
The Request for Advice

On 7 June 2002 the Health Council received a letter (reference: POG/ZP 2.294.354) containing the request from the Minister of Health, Welfare and Sport, Dr E. Borst-Eilers, for a review of the current level of knowledge of CFS. The request for advice reads as follows:

Dear Professor Knottnerus,

Chronic fatigue syndrome (CFS) imposes considerable limitations on the occupational, social and personal functioning of the people who suffer from it. The clinical picture is surrounded by ambiguity and uncertainty. Views as to the aetiology and possible causes are many and varied. No conclusive explanation has yet been found for the syndrome. In practice, the condition sometimes goes unrecognised (partly because the principal symptoms – fatigue and general malaise – are not specific to CFS and there is no diagnostic test available for determining whether someone has CFS). Consequently, some patients feel that their symptoms are being ignored, which inevitably causes frustration in the concerned individuals.

Absence from work, and virtual or complete exclusion from employment, are common in these patients. A complaint commonly heard from patients is that they are not eligible for social security benefit under the Disability Insurance Act (WAO) because the cause of their symptoms is said to be unknown, or because the limitations that they are experiencing are not deemed to constitute a disease or infirmity. Patients are reported to experience similar problems in connection with the allocation of other services and facilities (under the Act on Provisions for Handicapped People [WVG] for example, or the Law on the Reintegration of Disabled Persons [REA]) and as far as the assessment of their need for healthcare services is concerned.

This can adversely influence the financial position of the concerned patients.

According to an estimate made by the Free University of Amsterdam's Social Economics Institute in 2000, the social costs of CFS are in the region of Eur 250–600 million per year.

I request that you provide a review of current knowledge on CFS. I request that you give particular consideration to the following topics and questions in your recommendations:

- A description of CFS, including its clinical course. The definition of CFS that is applied in the clinical setting and in scientific research. The delineation of, and the overlap with, other somatic and/or psychological disorders and illnesses.
- An estimate of the incidence and prevalence of CFS, now and in the years to come – if possible, also broken down into relevant groups, such as children, adolescents, men and women, and people of different ethnic origin. Is there evidence to suggest that CFS is more prevalent in particular occupational groups?
- A comparison of the incidence and prevalence in the Netherlands with the neighbouring countries and a possible explanation of any differences.
- Possible causes of CFS and any factors (biological, social, sociological, psychological and societal) that influence the syndrome. What is known about the relationship between work and the development and/or persistence of CFS?
- Risk factors and possible preventive measures. Is it possible to prevent (or exert a positive influence over) the development of CFS by means of behavioural, lifestyle or other measures? Are there any known risk factors for CFS and is early detection of (predisposition to) the condition possible or beneficial?
- Possible therapeutic interventions. To what extent are possible therapeutic interventions applied in the Netherlands? What is known about cooperation between occupational physicians and GPs in relation to these patients?
- Gaps in our knowledge about CFS and promising avenues of research.
- Ways of promoting the dissemination of knowledge about this condition among care providers and ensuring that this knowledge is applied in practice.

For your information, I enclose the *Knelpuntenoverzicht* (Review of Problems), dated April 2002, which was compiled by the *ME-Fonds* (ME Fund) and the CFS patient organisations.

Yours sincerely,

The Minister of Health, Welfare and Sport, Dr E. Borst-Eilers

The letter was accompanied by the following discussion paper, compiled by the *ME-Fonds* in cooperation with the other CFS patient organisations:

Chronic fatigue syndrome/ME Review of Problems April 2002

Introduction

The Ministry of Health, Welfare and Sport has requested four organisations concerned with chronic fatigue syndrome/ME (hereinafter referred to as ME/CFS) to produce a survey of the problems associated with ME/CFS. These four organisations include the Dutch ME Association, the *Steungroep ME en Arbeidsongeschiktheid* (ME and Incapacity for Work Support Group) the *ME-Fonds* and the *Vereniging ME-huis in oprichting* (The ME-Home Foundation).

ME/CFS is a chronic, disabling illness that is accompanied by severe exhaustion and a host of other symptoms. Most patients are no longer able to lead a normal life. Going to school and working are frequently a problem. Besides being a personal tragedy, ME/CFS is also a tragedy for the community. The annual costs to society in 2000 were estimated by the Free University of Amsterdam's Social Economics Institute (research report entitled *Maatschappelijke kosten en ME* [Social Costs and ME]) at between 560 million and 1.34 billion guilders (Eur 254–608 million). The number of patients suffering from the illness in the Netherlands appears to have increased. Research conducted among GPs indicates that 27,000 patients were diagnosed in 2000, compared with 17,000 in 1993. This study found that the diagnosis of ME/CFS is often not made in cases where it is applicable. The ME organisations therefore estimate the number of ME patients in the Netherlands to be 30,000. It not unlikely that this will, within a few years, turn out to an underestimate. CFS stands for chronic fatigue syndrome. This illness was formerly known in the medical world as ME. This name is still used in the World Health Organisation's International Statistical Classification of Diseases (ICD-10) and can likewise be found in the widely-used classification for occupational and insurance physicians in the Netherlands. ME was originally an abbreviation of myalgic encephalomyelitis, which proved to be an unsatisfactory name for the illness. However, the abbreviation 'ME' (without the underlying medical terminology) is the name by which the condition is still most widely known among the general public. This is why we have predominantly used the abbreviation 'ME/CFS' in this document.

The problems experienced by ME patients lie in the medical, social and scientific sphere. They have consequently been divided into the following categories in this document:

- A. Problems relating to healthcare
- B. Problems relating to social position
- C. Problems relating to scientific research

A. Problems relating to healthcare

By contrast with many other people with chronic illnesses, ME patients have to contend with a number of specific problems:

1. Incorrect image among physicians, care-givers, those around them and the public

1. ME/CFS claimed not to exist

The United Nations' World Health Organisation (WHO) has recognised ME as a disease. ME has been included in the chapter on 'Diseases of the nervous system' in the 'International Statistical Classification of Diseases and Related Health Problems' (Tenth Revision, ICD-10, World Health Organisation, Geneva 1992, Volume 1, page 424, G93.3). As a Member State, the Netherlands is bound by this classification. In 1994 the ICD-10 was elevated to 'standard' status in the Netherlands at the recommendation of the National Advisory Council for Public Health (NRV). ME also features in the diagnostic codes for occupational and insurance physicians. ME/CFS is nevertheless still frequently not recognised as an illness in the medical world. To this day, there are medical practitioners who maintain that ME/CFS does not exist, and who refuse to apply the diagnostic criteria and make the diagnosis.

2. No diagnostic test available for ME/CFS

There is no laboratory test with which the diagnosis of ME/CFS can be confirmed. The diagnosis is made by testing whether the patient satisfies the diagnostic criteria and by subsequently ruling out every other disease that might possibly cause the symptoms. Although other diseases are diagnosed in the same manner, this nevertheless frequently gives rise to the misconception among practitioners that it is not possible to objectively make the diagnosis of ME/CFS.

3. ME/CFS is claimed to be caused and/or perpetuated by wrongful thinking and wrongful behaviour

Despite considerable evidence from scientific research to suggest that ME/CFS is associated with physical disorders, the precise cause of the illness remains unknown. In the absence of medical evidence, many physicians have been inclined to reach for psychological explanations rather than admitting that they simply do not know. This is definitely the case with ME/CFS. Psychological research into ME/CFS receives disproportionate attention in the Netherlands. A misconception has consequently come to preponderate among physicians and care-givers that the illness is the result of wrongful thinking and wrongful behaviour on the part of the patient. Psychological therapy is often erroneously regarded not as a way of learning how to better cope with the consequences of the illness, but as a way of curing it. There is insufficient scientific evidence to support this presumption. The patient is thus burdened with the idea that he himself is the cause of his illness and that he is himself to blame if the symptoms do not resolve.

2. Diagnosis

1. Usually made (far) too late

There are internationally established diagnostic criteria for the diagnosis of ME/CFS (see Appendix). These consist, in part, of ruling out other conditions that might account for the symptoms. There is insufficient knowledge of these diagnostic criteria both among GPs and among specialists, and the diagnosis is consequently made late and inaccurately. As a result, patients are left in uncertainty for an unnecessarily long time, they are given the wrong diagnosis, the diagnosis of ME/CFS is made where it is not applicable, or else another diagnosis is erroneously missed. This unnecessarily imposes an additional burden on the patient.

2. General practitioners

Virtually no attention is paid to ME/CFS in the training and continuing education of GPs and what little attention is, in fact, paid has a psychological bias. Moreover, research conducted in 2000 has shown that around 13% of GPs do not make this diagnosis or do not wish to investigate whether the diagnosis is applicable. The same research also reveals that 47% of the GPs did not make the diagnosis of ME/CFS in the case that was presented to them, which – according to the researchers – was definitely ME/CFS. The reasons cited for this include uncertainty, objection to the diagnosis and lack of knowledge.

3. Specialists

There are very few specialists who are able to make a proper diagnosis. Furthermore, there is no specific speciality to which ME patients can be referred. Patients who are referred to a specialist are frequently told: “I have not found anything in my field, so you are healthy”.

4. Insufficient diagnostic investigation

In some cases, the diagnosis of ME/CFS is used as an excuse for not conducting any further investigations. Other possible diagnoses may be missed as a result of this, with all the attendant consequences.

3. Treatment and support

1. Too few expert physicians

There are far too few physicians who are able and willing to treat ME/CFS. This also causes problems for the GP, who has few options for effective patient referral.

2. Too few treatment options

The biggest problem is that there is (still) no treatment that is aimed at tackling the cause of ME/CFS. Furthermore, the possibilities for symptom control remain insufficiently explored and exploited.

3. Imposed treatment

Patients sometimes have treatment imposed on them, which can lead to a serious deterioration in their state

of health. This situation applies at some university centres in the case of the combined use of cognitive behavioural therapy and graded exercise.

4. Cognitive behavioural therapy

There is undoubtedly a need for psychological support, in view of the apparently hopeless situation in which many ME patients find themselves. However, the support options are, for the most part, dominated by a controversial protocol (the Nijmegen CBT Protocol).

5. Rehabilitation is seldom (if ever) available

Rehabilitation should help ME patients to make the best possible use of their limited opportunities. Specific rehabilitation is, however, virtually non-existent. Where such facilities are available (e.g. at 'Het Roessingh' in Enschede), there are very long waiting lists and the capacity is extremely limited.

6. Limited reimbursement of treatment

Some regular treatments that can be effective in particular cases are not reimbursed (limited reimbursement for physiotherapy, no reimbursement for specialist treatment abroad). Many alternative treatments that benefit certain patients are likewise not reimbursed.

7. Home care problematic

Owing to the lack of residential treatment options, patients are currently mainly reliant on their GPs. However, GPs often have little opportunity to make home visits owing to lack of time. Furthermore, if the patient has problems, GPs assume that he is ambulatory and will come to them, whereas this is by no means always possible. Ultimately, patients are regularly left to fend entirely for themselves.

8. Patients poorly handled

Patients are frequently poorly handled by physicians, with their symptoms either not taken seriously or trivialised.

B. Social problems

1. Income and social security benefits

1. Exclusion from social security benefits

ME patients who have been declared unfit for work are still sometimes either excluded from receiving social security benefit under the Disability Insurance Act (WAO), the Invalidity Insurance (Young Disabled Persons) Act (Wajong) or the Invalidity Insurance (Self-employed Persons) Act (WAZ). Recent, as yet unpublished, research indicates that 28% of insurance physicians and 27% of occupational physicians are of the opinion that ME/CFS cannot be regarded as a disease or infirmity (a condition for entitlement to a WAO benefit). Plans are constantly being put forward to bar ME patients from receiving disability benefits on the grounds that the cause of the illness remains unknown or else based on a misrepresentation of its cause and of the possibilities for treatment.

2. Determining limitations

There is no sound and undisputed method available for objectively determining the limitations and the physical capacity of people with ME/CFS. The method that is used in the assessments leads to a great deal of arbitrariness.

3. Other financial problems

For the same reasons that lead to the problems with WAO benefits, ME patients can be faced with the prospect of non-payment of wages during the first year of their illness, dismissal without entitlement to unemployment benefit for refusal to work, refusal to pay out on private disability insurance policies, reduction or withdrawal of social security benefit due to non-fulfilment of the obligation to apply for jobs, and exclusion from entitlement to dependents pension.

2. Work

1. Labour disputes

Non-acceptance of the illness or of the associated limitations by an occupational physician, insurance physician and employer frequently leads to labour disputes and dismissal.

2. Modified work and reintegration extremely difficult

Problems with social security benefit frequently make modified work difficult or impossible. Anyone who is still capable of working will often require a drastic reduction of hours if they are to remain in their job,

something that employers usually find undesirable. Occupational reintegration is frequently impossible because no account is taken of the limitation of physical capacity.

3. Exclusion from services/facilities

People with ME/CFS are sometimes barred from receiving services/facilities under the Law on the (Re)integration of Disabled Persons (REA) because they are not regarded as 'disabled'.

As a result of the problems mentioned above, ME patients who could still be partially capable of working are often remain excluded from employment.

3. Education

ME/CFS is an important cause of prolonged sickness absence among pupils in secondary education. Students with ME/CFS in vocational training and higher education also frequently run into difficulty.

1. Adaptation of teaching impossible in practice

While it is possible, in theory, to adapt teaching to the capabilities and limitations of a student with ME/CFS, this often proves extremely difficult, if not impossible, in practice.

2. Education and support not available

There is no proper education and support available for students with ME/CFS.

3. School attendance officers

Pupils with ME/CFS and their parents sometimes encounter problems with school attendance officers who do not accept absence from school.

The result of all these problems is that children and young people with ME/CFS can fall seriously behind with their education. This is, in part, unnecessary and it is difficult to catch up at a later stage.

4. Services and facilities

1. Arbitrariness and exclusion

As far as services and facilities such as those provided under the Act on Provisions for Handicapped People (WVG), disabled parking facilities and home care are concerned, people with ME/CFS have to contend with the same arbitrariness and exclusion as arise in connection with WAO benefits.

5. Life situation

1. Isolation

People with ME/CFS often suffer from isolation, social exclusion and poverty.

Furthermore, they are continually forced on the defensive because those around them and all sorts of official bodies cast doubt on the severity of their condition.

2. Future prospects

Children and young people with ME/CFS miss out on normal development and have a wholly unclear and uncertain future.

6. Social consequences

1. High costs

Research has shown that ME/CFS currently costs society between €254 million and €608 million per year.

2. Increasing number of patients

Research among GPs has shown that the number of diagnosed ME patients in general practice rose from 17,000 in 1993 to 27,000 in 2000. One of the reasons for this increase may be that the diagnosis is less frequently being missed. A rise in the total number of patients is inevitable, however, since the percentage who recover is low and many new clinical cases are constantly emerging. It is likely that the estimate of 30,000 patients will turn out to be too low within a few years. An estimated 5,000 of these 30,000 people are under 25 years of age.

C. Scientific knowledge and scientific research

1. Methodological problems

1. Patient population

ME/CFS research does not appear to define the patient population in a uniform manner. Moreover, each

study delineates the patient population in a different fashion, making it difficult to draw comparisons between one study and another.

2. Control group

A great deal of the research has been conducted without an adequate control group. The results are consequently difficult to evaluate and verify.

3. Scope and duration

In order to make informed judgements regarding the reasons and risk factors underlying ME/CFS, research with large numbers of patients and controls is required. Furthermore, the research should be conducted over a longer period, given the prolonged nature of the illness. Research of this kind has yet to take place, owing to a lack of funding.

4. Biased research in the Netherlands

The research in the Netherlands is too biased towards the psychological treatment modalities and too little research is conducted into:

- the biochemical aspects of ME/CFS
- the (unknown) cause of ME/CFS
- effective therapies
- demographic and epidemiological aspects, such as the distribution of the condition in the Netherlands, incidence and prevalence, risk factors and the course
- methods for gaining a clear picture of the limitations and physical capacity of ME patients
- the implications of the illness for the patient and those around him.

2. Organisational problems

1. Multidisciplinary

The research conducted to date indicates that the cause of the illness is, in all probability, multifactorial.

This underlines how important it is that research into this disorder should be multidisciplinary in design.

This approach is difficult to achieve in the research arena.

2. Structured scientific approach

There is no specific professional group that is treating ME patients and taking the lead in the research field.

This explains why studies with a structured scientific approach fail to get off the ground and why worldwide research results have not been satisfactorily correlated.

3. Status of the research

Far too little funding has been available in the past to permit research into ME/CFS. This research has consequently not been held in high regard in the Netherlands. Many research plans have foundered due to their high-risk nature, and the lack of coordination and funding.

4. Knowledge exchange

There is no central location in the Netherlands where knowledge is available about research conducted within all branches of medicine at home and abroad into ME/CFS. An overview of ongoing domestic and international studies is also lacking. As a result of this, it is extremely difficult for physicians to gather knowledge on this topic. Knowledge exchange is practically impossible.

Conclusion

Some of the problems that have been discussed above are intrinsic to the illness. These will remain insoluble so long as there is no effective means of treatment and effective prevention remains impossible. This requires targeted research.

Other types of problems can, however, be resolved by means of specific measures. These problems are currently causing unnecessary damage to physical and psychological health, and are imposing an unnecessary burden and expense on society.