

and Baker provide a timely review, concluding that maternal transmission of scrapie and other spongiform encephalopathies is a myth.

There is no good evidence of maternal transmission in any of the human spongiform encephalopathies, in experimental spongiform encephalopathy in laboratory species, or in transmissible mink encephalopathy. The occurrence of bovine spongiform encephalopathy in cattle born after the feed ban in 1988 has led to speculation regarding maternal transmission of this disease, but epidemiological evidence provides little support for this hypothesis and concern about possible continuing contamination of cattle feed has led to new legislative measures. Maternal transmission in kudu, a breed of captive zoo ungulate, was originally suspected but is now uncertain.

Only with natural scrapie is there evidence of maternal transmission. However, Ridley and Baker question the evidence that shows infectivity in the placenta of sheep with scrapie, and they provide alternative interpretations of embryo transfer experiments and the important breeding experiments carried out by Dickinson and colleagues.¹ There are therefore cogent reasons for reconsidering the importance of maternal transmission in natural scrapie, although as Kimberlin has pointed out maternal transmission may be masked if there is a high level of lateral contagion.²

In scrapie, and other spongiform encephalopathies, the occurrence and phenotype of disease is influenced by host genetics and agent characteristics and by the level of infectious challenge and route of exposure. In scrapie, analysis of the prion protein (PrP) gene in sheep has shown that susceptibility to disease is linked to polymorphisms of the PrP gene. The genetics of scrapie susceptibility, summarised by Ridley and Baker, are complex, with the type of inheritance (dominant, partially dominant, or recessive) varying by breed of sheep. Crucially, there is also evidence of an interaction between host genotype and strain of scrapie agent,³ indicating that selective breeding for resistance to scrapie may be problematic and that

regarding scrapie as a simple genetic disease may be an oversimplification.

One of the peculiarities of the epidemiology of scrapie is that the large sheep population in New Zealand has been scrapie free for decades. If scrapie is primarily genetically determined, the current analysis of genotypes of sheep in New Zealand may provide important data—for example, it might be predicted that all tested sheep will be of “resistant” genotype. However, in 1979 Hourrigan and colleagues⁴ reported the development of scrapie in two out of 20 sheep imported from New Zealand following contact with flocks known to be affected by scrapie, and 39% of the progeny of the inbred New Zealand flock also developed scrapie.

Despite molecular biological research, the mechanism of transmission of natural scrapie remains unclear, and control of this important endemic disease cannot currently be based solely on genetic manipulation. Even if maternal transmission is a myth, the complexities of the interaction between host genetics and agent strain, together with the evidence for lateral transmission,^{1,2} indicate that eradication of scrapie may be difficult to achieve. One important implication of the paper by Ridley and Baker is that previous assumptions about scrapie may have to be re-evaluated, and further research on the epidemiology and transmission characteristics of the natural disease will be necessary if scrapie is to be eradicated. Even if the epidemic of bovine spongiform encephalopathy declines and disappears early in the next century as predicted, scrapie will remain as a potential source of animal zoonoses in many countries where it is endemic.

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Setting priorities for research and development in the NHS: a case study on the interface between primary and secondary care

Roger Jones, Tara Lamont, Andrew Haines

Since 1991 the NHS has attempted to identify and prioritise its needs for research and development in a systematic manner. This has not been done before and there is little evidence on which to draw. Multidisciplinary expert groups have identified priorities in different topics using explicit criteria and after widespread consultation within the NHS and research community to identify pressing problems and opportunities for research. This paper focuses on a review completed in 1993 to identify research and development priorities for the NHS in relation to the interface between primary and secondary care. The review covered several recent developments which require evaluation. The authors describe the process used to identify research and development priorities in this complex subject and examine the strengths and weaknesses of the approach. This case study should help to stimulate a wider debate on methods of identifying priorities, particularly those using participatory approaches, in research and non-research contexts.

Medical research has traditionally been investigator led, proposals for funding being judged largely on

scientific merit rather than in relation to health needs.¹ The NHS research and development programme is a new departure and the first attempt in Britain to establish a coherent research strategy to support the promotion of health and the provision of health care. The objective of the research and development strategy is to create a research based health service, in which reliable and relevant information is available for decisions on health policy, clinical practice, and the management of services. The broad scope and structure of the programme and the methods used to systematically review NHS activity were described in *Research for Health*.²

Setting research priorities

In an early attempt to identify a rational mechanism for setting research priorities Black and Pole devised five indices of disease burden and service activity to rank 54 disease categories and recommended that quantitative measures of burden should be used to identify funding priorities in biomedical research.³ Later Drummond *et al* applied retrospective cost-benefit analysis to decisions on research funding, using

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Summary points

- Research in the NHS has traditionally been "top down" and investigator led
- The NHS research and development strategy is the first systematic attempt to identify research priorities in relation to health need
- The NHS Central Research and Development Committee has set up a series of advisory groups to identify priorities for funding in key areas
- An advisory group met during 1993 to determine 21 priority topics for research and development funding in relation to the interface between primary and secondary care
- The process of priority setting enables the views of consumers as well as experts to be incorporated into a problem led approach to the identification of research and development priorities

the model of a clinical trial in diabetic retinopathy.⁴ They suggested that the cost effectiveness of different types of studies could be used prospectively to determine research priorities, though measuring benefit is difficult.

In 1989 Feachem and colleagues proposed a framework for setting research priorities in developing countries.⁵ They identified conditions that needed to be met for a health problem to generate research, including ignorance about its epidemiology, costs and consequences, and the effectiveness of interventions. Other workers have emphasised the importance of a participatory approach for setting priorities in research (as in health care),⁶ embracing the views of researchers, policy makers, funding agencies, service users, and practitioners.^{7,8}

Criteria of research and development potential

The process for setting research and development priorities within broad themes identified by the NHS Central Research and Development Committee has been developed and refined by successive groups. We report and reflect on the method of working of the fourth advisory group (set up to identify research and development priorities at the interface between primary and secondary care), which reported at the end of 1993.⁹ The following criteria for setting priorities were agreed by the advisory group. The criteria related to needs of the NHS and to research and development potential.

Criteria related to NHS need were likely benefit of research to the NHS and patient care; relevance to policy initiatives such as the *Health of the Nation* and the patient's charter; burden of disease; costs to the service and to patients; and practice variation, areas of large variation being given greater priority.

Criteria related to research and development potential were feasibility of research, including availability of existing methodology and resources, and the likelihood of research being implemented—that is, the degree of management commitment to the issue, study design, and participants.

These criteria were developed from the work of previous advisory groups and relevant publications and were distinguished by the incorporation of feasibility and implementation and by including practice variation as a proxy measure of uncertainty (and hence indicator of research need).

Primary-secondary care interface

The interface between primary and secondary care was selected as an important issue for identifying research and development need by the central research and development committee for several reasons. Probably the gatekeeper role of general practitioners

has contributed to comparatively low levels of expenditure on the NHS in comparison with health care systems abroad. The effectiveness of this system relies to a great extent on what happens at the interface between primary and secondary care. The balance of care between sectors has shifted in Britain and elsewhere; economic analyses in Britain and other European countries indicate reductions in the proportion of total health expenditure on inpatient care over the past decade.¹⁰ The primary-secondary care interface has also been highlighted because of concern about variations in referral rates by general practitioners to specialist care and the appropriate use of specialist services,¹¹ and also because of the development of new methods of commissioning and contracting which are likely to influence the type and quality of care provided in general practice.¹²

A series of policy initiatives also focuses on the relation between primary and secondary care. The document *Health of the Nation* sets targets for reductions of mortality and morbidity which will clearly require coordination of effort between primary and secondary care.¹³ The Tomlinson report and the government response *Making London Better*¹⁴ have emphasised the need to develop and enhance primary and community health services in the capital, with proposals to divert resources from the hospital sector. The patient's charter¹⁵ and related initiatives aim at making the NHS more responsive to users and imply the provision of a coordinated primary, secondary, tertiary, and community service appropriate to patients' needs, location, and circumstances.

Setting up the advisory group

The advisory group was set up in March 1993 and included interests from both primary and secondary care sectors and from a range of disciplines, including nursing and medicine (generalist and specialist), management (purchasers and providers), research, and consumer concerns. The group eventually consisted of 16 members as well as observers and secretariat support.

Panel groups

The advisory group formed three panels which considered entry to secondary care, exit from secondary care, and shifts in the balance of care. Box 1 shows the scope of these different panels. Panels were given the task of reviewing existing evidence, considering responses to consultation, and identifying the key issues within each of these three areas to forward to the advisory group.

Identifying problems

Professors David Wilkin and Martin Roland (Centre for Primary Care Research, University of Manchester) provided a critical overview of the evidence and suggested the framework for the three panels. The advisory group also needed to obtain the views of those working in and using the service on the most pressing problems which could be addressed by research. A total of 242 organisations were consulted in April 1993, including NHS staff (through regional directors of research and development), statutory agencies, professional bodies, consumer groups, academic centres, and research organisations. Evidence was received from 138 respondents.

In addition, workshops were convened in London, Leicester, and Newcastle to generate informal discussion of problems at the interface. A further workshop was held at the Department of Health to explore relevant policy issues. A small group of

Box 1—Panel structure

- *Entry to secondary care*
Encompasses different pathways by which patients enter secondary care for investigation, diagnosis, and treatment. This includes referral by general practitioners and other staff in the primary care sector (including dentists, optometrists, health visitors, and physiotherapists). Patients may also refer themselves for specialist treatment by accident and emergency departments of direct access clinics. Many issues centre on defining the appropriateness of a referral
- *Exit from secondary care*
Patients leaving hospital may receive care from the primary health care team, community health services, outpatient departments, family, and friends. Other agencies include social services and voluntary support networks. Key questions are centred on length of stay and follow up arrangements, particularly for groups such as elderly people, psychiatric patients, and those with physical disabilities
- *Shifts in balance of care*
Recent developments have shifted the balance of care along the continuum of primary and secondary care. They include different models of integrated care, shared care schemes (for instance, in diabetes and asthma), specialist outreach services, intermediate hospital care, and forms of substitution for hospital care (such as near patient testing in general practice). There is currently little evidence on the cost effectiveness of different patterns of care

informal carers also met to advise the advisory group on some of the issues facing individual users of the service. Further research topics were suggested by Wilkin and Roland's overview. In addition, advisory group members themselves nominated topics for the panels to consider.

Translating problems into subjects of research need

The letter of invitation asked for simple statements from those closest to the most pressing issues to be addressed, rather than fully worked up research proposals. The task of the panels was to translate these raw statements of need, totalling some 800 suggestions, into subjects suitable for research and to consider them in relation to the relevant available research evidence. A structured summary of the consultation material was prepared by the secretariat, and the panels used this to identify a series of provisional priorities, which were forwarded to the advisory group. Each priority was accompanied by a detailed, referenced supporting statement justifying the importance of the topic to the NHS to inform the prioritising of topics by the advisory group.

Identifying priorities

Twenty five topics were forwarded by the three panels to the advisory group. These were discussed and a master list of 21 agreed for scoring. The criteria of NHS need and research and development potential were agreed by the advisory group early in its deliberations. These were used as indicators of the likely research impact of a given topic. The group used a simple five point global scale based on these criteria. Experience from previous groups suggested that results of a global rating correlated well with individual

ratings against each of the criteria. Scores were entered by using an electronic system which highlighted variation in the group. Topics of low agreement were discussed and rescored, and mean scores were used to produce a list of topics in priority order (box 2).

These 21 priority topics were ratified by the central research and development committee in October 1993. The report of the advisory group was widely disseminated in the NHS and elsewhere as a policy document in spring 1994.⁹ All those responding to the consultation exercise received a copy of the report. Work is now being taken forward in these priority areas, with the establishment of a commissioning group and a full time programme manager responsible for commissioning a national programme of research and development on the interface between primary and secondary care.

Discussion

The process of setting priorities against agreed criteria is a rational response when competing demands are made on limited resources. The central research and development committee advisory group was given the task of identifying research and development priorities for the NHS related to the interface between primary and secondary care. This exercise attempted to identify priorities for research and development systematically after widespread consultation on the needs of the NHS. This "raw" information was then translated into topics suitable for research and relative priorities identified by means of an expert multi-

Box 2—Twenty one priority areas for research and development related to interface between primary and secondary care

Top priorities

- Transfer of information across interface between health care professionals and other agencies
- Evaluation of clinical guidelines at the interface
- Appropriate access, use, and location of diagnostic facilities and new technologies
- Impact on referrals and discharge of including patients and carers in decision making
- Appropriateness of outpatient follow up
- Evaluation of treatment by referral versus management in primary care
- Impact of purchasing arrangements on interface
- Aftercare: rehabilitation and community care for priority groups
- Prescribing across the interface
- Models of intermediate care

Further priorities

- Evaluation of specialist outreach schemes
- Patients' and carers' social needs
- Evaluation of shared care schemes
- Implications of day case surgery
- Changing skills and training requirements at the interface
- Implications of shorter length of hospital stay
- Characteristics of primary health care team and access to specialist care
- Availability and patterns of secondary care services affecting entry
- Effectiveness of inpatient discharge procedures
- Evaluation of first contact care provided outside general practice
- Relations of patterns of referral to health need

disciplinary group. The review was completed in six months and the time constraints may have affected the quality of the process, if not the outcome, of setting priorities systematically.

One problem for the advisory group was identifying priorities in a new and changing field. Defining the interface between primary and secondary care was itself no easy task, and the group spent much time mapping this complex subject. However, the fact that this was relatively uncharted territory was in many respects an advantage in drawing up a research agenda which was genuinely multidisciplinary and focused on the needs of the service. It would probably have been more difficult to obtain consensus in a topic which was more specialty bound.

There were, however, problems in identifying priorities by a multidisciplinary group. Despite the wide range of interests and experience in the group, coverage of different forms of expertise was not exhaustive and there was often difficulty in establishing the present state of research knowledge in a given aspect. Wilkin and Roland provided a broad overview, but decisions about particular topics were not supported by further systematic reviews of the evidence. Members and others had to rely on individual or shared awareness of published and current research. Indeed, a wider problem faced by the group was the difficulties of distinguishing between the importance of a topic (in terms of disease and cost burden) and the feasibility of research. Different kinds of expertise were needed to define the magnitude and nature of the problem and identify the research objectives which could be achieved. Identifying the appropriate balance of skills and experience while keeping the group to a manageable size was not easy.

CONSUMER INPUT

The group attempted to balance the need for professional expertise with lay input. However, the question of consumer input was particularly difficult. A separate consumer consultation exercise, in the form of a focus group of informal carers, was convened as one attempt to sample the views of ordinary users as opposed to the "professional" consumers who responded to the formal consultation exercise. This input was helpful to the advisory group as a prompt to consider the needs of individual users when setting priorities. However, it was not clear how generalisable were the findings of one small focus group.

The participation of consumers raises wider questions about participation and a problem of exercise. A key feature of the NHS research and development programme is the attempt to identify the needs of the NHS by widespread consultation. Individuals and organisations approached during the written consultation and workshops include those using services and other consumer groups, those providing care (including professional organisations), those managing services, and members of the research community. The case for involving stakeholders in defining a research agenda is based on both a theoretical notion of the democratic right of participation in public decision making and the belief that this process leads to better decisions which are closer to "real" research needs. However, the mechanism for involving consumers needs to be carefully thought through if it is to be more than a legitimising activity,¹⁶ and the central research and development committee is planning to set up a group to advise on and monitor consumer participation in the NHS research and development programme.

SOURCES AND VALUES OF RESEARCH EVIDENCE

One rationale for attempting a "bottom up" identification of need has to do with ownership of research

results and the belief that the resulting research is more likely to be accepted and taken up in the health service. Much expectation and awareness of what research could deliver on this subject was generated by consultation, particularly the regional workshops. Further methods are needed to develop a dialogue between the stakeholders and those carrying out research to ensure that research findings achieve a better uptake in the service.

Tensions remain in trying to reconcile a participative approach with the "top down" advice of an expert group. Were ideas generated by consultation exercises that would not have originated from an expert advisory group alone? How does an expert group balance "technical" information on burden and cost with the perceived need identified by consultation responses? How comprehensive is the group's awareness and use of research evidence? The potential tension between "investigator led" research and "problem led" research has once again been highlighted in the recent House of Lords select committee report on research in the NHS.¹⁷

This was a genuine attempt to identify the needs of the NHS for research and development in an area where clear answers are needed to inform purchasers and providers of the service, but the accommodation and weighting of different sources of evidence and the values attached to them still need to be more explicit. Careful evaluation of the process of setting priorities and the costs and benefits of such an exercise is also needed. An audit trail would be helpful to trace the path of suggestions made from consultation, through translation into topics suitable for research by the advisory group, to the decisions by those funding individual proposals in the priority areas. The final outcome is for research and development to contribute to improved patient care and cost effectiveness of services.¹⁸ The work of this advisory group cannot yet be fully assessed and will depend on the ultimate impact of the research programme on health services and patient outcomes.

The opinions expressed in this paper are our own and do not necessarily represent the views or policy of the Department of Health.

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Commissioning a national programme of research and development on the interface between primary and secondary care

Janet Wisely, Andrew Haines

The first call for applications to the NHS research and development programme on the interface between primary and secondary care was advertised in February 1994. A total of 674 outline proposals were submitted and 54 (8%) secured funding. Projects have been commissioned in 16 of the 21 priority areas and around £6m has been committed. Analysis shows that multidisciplinary applications are more likely to be funded and that the odds for a successful application are on average nearly doubled for each discipline represented up to five. A survey of applicants and peer reviewers found satisfaction with much of the commissioning process, but peer review and feedback were subject to criticism, particularly by unsuccessful applicants. The programme shows that it is possible to commission a large number of projects in an innovative area of research and development and has identified refinements that will further increase the efficiency and acceptability of the process.

The priority setting process for the national programme of research on the interface between primary and secondary care is described in the accompanying article (p 1076).¹ This paper details the commissioning process. We believe that the process should be as "transparent" as possible and that there should be opportunity for comment and constructive criticism to help refine the process.

Box 1—Rating system used by external reviewers to assess proposals

- 0—Unsupportable
- 1—Low priority for shortlisting
- 2—Good: could be shortlisted
- 3—Very good: recommend shortlisting
- 4—Exceptionally high standard: strongly recommend shortlisting

Commissioning process

Responsibility for the national programme of research and development on the interface between primary and secondary care was given to the research and development directorate at North East Thames Regional Health Authority (now NHS Executive North Thames). A commissioning group chaired by Professor Michael Clarke was established to advise on the programme. The commissioning process (figure) followed a two stage format established by previous NHS programmes.

CALL FOR RESEARCH PROPOSALS

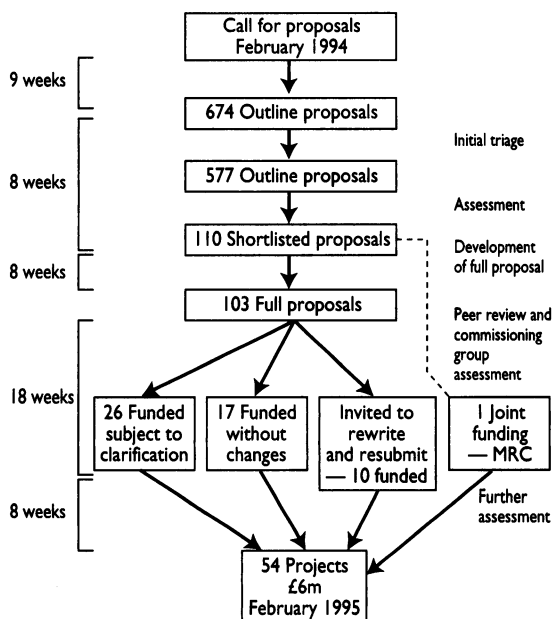
In February 1994 an advertisement was published in the national press, in health journals, and by the regional research and development directorate networks. Four page outline proposals were requested and the desirability of multidisciplinary applications mentioned. A total of 1455 requests for further information were made, and the application pack included a brief description of potential subjects for research within each priority sector.²

SHORTLISTING PROCESS

In total, 674 outline proposals were submitted. Initial triage identified 97 that clearly did not address a priority area, and these were excluded. The remaining 577 were then reviewed by a member of the commissioning group and an external reviewer. Reviewers, who were not allocated proposals from their own institutions, were asked to award each proposal a rating from 0 ("unsupportable") to 4 ("exceptionally high standard: strongly recommend shortlisting") (box 1). When awarding ratings the reviewers took into account the priorities of the programme and considered nine criteria (box 2).

Even weighting was given to each priority in the shortlisting process. The proposals were grouped according to their overall rating—namely, high (≥ 6 ; 68 proposals (12%)); borderline/mixed (5, 4 (4-0), (3-1), and 3 (3-0)); 104 proposals (18%); and low (4 (2-2), 3 (2-1), 2, 1, and 0; 405 proposals (70%)).

The commissioning group focused on the borderline and mixed proposals, and each was discussed in turn.



Outcome of commissioning process to February 1995 (one funded project subsequently withdrawn—see text)

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