applied widely we assumed that regional clinical genetics departments and those who care for people with cystic fibrosis, including general practitioners, would cooperate. We are confident that our counselling and that which individuals and couples would receive in other clinical genetics departments in Britain would be the same, and we are therefore happy to recommend widespread application of screening.

We accept that cascade screening will detect only about a quarter of carrier couples, as D J H Brock points out.² These "islands" of carriers and carrier couples are, however, easily accessible as a person with cystic fibrosis or a carrier is at the islands' centre and knowledge of the disorder is high among the people concerned. Whatever screening programmes health authorities introduce, cascade screening should be the starting point until the public starts to request population screening programmes, as Sandy Raeburn suggests as the ideal.³ The uptake of prenatal diagnosis per detected carrier couple will naturally be higher in Brock's screening programme than ours if Brock starts by offering the test in pregnancy. Most of the women in the carrier couples were not pregnant when our screening detected them; when pregnancy occurred only one declined tests. We do not think that the "sick family" syndrome is likely with the commonest autosomal recessive disorder, as Nadeem Quereshi claims.⁴ It is easy to reassure carriers who are detected that everyone carries a few recessive genes.

No one (except perhaps Marteau) believes that relatives should not be offered carrier screening. The only question that then remains is whether promotion of an active testing programme in relatives infringes their basic right not to know whether they are a carrier; we submit that it does not. Ideally we would like tests to be available for those who ask for them, whether relatives or not, with active promotion no longer necessary.⁵ This will be possible only when public awareness, even among relatives, is far greater than it is now. We hope that more people will now enter this debate.

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Screening in primary care is preferable

EDITOR,—We agree with the stance taken by Julian D A Cooper and Antony J Franks and by Nadeem Quereshi in response to the debate about population screening for carriers of the cystic fibrosis gene.^{1,2} We found in a survey that the general public is interested in becoming involved with the issues and showed a clear preference for screening to be offered through primary care.³

When we offered carrier testing with other health promotion through a prepregnancy care clinic in one general practice, although attendance

was low (about 1% of patients of reproductive age), interest in screening was high among participants (16/18 (89%)) and to date 10 of the 18 participants (55%) have been tested. A feature of this model was the variable length of time between learning about carrier screening and actually requesting the test—several months in some cases. The preconception approach is complementary to screening in pregnancy and will overlap with it. Although uptake may be small, assimilation of the implications of the test will be enhanced in the antenatal period.

Quereshi notes the need for adequate professional education if screening were to take in primary care.² With general practices increasingly finding it necessary to focus on activities that generate the highest income in the short term, there is little motivation to undertake many aspects of preventive health. So long as screening for genetic disorders retains its current low profile with purchasing authorities, efforts by specialist genetic centres to provide the background training and support for primary care will be wasted. We recently surveyed the 12 general practices in north Newcastle for their reaction to a proposal to develop genetic services in the community with the offer of appropriate staff support in a collaborative pilot project. Only two practices were clearly in favour. Most of the others were not in favour on the grounds of lack of time and adequate reimbursement rather than disagreement with the principles entailed.

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Audit improves neonatal (Guthrie) screening programme

EDITOR,—We conducted an audit of the neonatal screening programme for phenylketonuria and congenital hypothyroidism in the Northern region in 1993. Like Allison Streetly and colleagues, who looked at screening in south London,¹ we found that arrangements for monitoring the existing screening programmes were inadequate, though we do not have London's problems regarding mobility of families and we have a smaller proportion of people from ethnic groups than the London areas.¹ We found that in five of the districts no satisfactory mechanisms existed for checking that each baby had been screened and that in five of the 11 districts that make checks an inappropriate delay (several weeks) in checking could occur. Only six of the 16 districts had a timely failsafe mechanism in place for ensuring that all babies were screened. In addition only one district was confident that its coverage was 100%; other districts did not routinely monitor their coverage.

After the audit, information was fed back to each district with suggestions on how the screening programme could be improved. Six months later all districts except two had a timely failsafe mechanism for identifying babies who had not been screened. We agree with Streetly and colleagues that monitoring of coverage is essential and that explicit standards need to be set.¹ In our experience, audit has been a useful mechanism for reviewing the neonatal screening programme and has resulted in changes in most districts to improve the monitoring of the programme.

The audit was carried out with a grant from Northern Regional Health Authority.

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Two tier system for outpatient treatment

EDITOR,—For the second time a fundholding practice has referred a patient to me with a request for a consultation only. I understand that this means that the practice pays about £30 for the privilege of seeing me, but I am not allowed to do any investigations or obtain any x ray films. I am allowed only to see the patient and express an opinion.

Where does responsibility for the patient lie? If the problem is, say, backache, the x ray films are either inadequate or inappropriate, and I am concerned that there is a tumour and believe that further investigations are required, what do I do? The current system seems to be that I write to the general practitioner and pass the ball back into his or her court. If the general practitioner decides to take no action where does the responsibility lie? If the general practitioner decides that further investigations are required the patient has yet another trip to hospital. If the general practitioner does not act on the consultant's advice and a problem arises the consultant, in my view, has failed in his or her duty to the patient.

There already seems to be a two tier system for inpatient treatment and there may well be one for outpatient treatment too, which would operate to the financial advantage of fundholding general practitioners but, I suspect, to the disadvantage of patients.

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Waiting times in an ophthalmic outpatient department

EDITOR,—A M Ansons and colleagues describe some of the problems that a busy academic eye department is experiencing in meeting the patient's charter.¹ At around the time of the charter's introduction we conducted a survey of ophthalmic outpatient services in a district general hospital with three consultants and serving 250 000 people. Information was collected by six management students, who obtained details of 393 (95%) of the 414 patients attending clinics during one week.

Overall, 116 patients were seen by a consultant, 58 by the registrar, 190 by one of the three senior house officers, and 50 by one of the general practitioner clinical assistants. The clinics were usually fully booked but had an additional daily workload of between one and 15 unbooked patients (referrals from the accident and emergency department and urgent referrals from general practitioners) added to them. The senior house officers ran a daily casualty clinic but sought advice from a consultant or the registrar concerning 79 patients. Sixty six patients arrived later than their booked appointment (usually because of transport problems) and had to be fitted into the clinic out of

sequence. While 104 consultations lasted five minutes or less, 88 lasted over 20 minutes. The number of patients seen within half an hour of their booked appointment time, without correction for other factors, was 228 (55%).

The survey found hospital practices that caused delays; these have now been altered. Five of the 15 clinics started more than 10 minutes late because medical staff were completing ward rounds or attending lunchtime meetings. The computer booking system was programmed to book both old and new patients at five minute intervals.

As patients and their relatives have to wait in the outpatient department the facilities provided should be reasonable; 108 patients found the seats uncomfortable, 112 did not know where the toilets were located, and 50 did not know where to obtain refreshments. Surprisingly, only four patients complained specifically about waiting times. Altogether 277 patients were over retirement age, and their apparent lack of concern about waiting may reflect an older generation's acceptance of queuing and the fact that most were not taking time off work. Also, 201 were seen by nursing staff before their medical consultation in order to check visual acuity, dilate pupils, or test visual fields. They therefore probably considered that they received attention without much delay.

We agree with Ansons and colleagues that the current targets for waiting times are unrealistic for many busy eye clinics.

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Increasing the accessibility of data

EDITOR,—George Davey Smith's editorial on increasing the accessibility of data is too optimistic about the power of sharing data to arrive at "truth." Sharing data can be an impediment to truth. That happens when people start arguing in such detail about the "right" analysis of data that outsiders give up and conclude that there is no evidence.

An epidemiologist once told me that he would never share his data with an opponent since that person would almost certainly find some decision concerning the analysis of the data or some misclassified subject to decry in public—preferably to a mass media audience. The recent slandering of careers that has occurred as a result of some errors in data from clinical trials—which randomised trials tolerate—serves as a good reminder, even if the origin of the errors cannot be excused.² A notorious case in which not only truth and careers but also public health suffered was the reanalysis and reinterpretation of data from the first case-control study of Reye's syndrome and salicylates. It destroyed the study's credibility in the eyes of many. Only many years and many patients later did new and costly studies show that the original study had been right.³ It is no solution to say that the intention of a reanalysis should not be irresponsible or malevolent, because to the true Popperian the intention of an argument is not important, only the argument itself counts.

Davey Smith concedes that in observational research the ability to reach different conclusions based on the same data is recognised.⁴ Randomised trials do not, however, escape subjectivity. It suffices to read the literature published after the trial of global utilisation of streptokinase and tissue plasminogen activator for occluded coronary arteries (GUSTO) to see that even members of the same trial board, who had access to the same data, can greatly differ over their interpretation. This is not new in medicine and epidemiology. Some useful lessons about analysis of data were learnt from the protracted discussion about the university group diabetes programme trial: repeat analyses of data and discussions took more than a decade, only to arrive at deadlock.⁵

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Research into outcomes and effectiveness

EDITOR,—Trevor Sheldon's critique of the research into the outcomes and effectiveness of treatment carried out by the US Agency for Health Care Policy and Research and, in particular, the patient outcomes research teams, which are supported by the agency, is based on a fundamental misunderstanding of our programme, mission, and methods.¹ The US Congress created the Agency for Health Care Policy and Research in December 1989 to focus on a set of important issues that were not being addressed by existing research institutions. Specifically, the law establishing the agency and the medical treatment effectiveness programme mandates a new, hard look at the effectiveness and appropriateness of current health care practice in a wide array of clinical conditions. It requires examination of a broad range of outcomes (not merely mortality and morbidity but relief of symptoms, functional status, quality of life, and cost) as achieved in non-selected patients and practices.

Many of the agency's studies of outcomes focus on conditions and procedures that are not life threatening but that impact appreciably on the quality of life—that is, low back pain, cataract, benign prostatic hyperplasia, and hysterectomy. Because many of these conditions and procedures have not been studied before, the agency's first generation of studies invested heavily in fundamentals—documenting practice patterns, showing the need for better evidence, identifying the relevant outcomes, and developing research methods and measures of outcomes. These building blocks are important in their own right, and collectively they have led directly to the development of a set of rigorous studies to test hypotheses.

For example, work by the patient outcomes research team on prostatic diseases was critical in showing the weakness of evidence supporting current treatment for benign prostatic hyperplasia and in convincing the clinical and research communities of the need for a randomised trial. The team developed measures of the symptoms and treatment outcomes specific to the condition that are important to patients and a symptom scale for benign prostatic hyperplasia that has been

validated and adopted by both the American Urological Association and the World Health Organisation. In collaboration with the American Urological Association the team has designed and obtained funding for a randomised controlled trial that will compare directly the effectiveness of transurethral resection with two drugs (finasteride and doxazosin). Similarly, the randomised controlled trials in prostatic cancer, back pain, and cataract that are now under way in the United States, as well as a case-control study by the patient outcomes research team for cataract, would never have been undertaken without the preliminary analyses by the teams.

Systematic, critical reviews of the literature on their topics is another important contribution of patient outcomes research teams. The conclusions of these reviews, while often disappointing with respect to the quantity and quality of the literature, provide valuable documentation on the state of the evidence regarding the effectiveness of different treatments and have led to some fundamental changes in thinking about what is critical to study. Proponents of randomised controlled trials and traditional meta-analysis who have criticised attempts by the patient outcomes research teams to synthesise the findings of non-randomised studies should appreciate the contribution of the teams in documenting the non-existence of randomised controlled trials and, in some cases, the poor quality of trials that have been done.

In clinical areas where solid evidence of the benefit of treatment exists, investigators supported by the Agency for Health Care Policy and Research are determining whether these benefits are routinely achieved. The patient outcomes research team for acute myocardial infarction has shown that a significant number of patients eligible for thrombolysis do not receive it.² The agency's mandate in research into outcomes does not stop with the publication of a study showing effectiveness but requires analyses of practice to determine whether effective treatments are used routinely. If they are not, strategies must be developed to change practice.

For many clinical conditions, when multiple outcomes and alternative treatment strategies are considered, research (even randomised controlled trials) can seldom identify a single, "correct" decision regarding treatment. At best the evidence to guide decisions will be in the form of a set of probabilities associated with the various risks and benefits. Large simple trials offer little help, because the outcomes that need to be measured are seldom simple.

Unlike death, relief of knee pain or changes in sexual function, visual function, or affect are difficult to measure, and information has to be obtained from patients—a process that is neither simple nor inexpensive. In the absence of clear answers about best treatment the preferences of informed patients must play a major part in clinical decisions. Patient outcomes research teams have drawn attention to the critical dimension of patients' preferences and have shown the capacity of the teams' approach to obtain good information about these.

While we certainly do not believe that randomised controlled trials are a panacea, neither do we propose observational studies as a substitute for experimental methods. Observational methods can help to define the critical constructs and the hypotheses that need to be tested experimentally and determine whether practice patterns are consistent with evidence of effectiveness.

Experimental studies certainly have a place in research into outcomes too. This is because research into outcomes is not, as Sheldon suggests, a method; it is a conceptual framework for studying the relations between health care services and their outcomes. It uses multiple research designs and methods drawn from the full array of epidemiological and clinical research.

As a new theoretical and methodological enter-