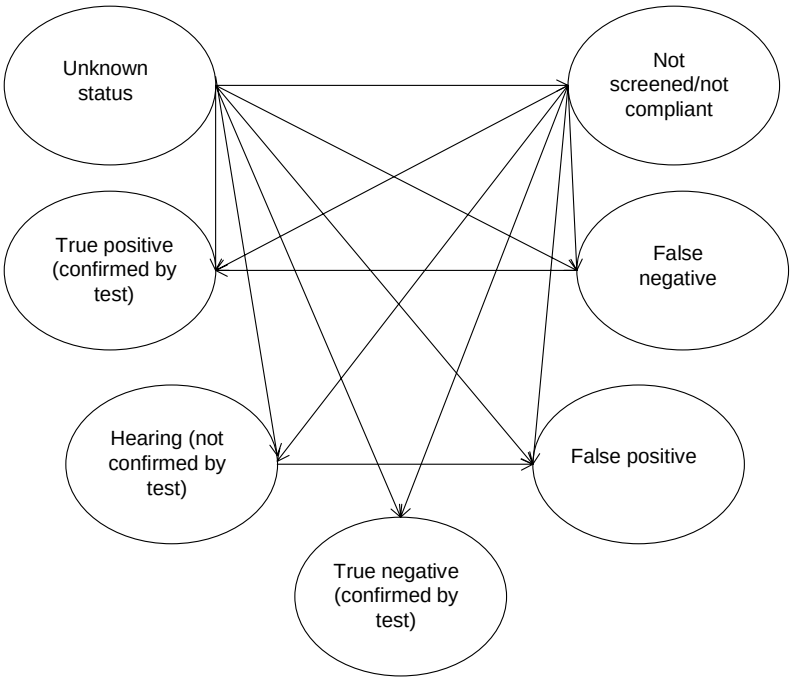


Figure 7.2. Model structure.

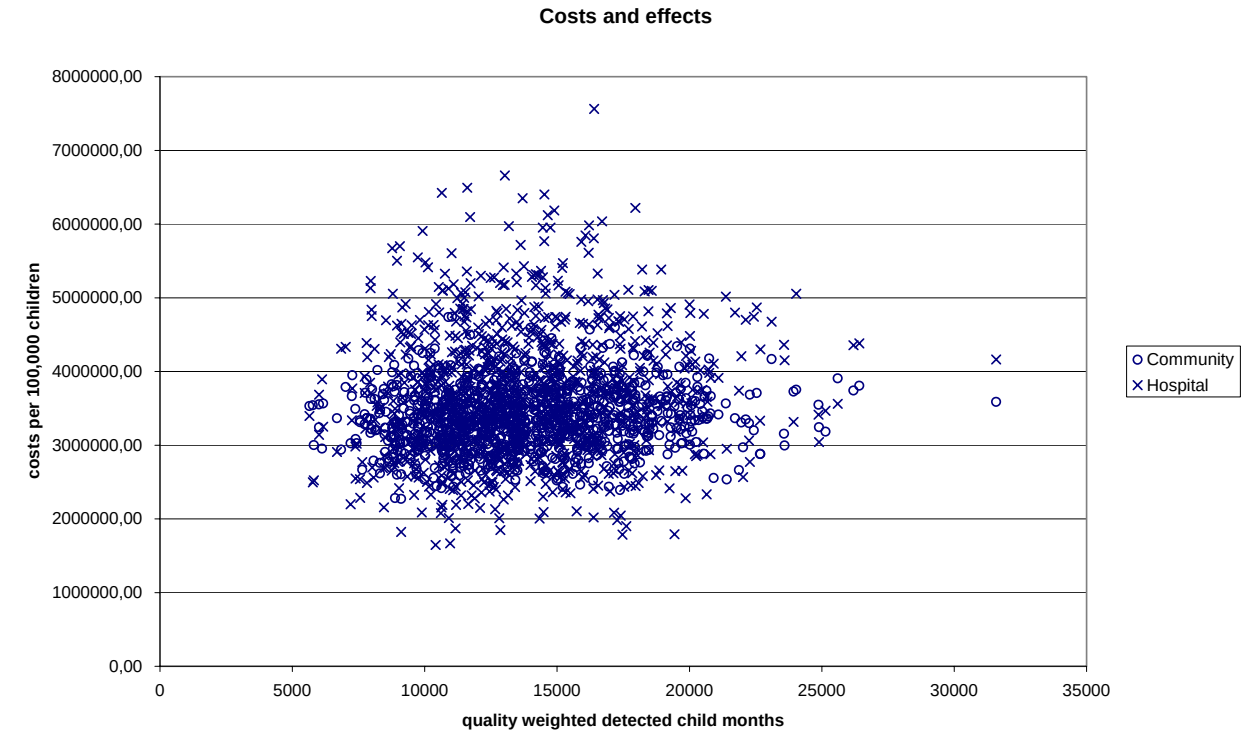


Figure 7.3. Costs and effectiveness of screening in hospital and community sites. Results of probabilistic Monte Carlo simulation (1000 trials).

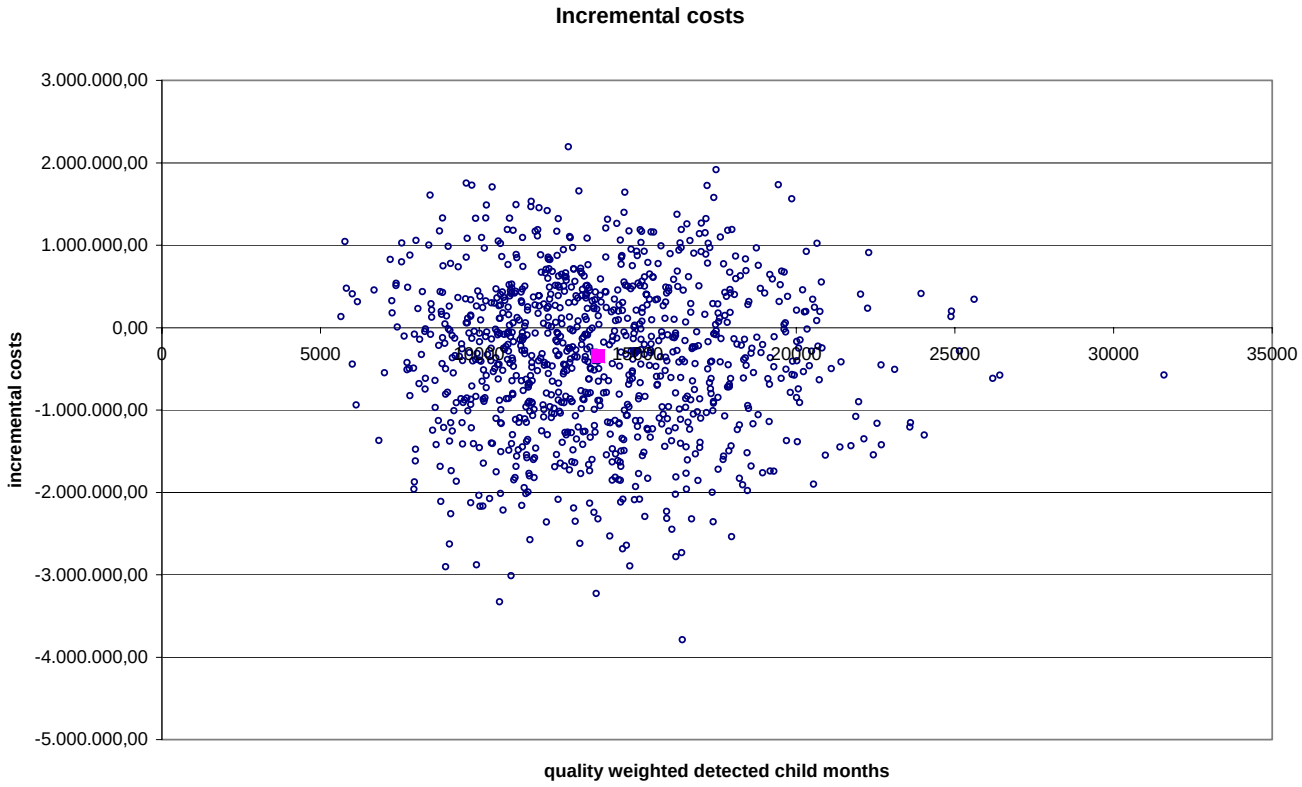
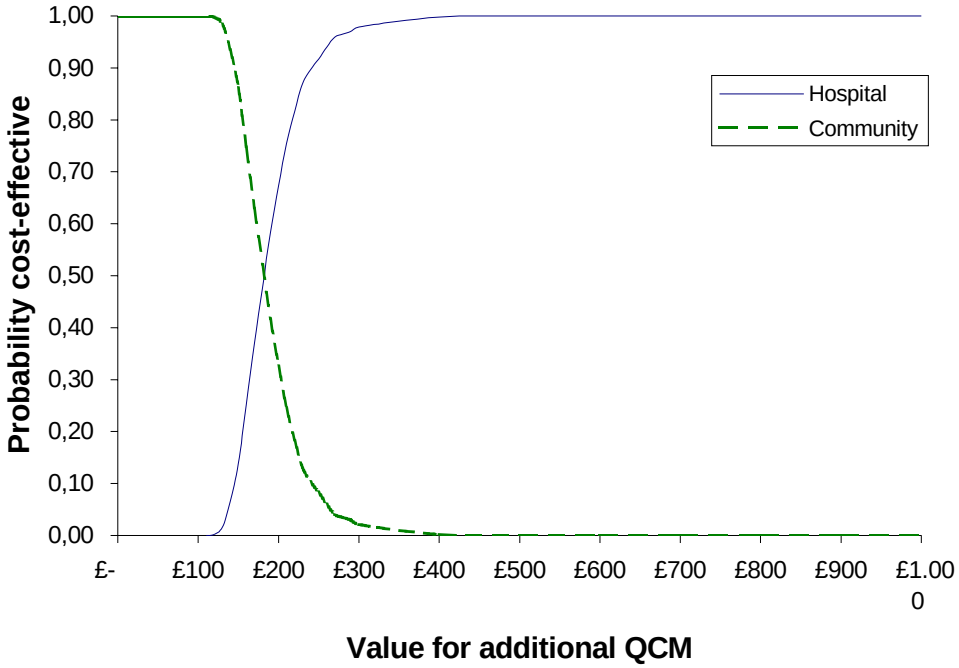


Figure 7.4. Incremental costs between hospital and community sites, Monte Carlo simulation with 1000 trials. Negative incremental costs indicate higher costs in community sites. The solid dot shows the base case result.

Figure 7.5. Cost effectiveness acceptability curve showing the probability that one setting is more cost-effective than another for a given ceiling value and for the assumption that prevalence in hospital sites is higher than in community sites. QCM = quality weighed detected child months.



7.4.3 Discussion

We applied a decision-analytic Markov model to empiric data of first stage implementation areas of NHSP in England to evaluate cost and effectiveness of different settings for newborn hearing screening. Base case assumptions with constant test parameters but cost difference between hospital and community settings yielded a cost difference of £25 per QCM: to detect one hearing impaired child one month earlier produced costs of £268 in hospital settings and of £243 in community settings. This cost difference, however, was not statistically significant. Probabilistic multivariate Monte Carlo simulation revealed that in 48% of 1000 simulated trials community settings would yield higher costs than hospital settings. The cost effectiveness of the two newborn screening models — hospital-based and community-based — did not differ significantly, assuming comparable screen performance for the two newborn screening models. Projected magnitude of costs per detected child was comparable to the costs found by other UNHS models (Nekahm *et al* 2002), suggesting the model gives results with external validity. As this is the first model to report costs per quality weighted child month, these results cannot be directly compared to other findings.

Extremes analyses showed that any statistically significant difference in prevalence, sensitivity, specificity and costs would result in significant differences in cost effectiveness between settings. Any further evaluation of cost effectiveness between different programme alternatives should evaluate in the first place if there is substantial difference in terms of these parameters.

Thus, long term best estimates of comparative cost effectiveness indicate no obvious differences between hospital-based and community-based models. It is the case that community-based model requires significantly higher set up costs (more screening devices, more people to be trained) than hospital-based model, but in the longterm analysis these costs become less significant.

7.5 Conclusions

- The NHS costs of NHSP and IDT screens in NHSP sites ranged from £26,384 to £55,874 (average £34,315) and £10,042 to £48,074 (average £25,170) respectively.
- NHSP appears to be a cost effective strategy for hearing screening when compared to IDT screening i.e. an average additional health service cost of £12,500 per additional case detected. Including family costs, NHSP is the dominant policy option i.e. cost saving and more effective (higher case detection rate). These findings support the findings of the UK HTA study of Davis *et al* (1997) and recent US cost effectiveness analyses.
- Based on preliminary data from NHSP sites, modelling indicates the costs and effects (i.e. yield) of community and hospital-based NHSP to be equivalent. However, further data are required to confirm this finding.

8. SUMMARY AND RECOMMENDATIONS

Background

The decision to implement a national newborn hearing screening programme and to phase out the existing 8-month infant hearing screen was taken in 2000, following the HTA review (Davis *et al* 1997). Implementation began in 2001 and is expected to be complete for England in 2005/6. A concurrent evaluation of the national Newborn Hearing Screening Programme (NHSP) took place between May 2001 and June 2004. The evaluation was based exclusively on the first phase of implementation, which covered 23 'sites' or service areas in England. This represents an annual birth cohort of about 120,000 births or about a fifth of the national birth cohort. Implementation of NHSP in the first phase sites began in January 2002, with the last of the sites starting screening by September 2002. Eighteen of the first phase sites used the hospital-based screening model (a new cadre of screeners trained to carry out the screen in maternity units before discharge), four the community-based model (existing Health Visitors trained to carry out screening at an early home visit), with one site a hybrid model based on a small cadre of specialist Health Visitors carrying out all screens in a community setting.

The evaluation was directed at screen performance, assessment and follow-up, psychological evaluation of the NHSP (including assessment of maternal anxiety), experience of the parents of true cases identified by the screen, the impact of the screen on related services, and costs and cost-effectiveness of the screen. The following paragraphs summarise the findings from each domain, and are presented as short summary statements for clarity. Further detail can be found by referring to the relevant chapter.

Screen performance in NHSP

1) A user-friendly tailored screening-management system is vital for managing and auditing the screening programme; eSP seems to fulfil that need, while the original systems did not.

2) 99.5% of all target babies were offered a screen; the draft minimum quality standard is 99%.

3) 97.5% of all target babies entered the screen; the draft minimum quality standard is 95%.

4) 96.0% of all target babies completed the screen; the draft minimum quality standard is 95%.

5) Refer rate decreased consistently from the beginning of the screen in 2002 to 2.7% averaged across sites by September 2003; the draft minimum standard is 3%.

6) 9.6% (95% CI 5.9-13.3%) of all referred babies had not been followed up by 6 months after referral; there is no direct minimum standard for 'lost-to-follow-up' although the draft minimum standard that 95% of referred babies should start assessment within four weeks of screen applies indirectly.

7) 11.5% (95% CI 8.7-14.3%) of all referred babies were identified with hearing loss.

8) Yield per 1000 babies screened is 1.64 (95% CI 1.27-2.01): 1.00 (95% CI 0.78-1.22) per 1000 screened for bilateral permanent hearing loss—this is similar to published prevalence rates; and 0.64 (95% CI 0.37-0.91) per 1000 screened for unilateral permanent hearing loss.

9) Aggregated screen performance data across all first phase sites were good, and met most of the current NHSP draft minimum standards; however, within these data were individual sites not performing at acceptable levels. Action is being taken by the implementation team; explicit process and procedures need to be in place to manage such under-performing sites.

Follow-up of true cases identified by NHSP

10) Based on data from true cases, median age at first follow up after screen referral was five weeks of age. Some 64% of well babies are likely to have their first audiological follow-up by 4 weeks of age. Ninety-five per cent of cases had had the first follow-up by 11 weeks of age. Reasons for the longer delays for well babies are mainly service-related and suggest the need for improvements in aspects of paediatric audiology services; efforts should be made to prioritise follow-up of screen referrals in order to shorten the waiting period to no more than four weeks, and clear explanations of the reason for the wait should be given; mothers of referred babies should be given an appointment date and time before discharge if at all possible.

11) The median age at identification of permanent bilateral hearing loss was 10 weeks which marks a major improvement compared to 18 months of age before the implementation of newborn hearing screening. Ninety per cent of the true cases identified via the screen were identified before six months of age; the draft minimum standard is 80%. Age of identification was independent of the severity of the hearing loss.

12) Age at follow-up and age of identification were not dependent upon severity of the hearing loss.

13) The median age of children who were fitted with hearing aids was 4 months which is a very considerable improvement compared to around 2 years of age before the implementation of newborn hearing screening. Eighty per cent of well babies were fitted with hearing aids by 6 months of age; including NICU babies, 90% were fitted by about 30 weeks of age (the draft minimum standard is 6 months of age). Babies with moderate hearing loss tended to be fitted later than those with severe or profound

loss, often because of parental choice. Efforts should be made to fit hearing aids, where appropriate, within four weeks of identification of hearing loss.

14) The very early fitting of hearing aids requires considerable skill and knowledge, particularly with the advent of DSP (digital signal processing) hearing aids. Systems for ensuring the quality of hearing aid fitting and management in very young infants need to be strengthened.

15) There were significant numbers of babies with unilateral hearing loss identified by the screen. Evidence-based guidelines for management are urgently needed.

16) 54% of all cases with permanent bilateral hearing loss are from an 'at-risk' population. 3/4 of these 'at-risk' babies have spent 48 hours or more in the neonatal intensive care unit. 36% of children identified with permanent bilateral hearing loss have additional conditions and/or disabilities.

17) It is not appropriate to screen babies with unilateral or bilateral meatal atresia; such cases should be automatically referred; this is now in the national protocol.

18) About 10% of the cases with bilateral hearing loss were cases of auditory neuropathy. Research into the causes, management and outcomes of auditory neuropathy is urgently needed.

Psychological evaluation of NHSP

19) Referral for diagnostic tests has a small but significant effect on mothers' emotional well-being in the first three weeks after screening; the effect is below the cut-off for clinical concern. This small but significant emotional distress following recall for diagnostic tests after newborn hearing screening is no longer evident at six months.

20) Ensuring good knowledge of possible reasons for referral seems to be protective against anxiety and thus suggests a potentially effective yet simple intervention to minimize the adverse emotional impact of this screening programme.

21) The results provide evidence to support the hypothesis that mothers of babies receiving a referral for diagnostic tests after screening experience less emotional distress if the screening is conducted in the community compared with the screening conducted in the hospital. This hypothesis awaits testing.

22) Newborn hearing screening does not cause more emotional distress than a test conducted some months later in infancy.

23) As well as its advantages in terms of sensitivity and specificity, newborn hearing screening is associated with higher levels of maternal satisfaction. Such satisfaction may help facilitate attendance for follow-up tests.

24) Hospital-based dedicated screeners expressed more job satisfaction than community-based Health Visitor screeners. Although the two groups differed in overall levels of job satisfaction, their satisfaction was influenced by similar factors. These factors need to be taken into account in continuing the effective implementation of newborn

hearing screening. Evaluation of the long term job satisfaction of hospital-based screeners is needed.

The true cases study—the experiences of parents whose children have been correctly identified as deaf through the screen

25) For parents, the defining experience of screening is how to interpret and how to respond to the inconclusive message that each stage of the process delivers. For about half of the parents in the sample, the inconclusive message gives little or no concern. This lack of concern is assisted by two main factors: the totally reassuring manner of the screener and the content of the explanation offered. Positive appraisal of screener manner was not just made on grounds of what they said, but also how they seemed as people – their character and their sensitivity.

26) The offering of an explanation why the baby had not passed the screen was important in reducing anxiety. Where explanations were vague parents were more worried. For some parents, an important element in that explanation must be an acknowledgement that deafness might be one of the range of explanations why the baby was not passing. This was of particular importance in situations where there were potentially other signs that the baby may be at higher risk (eg NICU history).

27) An explanation that set the screen outcome in a wider context was considered vital i.e. one that showed that few babies that were referred actually had a hearing loss. Where parents were told this, it was very helpful, where parents were not, it added to their growing concerns. There was evidence of the importance of checking that parents really have understood what the screen result implies rather than simply assuming that the reassuring message will of itself be adequate explanation.

28) A waiting time between the end of screening and the first appointment with audiology that was short was helpful for many families. In addition the possibility of receiving the appointment date immediately at the end of screening was especially reassuring. Knowing exactly why they were required to wait (e.g. giving time for fluid to clear from baby's ears) was also helpful. When the appointment followed on quickly it tended to be positively perceived as being part of the same process that was being handled efficiently by professionals who knew what they were doing. This routineness was linked by parents to helping to reduce stress/worry.

29) There were some examples of poor practice, and two cases raise particular concern: (i) the family who during the waiting time felt unsure whether they should communicate with their baby and if so how; (ii) the family who had received no information in their preferred language, an appointment letter in English that they could not understand and who waited three months for an audiology appointment without being sure if that was a usual period of time to wait or not.

30) Families made good suggestions about how to improve the transition to audiology for follow-up assessment; e.g. by setting aside slots of time on a regular basis for those who had been referred so that there were no unnecessary service-linked barriers to their progression through the system.

31) A minority of families would have appreciated active support during this waiting time.

32) Good explanations at follow-up assessments were a key component of what parents perceived to be good professional communication. In order for parents to positively appraise an explanation, it had to be thorough, using appropriate register or using examples that were connected to a reality with which they were familiar. Parents identified that being made a partner in the process was a key feature of good communication. One way of achieving partnership with parents is by engaging them in the testing procedures. Being approachable was identified as an essential component of professional manner. Those professionals described as unapproachable were generally those seen at the first audiological assessment.

33) The practicalities of the diagnostic process could be challenging for many families. However, having a professional that was accommodating helped to counter this. One way that professionals could be accommodating was by notifying parents of the duration of appointments so that they could prepare themselves and the baby appropriately.

Impact of NHSP on services

34) The advent of NHSP was seen to help improve inter-agency working between health (audiology services) and education (LEA support services for deaf children). Examples of improvements included increased frequency of contact, the use of IT to enable fast referral, the joint development of protocols to redefine roles and responsibilities, the inclusion of education staff at the point of disclosure, the establishment of joint care pathways, and the joint development of web-based resources.

35) Other national initiatives relating to young deaf children—MCHAS (Modernising Children's Hearing Aid Services) and ESP (Early Support Programme)—were noted to be having a significant impact on joint working.

36) Social Services rated their relationship with audiology to be good (65 per cent of services interviewed stated they were extremely satisfied with their links), but usually this is linked to their work with older deaf children, young people or adults, as opposed to deaf children 0-2 years of age. Some Social Services have no links with audiology or education services. Perceived reasons for this include Social Services workloads, lack of resources, the difficultly in establishing a specific contact point or person within Social services, lack of clarity about the role of Social services with young deaf infants and families, and strategic level barriers.

37) All three service groups (audiology, education, social care services) identified the need for appropriate training opportunities and linked this to their ability to provide a high quality service for very early identified deaf children and their families.

38) Out of the three groups of health professionals studied which have an awareness role in the NHSP programme (Health Visitors, midwives and GPs), HVs are the most

knowledgeable and GPs are the least knowledgeable about NHSP. Efforts are needed to improve awareness in these groups.

39) Almost all the Health Visitors and midwives who responded to questionnaires expressed some degree of satisfaction with the changes brought upon by NHSP; the views of non-respondents may of course differ.

40) The focus groups with D/deaf professionals indicated that these professionals have had little involvement in NHSP and it has had little impact on their working practices. Consideration needs to be given as to how to change the situation, and thus affirm D/deaf professionals as active and valued members of the early years team.

Cost and cost-effectiveness

41) The NHS costs of NHSP (universal newborn hearing screening) and IDT (the Infant Distraction Test screen at 8 months of age) in those NHSP first phase sites studied (16 sites for NHSP and 10 sites for IDT) ranged from £26,384 to £55,874 (average £34,315) and £10,042 to £48,074 (average £25,170) respectively.

42) NHSP appears to be a cost effective strategy for hearing screening when compared to IDT screening with an average additional health service cost of £12,500 per additional case detected. Including family costs, NHSP is the dominant policy option: cost saving and more effective (higher case detection rate). These findings support the findings of the UK study of Davis *et al* (1997) and recent US cost effectiveness analyses.

43) Based on the data from first phase NHSP sites, modelling indicates the costs and effects (i.e. yield) of community-based and hospital-based newborn hearing screening to be equivalent. However, further data are required to confirm this finding.

Overview and recommendations

Broadly speaking, the evidence from the evaluation points to a highly-competent implementation, delivering in the first phase sites good information for parents (via video and leaflets), well-trained screeners, an effective screen meeting most of the draft minimum quality standards. Within this aggregate picture, some screening teams (which tend to be urban with social and other challenges) have been under-performing; the implementation team is aware of these and has put procedures in place to manage the transition to acceptable screen performance. The implementation has in many ways been a model which other developed countries seek to emulate, and recent presentations (eg to the international meeting on newborn hearing screening in Como, 2004) suggest that the Newborn Hearing Screening Programme in England is regarded as a model of good practice, especially because it has been developed with a top-down public health perspective and on a whole-population basis, because a team has been funded to manage the implementation, because appropriate IT systems to support the screen have been developed, because the implementation covers intervention with health, education and social services as well as the screen itself, and because there has been a separate evaluation exercise.

Before making its recommendation for the introduction of a national programme of newborn hearing screening, the National Screening Committee (NSC) expressed concerns about the potential maternal anxiety engendered by a newborn hearing screen, particularly for the parents of those babies referred by the screen, about the ability of services in health (paediatric audiology) and education (LEA Support Services for Hearing Impaired Children) to assess accurately and manage effectively children identified very young, and about the role (or lack of) of social care services with families of true cases.

The evaluation of the first phase implementation has demonstrated, broadly speaking, that maternal anxiety is likely to be within acceptable limits, and that maternal satisfaction with the screen is generally high. There is evidence that not all parents received the screening leaflets at the time of screening, nor antenatally, nor had they seen the video; since knowledge is a protector against anxiety, this is a matter of concern. Parents who cannot read written English or understand spoken English require proper interpreter services, and it is not clear that these are fully available.

With regard to paediatric audiology services, it is clear that age of identification and age of hearing aid fitting for true cases suggest that in most cases paediatric audiology services are able to complete the follow-up assessments within appropriate time frames. Bamford et al (2001) reported on wide practice variability in audiology and education services for deaf children and families, based upon survey work undertaken in the late 1990s. It is clear from the current survey work with first phase NHSP audiological services that significant improvements have been made as a result of the implementation of NHSP, and that the ages at which identification and intervention take place is highly encouraging. However, age of identification and age of hearing aid fitting are no more than potential markers of service quality, and other sources than this evaluation do raise doubts about the quality of assessment and management of some audiological services. The Modernisation of Children's Hearing Aid Services (MCHAS) is a major NHS modernisation initiative that has used evidence-based guidelines to develop the skills and understanding in all paediatric audiology departments in England necessary to select, verify, evaluate and manage high quality Digital Signal Processing hearing aids for children in a service context that reflects not only good use of technology but also a 'family friendly' approach (www.mchas.manchester.ac.uk). Quality assurance studies carried out during the roll-out of this training show a significant number of sites where the procedures are not being used; in some cases there was evidence of unsafe practice (Sutton and Evans, internal MHAS/RNID report, 2004). This means that in these areas newly-identified deaf children would not be receiving optimal intervention, threatening some or all of the potential gains offered by newborn screening and early identification.

Judgements on the effectiveness of management of very young deaf babies by education services are more difficult to make, but it is clear that the NHSP programme has had a major effect by stimulating the development of the ESP (Early Support Programme—now no longer a Pilot), funded by DfES, which is producing a number of key materials to support teachers of the deaf, families, and others in the early management of deaf babies. The impact studies also indicate better cooperation and collaboration between audiology and education services as a result of NHSP implementation. The timing of these initiatives was such, however, that the benefits are probably to be found in the later phases of the implementation, and concern must remain about their uptake in the early phases of implementation.

The NSC's concerns over the lack of involvement of social care services has been borne out by the evaluation, and this is being addressed by the NHSP implementation team: a study has

been commissioned and draft recommendations made to develop the role of social care services, although resource issues represent a crucial barrier to progress in this area.

IT systems are key to the successful management, audit and quality of a screening programme, and to the facilitation of longer term strategic decisions. The early decision of the implementation team to use two different off-the-shelf IT systems was in retrospect unfortunate, since neither was user-friendly enough nor reflected the decision tree in the NHSP programme, and using two systems led to difficulties with merging data, but the situation was rectified relatively quickly with the development of the eSP system. This comprehensive system has met user expectations and is the first national system to be integrated with the central issuing system for NHS numbers (NN4B); it is important that the eSP is fully integrated with future systems and is not undermined by the introduction of the new NHS IT systems.

Another key to a successful screening programme is the use of agreed protocols. This is particularly the case with the screen itself. Some first wave sites unilaterally altered aspects of the protocol including in some cases performing a second screen on those not passing the first screen. For example, the use of an OAE test as an initial step for those referred is acceptable only if it is seen and explained to parents to be part of the follow-up assessments; if called a 'further screen' it will not accord with the information already given to parents, will be likely to cause increased anxiety, and will undermine people's understanding of what a screen is. Local variation must be avoided in order to preserve successful audit and quality assurance. That is not to say that protocols must never change; rather, they should be based upon evidence of gains (cost-effectiveness, increased benefits, reduced harm etc), and should be agreed nationally and implemented across all sites so that IT systems, and training and information to parents can be brought into line with the changes. Such changes should be based on robust evidence—the source of such evidence will be the national implementation itself, obtained through the ongoing quality monitoring and via agreed sub-trials of protocol changes (which should only be undertaken after full implementation has been achieved).

This raises the issue of the use of different models for the screen within a single national protocol. The brief to the Implementation team made clear that while the programme should in the main be a hospital-based programme, the National Screening Committee wished to accede to the request from proponents of community-based screens to include this as a model in a number of sites. This allowed the evaluation to compare aspects of the two models. The data from the evaluation suggest that, at least in the 18 hospital-based sites and the four community-based sites, both implementations meet the draft minimum standards for the screen performance. However, within these data the community-based sites had higher coverage, lower refer rates, higher positive predictive value and lower lost-to-follow-up rates. Caution should be exercised in generalising from these data since there may plausibly have been a selection bias in the sites selected for community-based screening. Levels of maternal anxiety associated with the screen were comparable and at clinically acceptable levels; the question of whether levels of anxiety for false positive referrals are less in mothers who have experienced the community-based screen than in those who have experienced the hospital-based screen remains open—we hope to address that with data collection from other community-based sites in the medium term. Finally, on the basis of the data available on screen performance, the cost-effectiveness comparison shows no significant differences between the two models.

On the basis of these somewhat limited findings, therefore, it could be argued that either model could be implemented. One issue which would argue against the community-based model is the extent of set-up costs, which although the effect lessens with cost modelling over a longer time frame, in the short term are significantly higher for the community model: training a large cadre of Health Visitors, and purchasing significantly more screening devices, would undermine the current resourcing of the implementation if that model were to be introduced more widely. There are of course a number of dedicated and articulate proponents of community-based screening, and it could be viewed as being in accord with the move from acute-based services; these professional colleagues deserve to be listened to. On the other hand, this cannot be allowed to prevent difficult decisions being made. Since IT systems are increasingly at the heart of service quality-assurance and future decision-making (*'the information system is the nervous system of a screening programme*' Muir Gray, NSC Programme Director's Report 2003-4), there is an argument that if there is nothing to choose between the two models, then the one option that should not be chosen is to mix the two, since this makes the quality assurance systems more difficult. (Note that hospital-based screening will have a variety of systems for covering those babies whose screen was not completed in hospital, and that some of these will be 'community-based' in the sense that the screen is carried out at home or in community clinics by the screeners; this is entirely appropriate and should not be confused with the argument here which is about all screens being carried out by Health Visitors as a small part of their routine workload).

The NHSP quality assurance working group has drafted a specification for the NHSP quality assurance (QA) services (see Appendix). The strategy for QA accords with the NSC proposals for QA of screen programmes, although since, as the NHSP Director has pointed out, newborn hearing screening is somewhat different from the other programmes in that it has arguably less need for assessment of method and protocol but more need for monitoring the post-screen activity in health, education and social care, it needs to cover pre-screen, screen, post-screen assessment and post-screen support. Acknowledgement of this wide QA brief accords with the original brief for the NHSP implementation. The QA specification is central to the future success of NHSP, and requires the appropriate infrastructure and staffing.

Three further points may be made about QA and monitoring of the programme. First, the continuing role of the NHSP Steering Group in the governance of the programme, alongside the role of the NSC, would benefit from clarification. Second, the draft QA specification has the support of the evaluation team, but would benefit from details of the processes that come into play when QA indicates problems. Third, there is lack of clarity about whether QA for NHSP could be delivered solely by quality assurance systems that are integral to service management, or whether there should be a separate central QA team in addition. Given the multi-agency nature of NHSP as a service, we favour the latter.

Relevant to QA are a number of outstanding issues, many of which have already been referred to in different parts of this report. It may be helpful to reiterate some of these:

- research is needed on the outcomes associated with mild hearing loss and babies identified with unilateral hearing loss, and on the appropriate management; this will have implications for the case definitions for NHSP.

- surveillance systems need to be implemented in order to remain alert to children with progressive, late onset and acquired hearing loss; guidelines are now available from the implementation team;

- work is needed on how best to provide families of children with hearing loss with informed choices;

- there is a significant shortage of specialised staff to work in audiology, deaf education and social care, and strategies need to be in place to address this; how to provide appropriate training for audiology, education, social, and D/deaf workers active with families of young deaf babies is a related issue;

- there are doubts about the quality of some paediatric audiology services in England, particularly with regard to post-screen assessment and the fitting and management of digital signal processing hearing aids; such services need to be identified, and support and training systems put in place;

- agreement needs to be reached on how better to integrate social care services within service delivery for families with deaf children, and the resource issues addressed;

- the factors relevant to job satisfaction for screeners need to be taken into account in continuing the effective implementation of newborn hearing screening.

Finally, a comment on the evaluation itself. The timing and aims of the NHSP evaluation have been atypical. The procedure to be followed when the research evidence suggests that a new screening programme should be introduced is usually to complete a pilot implementation, followed by full implementation if the results of the pilot are satisfactory. Pilots are 'a useful mechanism for testing the feasibility, public acceptability and cost-effectiveness of new screening programmes in practice' (National Screening Committee). However, the case for introducing newborn hearing screening, and for phasing out the existing poorly-performing 8-month Infant Distraction Test screen, was so strong that the NSC recommended immediate national implementation (on a phased timescale) in parallel with an evaluation of phase one of the implementation. The clinical and political imperative of such a decision in effect made this evaluation an evaluation of the *how* rather than the *whether* of implementation, and the recommendations reflect this context.

Should such a situation arise in future, with another screening programme, the following points should be considered:

The delays to the NHSP evaluation, and the consequent extension into 2004, arose in large part because of delays in the NHSP implementation caused by a number of issues but in particular the early problems with IT systems. It is probably too hopeful to expect a new

programme of this complexity to proceed from day one as planned, and so the evaluation should be timed to start some time after the start of implementation.

If an evaluation of a screen is planned, considerable thought needs to be given to how data are to be collected, and how reliable that would be at the beginning of a programme. In this case both the evaluators and the implementers realised early on that the IT systems were inadequate and changes were made. It could be argued that an evaluation ought to have separate stand alone reliable data collection systems—and for a strict pilot (i.e. where no decision has yet been made on implementation) this is probably so. But where as here the evaluation is coterminous with the implementation, this probably cannot be justified (in terms of professionals' time and resources).

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