Archives of Disease in Childhood 1990; 65: 1193–1198

ARCHIVES OF DISEASE IN CHILDHOOD

The Journal of the British Paediatric Association

Annotation

Hearing screening in children—state of the art(s)

General principles of screening

Many of the 10 Wilson-Jungner principles of screening[1] are now widely familiar, even among those who cannot recite them all or attribute the source. The principles refer to the need for acceptable tests that will predict with reasonable accuracy during its asymptomatic stage the presence of a disease (or of an impairment that will lead to a disability) to the necessary availability of acceptable effective treatments, and to various other ethical and practical prerequisites for screening asymptomatic populations. These principles have never been seriously challenged, although there is reason, when exploring fast moving fields or when the objective is the secondary prevention of disabilities, to avoid applying them over literally. Current reappraisals of child health surveillance[2] and further ethical considerations[3] urge four additional principles:

(xi) The incidental harm done by the screening, and by the information (correct or otherwise) that it gives, should be small in relation to the total benefits from the screening-assessment-treatment system.

(xii) There should be agreed guidelines on whom to divulge the provisional and the final results to, and on when and how this is best done; there should be transitional counselling support where necessary.

(xiii) All screening arrangements should be reviewed from time to time in the light of changes in demography, culture, health services, technologies, and the epidemiology of the target conditions.

(xiv) Because 'cases' are not homogeneous, the balance of costs, benefits, and risks from screening, assessment, and treatments has to be worked out on a stratified (demographic or case type) basis, and the definition of the target group has to be revised so that this balance is favourable for all strata within it.

Reflection on the 14 principles leads to a more cautious approach to screening than has recently prevailed in some quarters.

Subject to three qualifications of degree listed below, severe profound prelingual hearing impairment does satisfy the principles; screening for otitis media with effusion would require a lenient reading of several of them. Incomplete, but coherent and scientifically underpinned, evidence indicates that early fitting with hearing aids is advantageous for linguistic and cognitive development in permanent hearing loss.[4] Early detection is therefore an appropriate public health goal, potentially to be achieved by some form of surveillance such as a screen.

In practice the three most important principles affecting screens of hearing concern the availability of suitable screening tests, of audiological assessment, and of effective treatment. Test methods have to be predictive but simple, inexpensive, and robust. The high requirement for skilled interaction and the difficulty in achieving cooperation of young children entail that screening by behavioural tests between about 1 year to about 4 years of age cannot be robust; this has not stopped some authorities employing undertrained staff, or misemploying highly trained staff, in behavioural screening within this age range.[5] In post-screen assessment, measurement of the hearing of most children at any age is feasible, although even here obtaining accurate results on very young or uncooperative children is resource intensive, and not always an option, given the gross shortfall of trained paediatric audiologists. Until a network of properly staffed children's hearing assessment centres spans the country,[6] with technologically competent staff and ready access to a full range of techniques for fitting and verifying the performance of suitable hearing aids, it is questionable whether these three requirements are *fully* met. The basis for true diagnosis is rarely present; though there are valid hopes for the future, it is hard at present to justify attempting it, in terms of practical benefit. This has not stopped the term 'diagnosis' being widely misused to refer to the assessment stage where suspected impairments are confirmed or otherwise.

Public health and technique aspects

Late detections are rightly viewed both individually and statistically as serious failures on the part of preventive child health services. Data from screening trials and service monitoring question whether sufficiently early detection of hearing impaired children is being achieved by existing screens on a scale commensurate with their costs. There are five main problems with the screens done by health visitors at 7–9 months, and also with intermediate screens that may exist between 1 and 4 years (although the main problems are less pressing in neonatal and school screens where populations are captive). The five problems are: (i) difficulty in converting free ranging contingency responsive professionals such as health visitors into followers of detailed audiometric procedures[7]; (ii) great difficulties in ensuring coverage above about 95%, with often much lower coverage in deprived areas and among older children; (iii) high risk values for children in problem families not typically reached

by the arrangements[8 9]; (iv) a continuous 'background trickle' of detections due to professionals and relatives from birth onwards, on top of which it may be hard to achieve or to show much further yield from the screen; and (v) the high prevalence of fluctuating otitis media with effusion from about 6 months to 6 years of age, complicating any screen with a referral criterion milder than 40 dB hearing loss, and overloading the services with children, of whom only a small minority with permanent hearing loss or persistent otitis media with effusion can or should receive surgical intervention. These five problems conspire to reduce the actual incremental yield of cases from any particular screen.

Typical service review data include the age of detection of known cases, and numbers of cases referred by health visitors; these, plus the presence of multiple screening arrangements can give a false sense of security. Such data are insufficiently analytic to relate the effort and hence the cost of screening to indices of actual health output such as the yield. Some district record systems cannot even demonstrate what proportion of detections are directly due to the screens that occur locally. Equally, claims about sensitivity and specificity of the test procedure under research conditions may not be relevant to a general or particular implementation. For example, the distraction test screen by health visitors at 7–9 months is virtually standard. The *test* can be an excellent and accurate tool in skilled hands,[7] with acceptably high test sensitivity, and it can have positive predictive value over 80%,[3 8] but this is not the main point. In typical screening implementations, the noisy environments, 'rusty' skills, and low coverage lead in effect to much lower sensitivity and positive predictive value for the *system*. Recent studies have documented implementations that are clearly inadequate.[8 10] In the districts with least good services, the median age of confirmation is currently over 2 years—not significantly better than would be expected from that achieved by the 'background trickle' processes involving general practitioners and relatives; only 10–20% will have been detected by 7–8 months. Evidence that many of the difficulties can be overcome with special health visitor training programmes[3 7] may not apply to authorities where population involvement is low or staff recruitment difficult. As only about two to four in every 10 target children found are found by these screens and improvements in their quality, while laudable, can exert only a small effect on the mean age of detection, strategic effort might perhaps be first directed elsewhere.

The crux is whether existence of a screen materially reduces the mean age of detection of permanent hearing losses for the target group in question, compared with absence of that screen. Sensitivity is only one aspect; timing, coverage, and feasibility of rapid follow up are also important. For example, if neonatal screening had a yield of only 50% of the children requiring hearing aids and special habilitative support, this would lead to a materially better result than existing screens at 7–9 months give, because of the very early timing. Davis and Sancho calculated that neonatal screening on an at risk basis could exceed 50% yield, provided that test sensitivity was high. Cross referral of congenital dysmorphologies plus predocumentation of any family history would also need to feed into the screen, adding to presence of the infant in neonatal intensive care, as features defining the risk group.[11] In the event, implementation problems and an apparently lowered prevalence in recent years of impairments in graduates from neonatal intensive care units bring the yield to more like 40–45%. However even this will almost halve the mean age of detection and reduce the median age down to below 1 year when neonatal at risk screening is introduced.

After some false starts with insensitive and non-specific automated behavioural tests such as the auditory response cradle, neonatal at risk screening with sensitive physiological tests has reached the agenda. Child screening versions of tests based on the evoked otoacoustic emission (EOAE)[12] and simplified auditory electrical brainstem response (SABR)[13] show considerable promise of adaptation to the premature population, without excessive loss of sensitivity or specificity. (I do not quote precise figures here for detection age, sensitivity, or specificity because they are always relative to particular populations, and precision in these respects is often spurious in the light of the more influential shortcomings of implementation.) Stevens *et al* suggest that a sensitive screen with EOAE, followed by the more specific contingent sifting by SABR of the failing cases, would be optimal, and this deserves evaluation on very large numbers.[14] McLelland and colleagues in Belfast (R McLelland, *et al*; personal communication) have documented a reduction in median detection age from over 2 years to under 9 months, on the introduction of a neonatal at risk screening programme with auditory brainstem responses, confirming the predicted public health potential of neonatal screening. Although the general picture looks very promising, sufficient trial data have not yet accumulated on neonatal at risk screens to recommend optimal technologies, criteria, and procedures, or to document costs exactly. For congenital sensorineural hearing impairment, with a nominal prevalence rate (including moderate cases) around one per thousand, statistical significance is conspicuously absent from published data, because, to be adequately powerful, trials would need to run for years or to embrace whole nations.

Universal neonatal screening has not yet become credible from the logistical or cost point of view. If current development work were to succeed in making a test short, cheap, and robust enough, universal neonatal screening in 10 years' time would be just conceivable. The main obstacle is the false positives swamping the available assessment facilities, making it impossible to assess all the screen failing children within an acceptably short delay, and undermining the objective of early detection and confirmation! On an at risk basis, assuming that neonatal intensive care facilities are attached to maternity units approaching 5000 births/year, and that agreement is secured for a few simple, but systematic questions to all mothers to be about any family history of deafness, the notional cost per hearing impaired child detected is of the order of £4000 at current prices. This would deteriorate by a factor of about eight for universal screening, and there would be horrendous logistical problems in arranging testing with currently available equipment during the short stay now favoured in maternity wards when births are normal. In relation to the other high costs already incurred neonatally by many of those at risk, and in relation to other costs of rehabilitation and life long support necessary for late detected deaf children, £4000 seems small and highly cost beneficial. However, as the subsequent costs of non-detection fall on departments other than audiology and neonatology (that is, two which would have jointly to set up neonatal screening and the arrangements for assessment), these cost advantages for screening may not immediately win the argument for establishing neonatal at risk screening district by district. Directors of public health with their transdisciplinary perspective would have to take a leading role; the cost effectiveness of neonatal at risk screening should carry considerable force in gaining their support.

By school entry, the screening version of pure tone audiometry ('sweep') is straightforward, and the screening systems employing it are virtually standard. Screening in later childhood is unjustified epidemiologically.[3] In the age range 1–4 years, screening practice in the UK is arbitrary,[3 5] and with the exception of impedance testing for otitis media with effusion there are no proper published studies of opti-

mal method or applications to populations upon which to base a clear view of what should be done in this age range. Spreading recognition of the above five problems of current screens by health visitors with the distraction test, plus the difficulties of treatment and coverage in the intermediate years with existing tests, has led to reappraisal. Screening by test may occur in the future only for the neonatally at risk that it is practicable to test, and universally for school entrants; some other form(s) of surveillance would then have to be delivered for the intervening years. Research and development are required to specify what these alternatives and their performance indicators should be, and to determine how the numerous cases of otitis media with effusion that are non-persistent can best be routed past or out of the system. To deny a dilemma here, on the grounds that general practitioners will in future conduct appropriate surveillance under the new contract, is naive in the instance of hearing, because the problems of coverage, monitoring, training, acoustic environment, and public health control at present seen with health visitors will remain or could even increase. The nature of the work to be done is not only unspecified, but unclear as to desirable and feasible goals. Development of alternatives is urgent, because there are signs that some authorities are already abandoning health visitor distraction test screens in the first year.[15]

Implementation

Deficiencies in the objectives, control, and implementation of screening programmes are widespread, and have diverse causes. Chronic funding difficulties are compounded by problems of recruitment, training, and grading. Justifying the effort on solving these problems presupposes a good public health strategy. Screening programme managers need to harmonise with the directors of public health and with unit general managers to ensure that the goals of a screen are clear and that it is feasible. Where a screen is of long standing, aspects of technique and values of referral criteria may go adrift in the absence of monitoring. On new programmes, an inadequate number or level of staff may have been provided at the outset, or appropriate scientific audiological input may be lacking.[16] Beware the salesman and the accountant who suggest that 'automated' equipment can be run by an unsupported 'button pusher'; developers of equipment rarely achieve that declared goal. In the sharp environment of neonatal intensive care, for example, undergrading of the tester is a recipe for poor cooperation, low coverage, and unreliable results. Commitment to the value of the programme has to be secured, so that virtually all target children will receive the test, and staff sickness or other types of hiatus will not reduce coverage. This is especially important where the screening arrangements are new, and especially with neonatal at risk screening, where the activity is undertaken or hosted by neonatalogy, a discipline having priorities concerned with life and death, as well as with quality of survival. The wide range of staff whose cooperation is required suggests that published data documenting the effectiveness of screens elsewhere will be necessary, if not sufficient, to secure commitment of all parties. The most crucial step is for neonatologists to agree that they among others 'own' the problem of prelingual deafness.

Listing thus the problems of 'real world' implementation might appear a discouragement from instituting screens that could be made epidemiologically and audiologically valid. However, faith has to be tempered by realism. Creeping, under resourced 'improvements' are all too strong a temptation when the enthusiasm to do good is high but the funding prospects are low. Unfavourable reports of ineffective screens spreading out from under provided and faltering implementations can blight their prospects, however good the idea might be. Programme managers versed in policy, practice, and health service tactics are just as necessary as epidemiologists and bioengineers. The vocabulary of sensitivity, specificity, and positive predictive value was agreed and spread widely 20 years ago, when public health doctors and developers of test procedures recognised one another's arts. They now increasingly have to acknowledge a third art: the managerial art, which is essential for prioritising, establishing, and consistently delivering effective secondary prevention, amid the rival possibilities that compete for attention and funds. A major basis for that art is good research information on what is worth doing. While more such information is, as ever, required, this does not excuse failure to act on the best information currently available.

M P HAGGARD

MRC Institute of Hearing Research,
University of Nottingham,
University Park,
Nottingham NG7 2RD

1 Wilson JMG, Jungner G. *Principles and practice of screening for disease.* Geneva: WHO, 1968.
2 Hall DMB, ed. *Health for all children: a programme for child health surveillance.* Report of the Joint Working Party on Child Health Surveillance. Oxford: Oxford University Press, 1989.
3 Haggard MP, Hughes E. *Screening children's hearing: a review of the literature and implications of otitis media.* London: HMSO, 1990 (in press).
4 Markides A. Age at fitting hearing aids and speech intelligibility. *Br J Audiol* 1986;**20**:165–8.
5 Stewart-Brown S, Haslum MN. Screening for hearing loss in childhood: a study of national practice. *Br Med J* 1987;**294**:1386–8.
6 Haggard MP, Pullan CR. Staffing and structure for paediatric audiology services in hospital and community units. *Br J Audiol* 1989;**23**:117–22.
7 McCormick B. Hearing screening by health visitors; a critical appraisal of the distraction test. *Health Visitor* 1983;**56**:449–51.
8 Dhont G, Blontrock J. Results of screening for hearing disorders using the Ewing test. (In Flemish.) *Acta Otorhinolaryngol Belg* 1988;**42**:319–26.
9 Johnson A, Ashurst H, for the Steering Committee, Oxford Region Child Development Project. Screening for sensineural deafness by health visitors. *Arch Dis Child* 1990;**65**:840–5.
10 Brown J, Watson E, Alberman E. Screening infants for hearing loss. *Arch Dis Child* 1989;**64**:1488–95.
11 Davis AC, Sancho J. Screening of hearing impairment in children: a review of current practice in the United Kingdom. In: Gerber S, Mencher T, eds. *International perspectives on communication disorders.* Washington: Gallaudet University Press, 1988:237–75.
12 Cope Y, Lutman ME. Oto-acoustic emissions. In: McCormick B, ed. *Paediatric audiology, 0–5 years.* London, Taylor and Francis, 1988:221–46.
13 Mason SM. Automated system for screening hearing using the auditory brainstem response. *Br J Audiol* 1988;**22**:211–4.
14 Stevens JC, Webb HD, Hutchinson J, Connell J, Smith MF, Buffin JT. Click evoked otoacoustic emissions compared with brain stem electric response. *Arch Dis Child* 1989;**64**:1105–11.
15 Scanlon PE, Bamford JM. Early identification of hearing loss. *Arch Dis Child* 1990;**65**:479–85.
16 Davis AC. Neonatal hearing screening: part of an integrated health service for the 1990s. *Newsletter of the British Association of Audiological Scientists* 1990; **17**:36–42.