

SCREENING FOR HAEMOGLOBINOPATHIES – A SYSTEMATIC REVIEW*

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The haemoglobinopathies are serious diseases with reduced life expectancy and requiring life-long treatment. They mainly affect ethnic minority groups; Asian, black and Mediterraneans. About 10% of all births in the UK are to these higher risk groups and although they are concentrated in the major cities, about 30% of all affected birth occur in districts with less than 5% of ethnic minorities. Antenatal haemoglobinopathy screening is intended to identify pregnancies at risk of an affected fetus. If the mother is identified as a carrier, testing is offered to her partner, with a view to offering PND and TOP to carrier couples.

Neonatal testing is intended to identify newborns affected with sickle cell disease, and not already diagnosed through PND, in order to promptly institute penicillin prophylaxis and comprehensive care which reduce morbidity and mortality. Infants with presumed sickle cell disease are retested, and parents of affected and carrier infants offered counseling.

No formal randomised trial in the UK has shown directly that screening reduces morbidity and mortality. The effectiveness of neonatal screening in preventing morbidity and mortality depends on three steps: firstly the sensitivity of the screening and diagnostic tests as prerequisite for early detection. This depends on laboratory technique but with HPLC is approaching 100% (1); secondly the follow-up of screen positive children, and their compliance with penicillin prophylaxis, which varies between localities and is often not complete (2,3); and thirdly the effectiveness of prophylactic oral penicillin in reducing the incidence of pneumococcal sepsis. There is good evidence that administration of oral penicillin to infants and young children with sickle cell disease reduces the incidence of pneumococcal septicemia (which has a fatality of about 15%) by 84% (4).

Objectives of the review

- to review alternative options for antenatal and neonatal haemoglobinopathy screening programmes in the UK.
- to develop a decision model to compare cost-effectiveness of universal and selective screening strategies and apply it to estimates of local health district ethnic composition.

Characterisation of alternative strategies

In a universal antenatal screening programme all women are offered testing. In a selective programme testing is offered to all non-North European women, and to all women with low MCH (mean corpuscular haemoglobin) result regardless of ethnic status. An

alternative option, testing based exclusively on ethnicity regardless of MCH result, was also examined.

Neonatal screening would either be universal (all newborns not already diagnosed prenatally) or selective (undiagnosed babies of non-North European mothers) with selection independent of the antenatal programme. A targeted programme, which would take account of parental carrier results to reduce the number of neonates requiring screening, was considered in subsidiary analyses. It was assumed that neonatal testing would be based on newborn heel prick samples collected on filter paper for routine PKU and congenital hypothyroidism tests. “No antenatal testing” and “no neonatal testing” policies were examined in subsidiary analyses.

Methods

Models were developed to estimate the life-time treatment costs and life expectancy of children with haemoglobinopathies, and where relevant the effects of early diagnosis. A computer model predicted the fetal prevalence of haemoglobinopathies and calculated the costs and outcomes of each screening option.

The effectiveness of antenatal screening was measured by the expected number of women with affected fetuses who were offered choice over the outcome of pregnancy. The number of affected livebirths prevented by screening was examined in subsidiary analyses. The effectiveness of neonatal screening was measured by the number of late diagnoses of sickle cell disease prevented. Costs were based on a health service perspective.

This model was applied to ethnic composition data for district health authorities in the UK. The preferred screening strategy in each district was estimated using incremental cost-effectiveness ratios (ICERs), the additional cost of a universal compared to a selective programme per additional unit of effect.

It was assumed that districts would be willing to pay between £50000 and £150000 to offer an additional choice over the outcome of an affected fetus, based on an analysis of similar screening programmes; and between £10000 and £50000 to prevent an additional late diagnosis of sickle cell disease.

Results

Findings relevant to both antenatal and neonatal screening

- Neither antenatal screening of North European women, nor neonatal screening of their children, is cost-effective, even under extreme assumptions about frequency of sickle cell trait and inter-ethnic unions.
- The rationale for universal screening is therefore based on the presumption that it will result in a higher coverage among ethnic minority women and their children.
- Lowering the failure to screen rate in a selective programme is always more cost-effective than changing to a universal policy.
- Selective screening is highly cost-effective compared

to no screening.

- If costs of ethnic ascertainment and pre-test counselling are included, the case for universal compared to selective screening is slightly strengthened, but the case for selective screening compared to no screening is substantially weakened.
- Use of economic criteria alone to determine whether local screening policy should be universal or selective is not equitable: ethnic minority mothers and infants in lower prevalence areas would receive a lower coverage screening service than would be available to them in a high prevalence area.

Antenatal screening

- Universal antenatal screening costs were estimated to be in the range £35,000 to £145,000 per 10,000 antenatal population, and increased with prevalence. Selective screening costs were £30,000 less in low prevalence areas, and £18,000 less in high prevalence areas.
- Adverse screening outcomes (PND induced miscarriage, TOP of unaffected fetuses) would be very rare in both universal and selective strategies.
- If the purpose of antenatal screening was prevention of affected livebirths rather than offer of reproductive choice, universal screening would be difficult to justify in any district in the UK on the basis of costs averted, but selective screening would still be preferred to no screening.

Neonatal screening

- Universal neonatal screening was estimated to cost approximately £22,000 per 10,000 antenatal population. Selective neonatal screening costs range from less than £200 per 10,000 antenatal population, to £11,500 in an area with 50% ethnic minorities.
- Antenatal screening, even if universal, would not render neonatal screening redundant at currently estimated rates of PND uptake (approximately 15% in Black women). High (80%) uptake of PND would weaken the case for universal screening considerably, but would not affect the case for selective neonatal screening in preference to no neonatal screening.
- The costs associated with neonatally identified carrier infants are small in relation to overall programme costs, and do not alter the comparative cost-effectiveness of universal and selective strategies.
- Targeted screening of infants is a cost-effective alternative to selective screening, but would require robust information systems that have not yet been developed.

Conclusions

- Selective screening is cost-effective in comparison to no screening.
- Universal screening may be cost-effective in higher prevalence districts, depending on coverage of

selective screening and economic willingness to pay criteria.

- On baseline assumptions, if coverage among ethnic minorities in selective screening is 5% lower than in universal screening, a universal antenatal strategy would be cost-effective at a fetal sickle cell disease prevalence above 5-12 per 10,000 (7-15 districts out of 170). A universal neonatal strategy would be cost effective at a prevalence above 7-18 per 10,000 (2-14 districts).
- If selective screening obtained coverage only 1% lower than universal, universal screening would be required in at most 2 districts.
- Equity considerations require that:
 - All districts should adopt explicit selective or universal strategies for antenatal and neonatal screening
 - The same economic and prevalence criteria for local policy determination need to be applied nationally
 - Minimum standards for coverage of screening should be adopted and coverage should be routinely monitored
 - Procedures for selection based on ethnicity are standardised
- Antenatal and neonatal screening should be part of broader preventive and clinical services for haemoglobinopathies and these should be fully integrated.

Recommendations for research

- Development of specifications for information protocols that can routinely deliver statistics on the coverage of antenatal and neonatal screening, within ethnic group; and a pilot study in which such protocols are implemented.
- Research into prevalence of fetal haemoglobinopathies throughout the UK, and the frequency and causes of (a) failure to offer reproductive choice to mothers with an affected fetus and (b) of late diagnosis of haemoglobinopathies in children.
- Research into the relationship between timing of maternal carrier and couple testing and uptake of PND and TOP.
- Medico-legal and ethical studies to determine how much pre-test information about antenatal and neonatal screening is required, in order that consent to testing can be considered to be informed.

References:

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**The full review will be published under the title: Antenatal and neonatal haemoglobinopathy screening in the UK: review and economic analysis. Corresponding author: A.Ades@ich.ucl.ac.uk*

Commentary - Professor Sally Davies (London)¹: The two common technologies (IEF & HPLC) for screening come out at very similar costs, so the choice of method is not important for cost analysis. Neonatal screening programmes should have at least a twenty-five thousand sample throughput per year to be cost-effective.

Selective screening presents a major problem. At least twenty and up to fifty per cent of children in selective screening programmes are missed. Another thirty per cent are tested who did not need to be. Litigation costs for missed cases should probably be included in cost analysis. Ten years of Brent data demonstrates (as in America) that community screening programmes do not result in prevention of many sickle births, though they do result in prevention of most beta-thalassaemia major births.

If selective screening policies are set up then, with the increasing incidence of intermarriage and partnership between different races, this will have to be adjusted over the years.

Does selective screening work? Ethnic monitoring by midwives has been very poor, so there is a massive training issue if districts opt for selective screening. Ten years ago in London, where there were large numbers of ethnic minority women in localised areas, they did work, but as soon as you get away from very high prevalence areas then it appears to fail. Intermarriage is increasing and without universal antenatal screening the chance of missing somebody with mixed ancestry is too great.

Universal neonatal screening across the country would cost less than two million pounds annually. Each extra sickle patient identified would cost forty-one thousand pounds if you consider the country as a whole. Different prevalences however mean that in North Thames an extra sickle cost under eight thousand pounds to diagnose, whereas in Cornwall it costs nearly two hundred thousand.

Commentary - Professor Bernadette Modell (UCL, London): It is difficult to consider screening programmes in isolation. Antenatal screening crosses every boundary from primary to secondary care and even between different laboratories. There is a danger that we think we have to eliminate these diseases but this is not the public health agenda. The public health agenda is offering choice, which requires information in people's preferred language.

For thalassaemia, there's a very high uptake of prenatal screening among Cypriots (80%) and for Pakistanis there was 30% utilisation ranging from 0 - 70% by region - so there's a lot of inequity in the way that this service is delivered. However, an enquiry showed that stage of pregnancy makes a great difference. Forty per cent of mothers were not counseled in time for the offer of prenatal diagnosis. The uptake of prenatal diagnosis by British Pakistanis, when offered in the second trimester, is forty per cent with sixty per cent termination of affected pregnancies. When offered in the first trimester it is seventy-five per cent with over ninety per cent termination of affected pregnancies.

But antenatal screening as the only policy is inadequate in many ways. Knowing you are a carrier might influence your choice of partner, your choice of when and how many times to conceive and so forth. Yet people are identified and counseled in the first pregnancy only at eighteen weeks, when prenatal diagnosis is much preferable at eleven weeks. For sickle cell disorders this happens not just in the first pregnancy but also in subsequent pregnancies, meaning that people are not being offered the service that they find acceptable.

What other possibilities are there? It would fit in with the national curriculum to give information in schools about inherited disorders for which people may be screened, but one would have to include disorders which affect everybody including cystic fibrosis.

We must encourage greater involvement of primary care teams in screening for these disorders by encouraging pre-pregnancy testing, very early pregnancy testing and also promoting family studies. This needs community-based sickle cell and thalassaemia counsellors and they are capable of supporting the programme.

Editorial comment: The haemoglobinopathy review poses a difficult problem of widely different incidence in various parts of the country. Equity requires that a woman should not be penalised by being deprived of care, simply because she lives in a part of the country where there are few women with risk factors. Selective screening only of women from risk groups has had a dismal record so far, perhaps because "selective" has come to mean "optional". But will health authorities invest in universal screening programmes for conditions which they know to be very rare in their locality? One solution may be to re-examine the meaning of "selective". It is the process of selecting which is important. If instead we present this as two stage screening, in which the first stage is to ask questions that identify risk groups for testing, we have a process which can be standardised and audited, and failure to carry it out properly becomes a matter of professional practice. To make this work, better information for women is vital - see Professor Marteau's paper.

¹ Professor Davies heads a group undertaking a systematic review on haemoglobinopathies which had not been completed in December 1998.

COST-EFFECTIVENESS OF ANTENATAL HIV SCREENING IN THE UK

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Administration of zidovudine to the mother in pregnancy and at delivery, and to the baby, together with elective Caesarean section and avoidance of breastfeeding, can reduce the transmission of HIV from mother to child from 30-35% to as low as 2-5%.

UK guidelines recommend universal antenatal HIV testing in London & selective testing to women at higher risk elsewhere. However, in 1997 only half the maternity units in London had universal screening policies; 52% units elsewhere tested only women who requested it. We undertook a cost-effectiveness analysis of antenatal HIV screening in different parts of the UK. Decision analytic models of HIV disease progression and treatment costs in screened & unscreened women and their infants, a risk reduction model including bottle-feeding, zidovudine & mode of delivery, & a model characterising the size & prevalence of previously undiagnosed HIV in potential target groups in the antenatal population were constructed & UK data applied. Costs were discounted at 6% & life-years at 2% per year.

The prevalence of HIV among high risk women merits selective rather than on-request screening throughout the UK. In London the average prevalence of undiagnosed infection among Low Risk women merits universal testing under all scenarios. Results of the incremental analysis of selective vs universal screening were most sensitive to screening costs, maternal treatment costs incurred by an earlier HIV diagnosis and the difference in uptake of testing that could be achieved between universal and selective strategies.

However, results showed that with low test costs (£1.20 per HIV antibody test) and only 2-3 minutes of offer time, universal screening would be cost-effective everywhere, given a willingness to pay of £10,000 per life-year gained and high uptake of interventions to reduce mother-to-child transmission. Pooling of HIV tests could significantly lower test costs without compromising sensitivity. Universal antenatal HIV screening in the UK could be cost-effective, and avoids the equity issues associated with selective screening.

Commentary - Dr Graham Davies (Gt. Ormond St Hospital, London) and Dr Angus Nicoll (CDSC, Colindale, NW9): Experience in London showed that the majority of vertically infected babies were diagnosed only when they presented with features of AIDS, and several previous opportunities to identify these at-risk mother baby pairs had been missed. An inter-collegiate working party was convened to examine the reasons why the uptake of testing and the diagnosis was HIV and pregnancy were so poor in this country. They recommended that:

- Information is needed for all women antenatally;
- Education and training of staff, particularly midwifery and obstetrics staff, is important;
- Voluntary confidential HIV testing should be available without obstacle throughout the country;
- The tests should be strongly recommended to women who are at higher risk as a routine part of antenatal care;
- Women diagnosed as positive should receive urgent specialist care and support;
- Systems must be in place to monitor the uptake of testing and the diagnostic rate.

Since the report was published, the DoH in conjunction with the Royal College of Midwives has been working on culturally appropriate information for women and training packs for the midwifery services.

The NSC criteria for screening are satisfied. We know what needs to be done, particularly in London. But who, at a local level, should be responsible for making sure that this happens?

Points from discussion:

- The haemoglobinopathies and the HIV programmes have several things in common: the universal versus selective issue; ethnic monitoring is needed; the mother is under the care of an obstetrician – who will not be the one who subsequently cares for the mother or child.
- In other countries in Europe and in the USA, the incidence of HIV infection in children is declining markedly due to interventions aimed at prevention vertical transmission, and a high uptake of antenatal screening. Universal screening in the Rotunda Hospital in Dublin, and in Edinburgh, has been very successful. In the States public health policy has addressed this issue regularly and in other parts of Europe the obstetricians are more involved.
- The insurance companies say that simply having a routine test will not carry a penalty.
- Is it really true that the cost of pre-test counselling are virtually nothing? Some people have said that you need an hour's counselling before having a test. But AIDS exceptionalism put up a lot of barriers - if you're a woman living in the east end of London in temporary accommodation with four children and you're told "would you like an HIV test? – but you can't have one unless you come back for another hour's counselling on another day". It was not practical. HIV tests should be included with other tests.
- There is confusion with anonymous testing – some women (especially those from parts of Africa) think they are automatically tested for HIV in pregnancy and are surprised to learn they are not.
- Screening on a national or regional basis offers major economies of scale - in terms of laboratory work and staff training and education and the national education of the at risk populations. Compare the immunisation

programme - a national responsibility with local co-ordinators who are organised nationally and the education programmes and publicity are conducted nationally.

- HIV is increasing rapidly in many countries – for example, in India. The UK is peculiarly sensitive because 60% of all HIV infection world-wide is now in Commonwealth countries.

Editorial comment: There is clear evidence that identification of HIV in pregnancy has real benefits for the child, with a reduction in the risk of transmission to as low as 2% if all measures now shown to be effective are applied. Birth of an HIV infected child must be regarded as a largely avoidable disaster. The problem is making sure that the screening is done, particularly with a selective policy. As with haemoglobinopathy, the dilemma is that some districts have extremely low rates of HIV. Although the Review illustrates that screening can be cost-effective even at very low prevalences, screening all pregnant women for HIV (and for haemoglobinopathy) will not reach the top of the priority list in such districts unless there is clear central guidance. See also the editorial comment on haemoglobinopathy screening and the appendix on Hepatitis B.

SCREENING FOR CYSTIC FIBROSIS – SUMMARY OF THE SYSTEMATIC REVIEW FINDINGS

Murray J, Cuckle H, Taylor G, Littlewood J, Hewison J. (Leeds)

Cystic fibrosis (CF) is a common serious recessive disorder associated with considerable morbidity and high case-fatality. We have carried out a systematic literature review to decide whether screening should become routine and, if so, which strategy to adopt.

Background

In 1989 the transmembrane conductance regulator (CFTR) gene situated at 7q31, was shown to be responsible for the condition. To date over 800 mutations in the CFTR gene have been identified, although not all have been found to be disease causing. The most common UK mutation is $\Delta F508$, which accounts for 75% of carriers, and three commercial multiple-mutation assays are available that can detect about 86% of carriers in Scotland, Wales and the North, or 80% elsewhere. Different proportions apply to Asians (35%), Ashkenazi Jews (95%) and Blacks (41%). The UK birth prevalence is 1 in 2,400, which implies a carrier frequency of 1 in 24. A carrier couple have a 1 in 4 risk that each of their children has CF and this will be reduced to under 1 in 50,000 if neither has a detectable mutation. When only one is demonstrated to be a carrier the risk is about 1 in 500.

Genetic screening

This aims to reduce birth prevalence by identifying carrier couples who can have prenatal diagnosis and selective termination of pregnancy. Other options are to: avoid pregnancy; change partners; have artificial insemination using donor sperm or egg; and pre-implantation genetic diagnosis (PGD) to select unaffected zygotes. Carrier couples can be identified directly during or before pregnancy, or indirectly by determining the carrier status of everyone of reproductive age in the population. A third approach is systematic 'cascade' testing within CF families.

There have been 11 published studies reporting the results of antenatal screening pilot projects. The combined results on over 40,000 tests demonstrate the feasibility of the method and show the acceptability of screening (uptake 74%), and invasive prenatal diagnosis in carrier couples (uptake 89%).

Pre-conceptual screening has been tried in a family planning clinic setting with high uptake. Pre-nuptial testing is already available for orthodox Ashkenazi Jews. PGD is currently being carried out in 6 licensed UK centres although world-wide under 100 procedures have been done for CF.

Four general population screening studies have been carried out in general practice with a total of almost 11,000 patients. Uptake was only 8% when invited by letter and 48% when approached opportunistically in the clinic. Uptake was also low when screening was offered to school students (41%, 42% and 70% in three studies), in the workplace (21%) and as the result of a general community-wide campaign (8%).

There have been three studies of active cascade screening. Uptake was high and a large proportion of those tested were carriers. However, mathematical models have shown that under 15% of carriers in the population would be detectable this way.

Neonatal screening

This aims to bring forward the diagnosis of CF and so improve prognosis. The detailed experience of neonatal CF screening has been reported for 20 programmes including six in the UK. Protocols vary: single or repeat testing; fetal blood spots or meconium; IRT or DNA. In total more than 5 million neonates were screened with a low false-positive rate (0.5 per 1,000), acceptable detection rate (90%), and favourable positive predictive value 33%.

The ability of screening to alter long-term prognosis has not been conclusively proven. Two randomised trials of screening, five case-control studies, a study of sib-pairs and a trial of prophylactic compared with symptomatic treatment of early disease all provide relevant information. However, this is either predominantly short term or subject to strong statistical bias. Nevertheless there is some circumstantial evidence favouring a benefit.

Costs

We estimate the cost of antenatal screening to be about £46-53,000 per CF pregnancy detected, considerably less than the lifetime cost of treatment. Neonatal screening

costs about £4,400 per case detected or £6,400 for those that would not otherwise have had an early diagnosis; about £1,500 and £2,200 respectively when combined with antenatal screening.

Conclusions

We conclude that there is sufficient information to recommend: (1) Antenatal screening to be offered routinely; (2) Pre-conceptual screening for couples who request it; (3) Genetic screening for infertile men and for sperm donors; (4) Testing to be done in laboratories with an annual throughput of at least 5,000 CF tests; (5) Health authorities to consider introducing neonatal screening.

The full review will be published within the next few months and can be obtained by ordering from the following address: <http://www.soton.ac.uk/~hta/>

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Commentary - J.M.Littlewood, Hon. Consultant Paediatrician, St James Hospital, Leeds, England & UK CF Trust Since 1956, the median survival for people with CF has increased from around 2 years to over 30 years and a median survival of over 40 years is projected by the year 2000. It is already exceeded by those patients who manage to avoid chronic pulmonary infection with *Pseudomonas aeruginosa* (PA) or *Burkholderia cepacia* (BC). Early intervention and avoidance of lung damage are crucial in achieving better long term prognosis. There is a distinction between "CF disease", where the airways contain abnormal secretions but are uninfected and "lung disease" which follows infection and inflammation within these abnormal airways. After the onset of 'lung disease' there is a gradual deterioration of respiratory function, the speed of which is determined by the intensity of treatment and other factors. This transition between 'CF disease' and 'lung disease' is termed the "point of no return"(PNR). Gene replacement therapy will probably only be effective if given before the PNR.

- The prognosis of CF is related to the condition of the patients at the end of the first course of IV antibiotics.
- Respiratory function tests decline *after* chronic bacterial infection becomes established. They will usually remain stable for many years before chronic infection becomes established
- Survival depends on the infecting organism e.g. median survival if neither *Pseudomonas aeruginosa* or *Burkholderia cepacia* are present was 42 yrs; for those with chronic *Pseudomonas aeruginosa* infection it was 32 yrs and for those with *Burkholderia cepacia* only 14 years.
- 20% of US infants already have PA infection in infancy (6). Thirty percent of untreated Australian screened CF infants already have *Staphylococcus aureus* (SA) or PA infection of their lower airways by 6 months of age (13); inflammation follows infection rather than the reverse
- Chronic infection with SA can be prevented.
- Early PA infection can be eradicated

Conclusion All newborn infants should be screened for CF. The staff of a CF centre should be involved in evaluating and discussing the results of positive tests with the parents, in confirming the early diagnosis, in starting appropriate treatment and also be involved in the initial teaching of the family about cystic fibrosis.

Thirty years ago neonatal screening for CF was unlikely to show much benefit as the available treatment was of little value, potential new treatments were unlikely and screening was, quite correctly, not recommended. To day the situation is entirely different because early diagnosis and treatment are so vitally important to preserve the integrity of the airways and maintain the patient in the 'CF disease' phase pending the availability of new and better treatments resulting from the identification of the CF gene.

In the UK a detailed report on genetic screening for CF, both antenatal and neonatal, will soon be submitted to our National Screening Advisory Committee (33). While conceding the lack of absolute proof of a favourable effect on long term prognosis, if one considers all the available comparative studies all tend to show some advantage for the screened infants.

I would agree with Dr. Wilkens's conclusion that "the scales appear to be tipping in favour of neonatal screening" and would suggest that, with both the immediate and potential treatment possibilities they have now tipped even further to the extent that NCFS should be routine for all infants.

From the clinician's point of view, the following complete the now unassailable case for neonatal CF screening. The clinical facts discussed here, the present day improved treatment which must be introduced early before chronic damage, the imminent prospect of more specific even curative treatments (unlikely to help patients with advanced disease) and last but not least simple common sense and what we would wish for our own children and their children.

N.B.: This is a much shortened version of a paper prepared by Dr Littlewood for this meeting. The full paper sets out his experience and opinions on CF screening, supported by an extensive bibliography. It can be obtained from Dr Littlewood by e-mail: jlittlewood@dial.pipex.com

Commentary: Dr Hilary Harris (Manchester): A cystic fibrosis carrier screening research project ran in 8 General Practices in the North West from 1992 - 1995, co-ordinator Dr Hilary Harris. Patients were offered a cystic fibrosis carrier screening test by mouth wash sample at the booking appointment. Patients book early in pregnancy with their General Practitioner (average gestational age 6-8 weeks). There is therefore the opportunity to detect a carrier couple in the first trimester of pregnancy and to offer pre-natal diagnosis after due counselling. My own practice in Brooklands, South Manchester was the pilot practice for this project and has continued to offer cystic fibrosis carrier screening following the conclusion of the research programme. This is a small practice of 4,500 patients. So far we have carried out 258 cystic fibrosis screening tests

and have not yet detected a carrier couple. The uptake is about 95%. Twelve carriers have been detected, no carrier couples. Uptake figures are high and clinical genetic colleagues have raised the possibility of coercion. However, it is believed that this uptake which was reflected in other practices is a feature of the offer of the test being in a familiar place and by a doctor who has responsibility for ongoing care.

A positive cystic fibrosis carrier screening result will have implications for the family and for pregnancies in the future. Each result is recorded on the computerised patient record. That information will be available to their GP in the future and to the patient. It is important that the Primary Health Care team understand that beginning a screening process in Primary Care is analogous to stepping onto a pathway which may have the offer of pre-natal diagnosis and termination of pregnancy as an end point. Those patients for whom this process is ethically or morally unacceptable may choose not to step onto the screening pathway.

A Confidential Enquiry into Genetic Counselling by Non-Geneticists, co-ordinated by Rodney Harris, has looked at circumstances surrounding the birth of 46 children born with cystic fibrosis as the second child affected within a sibship. The birth of the second affected child was following the diagnosis in the first child and before 20 weeks of pregnancy in the second pregnancy. In many of the cases scrutinised there was no documented evidence of information and counselling given to parents who were already at high risk of producing an affected infant. Letters from paediatricians to general practitioners following the diagnosis of the first affected child with cystic fibrosis recorded treatment in 100%, mentioned the mode of inheritance in 40%, but only 10% mentioned that pre-natal diagnosis could be offered in a subsequent pregnancy.

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Commentary on general issues about biochemical and genetic screening - Helen Carter (RTMDC): Even where people have religious, moral or ethical objections to termination of pregnancy, they should be allowed the choice of stepping on the pathway of testing if testing is available. They need to be made aware of the potential risk to the developing fetus; but some families will nevertheless wish to undertake testing because the knowledge gained may help prepare parents, siblings, other families and friends; make better lifestyle decisions; and contribute to the health and wellbeing of the whole family.

The focus should be on informed choice. One needs to think carefully about the timing of any request for

consent. In the first days after birth, as one mother said, people are always "coming in and out, picking baby up, weighing her, sticking needles in her foot, and so on". For this mother the Guthrie was just part of that process, something of which she was only dimly aware. A case could be made for screening options to be discussed as part of the birth plan.

There are interesting issues about which diseases we should screen for, either where they aren't treatable or where even though there is treatment available the longer term prognosis is still not good. The established wisdom is that where treatment isn't available, one should not test and that's the position we've taken as an organisation. It's a position we are revisiting. Arguments against screening and points to consider include:

- The extent to which a test is predictive rather than foolproof
- The wish of some parents to enjoy the "good years" before the onset of a degenerative condition, without the diagnosis hanging over them as a sword of Damocles.
- The difficulty of choosing the "right" time to inform parents in the case of untreatable or late onset conditions.

Less often articulated but just as worthy of debate are arguments in favour of screening which include:

- The ability of parents to make decisions about the future size and shape of their family
- The potential for better lifestyle decisions - people say "If only we had known we would not have moved to that house. We would have moved to a bungalow".
- It allows a better quality of life, however brief, for the family and affected child because they are spared the inevitable hospital visits and tests in search of a diagnosis.

An update on biochemical screening of the new-born: Rodney J. Pollitt (Sheffield)

Two systematic reviews of neonatal metabolic screening were commissioned as part of the NHS HTA programme. Though differing in detail in their methodology and results, both reports concluded that screening by tandem mass spectrometry was likely to be practicable and effective but that a carefully monitored trial/pilot study was required before general introduction. These reports were discussed at last year's *Evolution or Revolution* meeting and generated considerable interest. The need for decisive action to prevent technology drift and piecemeal implementation was recognized, but a series of discussion meetings under different auspices during 1998 failed to agree a strategy or to identify funding. In the meanwhile a number of centres were taking pre-emptive action by acquiring the necessary instrumentation.

The systematic reviews both used *causal pathway* approaches and both pointed to the poor quality (in HTA terms) of the literature evidence. There is a considerable

gap between the level of evidence currently available and what the National Screening Committee wishes to see: evidence from high quality randomised controlled trials that a proposed screening programme is effective in reducing mortality or morbidity. Unfortunately RCT of neonatal metabolic screening by tandem mass spectrometry would be extremely expensive because of the rarity of the conditions involved. It would also raise major ethical problems because of lack of equipoise between the two arms. An intermediate course may be possible, using a Bayesian approach and collecting sufficient epidemiological and screening data to permit modelling, but stopping short of a full-scale trial. The matter may soon be brought to resolution in that the NHS R&D HTA Programme has included primary research to determine the cost-effectiveness of neonatal screening by tandem mass spectrometry in its latest call for proposals.

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Professor Chalmers (St George's, London; co-author of systematic review on biochemical screening)– asked what progress has been made in putting TMS screening in place? There is a risk of some labs doing their own thing – we should learn from the mistakes of the past, for instance in PKU screening. “Good enough” means just that. Strive for excellence and continuing improvement, but if we set unrealistic targets we'll never get off the starting block – and families will lose out while the academic debate continues.

Editorial comment – biochemical screening: The issue of TMS screening has become focused on MCADD screening as the first and most obvious target disorder. There are too many unanswered questions to recommend an immediate country-wide implementation of TMS screening. A bid for further research, constructed by a single consortium of biochemists, clinicians and epidemiologists and including the RTMDC, has been submitted to the HTA programme (March 1999). Medium chain acyl-CoA- dehydrogenase deficiency (MCADD) is a relatively common condition and the management consists essentially of special measures to avoid fasting whenever the child is unwell. There are however still doubts about the value of screening. Some cases remain asymptomatic, some die in the neonatal period, and some may decompensate so rapidly that intervention is impossible. The case for screening for glutaric aciduria type I and for other conditions is even more controversial. It is possible that much parental worry could be caused for little reward. It is hoped that the proposed research will address these issues.

A feasibility study has just been completed on the question of setting up an extended register or database for inherited metabolic disorders. This would be invaluable for further research on the quality and benefits of screening, the value of early intervention and the long term

outcomes. This study was conducted under the auspices of the Royal College of Paediatrics and Child Health and was funded jointly by the Research Trust for Metabolic Diseases in Children (RTMDC) and the National Reye's Syndrome Foundation of the UK. The report (by Rosemary Thornes) not only examines the role and value of such a register and the obstacles likely to be encountered, but also provides a detailed account of the new Data Protection Act 1998 which has far-reaching implications for all those whose research involves registers or patient databases. The report will be available soon from the Royal College of Paediatrics and Child Health.
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Editorial note – CF screening: The review is being considered by the Antenatal and Children's sub-groups of the National Screening Committee and a co-ordinated response will be presented to the full National Committee as quickly as possible.

GROWTH MONITORING IN THE UNDER TWO AGE GROUP

David Hall (Sheffield)

A group convened under the auspices of the Child Growth Foundation met in Coventry in the summer of 1998 to tackle the question of growth monitoring. It was not a complete systematic review – there are hardly any trials of growth monitoring. This paper reflects the results but it is not yet a true consensus.

Growth monitoring is a means to an end. The two main aims are to identify children with treatable conditions affecting growth so that they can be treated earlier; and to produce data of public health interest regarding the health of children and the differential between social classes. In the under twos, the issues are:

- measuring length in the newborn;
- monitoring length in the first two years;
- monitoring weight;
- monitoring head circumference.

Length in the newborn is easy to measure but hard to measure accurately. Parents like it - and for the midwife that may be reason enough. However, if we want accurate measurements that might be useful later on, midwives will need better training, and quality assurance checks - and this is only worthwhile if the measurement is useful.

Length is an essential part of the assessment of an infant who is of low birth weight or is dysmorphic in any way. In these situations, the examining doctor must ensure that an accurate measurement is taken as part of the neonatal examination (a procedure which itself needs to be re-examined). If there is a need for further monitoring of the baby's growth and progress - including length - this should be specified by the paediatrician.

We found no evidence, and little expert opinion, to support routine measuring of the length of all new-borns. The argument that it offers a baseline for the future is not convincing. Size at birth is mainly determined by maternal size, whereas size at age two is determined by the genetic potential inherited from both parents – so the baby's length will cross centiles to reach his genetic trajectory, limiting the value of the birth length when interpreting length measures.

Similarly we found no evidence and little expert support for routine *monitoring* of length. We felt that length should be monitored if there is concern about the baby's health or progress, but we have not defined what that concern consists of – however, if there is that level of concern, perhaps the baby should be under the supervision of a paediatrician or perhaps a dietician.

Weight at birth is an easy and accurate measurement. It is useful because:

- Birth weight reflects socio economic differences
- Low birth weight is a marker for many disorders and problems
- It is of prognostic significance.

- Birth weight is not a screening test but it should nevertheless form part of a minimum dataset for child public health.

The most difficult area is the question of weight monitoring in the first two years. Mothers like to weigh their babies, especially the first, as it is reassuring to see weight gain. But, it can also cause intense anxiety, due to misinterpretation of the chart – leading to unnecessary investigation and even to child protection proceedings. So, what is the optimum approach to weight monitoring?

Twenty years ago textbooks regarded failure to thrive as a diagnosis, usually associated with a list of organic causes, and needing investigation. But most babies whose failure to thrive is due to organic disease, will have other factors that cause concern, as well as their poor weight gain - and it will be these other factors that prompt referral. The investigation for organic disease of a baby whose only problem is his weight chart is unrewarding.

It would be unusual for a mother not to bring her baby for weighing and health visitors might be concerned to check that all was well; but we cannot say that regular weighing is a procedure of such importance and proven value that defaulters must be pursued. Child abuse is sometimes associated with poor growth, but poor weight gain is not usually due to child abuse. There is a relationship between poor weight gain and abuse – but it should not be over-emphasised.

The suggested best practice is that every baby should be weighed when the opportunity arises – at the times of immunisation and with the 8 month review. Together with the birth weight this offers six weight measures as a minimum.

Of course, the problem of weighing is that something must be done with the results, otherwise it is just a ritual. Babies who gain weight are not usually a worry, though some professionals give unsolicited and often wrong advice about obesity. Babies who have sustained weight loss need investigation. But what about babies who gain weight but more slowly than expected and cross centiles downwards? This is a complicated issue. Weight charts only show the distribution of babies weights at each age, they do not mean that babies must grow along the lines on the chart. It is normal for babies to cross centiles.

It is also a mathematical fact that on average big babies are more likely to cross centiles downwards and small babies to cross centiles upwards – but they can do the opposite and still be normal. Crossing centiles downwards is a phenomenon like being short – it may be normal or it may be a clue to some organic or social disorder. Charts can never tell us whether the baby is normal – but they could tell us how unusual the baby's growth pattern is – and the more unusual the more likely it is that there is something wrong. Tim Cole's chart and the Thrive Index (devised by Wright and colleagues in Newcastle) aim to address these difficulties. They have tried to define a level of "failure to thrive" which is sufficiently unusual to merit further enquiry, by identifying babies with the most extreme centile crossing pattern. Details are in the training manual which provides lots of clinical examples.

There have been relatively few intervention studies for FTT and most have been on clinic populations rather than community based samples. Most of those that have been done are under-powered - they may have failed to detect any benefits that did occur. The Newcastle study did show some benefit - though it also noted that many babies spontaneously "recover" or at least they return to a normal rate of growth.

There are still too many unknowns for us to suggest that this kind of monitoring and charting should be part of a national programme:

- We don't know the long term prognosis of these infants and whether it is materially altered by intervention; many are still short later in childhood and some also say the IQ is lower
- Possibly the most at risk babies are those who do not thrive in the first few months of life;
- Where does iron deficiency fit in?
- Several approaches to intervention might be equally effective;
- Is the real issue feeding and relationship problems? A study (presented at the RCPCH annual meeting in 1997) carried out in Leeds by Mary Rudolf and Pauline Rayner suggested that this might be so.
- These problems might be preventable by early parent support - like the Sure Start programme.

Perhaps our pre-occupation with the weight chart means that we only find those who are under weight - but are missing other babies whose feeding is causing just as much distress to the parents.

In summary, the consensus is:

- Weight at birth
- Length at birth for specified reasons only
- Monitoring of length for specified reasons only
- Weight at birth, 2,3,4,8 and 12-15 months.
- Training in weight chart interpretation

We suggest that HVs, SHOs and GP trainees should receive training in how to understand weight charts and weight gain and the assessment of dietary intake and feeding problems. HVs know much of this but the thrive index concept is not easy.

- For babies with no other complaints, community based intervention in first instance.
- Focus on intake, social interaction and feeding techniques
- Other features or actual loss of weight - investigate

Commentary - Dr Stuart Logan (ICH, London): We do have a problem. There are a number of babies who grow less well than we think they ought to. Very few have major underlying organic conditions or are abused although clearly these are important. Most are probably a mixed bag of normal children crossing centiles because they are moving to where their genes suggest they should be, some who aren't growing well due to intercurrent illnesses, some

with problems in parent/child relationships, some with oro-motor inco-ordination or perhaps, as in most cases, a mixture of lots of different elements. This may lead to longer term problems of short stature or possible IQ but the evidence of a causal relationship is unclear. The case definition that most of us use clinically is that, if a child is not growing terribly well and for some reason they "smell" a bit wrong, we're a bit worried about them and consider further investigation. Not the clearest definition in the world!

We can use statistical definitions of poor growth, yet the statistical definitions are neither terribly sensitive nor terribly specific and none of them are clearly linked to prognosis. We also have problems with interventions: there have been a number of case series describing intervention as an apparent success. However, in one randomised control trial, at least 55% of the babies who didn't receive intervention within a short space of time had moved out of the definition of failure to thrive. Case series are dangerous things on which to base practice.

The trials carried out in Newcastle and Leeds suggest some improvement of weight gain can be achieved through interventions - though the improvements are of fairly modest size. Unfortunately, we have no idea whether improvement of weight gain is of any relevance at all to the long term outcome for the children.

So, in summary, we have no proper case definition linked to either prognosis or potential to benefit from intervention, very little idea of the natural history of failure to thrive and little evidence that intervention is beneficial in terms of long term outcome for the children. If you took that along to the National Screening Committee and asked them to set up a screening programme it wouldn't take very long to sort it out! But the idea that we're going to stop people weighing babies is a fantasy. We can't stop people weighing infants and what is needed is to ensure that we do the least possible harm.

The other day a 13 month old baby was referred to me with "failure to thrive" who was growing quite nicely along the 3rd centile. The mother was in a complete state - incidentally she was a social worker. She had consulted a number of professionals and the main message that she had got from them - the reason that her baby had this "problem" - was that she was breastfeeding too long and that she must get on and stop breastfeeding.

We should have clear aims for the programmes that we are running and everyone should know what they are. We clearly need to weigh babies less often - this 13 months old child that I saw had been measured every two weeks! When we do weigh them, we need to do it properly - and we need to have a very clear idea about what we do with the weights we get. We must have local - and I emphasise the word "local" - protocols about who staff are going to refer, where those referrals go and, when they are referred, and the people they are referred to must have sufficient skills to make a sensible assessment and decide how to manage the family in the light of the enormous uncertainty that surrounds the whole area. Finally, we have to get the research right. We need an operational case definition and more good randomised trials, of those children who are likely to benefit from intervention.

Commentary – Dr Charlotte Wright (Newcastle).

Weight monitoring as a screening programme is already in place with large numbers of weights being collected on the great majority of children already. What is important is to make best use of the weights that are collected.

Failure to thrive precipitates a large number of referrals into general paediatric outpatients, while a much larger group of parents experience anxiety related to their child's weight gain, much of it probably unnecessary. We also know from our recent randomised controlled trial, that between 1/3 and 1/2 of children with unequivocal slow weight gain, appear not to have been properly identified in the community and certainly have not been referred for more detailed assessment.

Failure to thrive could be said to be a much commoner condition, but also with less sinister implications than we previously thought. The majority are living within unexceptional families but are nonetheless relatively undernourished as reflected in under weight for height and their strong tendency for catch-up weight gain, which seems to be faster and complete when intervention is offered. The long term consequences of FTT are considerable in terms of restriction of gain of height and weight but the impact on intelligence seems to be much less than previously thought.

All this leads to three basic conclusions:

1. That children should probably be weighed less often and more attention should be paid to the weights that are collected. The recommendation at present would be that children should be weighed at birth, 8 weeks, 3 & 4 months, with their immunisations, at 7-9 months and at around 1 year with their MMR. Most children with FTT drop away early in the first year, so that regular but not too frequent weighing would identify most cases. Obviously, children about whom there is clinical concern would need to be weighed for longer.
2. Explicit screening criteria should be applied to weight gain patterns, both to be able to reassure the worried well and equitably identify children with slow weight gain. Individual districts might differ in the screening criteria used, but the recommended criteria would be either a fall through 2 centile spaces, identifying the slowest gaining 5% or through 3 centile spaces, identifying the slowest gaining 1% of children.
3. Having identified children with relatively unusual weight gain patterns, the first stage of assessment and intervention should be based in primary care. Health visitors can undertake this work, if they have access to support from dieticians, and community paediatricians. Referral into secondary care should only take place after initial assessment and attempts at intervention have been made.

MONITORING HEIGHT

David Hall (Sheffield).

The debate about growth monitoring in children over two is mainly about height; there is currently little enthusiasm for monitoring weight to identify obesity as neither primary prevention nor treatment have had much effect. However, the "epidemic" of obesity and the increasing evidence for a genetic contribution to obesity mean that this issue will stay on the agenda.

Many conditions could be identified by height monitoring. Hypothyroidism should be excluded whenever a short child is being investigated; but the average height of a new case is only 1 SD below the mean. Only a minority would be found by height screening. The same goes for Crohn's disease. There is a slightly stronger argument for coeliac disease, but even here screening would only find a minority. Furthermore, many of the children with these conditions, even when identified by screening, have had complaints of relevant symptoms whose significance has been missed. Psychosocial causes of short stature are also important but we have not found any evidence to suggest that many cases are found purely by height monitoring.

Screening aims to identify truly pre-symptomatic conditions. We should focus on conditions which present with short stature and little else. Given the rarity of new cases of skeletal dysplasias at this age, the main target conditions are Growth Hormone Deficiency (GHD) and Turner's Syndrome (TS). A height measurement at school entry would find at least half the girls with TS - this would have definite health benefits. Similarly you would find many though not all of the cases with GHD - so a height measurement is useful for finding these two conditions.

There are two essentials for GM – growth charts and accurate measuring. The best charts for modern use are the nine centile charts which are based on a well defined data set and show the 99.6 and 0.4 centiles. Accurate measurement is vital. About 90% of the imprecision in measuring is intrinsic to the child – because humans are squashy rather than rigid structures and they do not have an exact height. In other words, even the best auxologist with the best machine will not be much more accurate. But avoidable errors due to carelessness must be eliminated, otherwise the whole exercise is completely worthless.

In thinking about height, we must distinguish between single measurements, and height velocity which needs at least two. Careful measuring achieves an accuracy in five year olds of about plus or minus 0.5 cm. In 2 year olds the precision is less, plus or minus 1 cm. measurement. If you identify only the children under the 0.4 centile, you will miss some cases but have fewer false positives. By shifting the cut-off to the 2nd centile the number of referrals would increase five fold - but you would find more cases.

Children with acquired pathology leading to a progressive impairment of growth will not be found if they started off normal or tall. They can only be found by monitoring growth over time, in other words growth

velocity. Since acquired pathology can affect children of any height, there is no logic in only re-measuring children who were short on a single screening measurement. So, should we monitor growth velocity to decide whether the rate of growth is within normal limits? The snag is that if we take two measurements within one year of each other, the imprecision of the two is compounded. The net result is that a growth velocity calculated at the 25th centile might be anywhere between the 9th, which is worrying, and the 50th which is fine.

What are the normal limits? We don't know - though it seems unusual to cross more than one centile channel. But TS rate of growth is well within the range of one channel crossing as can be seen if the Turner's chart is superimposed on the normal chart. Between five and nine a girl only drops half a centile channel.

One single height measurement at age five offers the best opportunity to identify those cases of growth impairment which have not presented clinically. It *may* also be worth taking height measures at two and 3 ½ and treating these as single height measurements, looking only for those below the 0.4 cut-off. There would be little benefit in trying to estimate growth velocity by taking measurements at two or three for comparison at five, because the imprecision of height velocity estimate at this age would limit their value.

The height of a child is correlated with that of his parents and siblings. If we could adjust the child's height measurement for his genetic background, we would identify more children who were short for parental and sibling height. And we could stop worrying about some very short children, on the grounds that this was just due to having short parents. But there are several snags:

- First, estimates are unreliable – they must be measured heights.
- Often only one parent is available - though even one parent height, or a sibling height, could be useful.
- Parents and siblings may be short for pathological reasons.
- Analysis of the Wessex data set suggests that you cannot safely dismiss short children whose parents are short. Several of the short children in that study who turned out to have pathology had short parents and would not have been investigated if the height had been corrected for parental height.

My own view at present is that the interpretation of family height data is too complex to be part of screening, and should be left to whoever is sorting out the referred children.

If there was a simple, cheap and safe package of tests for the short child, we need not worry about a large number of false positives. Unfortunately the only simple cheap tests are thyroid function and blood urea or creatinine. A karyotype for TS is simple for the child but not cheap. There are no instant tests for bowel disease or GHD. Indeed the diagnosis of GHD is a very complicated problem.

We concluded that at present we cannot recommend the use of serial height measures as a means of screening

for growth disorders. However, this does not mean that we should discard measuring altogether. We suggest the following:

- It is good practice to review a child's health and progress at age two - and a height measurement at this age could at least identify the occasional very short child.
- Every child should be measured at age five and the weight should also be recorded. These measurements should be plotted on the nine centile chart.
- The height and weight at age five should form part of the minimum public health data set for children.
- The nine centile charts should be adopted and the 0.4th centile cut-off should be used. Such children need a careful assessment as shown here – the GP can do this if they feel confident but otherwise the child must be referred.
- If not done at age 2, height should be measured when convenient - such as a visit to the surgery for minor illness or the pre-school booster.
- Growth velocity is not suitable for screening
- We do not know what to do about fat children
- Any child presenting with symptoms of ill health or poor growth must be measured as part of the clinical assessment.
- If the parent is worried about growth the GP must go through the same process as for the screen failures - or refer.
- Reassurance is usually correct in probability terms - but it is impossible to exclude growth disorders just by looking at the child.

N.B.: the above material is being submitted in modified form to "Archives of Disease in Childhood", in May 1999. *Anyone who wants a copy of the interim working papers (with references) on the Consensus is welcome to request this by e-mail: g.spriggs@sheffield.ac.uk but please note that this working paper is a consultation document going through successive drafts rather than a final or definitive statement.*

Commentary - Dr Tim Cole (London): The availability of charts with a 0.4th centile has made height screening at school entry a realistic prospect. The 3rd centile on the Tanner-Whitehouse charts was designed to screen in 3% of children, this proportion falling to about 1.5% by 1990 with the secular trend to increasing height. So the 0.4th centile picks up between one quarter and one eighth of the children that the 3rd centile would have identified. This improved specificity is clearly valuable at a time when resources are limited, but it does reduce the sensitivity as well. Our ignorance about the false negative rates of the 3rd versus the 0.4th centile at school entry is a concern.

At a time when the role of the school nurse is being questioned, height measurement at school entry is a clearly defined task which is well-suited to an assistant school nurse. It involves the measuring and charting of each child, and referring them if they fall below the 0.4th centile. If adjustment for familial height is to be included as well (see below) this would complicate the process

slightly, with the need to measure the height of another family member and plot the index child's height centile against it. This might require a higher grade nurse, with corresponding financial implications.

Height velocity in early primary school is not useful when measured in the community, for two reasons: centile crossing is rare in the age range 5-8, because height velocity itself is low at this age; and the poor quality of height measurement in the community (poor technique, poorly calibrated equipment and multiple observers) tends to swamp whatever information on centile crossing there is. This realisation, though disappointing in one sense, is also liberating for it means that short children free of obvious pathology should not be followed up for long periods - there is no benefit.

A key question to be answered is whether or not to adjust for familial height. Adjusting for family size means that short children from tall families are more likely to be referred than short children from short families. Is a short child less likely to have pathology if their family is short? Is the aim to identify short stature, or non-familial short stature? As it is, virtually all children below the 0.4th centile have parents who are shorter than average. So, if no adjustment is made, children with a growth disorder whose parents are even slightly taller than average are likely to be missed by the screening process.

Conventionally the familial adjustment is based on mid-parent height, using either a special chart or a formula. But there is no reason why it should not be based on the height of the mother or father alone, or even of a sibling. Simple charts can be constructed to make the adjustment, so technically this is not a problem. The issue is whether or not to do the adjustment at all.

The solution is to use two cut-offs in tandem: the (unconditional) 0.4th centile and in addition a familiarly adjusted 0.4th centile. This will pick up all very short children, plus some short children who come from tall families.

One practical issue is who should do the adjustment - the school nurse, or the GP after referral to primary care? If the GP does it, short children from tall families will not be identified by the school nurse, and so will not be referred. So if the familial size adjustment is to be done at all, it must be done as part of the primary screen. The decision on whether or not to adjust depends on balancing the extra cost of the higher grade nurse with the likely pick-up rate of short children from taller families. This is another area where we are short of hard evidence.

Commentary – Dr Hulse (Maidstone): Is there *at present* evidence to support a programme of school entry height screening for short stature for apparently otherwise healthy children? The answer is probably “yes” for growth hormone deficiency and the Turner Syndrome. For coeliac disease and hypothyroidism, the yield is low – this is secondary gain rather than a primary aim. Screening of tall children was not discussed at all. We should though think about Marfan's as this might save lives.

Most of us would be disturbed if all measurements in pre-school children were to be abandoned.

Height and weight are an important part of the clinical assessment of child. There is also evidence of the value of parental concern about growth, of the importance of growth monitoring in child protection, for certain socially disadvantaged groups in society, and for the care of children with known chronic disease such as diabetes or cystic fibrosis or where there is a history of family growth disorder. So perhaps we should be regarding pre-school monitoring as part of a local targeted programme rather than a national programme. But if you're going to measure it requires training, good equipment and schemes for quality control which simply don't exist.

A child becomes short by growing slowly but following velocity in practice has proved very difficult because it is difficult to sort our physiological variation from pathological variations in the early years. These come from measurement error, seasonal variation in growth, familiar short stature and children who are delayed.

We're left with two visions for the future. One is that every child has a personal, accurate, longitudinal record with, say, five data points on it. With the PCHR and the 1990 charts this is achievable. The problem lies in the interpretation of those charts and establishing robust programmes to determine appropriate referral criteria. So for the time being we have a more modest proposal which is universal height screening at the 0.4th% at school entry and selective growth monitoring for children who are at risk both medically or socially.

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REFLECTIONS ON HEARING SCREENING – CHANGING THE SYSTEM

**Adrian Davis (MRC Institute of Hearing Research,
Nottingham)**

Our systematic review presented last year showed that the infant distraction test finds about one in four children with a hearing impairment at a cost of at least £30000 per child. Neonatal screening could probably find around about eighty to ninety per cent of the children at a cost of £15000 per child. We made a number of recommendations regarding neonatal screening - but evidence and recommendations aren't necessarily easy bedfellows. John Bamford and I have given over fifty talks, published in quite a few journals and written a number of editorials. We've been consulted by districts who are keen to formulate business plans either for targeted neonatal screening or for universal neonatal screening. Policy has to be formulated on the basis of the evidence and the different models that you look at in terms of the cost, cost effectiveness and overall cost. One has to consider whether a local or a national scheme is best and I think that if a national scheme is chosen than the reviewer has to be prepared for additional work.

One of our *research suggestions* has been taken up by the HTA. We think it is worth screening for moderate, severe and profound deafness but in the States they also recommend screening for unilateral and mild hearing impairments. This needs further study.

But in *policy making* the review has created uncertainties. Individual districts see that they have failing services and want to take them forward. Researchers are not sure whether they should be doing more primary research or taking forward the implementation plans and strategies.

We need better co-ordination between the preparation and dissemination of reviews, and the policy implications - and that implies pre-arranged funding channels. We need a better understanding of how national screening programmes can be implemented, the time scales in which we're working, how those programmes are going to be funded and the wider context, the impact that a hearing screening programme might have on other parts of the child's health. Is a national policy always best? Do we need information to be given out in a uniform way? Districts might say - we agree to quality standards for information, but we want a local way of giving that information which may be context dependant.

Over the last month or so, we have given a great deal of thought as to how we are going to go forward with a national implementation of neonatal hearing screening and we plan an initiative to accredit high quality, family friendly hearing services that have a lot of the features that Theresa Marteau talked about this morning - considering the needs of the family both in terms of information and service.

Editorial comment: Professor Davis has highlighted a key issue – how do you move from a review that argues for a particular screening policy, to implementation? This is

much more difficult if one aims to do this on a national scale. On the other hand, the history of screening in child health, with a wide range of projects of varying and often indifferent quality, suggests that a national policy with agreed standards is desirable both for quality assurance and for equity.

The Children's Sub-group and the National Screening Committee have agreed a *policy statement* supporting a gradual move to universal neonatal screening, with the caveat that there are several unanswered questions - how to minimise anxiety caused by screening, how to ensure a streamlined responsive service, and what the true staffing and cost implications might be. Community screening might turn out to be a better buy than hospital based screening - but more work is needed to assess that possibility. The quality standards guidelines, which should be fulfilled before any district embarks on universal neonatal screening, are in preparation. The next steps are a review of which districts wish, and are able, to move to universal neonatal screening; a cost analysis of what implementation will cost; and a development programme to deal with issues such as information systems, and technical protocols for screening. The Children's Sub-group will monitor the process.

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Prospects in school screening for otitis media with effusion (glue ear)

**Mark Haggard (MRC Institute for Hearing Research,
Nottingham)**

Before addressing issues of screening appropriateness and method we must first ask:

What is the impact of otitis with effusion (secretory otitis, glue ear) - does it matter? The literature on this is heavily biased to medium-term sequelae in linguistic and cognitive development. These are small but this literature was not driven by the relevant public health policy question. The most universal way of measuring and scaling anything involving individual differences is in relation to the statistics of the population, population centiles, so we use the standard deviation of the normal population as the scale unit.

Imagine you're sitting in the middle of the population at the median or fiftieth centile; if you get a boost by one third of a standard deviation, you go up from the fiftieth to the sixty-first percentile, about eleven centile points. By this notion a wide variety of stakeholders can understand what is meant by a shift in difference, for example, of one third of a population standard deviation. The long-term post-remission, cognitive and behavioural sequelae of glue ear reported in the literature are mostly smaller than this – of the order of *0.15 up to about 0.35 of a population standard deviation*, depending on particular outcome measure and age. This is not a null effect, but neither is it a massive effect. It requires large numbers, good measurements, and well-controlled comparisons to talk meaningfully about such effects of modest size. The

fact that many studies have not met these criteria does not mean that nothing can be said.

To supplement understanding of disease impact given by developmental sequelae, we have been quantifying the magnitude of the *concurrent* impact, on symptoms, health, and quality of life, with a particular emphasis in effects on behaviour. As you might expect, *the effects while the child actually has the disease are generally larger*, by a factor of about two. Considering behaviour, parental quality of life and so on, our findings are of the order of 0.4 to 0.7 of a population standard deviation. That would take you from the median, (the 50th percentile) in the population, up to about the seventieth. That is a large enough disease impact to consider that there could be demonstrable beneficial effects of treatment and screening, and obtaining the funds to provide rigorous evaluation of them.

Timescale? Should we be worried about what happens *during* the disease, when the disease eventually remits in most cases? This remission issue, plus the interest in the critical period hypothesis, is perhaps what prompted the emphasis on developmental sequelae in the past. The typical duration for a child to be persistently affected by glue ear is from the second half of the first year through the next five years of life. The reference timescale for developmental sequelae must by definition lag, but can be thought of as starting at the beginning of schooling, through to the point where any sequelae become very hard to show, i.e. the second five years of life. Even in very large sample sizes, ten thousand or more, we can only just detect at conventional statistical significance levels a few cognitive sequelae lasting through to age ten. By that age the sequelae have largely been 'caught up' or swamped in extraneous variance. On the QALY principle of no time-discounting an effective 0.6 impact of a standard deviation in the first five years of life (concurrent), equates to a deficit of about 0.3 of a standard deviation over ten years of life (sequelae). In other words the magnitude offsets the shorter duration; we probably cannot disregard developmental sequelae but we certainly cannot disregard concurrent effects.

Evidence on treatment? The only treatments that are yet worth considering within the public health perspective on glue ear are surgical. Whether or not grommet insertion or adenoidectomy makes a difference to important outcomes is the subject of the TARGET national randomised control trial. The results have not been unblinded for publication purposes yet, so I cannot tell you anything here about the cost-effectiveness. The trial has appropriate outcome measures enabling us to determine whether operations (which certainly on average improve the condition of the ear) also improve the quality of life of the child and family. The trial involves not just proximal clinical measures, but also ultimate measures such as general health, behaviour and parental quality of life. The trial will tell us what we need to know, and whether the magnitude of improvement in the important variables is worthwhile.

School screening. What has all this got to do with current school screening? Firstly it does seem possible, if treatments are shown to be effective, that screening, largely

unquestioned for thirty years, will continue to be considered worthwhile. Secondly, one day we will have a biological marker for the persistent form of the disease. We need that because at some point or other, 83% of children are touched by this condition and it is not effective to see all of them in specialised clinics; we want to refer the hardcore. We don't yet have a perfect means of defining them in advance. Present case-finding and treatment policy is summarised in two words: watchful waiting. This involves a partial logical contradiction; it trades a presumed decrease in duration of benefit (via delay) in persistent justified cases for treatment, against the cost and inconvenience/risk of referral in non-persistent cases. Watchful waiting (i.e. past persistence) is so far the best predictor of future persistence, but it lacks specification of how to "watch" or when to stop "waiting", and it delays intervention in those that need it.

In the UK at present, many of the children who get into the system and receive treatment only do so by about age five. The major source of glue ear cases stems from school screening, whether they then go by the community or by the GP path. There are reasons for believing that school screening of hearing might have some value. It raises awareness among teachers and parents. It is well-known that in conditions that are complex and ill-defined, that are behavioural rather than organic, or which it is possible to view as elective, there is a marked social gradient in the uptake of healthcare. Mass screening (but not optional screening) tends to be more equitable than reactive referral systems. Hearing loss is such a condition, so a probable loss of equity must be considered in any removal of screening.

There are many unanswered questions beneath the policy issues in school-entry screening. How many new cases are left to find by the stage in question? Is there any effective basis for targetting sub-groups of children? What is the actual health gain for cases found? Could a reactive system work? For permanent hearing impairment, by the time children are five years old, the screen yield includes only unilateral and mild/moderate cases. For OME (glue ear) cases we are unsure about whether there is any benefit in detecting unilateral hearing loss or cases not already referred on a systematic basis.

The concept of incremental yield (IY) is useful here; it is the number of new cases that a screen finds. There are no good UK data on IY for school entry screening, but a screening study done in Sweden at age 7 provides a pointer. Having screened over 2,500 children, at age seven (they had been previously screened at age four), the number of new cases they found was very small. Of cases that were not previously known, or had missed previous screens they only found 6, which is an IY of 0.2%. Most of these children would have had a history - so the issue arises: could they have been found by a history-based questionnaire, without conducting on thousands of schoolchildren tests that are audiological and acoustically dubious in the real environment of our schools? We have developed such a questionnaire (in our epidemiological studies) and are letting others use it in a monitored fashion. The Childhood Middle Ear Disease and Hearing Questionnaire (CMEDHQ) covers the

manifestation of the disease, the consultation history plus hearing-related behaviours, and some risk factors. It is simple to score, and has a cut-off value that can be varied at will.

The cut-off value specified currently fails approximately thirty per cent of the age-group in winter. Although comparable with tympanometric surveys in other countries, that is rather a high percentage which could be reduced later. Here we err towards inclusiveness for research purposes in order to evaluate and improve the questionnaire, and avoid scandals of under-sensitivity at this early stage in evolution of the method. Varying the cut-off gives the classical trade between sensitivity and specificity. At one operating point they are equal; but if you really need to find most cases, then you will be prepared to incur poor specificity in order to achieve high sensitivity. From the questionnaire we have a continuous measure which generates that classical trade-off, allowing choice in policy. For audiometry, the degree of equivalent trade-off allowable is limited, chiefly by the noise floor in the testing room and the high prevalence of transient OME. False positives escalate dramatically for cut-offs below 20 dB, yet professionals do not like discharging children with this (or worse) hearing loss.

The MEDHQ has been evaluated in South West Cumbria on nearly three thousand children. Of those who fail the sweep screen test, two thirds are proven to have an abnormal conventional audiogram and go on to a further audiogram. Eventually 202 ended up at ENT and of these, about 40% were treated by an operation, a simple marker for an outcome that is of substantial concern as well as one that comes close to reflecting the population health gain from screening. All children had also been given the questionnaire. The comparison between the existing sweep test and our questionnaire found the instruments to be comparable in sensitivity and specificity. Of course not every parent is sensitised to their child's hearing problems, so, (depending on the way the index is computed) a questionnaire is generally expected to be less sensitive than a test. On the other hand, the specificity tends to be higher, unless the cut-off is purposely set to maximise one parameter at the expense of the other.

The favourable result with the questionnaire is both surprising and pleasing, and we think we know why it occurs. Screening should not seek to establish whether a child has fluid in the ear on a particular day, but whether they have a long-term history. Tympanometry is a very useful objective test method, but in relation to the above objective a single tympanometry fails to address the real issue. Existing tests don't tap into length of history, which is still the best predictor of a future history as a justification for intervention. Despite all the imperfections of questionnaires, they do integrate over time information from a close observer. The parent has been living with the particular child for perhaps five years. Of course identification of cases on this basis automatically calls into question whether watchful waiting would have a role in subsequent management of cases found in this way.

Conclusions So, what policy might work in this area? I do not think we can scrap the school-entry screen until we are sure that it's no use. We can't know that until

we know the answer about the effectiveness of treatment. Assuming that there is an effective treatment to follow screening, the questionnaire might enable community services to cut drastically the number of children who receive an audiogram, and give them a good audiogram rather than a bad sweep. Use of the questionnaire would also save by reducing the amount of repeated audiometry, because the questionnaire already taps persistence. This is one example of how resources could be redistributed to enhance effectiveness. It could also spread awareness and provide supplementary information for selective referral and even treatment.

We have established the *prima facie* case that a questionnaire is relevant and could be cost-effective. Next an independent research group needs to test a shorter form of the questionnaire applying a variety of different cut-offs in a complete design with 100% follow-up rather than (as here) follow-up only for failures on the sweep. The responding rates in inner cities are the chief concern. Response rate was well over 90% in South West Cumbria, but whether that generalises to Brixton or not, I do not know. That is one of the hypotheses to be tested, before a total system evaluation is done of costs, risks, and effectiveness. Decision on such a major screening study must await information on the effectiveness of surgical treatments to emerge early in 2000.

Editorial comment: Professor Haggard's paper presented data from work in progress – watch for the finished product. Other evidence from this group and elsewhere supports the notion that glue ear is significant, at least for some children, but does not have the devastating impact that many suspected in the past. Of course, for a very small number of individuals, extreme impact could result from synergy with other predispositions or pathologies. We need to find ways of identifying the persistent severe cases. I personally doubt that any *current* method of screening *in infancy* can help us, since an infant can develop severe glue ear at any time. Perhaps better parent education might help – in conjunction with education about communication in general.

As for school entry – the best advice is to continue with the sweep test for the present – we should have a better idea next year whether this should continue. We might continue the sweep, substitute the questionnaire or abandon school entry screening altogether.

Improving the distraction test A final and very important point – the distraction test will continue to be used until universal neonatal screening is implemented. Discussions with Professor Barry McCormick (who has given me permission to quote him) suggest that raising the screening level to 45 or even 50 dB might greatly improve the specificity of the test; it would reduce sensitivity especially for minor conductive hearing impairment; but the reliability of the responses, the significance of a failure to respond and the consequent reduction in repeat testing and referral would be major benefits.

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VISION SCREENING

Alistair Fielder (Imperial College, London)

Last year we discussed the Systematic Review on Preschool Vision Screening by Stewart-Brown and Snowdon (1997), which highlighted that there was a lack of good research on the natural history, disability and treatment of amblyopia. I have been asked to take stock of where we are at with vision screening – is there work in progress and have there been any advances?

Disability associated with amblyopia There are two aspects to consider. First, *what is the disabling effect of a deficit in one eye, or a mild deficit in two eyes?* As sighted individuals we might think we have an idea of what it is like to be totally blind - but given a moment's thought we realise simply cannot fully comprehend life without sight. We have absolutely no understanding of the disabling effect of a relatively mild deficit of one or both eyes; ie how it impacts on life's various activities such as social interaction, education, occupation and leisure. This is an important area for future research.

Second, *loss of the better eye.* Amblyopic individuals are well known to have an increased risk of losing the better eye through injury or disease, but this is based on only two reports. The British Ophthalmic Surveillance Unit of the Royal College of Ophthalmologists will in 1999 report its preliminary findings of a UK-wide snapshot survey of amblyopic individuals who lost the vision of the fellow eye during a 27 month period.

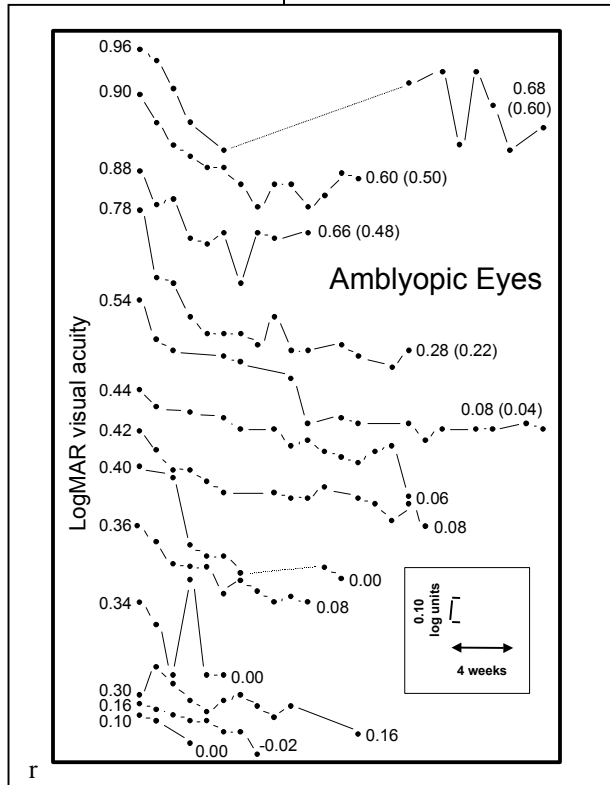
What about treatment of amblyopia? Two treatment trials (one a randomised control trial) have been funded and are due to start in 1999, so hopefully in a couple of years we will have a better understanding of amblyopia therapy. *Penalisation* is a treatment that's been around for many years which is enjoying a resurgence of interest. Unlike occlusion therapy which completely blocks out the vision of the better eye, in penalisation the vision of the fellow eye is blurred by atropine eye drops or ointment. This form of treatment may be less disabling to the child and according to the literature is approximately as effective as occlusion therapy

What is the natural history of amblyopia - does spontaneous improvement occur? The systematic review of preschool vision screening cited literature as supporting the possibility that spontaneous recovery might occur. Kurt Simmons, a US vision scientist, reviewed the same literature and came to the opposite conclusion. In my opinion the literature in question does not merit this close scrutiny and is not robust enough to form the clinical

evidence base. Simmons and co-workers also studied children with amblyopia who were tested more than once, many months apart, and who had not received any treatment and they reported no improvement (in press, British Journal of Ophthalmology). In conclusion, there is no substantial evidence that spontaneous improvement of amblyopia occurs.

The "dose" of occlusion Ideally, when embarking on a course of treatment, both the total dose and the dose rate of the treatment in question should be known – but they are not for amblyopia therapy. Determining therapeutic "total dose" and "dose rate" requires compliance data, which has only recently become possible by the design and manufacture of an occlusion dose monitor. Pilot studies are encouraging and a major study of compliance is soon to commence.

Correct refractive error first and wait until vision has stabilised Prescribed occlusion doses range from less than 20 to over 3000 hours – the latter surely is demanding for child and parents alike! Currently, occlusion is started either immediately or a few weeks after the refractive error is corrected by the prescription of spectacles. Recent work



has shown that improvement of visual acuity continues for quite a long time (up to 12-16 weeks) after spectacle wear has commenced (Figure). Thus "spontaneous" improvement will continue after occlusion therapy is conventionally started. This finding has immediate and practical implications. First, if occlusion treatment is commenced before vision has stabilised after spectacle wear has begun, it follows that some of the improvement will be falsely attributed to occlusion therapy as it would have occurred anyway with spectacle wear alone. Second, and important for the child: the worse the vision when occlusion therapy commences, the more unpleasant it is – being made to use only an eye which technically blind. So, to start occlusion after all spontaneous improvement has taken place, when the vision is stable, ensures a minimum of occlusion which is more comfortable for the child and less demanding for the family.

Screening for myopia? Should we be screening for other refractive states? In my opinion myopic children don't suffer greatly, for in contrast to the severely hypermetropic child who cannot see clearly at any distance, myopes (about 8% of six-year olds) see well by going closer to books, television, etc.

Functional consequences of refractive errors. The relationship between refractive error such as

hypermetropia and anisometropia to visual acuity and function is not understood. Here the limitations of measuring visual acuity alone become apparent. Professor Michael Tobin, Department of Special Education, University of Birmingham cites many examples of children with relatively mild abnormalities of vision, such as the child with a high refractive error, or nystagmus associated with albinism, who takes much longer to read and comprehend a piece of text compared to the perfectly-sighted child. Speed of information processing is clearly important educationally and yet is not measured in eye or child development clinics.

At risk groups We need better protocols for the follow-up of children who are born premature and those with neurodevelopmental problems. Both groups have a greatly increased incidence of strabismus and functionally significant refractive errors. Children with neurodevelopmental problems are also at risk of severe visual problems such as a cortical visual impairment, which not infrequently remains undetected for prolonged periods.

Fast-track community orthoptic and optometric clinics Another recent development is fast-track clinics run by orthoptists and optometrists using protocols drawn up by the ophthalmic team (ophthalmologist, orthoptist and optometrist). Such clinics, which can be based in the community or hospital, facilitate the simple identification and treatment of amblyopia and strabismus. The ophthalmologist need only become involved if the vision fails to improve spontaneously, or surgical treatment is required. Safeguards are required to ensure that the child harbouring a serious ocular pathology receives the appropriate care quickly.

Summary Last year, following the publication of the Systematic Review of Preschool Vision Screening, there was a natural display of professional defensiveness and confusion – “have we been completely wasting our time over the past few decades?”. We must now move on and I am pleased to report modest progress. Treatment trials are just about to begin and we will very soon know more about the lifetime risk of amblyopic people losing their better eye. We know that occlusion therapy need not – should not! – start concurrent with spectacle correction. Fast-track clinics offer a better and more convenient service for the child and family. The challenge of the next few years is to answer those critical “so-what” questions – what are the functional consequences of amblyopia, strabismus, and certain refractive errors.

Editorial comment: The Children’s Sub-group plans to re-visit the whole issue of vision screening in the summer of 1999. We need a working consensus while we await the results of trials in progress.

FIRST, DO NO HARM: The psychological aspects of screening and the importance of reliable information

Theresa M Marteau (Professor of Health Psychology at King's College, London).

This paper considers: the main psychological harms that have been documented in screening programmes; the role of information in reducing or avoiding some of these harms; why what we do know isn't actually implemented into practice.

I Psychological Harms

The three most frequently documented harms in screening programmes are:

- raised levels of anxiety, particularly documented amongst those who receive positive and, in particular, false positive results in screening;
- false reassurance, most often documented amongst those getting negative test results;
- uninformed decision making or uninformed choices.

Anxiety

Rothenberg and Sills (1968) described what they called the PKU Anxiety Syndrome. They were working in a hospital in Brooklyn when the PKU screening programme was first implemented and the false positive rate was about 10% at that time. They were seeing four or five parents a week who were not reassured by further testing which showed that the child wasn't affected and they were continuing to wait for the first signs of their children not developing.

To my knowledge there have been no systematic reviews of anxiety in neonatal screening programmes or indeed in antenatal screening programmes. We have recently completed a review of the emotional impact of screening in adults for a variety of conditions (Shaw, Abrams & Marteau, in press). There are differences in anxiety levels, within the first month of undergoing screening, between those who get a positive test result and those who get a negative test result. When we looked at anxiety from a month to a year, there was no difference between those testing positive or negative. But residual anxieties may well linger longer in neonatal screening programmes as suggested in several qualitative studies (McNeil, Sveger and Thelin, 1988; Tymstra and Bieleman, 1987).

False reassurance

This involves believing that your risk is lower than a test result suggests it should be. The most common is to receive a negative test result on screening and to believe that you have absolutely no risk whatsoever of you or your child having a condition – but usually there is a residual risk. A survey across six hospitals revealed that over 50% of women undergoing antenatal screening for Down syndrome do not understand that a negative test

result means low risk not no risk (Smith, Shaw and Marteau, 1994).

What happens to those who think everything is OK after they've undergone screening and then give birth to a child with Down syndrome? We recently interviewed parents of 260 babies with Down syndrome, a hundred of whom had received negative results in screening. One angry mother said "We couldn't have avoided conceiving the child, nothing that we could have done prior to conceiving the child but there is something we could have done after she had been conceived - but we weren't able to because the tests weren't done" (Hall, Bobrow and Marteau, 1997).

Uninformed decision making.

A survey in Wales asked women whose new-born infants had undergone the routine biochemical screening programme whether they'd ever heard of a number of conditions. The majority had heard of neonatal screening but only a tiny proportion had ever heard of PKU or Hypothyroidism, the conditions for which their babies had been screened.

II Role of Information

Responses to screening are a function of the situation (the screening that someone's just undergone) and the characteristics of the person (Marteau & Croyle, 1998). There are *three key characteristics* which will influence how people respond to screening:

- A person's mood before screening - the more anxious or depressed they are before screening, the more likely they are to be anxious and depressed afterwards.
- The way they cope with threatening information.
- Whether or not they're expecting to be given the test result that they actually get.

The information people receive about screening programmes is often unsatisfactory, though there is little experimental evidence to show that if we give people different kinds of information, or communicate in a different way, we can make a difference. The HTA systematic review of biochemical screening, for example, drew attention to poor communication and how it is associated with more adverse psychological outcomes for parents.

A group in Wales studied women who had undergone routine cervical cancer screening (Wilkinson, Jones and McBride, 1989). Of those recalled, 61% believed that their abnormal result meant they had cancer. They had high levels of anxiety measured on a standard scale. The group then prepared a very easy-to-understand leaflet explaining what the screening programme was all about. There was a dramatic reduction in the number who still believed they had cancer. That's a powerful example of how the right kind of information could lower anxiety.

There have been very few studies comparing different ways of giving risk information to clinical populations - the bread and butter of all screening

programmes. We know next to nothing about how best to give this kind of information.

What information should people be given? Probably they need to know what the screening test is purporting to do, how it performs, the process of screening, the benefits and limitations and the pros and cons of making different decisions. Nearly every survey of written information for patients finds that it's fairly poor. Information has to be written in a way that the majority of the people can understand -that includes writing in the language of the population who need the information.

If you frame the same information in different ways, you get different results. People are apparently very sensitive to the numerator so, for example, if you tell someone that there's a chance of a particular outcome of 100 over 10,000, they will think that it's more likely than if you say the chance is 1 in 100 - although actually it's the same. So it may be that one way of communicating the risk is to have a larger numerator. I put that up as a hypothesis which needs to be researched.

Telephone communication is often used in screening programmes, but may not be as effective as face-to-face communication. In a study of neonatal screening for cystic fibrosis in the States, parents' understanding of their test result following a false positive result was compared according to whether communication was face-to-face or over the telephone (Tluczek, Mischler, Farrell et al, 1992). Parents' knowledge was better when the communication was face-to-face than over the telephone. There's also an issue of *when* to give information and the need for different information for different stages of the screening programme.

III Implementing research findings in practice

Why is information provision still poor in many screening programmes? There are examples of good practice to be seen in the breast and cervical screening programmes. A deluge of reports all recommend that the provision of information written and also face-to-face could be made better. For the last twenty years these problems have been recognised but very little has changed.

I suggest five hypotheses for why communication of information in screening programmes is not improving despite evidence that it needs to:

- Some centres are unaware that there are harms associated with screening
- Some, while aware of the harms, are unaware that these can be avoided or reduced using effective methods
- Health professionals perceive that their *own* practice is unharmed unlike that of others
- Health professionals think that avoiding harms isn't a good use of finite resources - because to communicate does cost money and time which they consider better spent elsewhere
- The methods that are intended to *reduce* harm may be *seen as harmful* by staff. In a descriptive study, midwives and ultrasonographers were asked to comment on an "informed choice" leaflet on fetal anomaly scans, which explained that the scan wouldn't always pick things up, and also that a number of babies were aborted who were in fact

normal (Oliver, Rajan, Turner et al, 1996). Staff didn't want to use the leaflet, because they felt it would make women anxious.

Summary

Screening has the potential not only to benefit people, but also to cause harm. Some of those harms can be reduced. There's a little evidence that the process of communication, in particular written information, can be important in reducing or avoiding some of those harms.

Two areas require attention. There needs to be more research on effective ways of communicating and how to implement what we know already is good practice. Our notions of standards and quality need to extend beyond the laboratory to at least cover written information. This is at the heart of the National Screening Committee and I hope we'll see progress there.

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Commentary by Dr Aidan MacFarlane (Oxford): My concern is the over-medicalisation of people's lives. We give them pamphlets, video tapes, web sites with vast amounts of information about screening. There must be a balance between providing information and trusting your medical advisor. Having to read endless leaflets about the chances of this happening or that happening, may detract from the rest of our lives.

Commentary by Helen Carter: (Research Trust for Metabolic Diseases in Children).

I represent the patient's view point. Knowledge is good medicine – the more parents understand, the more likely they are to follow appropriate care strategies. I want to make the following points, based on a survey of our members from 1996:

- Parents want clear, full explanations and written information to return to time and time again. Written

information is no substitute for that one to one contact. But in a one to one contact, it doesn't all sink in. They want something that they can talk with their partner about and other members of their family.

- People need good, clear, simple language, the language of the man or woman on the Clapham omnibus. Do not assume prior knowledge; but remember that parents with little formal education *can* understand biochemical and biological concepts. There are examples of good practice in oncology – for example, patient information leaflets prepared in partnership with patient groups, and audio tapes of consultations in breast cancer clinics form a useful record of information communicated to the parent. They can take these home and reflect on it.
- To share information successfully, you have to deal with emotions. The fear, anxiety, anger, guilt, confusion or other emotions which can accompany a positive diagnosis can get in the way of successful data exchange and true dialogue and it is here, in particular, that written or taped information can provide a helpful backup.
- The way in which information is given - and the words used - is crucial and influences subsequent attitudes. Parents need information in ways which anticipate their questions, consider the likely impact, are linguistically and culturally sensitive, and allow them to make informed choices (this is a more parent focused concept than the more usual one of giving consent).
- We need to choose words carefully. Familiarity bred by routine can cause people to become sloppy with their use of language without considering its impact. One of our parents, whose child has PKU, was told when the test was done – “it’s very rare, it’s nothing to worry about, no one has it anyway”. I’m jolly glad I wasn’t that health care professional who had to go back and say, “um, well actually, you know I said it was rare and no one has it anyway, well, your child does.” I don’t think she enjoyed that very much and I know the parents didn’t. I asked a parent recently what they were told before the PKU test and he said – “they said it was to find out if my child would be mentally retarded.” Now if that test had been positive, what would be going on in those parents’ minds?

Timing is important It is no good giving half a story. Whereas everyone will need to proceed at their own pace it is important to have knowledgeable competent people ready to meet the parents’ information needs in full. This has training and resource implications. This story illustrates the point:

A mother was told that the Guthrie test was to screen for a rare disease and was “nothing to worry about”. The test was positive. The midwife said she knew little about the condition but it was something to do with diet and brain damage. Mother was told to wait and see her GP – but she couldn’t wait and looked up PKU in books she was given when pregnant. She found a small paragraph in a chapter headed “the mentally

handicapped child”. This immediately made her believe that “our son was doomed to a terrible life of suffering”.

The potential harms are sometimes used as a reason for not introducing screening. In terms of ethics, litigation and economics, that may make sense, but it doesn’t make sense to the parents whose children end up with life-limiting illnesses that could have been prevented. No-one suggests that because we don’t always get it right for PKU we should stop the screening programme. So why is the argument that we may sometimes get it wrong an argument for not starting screening for other treatable diseases?

I am not condoning poor practice. Good written information was described by one survey parent as “a lifeline”. Another said it was the “difference between night and day”. We should implement the lessons learned from the screening programmes already in place. We should also remember the 80:20 rule. If we wait for everything to be just right, children who could be leading fulfilling relatively normal lives will continue to endure unnecessary suffering. So let us argue for continuous improvement - rather than using imperfections in the system as a reason for deferring action in embarking on further screening programmes.

Other points made in discussion: It is easier to describe than to quantify benefit and harm in screening – value judgements have to be made.

Many people in this country are functionally illiterate – but most have a friend who’s literate, who will read for them – so it is worth producing written material. There are ethnicity and cultural aspects – in how many languages do you produce literature? Liaison workers and community support networks are also needed.

We need standardised information about neonatal, antenatal screening that could be the basis of local leaflets, so we can avoid what is happening now and harm being done. This has been going on far too long.

Editorial comment: Tackling the issue of high quality information is a priority task for the National Screening Committee.

PARENT SUPPORT: THE ROLE OF HEALTH VISITING

Hilton Davis (Professor of Child Health Psychology: Guy's, King's & St Thomas' Hospitals School of Medicine/Lewisham and Guy's Mental Health Trust, Bloomfield Centre, Guy's Hospital, London SE1 9RT)

Two short stories contain the essence of what I have to say. A mother was concerned about behaviour problems in her child and met her health visitor at home. After ten minutes the health visitor gave exact advice on what the mother should do and then left within about twenty minutes. Although the advice was sound and based on accepted behavioural principles, the mother took no notice of what she had been told, remained anxious and sought help elsewhere.

A second mother with a very similar problem was visited by another health visitor, who listened to her very carefully, treated her with respect, and helped her to explore the problem. She gave no instructions about dealing with the problem, but helped the mother to think about how to observe and try to understand it. A week later the problem had improved dramatically, as a result of the mother herself devising a strategy identical to that advocated by the health visitor of the first mother.

Parental support is determined not only by what potential helpers do, but by the ways in which they do it. If they communicate genuine respect and care for people and work with them (not on them), it is remarkable what parents can achieve themselves. Help and support is as much about making people feel valued, able, capable, important and strong, as it is about giving information and advice. In fact, these feelings may be a prerequisite for effective information and advice.

This paper will outline a model of support, which is possibly an essential and primary ingredient of any methods of helping families. Preliminary evidence for the effectiveness of the model will be provided, followed by consideration of its implications for health visiting.

Parenting is one of the most demanding tasks that one is ever likely to undertake. The way children are treated has major implications for the health, development and emotional well-being of the children themselves, for their life as adults, for subsequent generations and society in general. Support for parents would, therefore, seem to be essential, as is being recognised by the UK Government in, for example, the Green Paper entitled "Supporting Families".

Health visitors are an important part of the proposed strategies for the provision of parental support. A series of major problem areas (e.g. post-natal depression, marital problems, youth crime, child abuse, adult mental health problems, child mental health problems and the effects of disability and chronic illness) have in part their origins in the psychology of parents and the family, and have solutions that are psychological and social and relate to family support. A team working in North Southwark, one of the most deprived areas in the country, found that 37% of children had three or more significant psychological or social problems, and that 50% of the

families had three or more risk factors for child mental health difficulties, including marital difficulties, physical and emotional problems in the parents, alcohol problems and criminal convictions (Davis, Day, Cox & Cutler, 1999).

Parental support has been translated into a wonderful range of options including, for example, early years centres, play groups, nurseries, and mother and toddler groups. There is a plethora of parent training programmes from Mellow Parenting to Parent-Link. There are parent, family and child befriending schemes including Newpin, Homestart, and Friends United Network. There are home visiting schemes including the Parent Advisor Service (to be described below) and counselling schemes to tackle postnatal depression (e.g. Holden, Sagovsky & Cox, 1989). Health visitors run sleep clinics and there are programmes (e.g. PIPPIN) aimed at helping parents learn about being a parent as opposed to instructing them in changing nappies. This wealth of options is carried out by a range of people from volunteers and parents themselves, to health visitors, teachers, nursery nurses, social workers, psychologists and psychiatrists.

There is a growing literature on promoting adaptation, prevention and early intervention (Cooper & Murray, 1997; Cox, Pound, Mills, Puckering, & Owen, 1991; Olds, Henderson, Chamberlaine & Tatelbaum, 1986; Olds, Eckenrode, Henderson, et al., 1997; Rae Grant, 1991). Although there are many methods of support, common to them all and crucial for their effectiveness are the characteristics of the people providing the services and the relationships they develop with parents. These characteristics are essential to the helping processes, since the provision of information or advice is only of value if parents trust the helper, listen, understand, and decide to comply. It is not sufficient to give parents information without considering what parents come to think and feel about themselves as a result of their interactions with professionals.

We have been developing models derived from counselling theories (Davis 1993). Briefly, these models assume that the process of support involves a set of tasks or stages, which begin with the development of a mutually trusting relationship, that allows open exploration of the problems or difficulties facing the parent so that a measure of clarity or understanding may be derived. Mutual aims can then be decided, strategies planned and then implemented, followed by a review of progress returning to previous stages as necessary depending upon the outcomes.

The development of a mutually respectful relationship is essential - if you are not trusted, parents will not talk openly, and you will not discover the true nature of their problems and therefore be unable to help them. Since the tasks are undertaken together, we see the model of the relationship as a partnership, which can be powerfully supportive in its own right. If a parent is regarded with esteem by a respected helper (usually perceived as in a relatively powerful position), then he or she is likely to feel valued, to increase in self-esteem, and to consequently be more able to adapt to or manage perceived difficulties more effectively. This implies, therefore, that the helper must endeavour to make parents feel vitally important and

certainly equal to the professional. That this is not often the case is apparent from parents' own statements such as "I am *only* a parent." Partnership implies working closely together, deriving common aims, negotiating, communicating effectively, and contributing different, but complementary expertise.

A set of qualities and associated skills by which these are demonstrated are required of the helper whatever his/her profession. These include respect or what Carl Rogers called unconditional positive regard. This can be conceptualised as valuing parents as people and assuming competence and strength, not weakness and incapacity. The second quality is genuineness, which implies attempting to be yourself, honestly and openly, and not being closed and defensive, not hiding behind a professional façade. If you really care, parents will know, and will equally spot pretence with its obvious implications for the helper-parent relationship. A third quality is empathy - the attempt to understand the world from the viewpoint of the person you are trying to help, as opposed to the imposition of the helper's own views. Humility allows the person who is seeking help to contribute to the process and not to simply be in the hands of someone who is all-knowing and all-powerful. This does not imply weakness, but is, in fact, derived from an inner strength, which is also essential for the helping role; helpers need to be able to think independently, which is why supervision is a prerequisite of the capacity to support others.

Evidence in support of these models includes parents' own views of their needs, satisfaction studies, observations of professional communication skills, treatment adherence research, and outcome studies (e.g. Davis & Fallowfield, 1991). Training courses to facilitate the development of these models and skills produce considerable improvements in the confidence and competence of a range of professionals to relate to and support parents (Rushton & Davis, 1992; Davis, Spurr, Cox, Lynch, von Roenne, & Hahn, 1997). People trained in this way facilitate considerable benefits for parents and for children with problems ranging from intellectual and multiple disabilities to emotional and behavioural adjustment (Avon Premature Infant Project, 1998; Davis & Rushton, 1991; Davis & Spurr, 1998).

For example, Davis & Spurr (1998) trained health visitors and CMOs to work on a home visiting basis with families of pre-school children. Referrals to this new service commonly exhibited multiple problems in the parents (e.g. depression and social isolation), in the children (e.g. aggression, non-compliance) and in family relationships (e.g. parental conflict and overt violence). Nevertheless, compared to a matched group of families who did not receive the service, those in the intervention derived significant benefits within four months of the initiation of support. The mothers rated their children's problems as significantly less severe and distressing. There were significant improvements in the mothers' self-esteem; in parenting stress; in the child's environment and interactions with the mother; and in emotional and behavioural problems, as indicated by the Child Behaviour Checklist. There were also reduced contacts with other health professionals, and the intervention was valued

greatly by the mothers, who felt respected and valued by the Parent Advisers.

In conclusion, health visitors are potentially in an extremely good position to support families. By doing so they can help improve the psychological and social adaptation of parents, promote children's development, health and well-being, prevent difficulties arising, and intervene early if necessary. The government is right to stress their importance, should substantially increase resources available for health visiting, and reverse the cuts that have been occurring. Their effectiveness depends on a broad policy that allows them to focus on the psychological and social aspects of children and families, and not just the physical. It also depends on the skills and qualities described here. These are not frequently discussed and may be wrongly assumed to exist naturally in us all. Yet they are a core element of clinical effectiveness; if health visitors are not respectful, empathic and genuine, they will not be respected in turn by parents. If they are unable to establish working partnerships with families, parents will not talk openly, will not themselves listen, and will not therefore derive help.

These skills and qualities depend upon training and subsequent skilled supervision. Unfortunately, most professional helpers, including health visitors, have never had such training and rarely receive systematic and ongoing supervision. Consequently, there are many who lack listening skills; they give advice without really understanding the difficulties and without including the parents as a partner in the planning process. As a result, their effectiveness is substantially reduced. Clearly there is a range of ability in all professions, from the very bad to those who are naturally skilled communicators. However most of us can be helped to do better with initial training. Nevertheless, the benefits derived from training must be maintained by regular supervision; when you listen to people carefully, you tend to hear distressing issues, and need support to deal with this.

Health visitors are a major resource, especially in the new Sure Start proposal. However, the needs in inner city areas outstrip the resources available, and there never seems to be sufficient help. There is also a requirement for continuity throughout the life cycle. All professionals or volunteers who works with parents or families, whatever the age of the children, are in a position to affect their well-being. Fortunately, these skills and qualities are not profession dependent; they are, in fact, just as important for the teacher as the social worker, GP or nurse. This should be acknowledged and appropriate training provided, and supervision should be in place to prevent burnout.

The skills and qualities considered here enable the helper to earn respect, and not just to assume it by way of one's position. They enable one to begin to emulate the wisdom of Gibran (1992) whose description of the teacher applies equally well to the skilled helper: "If he is indeed wise, he does not bid you enter the house of his wisdom, but rather leads you to the threshold of your own mind." Thank you.

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Commentary - Denise Kendrick (Senior Lecturer in General Practice, Nottingham). Our team from Nottingham have completed a systematic review of the effectiveness of home visiting. Among other things, we looked at 112 studies, more than thirty of which covered outcomes relating to parenting and the quality of the home environment. The analysis showed that home visiting was effective in improving measures of parenting. However, few studies used UK health visitors and most studies included participants from socio-economically disadvantaged backgrounds – are the benefits equally applicable in the UK and across differing levels? Very few studies made any mention of parent satisfaction or acceptability - Hilton's given us some pointers to how we make our programmes acceptable.

Questions that need answering - can UK health visitors achieve similar outcomes in terms of improving parenting? How cost effective is this and do professionals achieve better outcomes than lay people; and which professionals achieve the best outcomes? What is the duration of positive outcomes? Most studies had short follow-up periods except for David Old's work. Can the duration of positive outcomes be increased? Which aspects of the intervention provide the greatest benefits and

is that specific to different groups of parents? How does supporting parents compare with specific parent training programmes? How can we maximise the acceptability of these programmes of interventions to parents?

HEALTH VISITING AND SOCIAL SUPPORT

ANN OAKLEY (Director, Social Science Research Unit, University of London Institute of Education)

What is social support? The American epidemiologist Lisa Berkman said: 'From shopping bags in California bearing "Friends make good medicine" to editorials in the *Journal of the American Medical Association*, "A Friend, Not an Apple a Day will Help Keep the Doctor Away", the message is that social support is both good preventive and curative medicine. Like chicken soup, its powers are believed to be persuasive, the reasons for its effects are unknown, and knowledge of its qualities is widespread and based on folk wisdom...' (Berkman 1984:413).

Health visiting started out with the Ladies' Sanitary Reform Association which in 1862 first started to employ middle class women to visit working class mothers with an emphasis on physical hygiene and carbolic powder (Owen 1983). When the NHS arrived in 1946, health visiting became a statutory responsibility of local health authorities. Today health visitors have associated with health screening and the protection of children. They are caught between one model of care which is about supporting mothers, and another which is about risk assessment and which sees parents primarily as vehicles of risk.

There is a considerable literature on what parents think about health visitors. In six studies reported between 1979 and 1982 most mothers made positive comments, but between one in ten and two fifths of mothers did not consider health visiting a useful form of help (Bax et al. 1980; Foxman et al. 1982; Field et al. 1982; Graham 1979; North Camden CHC 1979; Orr 1980). A more recent study carried out by Mayall and Foster (1989:68-9) in London included the following comments:

- 1) Interviewer: Can I ask you why you think health visitors come round?
Mother: ...Well, you hear so many bad things about what people do. I think why they come to your house is to check up, to make sure your baby's healthy...and that you're treating it right.
- 2) Interviewer: Health visitors come to families with a baby or small child and ask to come in and talk with you. Do you think that's all right?
Mother:...I don't mind as long as they don't start telling you what you should be doing, especially if they have not got any children of their own...'

At the centre of these tensions is the home visit, particularly the unsolicited home visit. While many health visitors may think mothers are more relaxed in their own homes, mothers more often feel obliged to let the health visitor in for a statutory inspection of the premises, and do not expect it to be a supportive experience. The term 'home visiting' may mislead us precisely because of its general 'freshly baked bread' air - it *must* be a good thing. Health

visiting has the capacity to provide social support, but it does not always do so.

There is as yet no published review of the effectiveness of health visiting, though one is about to appear. There is also no review of the effectiveness of the other professionals who promote family welfare, or of most voluntary organisations working in this field. There are reviews of the components of what health visitors do: home visiting; family support; and parent education.

The Cochrane review of home-based support for socially disadvantaged mothers examined evidence from 11 trials, mainly American, of postpartum home visiting published between 1979 and 1994 (Hodnett and Roberts 1997). Six reported lower injury rates in the visited group. Eight looked at child abuse or neglect, and in four it was lower and in four higher in the visited group. Six trials examined the rate of child immunizations, four finding a lower rate of incomplete immunizations in children of visited mothers. Four trials reported on hospital admissions, and in all four the rate was lower in the visited group.

A second, non-Cochrane, review examined 31 trials, of which seven covered the period before birth (Olds and Kitzman 1993). Most of the trials were carried out in North America with disadvantaged populations, and home visitors were mainly 'paraprofessionals'. Olds and Kitzman concluded that home visiting offers a promising approach to the promotion of child health, and they came down on the side of well-trained professionals.

There are three reviews in the Cochrane library that cover support from caregivers during pregnancy, childbirth, and the postpartum period (Hodnett 1997a; Hodnett 1997b; Ray and Hodnett 1997). These reviews concluded that offering additional social support for at-risk pregnant women was not associated with improvements in medical outcomes, but there was convincing evidence of improvement in psychosocial outcomes; the evidence on social support and childbirth was persuasive, as was that for the potential of social support to reduce postpartum depression.

Jane Barlow carried out a review of the effectiveness of group-based parent training programmes in improving behaviour problems in children aged 3-10 (Barlow 1997). This approach was generally associated with improvements in children's behaviour as assessed by parents. One study used a 'placebo' control group of mothers talking about their experiences, and this was just as effective as professional-led didactic parent training.

The lack of well-designed studies in this country is notable, but it would be wrong to conclude that we know nothing. The Acheson report on Inequalities in Health (1998) commented that parents caring for children in disadvantaged circumstances are likely to need additional support if they are to protect their children from the effects of disadvantage. The report mentions two support trials with 7 and 15-year follow ups as evidence for the long-term value of enhanced support (Oakley et al. 1996; Olds et al. 1997; Olds et al. in press). In the Olds' trial, socially disadvantaged mothers who were visited had fewer subsequent births, less time on welfare, less substance abuse, and fewer arrests than mothers who were not

visited, and their children smoked and drank less, ran away less, had sex less and were arrested less than the children of mothers in the control group.

These findings are similar to those of the Perry pre-school project, also in the United States, which evaluated the effectiveness of good quality day-care combined with home visits. The 27 year follow up showed that children who had been in daycare had as adults a significantly higher level of schooling, higher monthly earnings, a lower use of welfare services and significantly fewer arrests (Schweinhart and Weikart 1993).

Investing in children as securing payoffs in later life as well as yielding short-term health gains. Health visitors in this country still provide a universalist service and are therefore in a key position to operationalise this investment. One crucial unanswered question is about the relative effectiveness of professional versus non-professional support. David Olds in Denver and my own group are both currently conducting trials on this.

Whatever work is done in future should preserve sensitivity to people's experiences and to different sociocultural contexts. One home visiting study had a programme objective of encouraging parents to have a 'future orientation' in their childrearing practices (Kitzman et al. in press). But the study was difficult to do because many homes lacked clocks. So the researchers had to buy clocks in order to encourage active thinking about the future. Material resources are also a crucial aspect of 'successful' parenting.

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Commentary – Professor Jane Robinson (University of Nottingham): Our systematic review will be with the NHSE by the end of January. It found that home visiting by British health visitors or their overseas counterparts is effective in improving parenting skills and the quality of the home environment, ameliorating child behavioural problems, improving child intellectual and motor development, especially among children with low birth weight or failure to thrive, increasing the uptake of childhood immunisation, reducing the use of emergency medical services, reducing the frequency of unintentional injury and to a lesser extent the prevalence of home hazards, improving the detection and management of post-natal depression, enhancing the quality of social support to mothers, improving rates of breast feeding and limiting family size.

Where there was insufficient evidence of effectiveness, recall that “no evidence for effectiveness is not evidence for non-effectiveness”. However, on child abuse and neglect there were equivocal findings. There is observer bias in the studies - families targeted for home visiting may appear to have a greater incidence of child abuse.

Much of the literature even remotely eligible for inclusion in any meta-analysis was non-British and based on targeted services rather than a generalist universal service provision. There are a number of groups visited by British health visitors who have never been entered into any form of experimental study. School nurses have never been entered into any form of randomised control trial - neither has the visiting of the homeless or, for example, traveller families by health visiting on which there are a number of papers written by health visitors. Health visitors are doing an enormous amount of work with the marginal in our society. This is not properly evaluated. The work they do is so insufficiently recognised and referrals to other agencies are a highly precarious business to put it at best.

Historically health visiting has always been determined by whatever was politically in fashion and whatever particular theories about child rearing were in vogue. We need to take that into account when we discuss the evaluation of services like this and indeed medicine and other occupations too. There are many ethical dilemmas for health visitors, particularly in the area of child abuse – they go into homes as friends of the family and yet at the same time have to go to case conferences and

spill the beans about what a family's been up to. These issues were raised at least fifteen years ago and yet have never ever been addressed by policy makers, by health service managers or academics.

We need to consider how we examine the relationship between social and psychological forces and biological functioning. How do these three areas interact and how do we use the research methods from these three areas constructively together in order to build the larger picture. As I am now retired, I wish you well.

Anonymous Comment: Most districts have a written policy about child health surveillance, but fewer districts have any kind of written policy about promoting child health. [Chair asked by show of hands how many delegates did have such a policy – only a minority did].

Delegate from Scotland: Scotland has the pregnancy booklet which is a national document and there's health promotion aspects in it...[Chair replies:] I can't imagine that dumping a book on the kitchen table in a high rise flat in a very poor area has the slightest impact on health. One clearly has to do a lot more - that's what I'm asking about.

Comment: In my part of the world, written materials, often immigrant families do not read their own language - so written materials do not provide much of an answer in health promotion.

[Chair:] Do you have a policy of a health visitor going through a list of hazards in a home, arranging that poor parents can borrow fire guards from an equivalent loan store, arranging peer support group for a mother who's trying to get breast feeding going and so on. If you're going to have much influence on the maintenance of breast feeding, peer group support is at least as good as professional support and may be better. Targeting - the evidence suggests that you get far greater benefits, far greater health gain per pound spent if you target your resources to certain specified groups than if you try and give the same service to everybody across the board.

We see child health surveillance as a core programme giving the opportunity of a minimum number of contacts whereby you can assess need and detect problems that arise - but certain clients will need a much greater level of input if you're going to achieve the sort of objectives that you're setting. There's a roughly fifty/fifty split between those who practice some form of targeting as a stated policy in their district and those who essentially, as far as their policy is concerned, were giving the same to everybody. People worry about whether having a health visitor is going to become stigmatised like having a social worker. Some feel that there should be a universal programme for everybody.

[Comment:] The single major effect on health is the quality of distribution of the gross national product in the country as a whole. We have small fingers in very large holes in the dyke which are created by the society which the government put forward to us.

Editorial comment: The papers by Davis and Oakley together with the comments and discussion suggest the direction for future development and research by health

visitors. The role being described here is in keeping with SureStart thinking, and is in keeping with changing evidence and opinion - but it moves even further from the traditional "medical" model with its emphasis on defect detection. This has implications for the concept of a core Child Health Surveillance programme, which will need to be considered by the next Joint Working Party on Child Health Surveillance.

SCHOOL NURSING: DO WE NEED A HEALTH WORKER IN SCHOOLS?

Jane Lightfoot (Social Policy Research Unit, University of York)

There is no consensus as to the role of school nurses in child health. Factors contributing to this uncertainty include the changing health needs of children, relatively little research into the role and activities of school nurses, and the potential for developments in primary health care to influence models of service delivery. An important question is whether, nearly one hundred years after the creation of a universal school medical service, we continue to need a health worker in schools. Two qualitative research studies carried out over the last four years by the Social Policy Research Unit include evidence relevant to this question, which is presented here. The first study, funded by the Department of Health and carried out between 1994 and 1996, investigated the role of the school nurse in meeting health needs potentially common to all school age children. The second study, funded by the NHS Executive's Mother and Child Health R&D Programme and carried out between 1996 and 1998, investigated the support needs of pupils in mainstream schools who have a chronic illness or physical disability. We obtained the views of a wide range of people on school nursing and/or on health needs at school: nurses and other health professionals; Trust managers; commissioners; Local Education Authority staff; and service users - that is, pupils, parents, Heads and teachers.

This paper examines the evidence about the current role of the school nurse and arguments for school as a setting for meeting health needs. It considers constraints on school nursing in meeting its potential in schools and suggests how these might be addressed by adopting a more strategic approach.

What is the role of the school nurse? Concerns over major health threats to the survival of children at the turn of the century have been replaced by other concerns, in particular emotional and behavioural difficulties, the impact of unhealthy lifestyles, and the needs of increasing numbers of children who now survive and manage a range of

chronic illnesses and disabilities. Against this background, what is the role of the school nurse?

Although practice varies, typically most of the school nurse's time is still spent on traditional routine surveillance activities and immunisations. The reason for a continued emphasis on surveillance appears to be historical patterns of contracting, rather than evidence of effectiveness: indeed, the type and frequency of surveillance activities varies considerably between Trusts. Child protection casework is part of the school nurse's role which is linked with surveillance. Aside from this contractual work, school nurses have a good deal of professional autonomy to develop their role, in negotiation with school staff. Although it follows that practice varies widely between individual nurses, it is possible to identify four key components of the school nurse role other than surveillance: health promotion (mainly health education rather than school-wide policy development); a pupils' confidante (a growing number of nurses offer confidential 'drop-in' sessions for pupils); family support; and an over-arching role as health adviser (to pupils, parents and school staff).

School as a setting for meeting health needs There are two broad arguments supporting school as a setting for a health worker. First, a growing number of pupils with a chronic illness or physical disability are now educated in a mainstream setting and may have special health needs requiring a response at school. Our research findings suggest that services may not be keeping pace with this growing area of need. In particular, pupils regarded teachers' understanding of their health condition *and* its impact on school life crucial for good support, yet teachers reported that their need for information and advice from health professionals remained largely unmet, despite some examples of good practice. Furthermore, responsibilities for medical care at school are still poorly defined.

A second argument for meeting health needs at school is where school offers advantages as a setting. Research points to three particular advantages: first, school brings together local populations of children, which is efficient for surveillance activities and offers scope (although currently under-developed) for population health needs assessment. Secondly, school is an educational environment, and so arguably an appropriate context for health education, which can benefit from input from health professionals. Thirdly, since most children spend their days there, school offers scope for easy access by children to a health professional in confidence.

Constraints on school nursing Although it appears that school nurses are developing their role in ways consistent with the advantages of the school setting, constraints are apparent. Since systematic local health needs assessment for school age children has generally been weak, contracts continue to favour traditional surveillance activity over other potentially beneficial ways of using resources for school nursing. Aside from surveillance, wide variation in practice between nurses means that potential service users (teachers, parents and pupils) report being unclear as to what the nurse might offer. It follows that best use may not

be made of school nursing services, particularly where these rely on self-referral, such as confidential 'drop-in' sessions. Lack of understanding on the part of school staff may also contribute to their limiting or denying access of a school nurse to pupils, since individual schools are 'gatekeepers' to allowing nurses to work on the premises. School nurses themselves report gaps in training to meet contemporary needs, for instance in teaching and counselling skills.

The need for a strategic approach The constraints on school nursing suggest that a more strategic approach is needed. Our research suggests a number of key components within such a strategy. Fundamentally, a systematic knowledge base about the health needs of children is required. Services appropriate for delivery in schools can then be established within this broader framework. In doing so, more knowledge is required about children's own views on their needs and on existing services. Children are service users in their own right and research shows that their views are not necessarily the same as adult 'proxies'.

A consensus on the contribution of school nursing to child health is needed, so as to clarify their role for potential users and to identify core knowledge and skills for training. Finally, any strategy for school health must be developed in partnership with colleagues in education. Although the 'gatekeeping' role of schools is one (negative) argument for doing so, a more positive side to partnership is to take into account the knowledge that school staff have about the health of their pupils and the active part they themselves play in meeting health needs through the curriculum, pastoral care, and medical tasks. School nursing will only ever play a part in meeting the health needs of children: the challenge is to identify and deliver this *distinctive* role.

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Commentary - Zarrina Kurtz (London) School nursing is a different sort of issue to the subject of the other topics covered in today's meeting, i.e. the effectiveness of various different services. The services covered relate to specific disease entities; school nursing is

a professional discipline or activity. School nurses, individually, work in a variety of different ways. The aims of any one service may be quite different from those of another. Before the effectiveness of school nursing can be evaluated, its role in children's health must be defined, and delineated from that of the school doctor, as well as other disciplines. My recent work has made me realise how important school nurses are, or could be, as part of Tier 1 services for children's mental health. But school nurses currently are inadequately trained for this work, and are poorly supported by specialist and other colleagues; furthermore, because they are so thin on the ground, they are not properly linked in to the primary care services network.

What children want above all from the school nurse is that she (or he) is a constant and available presence in the school. In very few situations can school nurses manage to fulfil this role. Many of the roles that they perform are also undertaken by others, such as health promotion, counselling, advocacy. For a number of these roles, it is questionable whether a nursing qualification is necessary.

There is a need for appropriately skilled health professionals - such as the school nurse - to work in a closely integrated way in the education system, to inform and link with the network of health promotion, primary care, and specialised services, such as Behaviour Support Teams, Youth Offending Teams, Drug Action Teams. Within primary care, the school nurse could take on a much greater role in screening, as, for example, in the model developed by the National Pyramid Trust* for emotional and behavioural problems.

*204 Church Road, London W7 3BP, telephone 0181 579 5108. One of several voluntary sector organisations that have sprung up around meeting children's needs, emotional needs, in school. The National Pyramid Trust have developed a model whereby school nurses would be taught the skills to screen for emotional and behavioural disorders in children - a whole new role for which they need particular skills. We need to know if school nurses want to do this work, what they find and whether it is a cost-effective service.

Commentary - Dr Sarah Stewart-Brown (Health Services Research Unit, Oxford): We reviewed the literature on the school entry health check. We found studies where they've simply counted how many children with "conditions" were found in different ways of doing medicals - all making the assumption that finding children helps them. None assessed what benefit accrues to these children from being found. Few studies defined what they were looking for or the severity. So, we don't know how valuable this is.

We hoped to find some studies on the impact of liaison between school nurses and teachers. For example, at the school entry health check, the nurse finds a child who has a particular problem and then goes and talks to the class teacher about how s/he might manage it. We also looked for studies on the effectiveness of health education undertaken by parents as part of the school entry health check. We couldn't find any.

My impression was that school nurses' work includes some work which somebody has told them needs to be done; for example, vision screening, hearing

screening, school entry, health checks. These are a hang-over from the time when it was important and statutory to do these things. And there is other work which they do because they feel it is helpful. This raises an important issue about the value of experiential knowledge in research. I have sympathy with school nurses who say "well why do we need a randomised control trial to know that this is valuable". You need trials because people fool themselves that what they're doing is useful - but its my hunch that people fool themselves less when they're doing something that they've discovered is useful for themselves. We need a debate about the value of people's knowledge and experience versus what's been discovered in trials, particularly when nothing much has been discovered in trials for school nursing.

Health promotion in schools has had a lot of bad press. We found that most programmes impact on health and health-related behaviour, but can't be shown to be doing it reliably, so something else is going on that we don't understand about the way these programmes are implemented. Successful interventions are likely to be multi-faceted - not just working in the classroom but working in the community or with parents, and whole school approaches like "health promoting schools" are more effective.

Nutrition and exercise programmes were the most effective in changing health and health related behaviour, but some of these programmes can be quite coercive. If you insist children go for cross-country runs, they will get fitter. If you only provide healthy food, then the children have to eat healthier food. But you're working in a coercive way, you're not offering choice. Many of us were forced to do physical education at school and gave it up as soon as we left! So although you can show a short term benefit in schools, do programmes that work in a coercive way actually have a long-term health gain?

I suggest that the optimal model for health promotion is personal development in the classroom, emotional awareness and understanding that contributes to mental health promotion; personal development in the staff-room, which enables teachers to listen and to treat children with more respect and not to use emotional violence as a system for keeping control in the classroom; changing the school ethos.