

Unhelpful Counsel?

MERGE's response to the Chief Medical Officer's Working Group report on CFS/ME

April 2002

“Brutalised by their reception in doctors’ examining rooms, they ceased consulting doctors, preferring instead to wait out their disease away from the medical profession’s unhelpful counsel.”

Hillary Johnson, Osler’s Web

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Preface by The Countess of Mar

This document has been prepared by MERGE in response to the great interest awakened by the Working Group's report to the Chief Medical Officer on CFS/ME in January 2002. The CMO's report has been widely seen as a positive step for ME patients in terms of the recognition of the illness and the need for the provision of medical and social care. Yet, there are several aspects of the report which have been of concern, not only for ME patients and their carers, but also for independent professionals with an interest in social and medical policy. Following several representations and after careful consideration, a decision was made by MERGE to prepare a response highlighting these concerns while pointing out the positive aspects of the report. At the back of our minds was the Joint Royal Colleges Report of 1996, a controversial document which went formally unchallenged though criticism was voiced in several quarters at the time. Given this example, it is important for MERGE to put on record in a formal document - if only for historical reasons - several of the key issues surrounding the deliberations and production of the Working Group's report.

As patron of MERGE, it is a pleasure for me to endorse this analysis of the report on CFS/ME to the CMO. I hope that it gives voice and some hope to many ME patients, some of whom have mixed feelings about the recommendations in the report and others who are frankly antagonistic to the underlying psychological philosophy that coloured the deliberations of the Working Group.

MERGE's report formed a necessary background to my statement on CFS/ME in the House of Lords on Tuesday 16th April 2002. In this statement, I pointed out that despite the fine aspirations in the report, its effect might be to "compound inaction, ignorance and even denial: inaction in not investigating the patient's illness or not providing any treatment - management is not the same as treatment; ignorance by promoting inappropriate and possibly harmful interventions; and denial of the true nature of ME." A contributor to the ensuing debate pointed out that in the three months since publication there "appears to have been a deafening silence... I remind the Minister that that work was described as "urgent" by the Working Group."

I sincerely hope that MERGE's analysis will help to reawaken interest in the research and treatment of this disabling illness, and that the professional and wider communities will at last come to understand just how disabling this illness is. Whilst treatment and cure might still be distant dreams for ME patients, I hope there will be a rapid sea change in the public perception of ME, and that there will be encouragement and support rather than scorn and derision for ME sufferers.



The Countess of Mar

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Executive Summary

The report of the Chief Medical Officer's Working Group on CFS/ME of January 2002 is an advance on the widely-criticised Joint Royal Colleges report on CFS (1996). It gives an authoritative statement that CFS/ME is a genuine illness which imposes a substantial burden on the health of the UK population, and stresses that improvement of health and social care for people affected by the condition is an urgent challenge. Importantly, it states that CFS/ME can be clinically recognised for treatment purposes; lists the initial battery of tests that should be performed; and is clear that inaction by healthcare professionals due to ignorance or denial of the condition is not excusable. These, and many more, positive aspects of the report represent progress in terms of recognition of the illness and its consequences. However, for a variety of reasons, the report is inadequate.

Most importantly, the narrowness of the remit, concerned primarily with *providing best practice guidance on the healthcare management* of the illness, has ensured that the focus is on containment and coping, rather than on solving the clinical conundrum, namely, what actually *causes* the illness and what steps can be taken to elucidate the cause. The constraint of the remit has several important consequences, some of which are discussed below.

It has permitted the Working Group to side-step the central issue which energises every discussion about CFS/ME. This concerns the diagnostic construct 'CFS' which most probably includes heterogeneous patient groups (one of them with classically-defined ME), limiting the generalisability of any specific management strategy or therapeutic intervention. The question, "what's in a name?", has particular poignancy in the case of this illness: one increasingly plausible answer is "everything" since *de facto* misdiagnosis not only complicates the interpretation of clinical trial evidence, but obscures treatment options and, in the long run, influences management and practice for the worse. Given that there is a growing number of experts who consider that there is a strong case for unpacking the term 'CFS' and reclassifying and renaming in accordance with more specific clinical criteria, a opportunity for the Working Group to seriously address this issue has been lost.

As the Working Group was concerned with therapeutic management, it sought evidence from a systematic review of forty-four randomised clinical trials on a range of clinical interventions. Two clinical "therapeutic interventions" were identified, cognitive behavioural therapy and graded exercise therapy. By conventional standards of literature reviewing, formal evidence for the use of either in a general patient population is rudimentary, a fact indicated by the authors of the original review. The specific efficacy of neither is convincingly supported by the systematic review evidence, and nor is the pragmatic efficacy supported by survey reports from CFS/ME patients. As regards cognitive behavioural therapy, five interpretable trials were identified (three "positive" and two "negative"), a less than conclusive evidence base for an intervention which is also non-curative, expensive, beyond the resources of Health Authorities to fund given the scale of the problem, and has been found helpful by only a small minority of patients surveyed by patient organisations. For graded exercise therapy, only three positive clinical trials were identified, none with a fully comparable control group and all consisting of patients classified by the Oxford criteria which does not diagnose 'ME' or 'CFS (1994)' exclusively. Again, the true usefulness of this therapy to the general population of patients is unclear given that the effectiveness of such motivational interventions is by no means established; that deconditioning may not, in fact, be an important factor limiting the activities of many CFS/ME patients; and that around half of patients surveyed indicated that graded exercise therapy actually worsened their condition. The third recommended intervention, pacing, is intuitively sensible but hardly warrants the status of a separate therapy within a healthcare management program.

Despite part of the remit “to make recommendations for further research,” the Working Group has dealt with the research findings in 639 words out of a total of some 34,600 in the main report. A large body of research literature on CFS/ME exists, however, and numerous biological abnormalities have been reported, although the aetiology of the illness remains elusive. By systematically assessing the significance of these, the Working Group may have been able to recommend a specific direction for future research. Instead, the message presented to the media, the public, and opinion formers is that the best that can be done is to manage symptoms, most prominently with psychological strategies. Interestingly, given the volume of published research evidence on pathophysiological mechanisms, the range of clinical signs and symptoms exhibited by CFS/ME patients, and a large body of professional opinion supporting a ‘biomedical’ model of the illness, it seems negligent that clinical opinion on the Working Group was proportionately over-represented by professionals who tend to subscribe to the use of biopsychosocial interventions for CFS/ME. Since four of their number resigned from the Key Group shortly before publication, after several years of participation, on the grounds that the report paid too little attention to biopsychosocial aspects of the illness, a question is raised about the role of the initial composition of the Working Group in colouring the final report.

At several points, the report mentions the problems of the most severely-ill patients. Yet, a database of information collected and analysed on behalf of severely-ill sufferers by The 25% ME Group, presented to the Working Group, was not used to full advantage in the final report, though other patient data was produced. MERGE’s summary of this data shows that 25% of these patients described themselves as bedridden, and 57% had been either housebound or bedridden for more than six years, illustrating in numbers rather than words that morbidity in CFS/ME can be substantial, despite the opinion of many healthcare professionals to the contrary. The management strategies recommended by the Working Group are inappropriate for this group of sufferers whose care remains a neglected challenge.

As regards social care, the Working Group set out to consider how the NHS might best provide care for sufferers. Yet, the NHS is only one agency among many providing care, and the responsibilities of other agencies involved in both care planning and direct service provision could have been usefully identified. Overall, the comforting statements from the Working Group about the services CFS/ME sufferers *should* receive in the community are little more than aspirations: without the full support and practical backing of local social work departments, sufferers will struggle to see their needs met either fully or appropriately.

Importantly, the report does not describe *how* or *when* change is to occur. The Working Party had no executive power and brought no additional funding to stimulate change. Even in its advisory role, it does not evaluate the cost implications of its recommendations, call for directives to be issued to the various health agencies or professional bodies, or propose any mechanism for checking that changes will be made. Crucially, it gives no indication that the illness will be looked at again, in the medium to long term, to assess whether real, meaningful change has come about.

In summary, while the Working Group’s report may go some way towards improving recognition of the illness, MERGE considers that it has avoided serious consideration of the important issues surrounding the diagnosis and treatment of ME/CFS; that it has given undue emphasis to management strategies of limited applicability; that practical recommendations for social care provision are lacking; and that, consequently, an opportunity to effect real change has been lost.

1. The report - background and content

1.1 Background to the report

In 1998, after much debate, the Working Group on CFS/ME was established to “review the practical care and support for patients, carers, and health care professionals alike.” Its brief was to “review management and practice in the field of CFS/ME with the aim of providing best practice guidance for professionals, patients, and carers to improve the quality of care and treatment for people with CFS/ME.” In particular, the Group aimed to:

- develop good clinical practice guidance on the healthcare management of CFS/ME for NHS professionals, using best available evidence,
- make recommendations for further research into the care and treatment of people with CFS/ME,
- identify areas which might require further work, and make recommendations to CMO.

Evidence and opinions were sought from many quarters, and a systematic search of the international evidence on CFS/ME commissioned. Unlike previous reports, the focus was “to provide *advice on clinical management*,” in accord with the NHS Plan of July 2000 which strives to increase the involvement of public and patients by promoting self management, improved information to and communication with patients, and greater choice of healthcare options to support the concept of individualised care.

The Working Group consisted of three groups: the Key Group which was responsible for surveying the evidence, developing the main report, and agreeing the final recommendations to the CMO; the Children’s and Young People’s Group; and the Reference Group which had an *ad hoc* advisory/consultative role, and whose members had a wide range of expertise and opinion.

1.2 Content of the report

The report, published in January 2002, consists of six chapters, of which chapters 2 to 5 form the policy-making information. The original “key message” summary of each chapter are given in the boxes below.

Chapter 2 of the report summarises the patient evidence presented to the Working Group. It covers the general themes of recognition, diagnosis, acknowledgement, and acceptance of CFS/ME by the public and healthcare professionals. It reports patient concerns about the need for information, and the need for professional education about the condition among healthcare professionals, both in the primary and further care sectors. It discusses the special needs and problems of people who are severely affected, of children and young people, and of carers:

Chapter 2 - Evidence from patients: Key messages

- Patients’ voices are not being listened to and understood.
- People affected by CFS/ME indicated improvements needed in three main areas: recognition, diagnosis, acceptance, and acknowledgement; healthcare service provision; care of groups with special circumstances.
- Patients reported the need for more healthcare professionals who know about and understand CFS/ME. Public awareness campaigns, professional education, and information for patients and carers are accorded high priority.
- Experiences of primary care are polarised. Positive experiences are characterised by: willingness of clinicians to treat the patient as an equal, supportive attitudes, belief in the patient’s experiences, and early recognition and diagnosis.
- Experiences of further care are predominantly negative. Needs identified include access to specialists and respite-care services.
- Those severely affected by CFS/ME (up to 25% of patients) feel “severely overlooked” by services. They experience isolation, lack of understanding, and particular barriers to accessing all forms of care.
- Children and young people are profoundly affected by public and professional uncertainties over the illness. Young people also suffer from impact on their families and from lack of support and expertise within the education system.
- Individuals with CFS/ME from disadvantaged class or ethnic groups face special difficulties, yet they are under-represented in research.
- Carers, particularly of young people, need more recognition, support, and respite.

“Patients are not being listened to or understood. Those severely affected feel isolated and overlooked.”

Chapter 3 describes the nature and impact of CFS/ME. It outlines the decision to use the term ‘CFS/ME’ in the report, and summarises what is known about the aetiology, pathogenesis, and disease associations; predisposing factors; triggers; maintaining factors; and possible disease mechanisms. It discusses the spectrum of illness, subgroups, symptom profiles, severity, and the socioeconomic impact of the illness:

Chapter 3 - Nature and impact of CFS/ME: Key messages

- CFS/ME is a relatively common condition of adults and children that is clinically heterogeneous and lacks specific disease markers, but is clinically recognisable.
- The broader impact of the disease, even in its milder forms, can be extensive; people who are severely affected and/or with long-standing disease are profoundly compromised, and improvement of their care is an urgent challenge.
- The aetiology (cause) of CFS/ME is unclear, although several predisposing factors, disease triggers, and maintaining factors have been identified.
- The pathogenesis (disease process) underlying CFS/ME is also unclear. Research has demonstrated immune, endocrine, musculoskeletal, and neurological abnormalities, which could be either part of the primary disease process or secondary consequences.
- One highly heterogeneous disease might exist that encompasses CFS/ME, or several related pathophysiological entities may exist; these distinct hypotheses should be studied.
- Current evidence does not allow complete distinction between CFS and ME, or useful delineation of subgroups. Every patient's experience is unique, and the illness should be managed individually and flexibly.

Chapter 4 puts forward suggestions for the management of CFS/ME, based on recognition, acknowledgement, and acceptance of the condition by healthcare professionals. It suggests approaches to patient management, diagnosis, and clinical evaluation. It stresses the need for information and support, and for systems to be put in place to facilitate ongoing care. Controversially, on the basis of the York Review and clinical experience, it identifies graded exercise, cognitive behavioural therapy, and pacing as interventions that might be useful for patients. It also discusses models for improved service provision:

Chapter 4 - Management of CFS/ME: Key messages

- Initial professional responses to CFS/ME can have major impact on the patient and carers. Clinicians should listen to, understand, and help those affected to cope with the uncertainty surrounding the illness.
- Early recognition with an authoritative, positive diagnosis is key to improving outcomes. Symptoms are diverse, but increased activity

frequently worsens fatigue, malaise, and other symptoms with a characteristically delayed impact.

- All patients need appropriate clinical evaluation and follow-up, ideally by a multidisciplinary team. Care is ideally delivered according to an agreed flexible management plan, tailored from a generically applicable range of options.
- Therapeutic strategies that can enable improvement include graded exercise/activity programmes, cognitive behaviour therapy and pacing; intrusive symptoms and co-morbid conditions may also require specific management.
- The overall aim of management must be to optimise all aspects of care that could contribute to any natural recovery process. Management strategies need regular review to guide their application and adaptation to the individual.
- Education and support, plus measures to tackle the broader impact of the disease, need to be initiated as early as practicable. Much support is provided by the voluntary sector. Patients can be empowered to act as partners in care.
- Review of the evidence highlights the lack of good quality research to support effectiveness of various therapies. Patient responses suggest that no approach is universally beneficial and that all can cause harm if applied incorrectly.
- The goal of rehabilitation or re-enablement will often be adjustment to the illness; improvement is possible with treatment in the majority of people.

“Early recognition with an authoritative, positive diagnosis is key.”

Chapter 5 focuses on the nature and impact of CFS/ME in children and young people, its clinical profile, social impact, and management. It also discusses the importance of education and child protection, and the impact on family/carers:

Chapter 5 - Children and young people: Key messages

- CFS/ME represents a substantial problem in the young - “children do get it,” though many recover, even after prolonged illness.
- Important differences exist between children and adults in the nature and impact of the disease and its management.

- The condition potentially threatens physical, emotional, and intellectual development of children and young people, and can disrupt education, and social and family life at a particularly vulnerable time of life.
- Clinicians face additional difficulties in supporting and managing the younger patients and their families and parents/carers.
- An especially prompt and authoritative diagnosis is needed in the young, while the possibility of other illnesses and complications must also remain in mind.
- Ideal management is patient-centred, community-based, multidisciplinary, and coordinated, with regular follow-up. Community paediatric services need to be available for most children, and for all with prolonged school absence.
- The clinician who coordinates care needs to consider educational needs and impact on the family and parents/carers as early as practicable.
- Care is best delivered according to a specific, flexible, patient-focused treatment plan, designed and reviewed regularly with patient and family.
- Future services need to be developed around the needs of the child or young person and their family.

Chapter 6 presents the recommendations of the Working Group, dealing with recognition and definition of the illness, treatment and care, health service planning, education and awareness, and research:

Chapter 6 - Recommendations of the Working Group

CFS/ME is a relatively common clinical condition, which can cause profound, often prolonged, illness and disability, and can have a very substantial impact on the individual and the family. It affects all age groups, including children.

The Working Group has encountered extensive evidence on the extent of distress and disability that this condition causes to patients, carers, and families. It has examined the evidence on the effectiveness of interventions used in the management of this condition. The Working Group is concerned about several issues. Patients and carers often encounter a lack of understanding from healthcare professionals. This lack seems to be associated with inadequate awareness and understanding of the illness among many health professionals and in the wider public. Many patients complain of the difficulty of obtaining a diagnosis in a timely manner. There is evidence of underprovision of treatment and care, with patchy

and inconsistent service delivery and planning across the country.

Finally, there is a paucity of good research evidence and very little research investment for a serious clinical problem that in likelihood has a pervasive impact on the individual and the community. Insufficient attention has been paid to differential outcomes and treatment responses in children and young adults, the severely affected, cultural, ethnic and social class groupings. The Working Group has identified measures that should be taken with some urgency to address the current situation.”

6.1 Recognition and definition of the illness

- The NHS and healthcare professionals should recognise CFS/ME as a chronic illness that, despite uncertain aetiology, can affect people of all ages to varying degrees, and in many cases substantially.
- In view of current dissatisfaction among some groups over the nomenclature applied to this illness, we recommend that the terminology should be reviewed, in concert with other international work on this topic.

6.2 Treatment and care

- Patients of all ages with CFS/ME must receive care and treatment commensurate with their health needs and the disability resulting from the illness.
- Healthcare professionals should have sufficient awareness, understanding, and knowledge of the illness to enable them to recognise, assess, manage, and support the patient with CFS/ME. Healthcare workers who feel they need extra skills should seek and receive help from those experienced in this area.
- General Practitioners should usually be able to manage most cases in the community setting, but must be able to refer patients for specialist opinion and advice where appropriate (e.g. because of complexity in diagnosis and treatment).
- CFS/ME of any severity in a child or young person – defined as of school age – is best coordinated by an appropriate specialist – usually a paediatrician or sometimes a child psychiatrist – in concert with the GP and a paediatric or CAMHS multidisciplinary team.
- Sufficient tertiary level specialists in CFS/ME should be available to advise and support colleagues in primary and secondary care.
- Management should be undertaken as a partnership with the patient, should be adapted to their needs and circumstances, and should be applied flexibly in the light of their clinical course.

- The support of the patient with CFS/ME and the management of the illness should usually extend to the patient's carers and family.
- Clinicians must give appropriate and clear advice, based on best national guidance, on the nature and impact of the illness to those involved in providing or assessing the patient's employment, education (primary, secondary, tertiary, and adult), social care, housing, benefits, insurance, and pensions.

6.3 Health service planning

- Service networks should be established to support patients in the primary care and community setting, to access when necessary the skills, experience, and resources of secondary and tertiary centres, incorporating the principles of stepped care. Services should be configured so that individual professionals and aspects of the service can meet individual needs, particularly in the transition from childhood to adult life.
- Health service commissioning through primary care organisations, supported by health authorities or wider consortia, must ensure that local provision for these patients is explicitly planned and properly resourced, and that health professionals are aware of the structure and locale of provision. Health commissioners should be requested to take immediate steps to identify the current level of service provision for CFS/ME patients within their locality.
- Each Strategic Health Authority should make provision for secondary and tertiary care for people with CFS/ME, based on an estimated annual prevalence rate of approximately 4,000 cases per million population in the absence of more refined data.
- People who are so severely affected that their disability renders them housebound or bed-bound have particular constraints in regard to their access to care. These specific needs must be met through appropriate domiciliary services.
- The NHS should make use of the wide range of support and resources available through partnership arrangements with voluntary agencies, enabling suitable self-management by the patient.

6.4 Education and awareness

- The education and training of doctors, nurses, and other healthcare professionals should include CFS/ME, as an example of the wider impact of chronic illness on the patient, on carers and family, and on many aspects of society.
- Healthcare professionals, especially in primary care and medical specialities, should receive

postgraduate education and training so that they can contribute appropriately and effectively to the management of patients with CFS/ME of all ages.

- GPs and medical specialists should consider CFS/ME as a differential diagnosis in appropriate patients, and should at least be able to offer initial basic guidance after diagnosing this condition (Annexes 6 and 7).
- Awareness and understanding of the illness needs to be increased among the general public, and through schools, the media, employers, agencies, and government departments.

6.5 Research

A programme of research on all aspects of CFS/ME is required. Government investment in research on CFS/ME should encompass health-services research, epidemiology, behavioural and social science, clinical research and trials, and basic science. In particular, research is urgently needed to:

- Elucidate the aetiology and pathogenesis of CFS/ME,
- Clarify its epidemiology and natural history,
- Characterise its spectrum and/or subgroups (including age-related subgroups),
- Assess a wide range of potential therapeutic interventions including symptom control measures,
- Define appropriate outcome measures for clinical and research purposes, and
- Investigate the effectiveness and cost-effectiveness of different models of care.

The research programme should include a mix of commissioned or directed research alongside sufficient resource allocation for investigator-generated studies on the condition.

The report also contains seven annexes containing evidence presented to the committee. These comprise:

1. Epidemiology
2. Prognosis
3. Patient evidence
4. General concepts and philosophy of disease
5. Management of CFS/ME - research evidence
6. Management of CFS/ME - report summary
7. Management of CFS/ME - children and young persons' summary.

Annexes 1 to 5 have not been published but are available at www.doh.gov.uk/cmo/publications.htm. Annexes 6 and 7 have been published as a separate document.

2. Positive aspects of the report

Since the publication of the heavily-criticised 1996 Joint Royal Colleges report on CFS, there has been a black hole in the professional and public recognition of the illness. The CMO report advances current thinking in several key aspects.

2.1 Recognition of CFS/ME as an illness

The report gives an authoritative statement that CFS/ME is a real illness which requires professional help, and has particular problems owing to the controversy which surrounds it. It consolidates the acceptance of the disorder previously expressed by the Departments of Health and Social Security and by the BMA as long ago as 1988:

“CFS/ME is a genuine illness and imposes a substantial burden on the health of the UK population. Improvement of health and social care for people affected by the condition is an urgent challenge.” (1.0)

“Patients, their carers, and healthcare professionals encounter different levels and varying manifestations of disbelief and prejudice against people affected by the condition.” (1.0)

“CFS/ME is a genuine illness. It should cease to be a waste bucket for heart-sink patients”

2.2 Recognition that CFS/ME can be clinically diagnosed

The report makes it clear that CFS/ME can be clinically recognised for management purposes, and lists in Annex 6 the clinical assessment investigations that should be performed. These include:

- Full clinical history
- Physical examination
- Mental health evaluation
- Sleep evaluation
- Basic screening tests, which can involve
 - Full blood count
 - C-reactive protein (CRP) concentration
 - Blood biochemistry tests including concentrations of creatinine, urea, electrolytes, calcium, phosphate, glucose, liver enzymes, and markers of thyroid function
- Simple urine analysis
- Other tests determined by history or examination

The diagnosis is based on the characteristic pattern of symptoms, once alternative diagnoses are excluded.

“Although the disorder is clinically recognisable, CFS/ME assumes many different clinical forms and is highly variable in severity and duration, but lacks specific disease markers.” (3.0)

2.3 Importance of a positive diagnosis

The report stresses that CFS/ME should cease to be a waste bucket for ‘heart-sink’ patients.

“A diagnosis of CFS/ME relies on the presence of a set of characteristic symptoms together with the exclusion of alternative diagnoses.” (4.2.1.1)

“A positive diagnosis of CFS/ME is needed, rather than one of exclusion. Without a validated test for the illness, diagnosis is based on recognition of the typical symptom pattern together with exclusion of alternative conditions. Thus, a positive diagnosis can usually be made from clinical history, examination, and a few appropriate laboratory investigations, as in other chronic illnesses of uncertain nature.” (4.2.1)

2.4 Need for action

The clear emphasis on the need for health and social care provision in CFS/ME is perhaps the most important thrust of the report.

“Appropriate management and service provision for patients with CFS/ME and their carers are urgent priorities.” (4.0)

In the view of the report, ‘action’ includes further research, especially concerning the severely affected.

“We suggest that the prevalence and impact of severe disease, the pathways to chronicity and to becoming severely affected, and strategies that would benefit such individuals urgently need further study.” (4.4.1)

2.5 Estimation of prevalence and service need

Given the wide variation in estimates of prevalence – partly a function of differences in case definition – the report comes up with a sensible estimate:

“On the basis of a reasonable estimate of adult population prevalence of 0.4%, a general practice with a population of 10,000 patients is likely to have 30 to 40 patients with CFS/ME, about half of whom may need input from services. The proportion of the latter patients who are severely affected by the disease is thought to be up to 25%.” (4.5.1)

2.6 Suggestions for best practice

At present, there are no guidelines on what clinicians should *do* with CFS/ME patients. The report has attempted to fill this gap.

“The incremental development of a locally based service, including provision of domiciliary care for severely affected patients, would significantly improve care for all patients with CFS/ME, but especially for this most disadvantaged of patient groups. The general components of such a service are: medical care, support for adjustment and coping, facilities for energy/activity management, and nursing and personal care.” (4.5.2)

2.7 Instruction to clinicians on patient management

In contrast with sensational media reports about the benefits of cognitive behavioural therapy and graded exercise, the report is clear about the limitations of current management strategies:

“No management approach to CFS/ME has been found universally beneficial, and none can be considered a cure.” (4.1.2)

Chapter 4 of the report details the general principles and some specific advice for management of the condition by GPs and healthcare professionals. Its key messages are important and are worth restating:

- Initial professional responses to CFS/ME can have major impact on the patient and carers. Clinicians should listen to, understand, and help those affected to cope with the uncertainty surrounding the illness.
- Early recognition with an authoritative, positive diagnosis is key to improving outcomes. Symptoms are diverse, but increased activity invariably worsens fatigue, malaise, and other symptoms with a characteristically delayed impact.
- All patients need appropriate clinical evaluation and follow-up, ideally by a multidisciplinary team... The overall aim of management must be to optimise all aspects of care that could contribute to any natural recovery process... Patients can be empowered to act as partners in care.
- Although care packages need to be individually tailored, where appropriate they should include visits from primary care teams, and assessment and provision of equipment practical assistance. (4.1.2)

2.8 Description of CFS/ME in children

The section of the report dealing with children and young people with CFS/ME is particularly well-written. There is a clear description of the impact of the illness on the child, the family, and the community. The development of an

integrated and multidisciplinary package of services is recommended as a matter of urgency, and the statement of rights is particularly welcome:

“Children’s rights are safeguarded by UN convention and need to be respected at all times by professionals and parents/carers. The rights to be heard, to have their views taken into account, to access quality medical treatment, and to be protected from abuse both by individuals and by systems need particular attention.” (5.0)

2.9 Recommendations to healthcare professionals about benefit provision

The report encourages healthcare professionals to be sensitive about their role as facilitators of welfare provisions:

“Negotiations with insurance companies and the Department of Social Security about proportional and rehabilitation benefits and therapeutic work can improve outcomes, and health professionals have an important role to play by providing support and advice in these negotiations. The same level of understanding needs to be shown by medical advisors to insurance companies and the Benefits Agency about the condition, its natural course, prognosis, and range of available approaches to recovery.” (4.4.5)

“It is not appropriate that participation in a particular treatment regimen is made an absolute condition for continuation of sickness/disability payments.” (4.4.2)

“No management approach has been found universally beneficial.”

2.10 Appropriate attitude for healthcare professionals

There are also some clear warnings for healthcare professionals:

“Healthcare professionals should adopt an understanding attitude and should not get into disputes with patients about what to call the illness, or about the belief that it doesn’t exist.” (4.1.1)

“Treatment should always be a collaboration between the patient and the clinician, and not something imposed. Good communication and a good therapeutic relationship can make an appreciable difference.” (4.4)

“...our conclusion is that clinicians need to apply current knowledge despite the remaining uncertainty [about disease cause or process]; *inaction due to ignorance or denial of the condition is not excusable*.” (4.1)

“CFS/ME should be treated in the same way as any other chronic illness of unknown aetiology. The aim is to develop a supportive relationship, and provide information and education to assist the patient, families, and carers towards self management with support.” (4.1.2)

“All interventions need to be administered with thought and care, and in accordance with revised Department of Health recommendations on informed consent.” (4.4.2)

2.11 Importance of patient consent for management strategies

The need for the active consent of patients to therapeutic interventions is stressed at various points in the report.

“The decision to recommend a particular approach is best guided by the individual’s illness and circumstances.”

“The content and development of any such approach should be mutually agreed by both clinician and patient and informed by up-to-date specialist knowledge.”

“It is not appropriate that participation in a particular treatment regimen is made an absolute condition for continuation of sickness/disability payments.” (4.4.2)

“Management strategies supervised by a therapist, including activity management, cognitive behavioural therapy, and so on, can be beneficial, *provided that they are agreed* and viewed as a partnership.” (4.1.2)

2.12 Needs of the severest sufferers

One welcome note in the report is the recognition of the particular needs of severest sufferers. While the Working Group chose not to highlight valid data collected by the 25% ME group, which represents the severest sufferers, in the final report, it nevertheless seems to have recognised the plight of these patients.

“A minority of those with CFS/ME remain permanently severely disabled and dependent on others... Current provision of services falls well below what is needed for the vast majority of severely and very severely affected patients... Yet, even if we lack easy solutions, professionals can still support, care, and provide for many patients’ needs by reaching such patients in their homes, maintaining contact, and continually exploring potential options.” (3.4.3.1)

“In general, this group is excluded from research, so they may not fulfil criteria used to test evidence-based approaches. For example, many comment on the inappropriateness of extreme exercise regimens that have been studied in less adversely affected patients... Care is an urgent challenge that must be addressed in appropriate and imaginative ways, drawing from service models applied to other severe chronic disabilities.” (4.4.1)

“The Working Group is concerned that it is necessary to make these points [about severity and its consequences] for CFS/ME, when such considerations are self-evident and part of usual clinical practice for other disorders that are better recognised.” (3.4.3)

“In many chronic illnesses, daily functioning, including mobility, cooking, cleaning, dressing, personal care, and social support, can be improved dramatically by sympathetic provision of appropriate practical assistance.” (4.3.3)

“Inaction due to ignorance or denial is not excusable.”

2.13 Voicing of patient and carer concerns

Use of patient voice throughout the report strengthens the narrative. They give voice to the patient concerns, and - importantly - justification to the antipathy towards health professionals felt by many people with CFS/ME. The chief points arising from the patient voices are poor recognition of CFS/ME or by professionals, difficulties that arise over diagnosis, and lack of professional and public acceptance and acknowledgement.

“Participants felt that the widespread lack of understanding of the condition is not specific to clinicians but includes other healthcare and social care professionals. This lack of knowledge was identified by the majority of those consulted, together with a lack of communication and advice, especially in the early stages, on how to cope in general with long-term illness for families and sufferers.” (2.2.2)

“There is evidence that some patients ‘fight’ for referrals, and in general GPs are confused over where to refer patients... The overall experience of specialist and hospital services among participants was predominantly negative... Some patients find themselves in geographical ‘black holes’ that lack specialist provision.” (2.2.4)

“Severely ill are severely overlooked; just ignored and invisible... Some report that they want to believe doctors and feel ‘frightened to say no’ or that they do not have the energy to disagree. Fears were also expressed over: being ‘branded’ as a ‘difficult patient’, losing benefits, letting people down, not trying, losing the love of the family, and being labelled as mentally ill.” (2.3.1.1)

“Some carers were clearly distressed about being ignored by GPs, and some reported unpleasant behaviour.” (2.3.4)

“A proportion of patients feel alienated from clinical professionals by early responses to their symptoms, illness experience, and disability.” (3.5.2)

“People with CFS/ME frequently experience problems with accessing state benefits.” (3.5.1)

2.14 Summary

The positive aspects of the report listed above represent an advance in terms of recognition of the illness and its consequences. Sufferers and their carers can now state, not only that CFS/ME is a genuine illness which can be clinically diagnosed, but that the elements of best practice and management have been sketched out and that inaction by healthcare professionals due to ignorance or denial of the condition is not excusable.

However, the report does not describe *how* or *when* change is to occur. It does not describe the cost implications, it does not call for directives to be issued to the various health agencies or professional bodies, and it does not propose any mechanism for checking that changes will be made. Crucially, it gives no indication that the illness will be looked at again in the medium to long term, to assess whether real, meaningful change has come about.

3. Limitations of the report

The positive aspects of the report listed in Section 2 concern the recognition of ME and the need for illness management in a variety of forms. However, the report has limitations. Some, such as the constrained remit, are obvious, but others are apparent only to those *au fait* with the issues, whether the research evidence or the deliberations of the Working Party.

3.1 Constrained remit

The report describes its own remit clearly:

“To review management and practice in the field of CFS/ME with the aim of providing best practice guidance for professionals, patients, and carers, to improve the quality of care and treatment for people with CFS/ME, in particular to: develop good clinical practice guidance on the healthcare management of CFS/ME for NHS professionals, using best available evidence; make recommendations for further research into the care and treatment of people with CFS/ME; identify areas which might require further work and make recommendations to CMO.” (1.1)

Given the controversy surrounding the illness, this remit seems primarily designed to contain and manage the clinical problem. By concentrating principally on management and ‘guidance’, the report has ensured that the focus is on containment and coping, rather than on addressing the clinical conundrum of causation. Though the cause of CFS/ME has yet to be elucidated, it is important to consider the various possibilities at length within the context of management options. The narrow focus also neatly sidesteps the problem of the preference of some clinicians for the umbrella term ‘CFS’, obscuring specific diagnosis and possibly, in the long run, influencing management and practice for the worse.

The result is that the Working Group has taken three years to uncover the obvious - that, for a variety of reasons, the available ‘management strategies’ are cognitive behavioural therapy and graded exercise therapy, both with a very weak and rudimentary evidence base (Whiting *et al.* 2001), and ‘pacing’, which is little more than a commonsense approach to physiological limitation. Welcome though recognition of the illness is, we should not forget that for patients and carers little if anything has changed, or probably will change, as a consequence of developing ‘best practice guidance’.

3.2 Unbalanced composition of the Working Group

The final Working Group consisted of Professor Allen Hutchinson (Chair), Dept. of Public Health, Sheffield; Professor Anthony J Pinching (Deputy Chair), Dept. of Human Science and Medical Ethics, St Bartholomew’s Hospital, London; Dr Tim Chambers (Chair of the Children’s sub-group), Southmead Hospital, Bristol; and

three groups – the Key Group (responsible for surveying the evidence, developing the main report, and agreeing the final recommendations to the CMO), the Children’s and Young People’s Group, and the Reference Group (with an *ad hoc* advisory/consultative role). The Report also concedes the input of un-named others not included in the above groups. The breakdown of the Working Group by professional interest is shown in Table 1 below. Due to a misprint in the final CMO report (page 72), the members of the Children’s Group are designated as “Key Group Observers”, though the existence of the Children’s Group is mentioned at several points in the text. Table 1, however, shows the composition of both groups.

Table 1. Composition of the Working Group - excluding the Chair and Deputy Chair - by professional interest at September 2001

	Key group	Children's group
Patient representatives	3 (2*)	1
Carers	1	2
Representatives of ME associations	3	4
GPs	1	0
Psychiatrists/psychologists	3 (3*)	1 (1*)
Public health specialists	2 (1*)	0
Paediatricians	0	3
Nurses	0	1
Council/NHS	0	1

(*In parentheses are the numbers of this professional interest group who refused to endorse the final Working Group report in January 2002. Four of these were professionals with an affiliation to psychiatry/psychology, one was a consultant in Public Health Medicine, and two were patient representatives.)

Given the range of clinical signs and symptoms exhibited by CFS/ME patients, the volume of published research evidence on pathophysiological mechanisms, and a large body of professional opinion supporting a ‘biomedical’ model of the illness, it seems negligent that three of the clinicians on the Key Group should have been psychiatrists/psychologists and that four should have been co-authors of scientific papers supporting the use of biopsychosocial interventions for CFS/ME. Interestingly, the Deputy Chair of the Working Group in a recent paper (Pinching, 2000)

advocated the use of the management strategies - cognitive behavioural therapy and graded exercise - finally identified as the therapeutic interventions of choice by the Working Group. The composition of the Children's Group was less skewed towards the psychiatric or the psychological, both of which are generally recognised to be far less appropriate models in children.

In total, six members (46%) of the Key Group refused to endorse the final report, a remarkable attrition rate for a Working Group reporting to the Chief Medical Officer of England. The four professional resigners from the Key Group argued that the report paid too little attention to biopsychosocial aspects (Clark *et al*, 2002). In Hospital Doctor, 17th Jan 2002, one of them, Dr Alison Round, was reported as saying that the report neglected the "biopsychosocial" aspects of the illness. Another, Dr Peter White, was reported to say, "All the evidence taken together suggests that the condition is biopsychosocial - both physical and mental factors are involved." This kerfuffle has not been universally welcomed: in a recent debate on CFS/ME in the House of Lords, Lord Clement-Jones said, "Some recent articles written by doctors in the wake of the report are absolutely disgraceful and ignorant. I feel strongly about some of those reactive reports" (Hansard, 2002). Patients and carers can only speculate on the kind of report that might have emerged (and the different emphases that might have been placed on psychological strategies) if these professionals had 'resigned' at the beginning of the process rather than at the end. As a patient said wistfully, "After all that... it's like cuckoos leaving their trademarks but not their signatures."

3.3 Problem of diagnosis and use of the composite term CFS/ME

Terminology is *the* 'hot' issue in ME and CFS: it energises the debate between patients and healthcare professionals, particularly psychiatrists. It also impacts on patient management and clinical practice since the results of clinical trials are determined by entrance criteria used to recruit patients to them.

The issue can be simply put. The original case description of the illness, 'ME' (Acheson, 1959; Dowsett *et al*, 1990) described a condition, commonly of infectious onset, characterised by:

- Exercise-induced fatigue precipitated by trivial exertion (physical or mental).
- Neurological disturbance, especially of cognitive, autonomic, and sensory functions. This could include impairment of short-term memory and loss of powers of concentration, usually coupled with emotional lability, nominal dysphasia, disturbed sleep patterns, dysequilibrium and/or tinnitus.

- An extended and relapsing course with fluctuation of symptoms, usually precipitated by either physical or mental exercise; typically, the symptoms vary capriciously from hour-to-hour and day-to-day with varying involvement of the cardiac, gastro-intestinal, and lymphoid systems.

Since the late 1980s, however, the medical profession has been urged by some of its members to adopt the term Chronic Fatigue Syndrome (CFS), a more wide-ranging diagnostic category which includes patients whose dominant symptom is medically unexplained, on-going, or chronic fatigue (in conjunction with several other physical or psychological symptoms) who would not necessarily fulfil the criteria for ME.

There are now several definitions of CFS. In the USA, the 1994 CDC case-definition of CFS is currently utilised (Fukuda *et al*, 1994), supplanting its predecessor, the 1988 CDC criteria, and has similarities with - but is not identical to - the classical description of ME. However, in the UK, a frequently-used case definition is the 'Oxford criteria' which includes patients with no physical signs and inadvertently selects subgroups of patients with high levels of psychiatric diagnoses (Katon & Russo, 1992; Freiberg, 1999). These definitions have been used to recruit to randomised clinical trials, including some of the trials of 'psychological' interventions, cognitive behavioural therapy, and graded exercise therapy, which form the basis of the management strategies uncovered by the Working Group report. Since the adoption of a particular case-definition of CFS will greatly influence the outcome of particular studies, it is perhaps no surprise that psychiatric research groups researching biopsychosocial strategies in these patients should find some encouraging results. However, as many patients and carers in CFS/ME support groups in the UK invariably point out:

- Fatigue is not their primary problem: musculoskeletal pain and post-exertional myalgia along with other physical signs are far more prominent, corresponding more closely to the classical definition of ME.
- The World Health Organisation International Classification of Diseases (ICD) has, since 1969, classified ME separately as a neurological problem (ICD 10 93.3), with 'CFS' incorporated into the current ICD as a sometime synonym for ME. The chronic fatigue states *per se* are listed under mental and behavioural disorders (F 48.0), a category which specifically excludes ME/PVFS/CFS.

For these reasons, many CFS/ME patients - particularly the most severely affected - resent being provided with non-curative coping strategies, such as cognitive behavioural therapy, by healthcare workers who have no interest in their particular symptom complex. In this, they are supported

by a growing number of experts who consider that there is a strong, perhaps overwhelming, case for unpacking the term 'CFS' and reclassifying and renaming in accordance with more specific clinical criteria (De Becker *et al*, 2001; Tan *et al*, 2002), such as the criteria for ME described above. The report alludes to the problem:

“The issue of subgroups or discrete entities within CFS/ME was the subject of much debate by the Working Group. We are conscious that some sectors strongly hold the view that the term ME defines a subgroup within CFS, or even a distinct condition. The Working Group accepts that some patients' presentation and symptoms align more closely to the original clinical description of ME.” (3.4.1)

To complete its task, the Working Group side-stepped the issue:

“We recognise that no current terminology is satisfactory, so *in line with our original terms of reference* [MERGE emphasis] we have used the composite CFS/ME for the purposes of this report, acknowledging that CFS is widely used among clinicians and ME among patients and the community.” (3.2)

“For how much longer will anomalies in nomenclature complicate and obscure clinical care?”

The issues surrounding the establishment of CFS as a diagnostic category, and the inaccurate and biased characterisations of CFS that have subsequently arisen, have been well reviewed by Jason *et al* (1997):

“Over the past ten years, a series of key decisions were made concerning the criteria for CFS diagnosis and the selection of psychiatric instruments, which scored CFS symptoms as medical or psychiatric problems. At least some of these decisions may have been formulated within a societal and political context in which CFS was assumed to be a psychologically determined problem (Manu *et al*. 1988). Many physicians and researchers believed that CFS was similar to neurasthenia and that CFS would eventually have a similar fate once people recognised that most patients with this disease were really suffering from a psychiatric illness. Psychiatrists and physicians have also regarded fatigue as one of the least important of presenting symptoms (Lewis & Wessely, 1992). These biases have been filtered to the media, which has portrayed CFS in simplistic and stereotypic ways... One major consequence is that many CFS patients feel dissatisfied with their medical care... and have gone outside traditional medicine to be treated for their illness...”

“A significant complicating factor in understanding the dynamics of this illness is that there are probably different types of illnesses now contained within the CFS construct... We believe that it is crucial for CFS research to move beyond fuzzy recapitulations of the neurasthenia concept and clearly delineate precise criteria for diagnosing pure CFS and CFS that is comorbid with psychiatric disorders. It is also necessary to better differentiate CFS from other disorders which share some CFS symptoms but are not true CFS cases.”

One of the most poignant sentences in the report is:

“The severely ill reflected strong loathing of the name CFS because fatigue is often not perceived to be their main problem; ME is a preferred term by many.” (2.3.1.1)

For these and other patients, the question is how much longer anomalies in nomenclature will be allowed to complicate and obscure clinical care. Given that the term 'CFS' most probably groups different kinds of patients under one umbrella, management recommendations are likely to be inadequate and probably misleading.

3.4 Choice and interpretation of best management strategies

It is important to realise that the Working Group was empowered to identify evidence for “management strategies” not treatments, since it is clear that none of the forty-four randomised clinical trials found and reviewed (Whiting *et al*, 2001) supplies convincing evidence of treatment efficacy for a specific symptom or condition. At first sight, the process of identification of 'useful' management strategies appears clear:

“Where research evidence exists we have been guided by it. (1.0) ...We used a trident approach to review and synthesise three lines of evidence: research findings, patient reports, and clinical opinion... Members of the Working Group expressed widely differing opinions on the potential benefits and disadvantages of these approaches. However, we agreed that all could be considered as management options in line with general principles outlined here... The Working Group agreed that there is no cure for CFS/ME but identified three specific strategies as potentially beneficial in modifying the illness: graded exercise, cognitive behavioural therapy, and pacing.” (4.4.2)

Though the report contains several caveats about all three 'management strategies' - perhaps as a sop to the non-biopsychosocial opinion on the committee (i.e., patients and carers) - it is the choice of these which, in the end, provides justification for the existence of the Working Group and the money spent (including that provided by the Linbury Trust) on the CMO report. Yet how each of

the three strategies was determined to be “potentially beneficial” is not as clear as it might appear. As regards the published evidence, the thorough review by Whiting *et al* (2001) state that it is very difficult to draw overall conclusions (from the forty-four randomised clinical trials) since very little information is available on baseline functioning. Most of the interventions were evaluated in only one or two studies, so the validity of generalising the findings is limited. Since there are few patient reports favouring cognitive behavioural therapy, and a sizeable proportion of patients feel that graded exercise therapy worsens their condition, the inference must be that the major recommendation for the use of cognitive behavioural therapy and graded exercise therapy was *clinical opinion*, the only other source of evidence left to the Working Group. If this is the case, then the professional composition of the Key Group was the crucial factor in determining the strength of recommendation for particular “potentially beneficial” management strategies.

It is also important to realise that research funding is critical to whether or not evidence is available. There are indications that psychiatric and psychological research groups conducting trials of cognitive behavioural therapy and graded exercise therapy have been particularly well funded (Abbot & Spence, 2002); hence, the forty-four trials available for analysis by Whiting *et al* (2001). This funding bias is itself worthy of examination as it informs us that the research agenda in CFS/ME has been driven, in the main, by a relatively small number of clinicians with a professional interest in exploring biopsychosocial models of illness. These clinicians were proportionately well-represented within the Working Group.

3.4.1 Cognitive behavioural therapy

The issues surrounding the true usefulness of cognitive behavioural therapy for CFS/ME patients have been widely discussed (e.g. Lancet 2001; 358: 239-41) but can be summarised as follows:

- Of the forty-four randomised clinical trials identified, only five involved some variant of cognitive behavioural therapy, and of these, three had a ‘positive’ result and two a ‘negative’ result.
- Two of these trials used the Oxford criteria which greatly limits the applicability of the findings as far as ME and CDC-defined ‘CFS’ is concerned.
- Dropout rates were high - 40% in the active arm (vs. 20% in the control) of the flagship trial on cognitive behavioural therapy by Prins *et al* (2000). As Whiting *et al* (2001) state in their review: “Dropout rates may be an indication of the acceptability of an intervention” and “cognitive behavioural therapy may be acceptable to only a small number of patients, limiting generalisability.”
- As is the case with most clinical trials, the results cannot be extrapolated to apply to the most severely ill (up to 25% in CFS/ME), nor to children or young people. Both categories having been excluded from these trials.

- While cognitive behavioural therapy most likely has some role in helping some patients to better cope with their symptoms until a cure is found, this role is limited (as it would be with cancer patients) and non-curative.
- Cognitive behavioural therapy is expensive and, with such a variable outcome, the cost-benefit ratio is problematic.
- As well as the limitations of the clinical trials in CFS/ME patients, there are doubts even among professionals about the specific efficacy of cognitive behavioural therapy. As a recent review commented: “...the foundations on which it rests are not as secure as some of its proponents would have us believe.” (Holmes, 2002).

“The foundations of cognitive behavioural therapy are not as secure as its proponents suggest.”

Though the CMO report states that “application of a cognitive behavioural model to CFS/ME has been found successful in most patients in the trials” (4.4.2.2), this bald statement is almost certainly untrue: of five randomised controlled trials, two were negative, dropouts were high, and some ‘improvements’ were seen in the control groups, indicating that not all improvement can be ascribed to CBT. The same section of the report contains a remarkable statement:

“The Working Group accepts that appropriately administered cognitive behavioural therapy can improve functioning in most patients with CFS/ME who attend adult outpatient clinics.” (4.4.2.2)

This is a masterful piece of drafting which skilfully suggests great benefits of cognitive behavioural therapy while leaving several exits in case of attack. What is “appropriately administered” cognitive behavioural therapy? What aspect of ‘functioning’ is meant? How can the Working Group accept that ‘most’ patients improve on the basis of the extrapolation of the results of three positive and two negative trials to the whole population of CFS/ME patients in the UK?

There are several quotations in the report which - probably unwittingly - go to the heart of the matter:

“Cognitive behavioural therapy for people with CFS/ME is currently unavailable or very difficult to obtain in much of the UK.” (4.4.2.2)

“There was disagreement among clinicians as to the precise value and place of cognitive behavioural therapy, which partially reflected the varying models of the therapy and disease.” (4.4.2.2)

“We also noted that misunderstanding, misplaced concern, and poor practice in this area could potentially undermine the beneficial application of this therapy or its principles in patients with CFS/ME.” (4.4.2.2)

“In one patient-group survey, only 7% of respondents found the therapy [CBT] ‘helpful’, compared with 26% who believed it made them ‘worse’. The remaining 67% reported ‘no change’.” (4.4.2.2)

As these quotes help to illustrate, cognitive behavioural therapy is non-curative (Wessley, 2001); is expensive and time-consuming, and beyond the resources of Health Authorities to fund; has an irrecoverably poor reputation among ME patients, especially the severely ill whom it incenses; has been found helpful by only a small minority of patients surveyed; and requires skilled therapists who need the consent of malleable patients rather than irate unwilling ones. As a recent commentary in the British Medical Journal stated: “Until the limitations of the evidence base for cognitive behavioural therapy are recognised, there is a risk that psychological treatments in the NHS will be guided by research that is not relevant to actual clinical practice and is less robust than is claimed.” (Bolsover, 2002). Or, as one patient has said, cognitive behavioural therapy is “not curative, not cheap, not accepted, and not the answer for everyone.”

3.4.2 Graded exercise therapy

Graded exercise therapy was the other “potentially useful” therapy identified by the Working Group on the basis of the three positive clinical trials out of the forty-four identified. The limitations of these trials have been discussed in depth elsewhere (BMJ 1997; 315: 947 and electronic responses to BMJ 2001; 322: 387), but the main points can be summarised as follows:

- The success of randomised controlled trials depends on strict comparability of control to treatment groups. In these trials there was not the same contact with the controls and patients, raising the possibility that factors other than treatment were involved in the “positive” outcome.
- All three trials consisted of patients classified by the Oxford criterion which does not diagnose ME or the CDC-CFS criteria (Fukuda *et al*, 1994) exclusively. The weakly-positive trial results may reflect this bias, have little relevance to CFS/ME patients, and have no relevance to the large numbers of severely affected or young sufferers.
- Graded exercise therapy involves a patient-motivation component to encourage compliance with the exercise regimen. However, the true usefulness of such programs is by no means clear (Harland, 1999).

- Its use is predicated on the belief that deconditioning is a factor in the perpetuation of illness in CFS/ME patients. However, there is good evidence that deconditioning is not a significant factor (Brazelmanns, 2001; Van der Werf, 2000) and that it cannot account for delayed post-exertional symptoms or the documented changes in muscle metabolism (Lane *et al*, 1998; Lane, 2000).

None of these is successfully dealt with in the CMO report, though some limitations are alluded to:

“One key controversy that exists over graded exercise rests on whether the nature of the treatment is appropriate for the nature of the disease, at least in some individuals. Existing concerns from voluntary organisations and some clinicians include the belief that some patients may have a primary process that is not responsive to or could progress with graded exercise, and that some individuals are already functioning at or very near maximum levels of activity.” (4.4.2)

“Voluntary organisations, as well as the Sounding Board events, note that graded exercise therapy can be effective in some individuals, but substantial concerns exist regarding the potential for harm.” (4.4.2.1)

Fortunately, some hard evidence from patient surveys is shown in the Working Group’s report, albeit in Annexe 3. This showed that of 1,214 patients using graded exercise therapy, 34% found it helpful but 50% (610 patients) reported that it made them worse. Graded exercise therapy had the greatest number of ‘worse’ reports of any therapy.

Clearly, as a management strategy, graded exercise therapy has its limitations for CFS/ME patients: “Best practice in this area indicates that the initial stages of any graded exercise programme should only be carried out by therapists (i.e., occupational therapists, physiotherapists, exercise physiologists, sports therapists, etc.) who have the necessary expertise to manage CFS/ME patients.” (4.4.2.1) At present, very few therapists are available with such expertise.

3.4.3 Pacing

In contrast with the two professionally-dictated interventions, pacing has been included as a ‘management strategy’ in response to patient experience - an example (some might say) of patients voting with their feet. Pacing allows patients to choose their own acceptable level of activity in accord with their fluctuating symptoms. It accepts that in the rehabilitation of sufferers, rest and relaxation also have an important role to play (Shepherd, 2001). The report clearly states the rationale for pacing:

“Clinical wisdom suggests that management of limited energy and supervision of any increases in physical or mental activity are an essential part of ongoing care for individuals with CFS/ME.” (4.4.2)

“A survey of more than 2,000 members of a voluntary organisation (Annexe 3, section 3) who were or had been severely unwell showed that 89% of group members found pacing ‘helpful’.” (4.4.2.3)

While pacing is intuitively sensible, its status as a clinical management strategy chosen after three years of deliberation by a Working Group is debatable, and there is a lingering suspicion that it has been recommended by the Working Group only as a concession to patient-based opinion. Whether sufferers will be allowed by healthcare professionals to choose this “recommended” therapeutic strategy in preference to psychological strategies is an open question. Indeed, almost as soon as the Working Group’s report was published, an item in the British Medical Journal commented: “The clinicians argued that the psychosocial side of the condition should have had greater emphasis and were concerned that ‘pacing’... was included as a form of treatment,” and quoted one professional as saying that “...doctors would not accept pacing just because it was recommended in the report” (Eaton, 2002).

3.4.4 Conclusions about the choice of management strategies

The preamble to the CMO report was explicit in its aims:

“Throughout, we have aimed where possible to base our commentary and recommendations on the best quality evidence, and from a range that includes randomised controlled trials and clinical anecdote. In the absence of research evidence to inform many issues, the bulk of the report is derived from a synthesis of patients’ and clinical experience. Where some data exist, albeit incomplete and not fully agreed, we considered the trident approach together with the likely resource implications to inform our conclusions.” (1.3.3)

How far have these aims been achieved? By conventional standards of literature reviewing, formal evidence for the use of cognitive behavioural therapy, graded exercise therapy and pacing is rudimentary. The fact that a few more clinical trials exist for cognitive behavioural therapy and graded exercise therapy than for any other intervention merely reflects the funding support which the interventions attract in the UK (Abbot & Spence, 2002). Patient evidence suggests that a small subgroup of patients might find either cognitive behavioural therapy or graded exercise therapy helpful (7% and 34% respectively) - possibly reflecting the heterogeneity of the patient grouping inside the construct ‘CFS’ - but that a substantial proportion (93% or 66% of patient responders, respectively) either find them ineffective or harmful. Pacing is nothing more than a commonsense approach enforced on most patients by their circumstances, and can hardly be described as a therapeutic management strategy. To use an analogy, pacing could describe the ability

of an amputee to hobble around in difficult circumstances: a “therapeutic management strategy”, however, might include a new prosthesis individually designed. Strangely, in a recondite section (but not in the easily-accessible overall conclusions) the report itself admits the truth:

“Review of the evidence highlights the lack of good quality research to support effectiveness of various therapies. Patient responses suggest that no approach is universally beneficial and that all can cause harm if applied incorrectly.” (4.0)

3.5 Failure to highlight data on the most severely ill patients

At several points, the report mentions the problems of the most severely ill patients:

“Severely ill are severely overlooked; just ignored and invisible.” (2.3.1)

“In general, this group is excluded from research, so they may not fulfil criteria used to test evidence-based approaches. Some report that they want to believe doctors and feel ‘frightened to say no’ or that they do not have the energy to disagree. Fears were also expressed over: being branded as a ‘difficult patient’, losing benefits, letting people down, not trying, losing the love of the family, and being labelled as mentally ill.” (2.3.1.1)

“Not enough is known about severe forms of the condition CFS/ME that are reported to affect up to 25% of patients.” (4.4.1)

Yet, a database of information collected and analysed on behalf of severely-ill sufferers by the 25% ME Group, which was presented to the Working Group, has not been used to full effect, and remains unmentioned in Annexe 3 (Patient Evidence). MERGE takes the opportunity of highlighting it in Table 2 below.

The 25% group, in a questionnaire report (25% ME Group, 2000), revealed that of 215 questionnaires returned some interesting observations could be made: 55% of respondents had been ill for more than ten years, and 50% of them had taken more than two years to obtain a formal diagnosis of CFS/ME. Twenty-five percent of respondents described themselves as bedridden, and 57% had been either housebound or bedridden for more than six years. As regards appropriate medical advice or treatment, 29% reported that none had been offered during the course of their illness. Only 25% of respondents felt that their condition was improving, or had improved from an even more chronic level. Important additional findings were that 76% (162/212) of respondents felt that the lack of a diagnosis or appropriate advice in the early stages of their illness had impacted on the severity and longevity of their symptoms; that 38% (81/212) described themselves as totally dependent on others; and that 48% (104/215) reported no regular assessment or management of their

condition. The management strategies recommended by the report are inappropriate for this group of sufferers, whose continued ill health - its aetiology, perpetuation and cure - remains a neglected challenge.

Table 2. Survey of severely affected CFS patients, reproduced courtesy of the 25% group

Age (years)	Number (%)
<20	5 (3)
20-39	70 (36)
40-59	90 (47)
>60	28 (14)
Time housebound/bedridden (years)	
<2	10 (5) / 10 (5)
2-5	49 (24) / 19 (9)
6-10	59 (29) / 16 (8)
>10	35 (17) / 6 (3)
Present condition	
improved/improving	53 (25)
stable at low level of functioning	105 (49)
slowly deteriorating	56 (26)
Duration of illness (years)	
2-5	31 (14)
6-10	66 (31)
10-14	49 (23)
>15	68 (32)
Illness onset	
sudden	104 (49)
gradual	110 (51)
Time to formal diagnosis (months)	
<12	76 (36)
13-24	28 (13)
25-60	53 (25)
>60	53 (25)
Time to appropriate advice/treatment (months)	
<12	66 (32)
13-24	14 (7)
25-60	40 (20)
>60	24 (12)
none given	60 (29)

3.6 Undue prominence given to the 'biopsychosocial' model of the illness

From the report of the first recorded outbreak in 1934 until the late 1980s, the emphasis was on the elucidation and treatment of the biomedical aspects of the illness (e.g., Acheson, 1959). Since then, a "biopsychosocial model" has been proposed - primarily by psychiatric/psychological professionals - defined by the report as:

"The biopsychosocial model of pathophysiology, applicable to all disease, suggests that once an illness has started its expression is affected by beliefs, coping styles, and behaviours, while consequential physiological and psychological effects act in some ways to maintain and/or modify the disease process." (3.3.4)

"Illness beliefs - The way in which abnormal illness behaviour and illness attributions (especially about cause) may be perpetuating ill health and disability in some CFS/ME patients remains a contentious issue." (3.3.3)

Psychological factors do, of course, accompany chronic illness - every patient has a mind and feelings which are affected by the experience of disease. The problem concerns the ascription of causation. The view that "psychosocial factors" either precede (cause?) CFS/ME, or play a major role in maintaining the illness after it has developed, has taken root among some, but not all, members of the medical profession, and has influenced the perception of CFS/ME in the media and among the general public. Naturally, patients have come to feel stigmatised and alienated, and perceive the influence of the model, particularly among medical practitioners, to have a pernicious effect on their care. To complicate matters, patients' beliefs that their illness is "physical" are seen by proponents of the biopsychosocial model as a sign of psychological dysfunction. Such psychologising of patients illness experience is not unique to CFS/ME patients. A recent study on Gulf War Syndrome was entitled: "Prevalence of Gulf war veterans who believe they have Gulf war syndrome" (Chalder *et al*, 2001). The principal author of this study was one of the Key Group members of the report. Many CFS/ME patients await with interest the next study in the series: to continue the analogy of the amputee used above, it could perhaps be on amputees (with or without CFS/ME) who believe that they have lost a limb. The CMO report itself - in select sections possibly written to assuage its lay members - does state the central problem with this model succinctly:

"Although they may have speculated about causation, mostly what has been demonstrated is an association. For example, the various psychological factors claimed to be causal may be a consequence of severe, prolonged CFS/ME." (4.2.1.4)

"Certain strongly held attitudes to the illness and coping mechanisms do seem to be associated with a poorer prognosis, but studies done so far have not enabled the direction of causation to be determined. Some have inferred that a poorer prognosis may be caused by such attitudes, but it can equally be argued that severe, prolonged illness may have a negative impact on attitudes and coping mechanisms... the various psychological factors claimed to be causal may be a consequence of severe, prolonged CFS/ME, and for the most part the study designs adopted would not enable the question of causality to be resolved." (4.2.1.4)

“However, it seems likely that cognitive dysfunction in CFS/ME cannot be explained solely by the presence of a coexistent psychiatric disorder.” (3.3.4)

“The biopsychosocial model of CFS/ME has influenced its perception among the general public.”

Nevertheless, peppered throughout the remainder of the report are examples of classical biopsychosocial modelism, despite the resignation of its supporters from the Key Group on the grounds that “the condition’s psychological aspects were being underplayed” (Hospital Doctor, 17th Jan 2002). Thus,

“An individual’s symptom profile is modified by the impact of illness on the person affected and those around them.” (3.4.2)

“Re-enablement should encompass cognitive, emotional, and social aspects as well as physical aspects.” (4.1.2)

“Ideally, services would be patient-centred, and adopt a biopsychosocial model or a holistic view of care.” (3.3.4)

“It is thought that certain strongly held beliefs about the cause of the illness can impede progress. These include the view that the illness is entirely physical or is caused by a persistent virus. These beliefs could be partially correct - e.g., a virus could have provoked a persistent or prolonged change in physical functioning. However, they could also act as obstacles to recovery or to necessary treatment.” (3.3.3)

Given that the evidence of efficacy for these interventions in CFS/ME sufferers is weak (Whiting *et al*, 2001), the relevance of these statements in the Working Group report is questionable. Why should the “ideal” service (which patients and their carers are paying for through their taxes) be one which adopts a biopsychosocial model, given the available evidence? More generally, how would it be if the same statements were applied to either asthma or angina, both of which have psychosocial elements yet are recognised as predominantly physical illnesses? As Susan Sontag says in her book, *Illness as Metaphor* (1978), “Theories that diseases are caused by mental states and can be cured by will power are always an index of how much is not understood about the physical terrain of a disease.” Some consider this insight to be particularly apt in the case of CFS/ME at the beginning of the 21st century.

3.7 Downgrading of relevant research findings

At points the Working Group’s report mentions its role in assessing research evidence: “...we sought to bring together

knowledge on CFS/ME to support initiatives to improve care for patients. This has been an intricate process, drawing on research evidence, the experience of patients and diverse clinical opinion.” (Foreword) “... make recommendations for further research into the care and treatment of people with CFS/ME.” (Remit, 1.1)

Yet, despite this, the main body of the CMO report deals with the research findings in 639 words (section 3.3.4) out of a total of some 34,600 in the main report. However, there is a large body of research literature on CFS/ME. As the CFIDS Association of America makes clear, though the aetiology of the illness remains elusive, numerous biological abnormalities have been reported in:

- Immune function - in the form of cytokine overproduction or poor cellular function (Patarca-Montero *et al*, 2000; Patarca-Montero *et al*, 2001).
- Brain and CNS - with possible involvement of the basal ganglia (Chaudhuri & Behan, 2000) or the functioning of the blood-brain barrier (Bested *et al*, 2001).
- Muscle - in the form of oxidation defects (McCully & Natelson, 1999) or post-exertional deficits (e.g., Lane, 2000; Paul *et al*, 1999).
- Autonomic functioning - as neurally-mediated hypotension (e.g., Bou-Holaigah *et al*, 1995).
- Hormonal function - most prominently at the hypothalamic-pituitary-adrenal axis (e.g., Scott & Dinan, 1999).
- Cardiovascular integrity - endothelial sensitivity to acetylcholine (e.g., Spence *et al*, 2000).
- Neuropsychological functioning - including impaired working memory and information processing unrelated to psychiatric illness (review: Michiels & Cluydts, 2001).

“The research literature contains several hypotheses and proposals to explain how CFS/ME may be caused or maintained. The quality of the evidence is variable, however, and many suggested mechanisms are as yet based on associations rather than cause or linkages.” (3.3.4)

Interestingly, these reasons for bypassing a full consideration of the research evidence, namely, the variable quality and lack of causal evidence, could also apply to the evidence for the choice of management strategies (cognitive behavioural therapy, graded exercise therapy, and pacing) and, it could be plausibly argued, to the biopsychosocial model itself.

The downplaying of the research evidence partly reflects the constrained remit, which was restricted to management strategies. With a different remit, the report might have been able to recommend a direction for future fundamental research after a thorough review of the literature. Instead, the message presented to the media, the public and opinion formers is that the best that can be done is to manage symptoms, most prominently with psychological strategies.

3.8 Inadequate coverage of social care and welfare issues

The foreword to the report states that: “In 1998, the Working Group on CFS/ME set out to consider how the NHS might best provide care for people of all ages who have this complex illness.”

While the NHS is the major player in care provision for patients, it is only one agency among many providing care for people. By focusing so closely on one agency, the CMO report has missed an opportunity to highlight more clearly the responsibilities to CFS/ME patients of other agencies and the professionals who work for them. The nature of the illness and its practical consequences, particularly for the severest sufferers, are such that social services should be closely involved in both care planning and direct service provision. Consideration should be given to those most severely affected, identifying them as a special interest group in terms of joint community care planning and in planning for children and young people’s services.

“The report’s message is that the best that can be done is to manage symptoms.”

The report recognises that, on the ground at present, the range of services are not ‘joined up’.

“Beyond primary care level, the issue that causes most concern is the lack of specialists and services... Some patients find themselves in geographical ‘black holes’ that lack specialist provision.” (2.2.4)

“Patients can encounter arbitrary and poorly informed decision-making on other issues such as home help and mobility badge schemes, as well as sheer resource limitation. Failure to access appropriate support from social services can be compounded if doctors fail to provide clear guidance about diagnosis and need.” (3.5.1)

While the recommendations on equipment and practical assistance (4.3.3) and the call for service networks (6.3) is welcome, statements about the services CFS/ME sufferers ‘should’ receive in the community are little more than howling for the moon: without the full support and practical backing of local social work departments, sufferers will struggle to see these needs either fully met or met appropriately. For example, a recommendation that clinicians should inform patients about local services is one thing, but providing clinicians with the ability to refer patients to the relevant agencies themselves would be truly useful. Indeed, with the advent of joint social work/health teams, this is no longer impractical. As regards recommendations to employers - even the NHS itself - the Working Group’s report is light on the provision of practical

advice about how the illness should be managed in the workplace. A fuller exploration of this issue would have been a welcome extension to the report, as would advice on good employment practice to all tax-funded employers.

It is re-assuring to see that Welfare Benefits have been accorded their own priority by the Working Group:

“A small subgroup of the Working Group was established to produce a paper on CFS/ME and the benefits system. This working paper was then submitted to the CMO in April 2000. Professor Donaldson formally copied the paper to the Chief Medical Advisor of the Department of Social Security to inform that Department’s Working Group, which was established to review the benefits system for people with chronic illness.” (1.3)

Yet, how useful it would have been, for patients and carers, to have had this information summarised in the main report, and attached in full as another Appendix.

The journey through public service provision is often a daunting one that can leave individuals feeling powerless and damaged by the very system that is supposed to support them. The experiences of patients in the health service, service users in local authorities, and claimants in the welfare benefits system, continually highlight the need for more independent advocacy services to ensure that people receive the services and support to which they are entitled, and to receive them with their dignity intact. Unfortunately, the Working Group report barely addresses these issues.

3.9 Words are not action - will anything actually change?

Though the CMO report makes some heroic suggestions for improving the quality of the patient-provider interaction, insisting that “Patients can be empowered to act as partners in care” (4.0), it carries with it no executive power, no funding to stimulate change, and no commitment to reconvene at a future date to report on the changes which may have been implemented. This severely limits its usefulness.

Given this, several aspects of the situation on the ground make significant beneficial change unlikely in the short to medium term. First, a significant number of patients have not been well served by healthcare professionals. For example, section 3.5 (above) has shown that 61% of the most severely ill patients report waiting more than 2 years for appropriate advice and symptomatic help (there is no ‘treatment’). Although the Working Group is, in places, upbeat about the prognosis for patients with the illness, e.g., “The likelihood is that most patients will show some degree of improvement over time, especially with treatment... Gradually progressive deterioration is unusual in CFS/ME.” (1.4.3), research studies on prognosis (e.g., Bombardier & Buchwald, 1995; Hines *et al*, 1993; Vercoulen *et al*, 1996) are less optimistic: around one third of sufferers regain up to 80% of their premorbid levels, but the remainder

experience remissions and relapses, albeit at a 'stable' level of functioning, often for years, or steadily deteriorate into severe incapacity and dependency. This often occurs without any support or significant help from healthcare professionals: without teeth, the recommendations of the Working Group are unlikely to alter this unfortunate picture. Again, research reports have shown that a substantial proportion of GPs do not believe they are dealing with a distinct clinical entity when they see CFS/ME patients (Stevens *et al*, 2000; Ho Yen & McNamara, 1991). A MERGE in-house analysis found that 20% of patients

“The report carries no executive power, funding, or commitment to follow up its recommendations.”

reported changing GP at some stage during their illness, and that roughly one third found their GP's attitude to be at best non-committal and sometimes openly sceptical. In a recent development, “Chronic fatigue syndrome/Myalgic encephalomyelitis” was voted by 12.6% (72/570) of respondents to the website of the British Medical Journal as one condition that best fitted the description of a “non-disease” (BMJ 2002; 324: 7334, data supplement). Published items of in-house literature for doctors perhaps clearly reveal how some feel about these patients:

“Never let patients know you think ME doesn't exist and is a disease of malingerers. Never advise an ME patient to make a review appointment. At the end of the consultation, I say goodbye, not *au revoir*.” **Dr Mary Church (a member of the BMA Medical Ethics Committee) quoted in the GP magazine, Pulse. 20th October 2001.**

“Question: What would be your initial response to a patient presenting with a self-diagnosis of ME?
Possible answers:

- a) Are you by any chance a teacher?
- b) Thank you for making the effort to come along. I am sure we will be able to help.
- c) For God's sake, pull yourself together, you piece of pond life.
- d) Well, lets just explore that, shall we?”

Dr Tony Copperfield (a pseudonym), described as being a GP in Essex, in Doctor magazine, 2000. (The 'correct' answer was (c).)

“I have every symptom of the disease. The pathogenesis of ME is increasing workload; being undervalued socially, politically, and financially; and being abused by those I try to help. You just have a get on with life.” **Name and address withheld. Doctor magazine. 18th March 1995.**

“If they really insist on a physical diagnosis tell them chronic fatigue syndrome is a complex disorder in which multiple biopsychosocial factors are mediated via the anterior hypothalamus - in other words, it's all in the mind.” **Dr Douglas Carnall, Bluffer's Guide: Chronic Fatigue. 12th January 1995.**

“ME is usually (in my surgery, always) a self-diagnosis: somebody comes in, sits down and says, 'I think I've got ME, doc'. This is what we in general practice call a '*heart-sink encounter*.'” **Dr Michael Fitzpatrick, “The making of a new disease”. The Guardian 7th February 2002.**

These quotes sit uneasily with the aspiration in the CMO report:

“The doctor's job should be to 'heal sometimes, relieve often, comfort always'.” (4.1.2)

“Positive attitudes and cooperation based on mutual respect seem likely to produce best outcomes.” (3.3.3)

Rather than promoting a culture in which CFS/ME patients and their carers can begin to be 'partners in care', a more likely outcome is the imposition of cognitive behavioural therapy and graded exercise therapy on some patients due to the media spin surrounding the report's conclusions. Patients should remember, however, that doctors have a duty to prescribe cognitive-behavioural interventions or exercise regimens with *as much care* as they prescribe drugs, and that CFS/ME patients who experience adverse effects or relapse - as indicated by patient reports of graded exercise therapy - may well be entitled to redress through the courts.

End piece - Patient voices



In the plethora of views about the research and management of this illness, the authentic voice of the sufferer is rarely heard. For this reason some individual poignant experiences are given below.

“I was eighteen years old when I was struck down with severe, virally-induced ME. I am now thirty-three. It has destroyed my quality of life. My feelings of loss and helplessness are often overwhelming. My parents have to care for me and the illness has deprived me of a career, a social life, and the possibility of marriage and children. I am 90% bed-bound and feel wretchedly ill every waking moment. At worst I am unable to hold a conversation, watch TV, or even read. My only hope is for a research breakthrough in this illness. More than anything else, I want to see ME recognised and a treatment found.” **Clare**

“The worst thing about having ME is, obviously, having ME. It is spending three years in your bedroom looking at the walls, in pain, isolated, unable to read, write, or talk, with a brain like spaghetti. The worst thing is having a brain which no longer works and which I can't do anything about. It's like being in solitary confinement, except that I haven't done anything wrong.” **Josh**

“The feelings of pain and sickness are with me all the time. The illness has changed my life. I can do none of my former hobbies, and am left hanging around on the fringes of a no man's land between the dying and the well. It's a double torture - having the illness and having it unrecognised. It has been said that patients like me should just move on, but after twenty years it seems to me that the only things moving on in this illness are professionals - medical and charitable - making careers out of my misery. A little humility and some humanity by those in the so-called 'caring professions' would go a long way towards helping me cope with what has been a truly awful experience.” **Alex**

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Postscript by MERGE

This document is the creation of the MERGE staff, Dr Neil C. Abbot and Dr Vance A. Spence. A substantial contribution, including to the production and design, was made by Dr David J. Newton. The sections dealing with social care were prepared following expert advice from Mr William Dockery.

MERGE has links with the 25% ME Group which represents the severest ME sufferers in the UK and this analysis of the CMO report has, in part, been carried out with this particular group in mind. We acknowledge the many patients, carers and concerned professionals from the *Friends of MERGE* scheme for their contributions and support to the production process.

MERGE exists to fund scientific investigation into the causes and treatment of myalgic encephalomyelitis (ME), to provide information and education about the condition, and to support sufferers. The charity was founded by Dr Vance Spence and Mr Robert McRae, both ME sufferers forced to retire early from their professions. With Roger Jefcoate CBE as its founding patron, and The Countess of Mar as its patron, MERGE obtained charitable status in April 2000 and, after establishing itself successfully, commenced its five-year plan of expansion from May 2001. Ambitiously, we aim to commission and fund a variety of research projects into the pathophysiological basis of the illness, and to establish a social care programme.

I want to help MERGE fund research and support sufferers

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- Information about the Friends of MERGE scheme

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Appendix

Following publication of the Working Group's report to the CMO, there was some debate about the future direction of research into CFS/ME in the UK. In particular, great reliance was placed on the "research" evidence documented in the National Research Register.

During a debate in the House of Lords on the Working Group's report on CFS/ME (16th April 2002), mention was again made of the role of the National Research Register in informing policy in this area. Accordingly, MERGE has included the executive summary of its document, *Research into ME/CFS in the United Kingdom: Can the National Research Register inform future policy*, in this Appendix. Electronic copies of the full document (50 pages, with 39 pages of tables) are available from merge@btopenworld.com.

Research into ME/CFS in the United Kingdom: Can the National Research Register inform future policy?

An analysis by MERGE, February 2002

Dr NC Abbot and Dr VA Spence

Executive Summary

There is presently a debate in the United Kingdom about future direction of public policy regarding research into Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS). Energising the debate is an apparent increase in the scale of the problem nationally and, recently, publication of a report by an independent working group to the Chief Medical Officer of England. However, policy must be guided by good data and great reliance has been placed on the UK National Research Register (NRR) of completed and ongoing medical studies as a resource for informing debate. This register is a database of ongoing and recently completed research projects funded by, or of interest to, the United Kingdom's National Health Service. This analysis of the information on ME/CFS contained within the NRR was designed to answer a specific question: given the interest in the development of a research policy for ME/CFS in the medium to long term, is the information contained in the NRR records robust and accurate enough to inform policy-makers?

The total raw number of studies on ME/CFS retrieved from the NRR was 28 ongoing and 133 completed studies (partial records are presented in a 35-page Appendix to this report). From each, the following key data were extracted: Title; End date; Contact person; Principal research question; Sample group description; Funding source and amount. Each record was assigned to an *ad hoc* "research category" (of interest to researchers), and a "clinical category" (of more interest to the public and policy-makers) on the basis of the professional and/or departmental affiliation of the "contact person".

Of the 161 NRR reports retrieved, 10 appeared not to involve ME/CFS patients directly, and 12 appeared to be duplicates of existing reports. Thus, only 139 (23 ongoing and 116 completed) could be classed as "relevant" reports - representing 0.17% of the 80,000 on the entire NRR database. Eighteen reports (5 ongoing and 13 completed) concerned research in Scotland. Many reports were incomplete: 35% and 31% of ongoing and completed study records, respectively, had missing descriptions of the proposed sample group; 22% and 28%, respectively, had missing details of sources of funding; and the amount of funding received was not stated in more than a half of all entries. In addition, some records had very similar content, despite a difference in "end dates" which varied by up to 18 months, raising the possibility that some records describe extensions of an existing project rather than separate discrete investigations.

When classified by clinical category, 41% of reports had “contact persons” whose professional association was with “psychiatry, psychological medicine or mental health”. The second and third largest categories were neurology, neurosciences or neurophysiology (13%) and general medicine/medical care research (12%), respectively. When classified by research category, investigations with some scientific rationale and some relevance to the pathophysiology of the illness constituted the largest group of records (43%), but many of these were smaller exploratory studies (evidenced by relatively small sample sizes) that are unlikely to have given a definitive answer to the initial research question. The main other categories contained clinical trials or other investigations of essentially biopsychosocial interventions (17%), followed by surveys pertaining to biopsychosocial interventions (14%), and surveys of welfare or social aspects (9%).

Given that the amount of funding received was not stated in more than a half of all entries, no definitive conclusions can be drawn from the information on source or amount of funding. However, the clinical category “psychiatry, psychological medicine or mental health” is the most successful in attracting research funding. Overall, however, few public resources (NHS or Research Council) have been directed towards researching this illness.

In conclusion, the NRR records tend to be incomplete; to contain inadequate descriptions of the research proposed; and to have no cross-reference to the results emanating from the research. The records relating to ME/CFS reveal that comparatively little research has been done given the scale of the problem in the UK and that few public resources have been directed towards research, particularly into the pathophysiological basis of the illness. Much of the research undertaken has been led by investigators with a professional or departmental affiliation to Psychiatry, Psychological Medicine and Mental Health, and none of the 139 studies were conducted on the most severely-ill patients.

Given the recent recommendations of the Chief Medical Officer of England that government investment in research on ME/CFS should be comprehensive and include a range of studies designed to “*elucidate its aetiology and pathogenesis, clarify its epidemiology and natural history; characterise its spectrum and/or subgroups; and assess a wide range of potential therapeutic interventions including symptom control measures*”, **we conclude that the NRR is not robust enough, as an information source or as a research resource, to inform the direction of future policy.**