ScotPHN report

Health Care Needs Assessment of Services for people living with ME-CFS

Scottish Public Health Network (ScotPHN)

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APPENDIX 1: Stakeholder Events

http://www.scotphn.net/projects/current_projects/care_needs_for_those_experiencing_cfs_me/

APPENDIX 2: Literature Review

http://www.scotphn.net/pdf/Appendix_2_Draft_pdf.pdf

APPENDIX 3: The Three Tier Service for ME-CFS

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FOREWORD

When the Scottish Government commissioned the Scottish Public Health Network to undertake a formal health care needs assessment of services for those people who are living with ME-CFS, it was recognised that it would be a challenging endeavour and so it has proved. There are passionately held views on all aspects of this topic held by professionals, patients, their carers and the organisations that have been set up to support and represent them and to lobby on their behalf. Many of these views are mutually exclusive and compromise has been unachievable and, indeed, may be undesirable.

As with any such assessment, this health care needs assessment can only ever be as good as the evidence which is used to underpin it. Even in this regard I have been struck by the strength and depth of the debate concerning the validity of, and the interpretation of, the evidence base surrounding ME-CFS. The evidence is slowly building and the debate continues and develops. This document, therefore, cannot be the final word on these matters.

Given these difficulties, the very fact that this task has been completed, albeit to a longer timeframe that initially intended, should be seen as an important step forward.

Over the last two years, as Chair of the Project Group, I have been impressed with the hard work, care and commitment which has gone in to producing this final document. I want to thank the members of the Project Group for the essential contribution that each and every one of them has made to this assessment. Importantly, however, I also want to acknowledge the sheer determination that has been shown by so many individuals, organisations, services and agencies, all of whom have contributed in very important ways to the creation of this document. In particular, I wish to thank those individuals suffering from ME who worked hard to provide scrutiny comments or review the consultation draft of the report; without this input, we would not have been able to produce as robust a report as we have.

Lastly, but importantly, Phil Mackie and Ann Conacher are to be congratulated for the way in which they have dealt with great sensitivity with individuals and organisations who have contributed to the development of this work and it is to their credit that this document reaches publication in a way that, whilst not being the last word on the matter, has the potential to be an important driver to change things for the better for people living with ME-CFS in Scotland.

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- All those who attended the service provider and service user stakeholder events on 9th and 23rd June 2008 respectively.
- The members of the Patient Scrutiny Panel.
- All those who participated in patient focus groups and interviews undertaken by Action for ME.
- The local contact in each NHS Board.
- Action for ME.
- The Bristol and Bath CFS/ME Coordinating Centre.
- David Brewster, ISD.

Where appropriate, individuals are named in Appendix A.

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GLOSSARY

ScotPHN	Scottish Public Health Network
ME	Myalgic Encephalomyelitis
CFS	Chronic Fatigue Syndrome
HCNA	Health Care Needs Assessment
NICE	National Institute of Health and Clinical Excellence
MCN	Managed Clinical Network
CMO	Chief Medical Officer
CPG	Cross Party Group
DWP	Department of Work and Pensions
ICD	International Classification of Diseases
CDC	Centers for Disease Control
NMH	Neurally mediated hypotension
POTS	Postural orthostatic tachycardia syndrome
QOF	Quality and Outcomes Framework
NZGG	New Zealand Guidelines Group
GET	Graded Exercise Therapy
CBT	Cognitive Behavioural Therapy
SGPS	Scottish Good Practice Statement on ME-CFS
SIGN	Scottish Inter-Collegiate Guideline Network
MRC	Medical Research Council
MDT	Multidisciplinary team

EXECUTIVE SUMMARY

Introduction (Chapter 1)

- Formal assessment of health care needs in relation to Myalgic Encephalomyelitis – or Encephalopathy – (ME) and the broader spectrum of conditions that have been grouped together under the term Chronic Fatigue Syndrome (CFS) is a challenging exercise.

- The growing evidence base for both ME and CFS highlights that they are likely to be part of a range of conditions with differing underlying causes that can present in similar ways. However, at this time it is not simple to distinguish clearly between the two, not least because there is no unambiguous diagnostic test (or tests) for either condition.

- Diagnosis is usually based purely on symptoms, using criteria-based diagnostic tools that have been developed using a blend of research evidence and expert consensus.

Health care needs assessment (Chapter 2)

- The process of Health Care Needs Assessment (HCNA) should, insofar as there is appropriate data available, describe the capacity of the population to benefit from a service or intervention and to make suggestions as to how such benefits can be realised. It should be undertaken using methods which are appropriate to the types of need being established. Clearly the aim is to use data collection mechanisms that are valid and robust. In general this requires a mixture of qualitative and quantitative data collection techniques. Such robust methods, within the limitations of the available data, have been used in developing this HCNA.

Definitions of ME and CFS (Chapter 5)

- Overall, it was the view of both the HCNA Project Group and its Steering Group that the review of the evidence did not suggest that a single definition of ME-CFS was useful. Similarly the two workshops did not achieve a consensus on a definition.

- A pragmatic solution is therefore required with separate definitions for ME and for CFS. It is recommended that the clinical, symptomatic definition of ME outlined in the Canadian Guideline be adopted in Scotland. It is further recommended that a symptomatic definition of CFS based on that proposed in the NICE guideline be adopted in Scotland.

- In adopting this two definition approach, the HCNA acknowledges that the Scottish Good Practice Statement provide the recognised clinical guidance on the diagnostic approach to ME-CFS. This is important as the guidance also addresses the concerns raised by the Scottish Neurosciences Council regarding the use of diagnostic tools.

Epidemiology of ME-CFS (Chapter 6)

- The absence of an accurate case-definition for ME-CFS has made describing the epidemiology of ME and of CFS problematic. This is an international problem which is yet to find an adequate resolution. A review of the epidemiological research literature published since 2003 has highlighted that there has been no study undertaken specific to the Scottish population.

- Recent guidance cites the overall prevalence of ME-CFS in the adult population at between 2 per 1000 and 4 per 1000. This is an estimated range based on published research studies from across the world and considered to provide a “best estimate”. Of these, it is estimated that 1 in 4 people with ME-CFS will be severely affected. There is very little data on service use, whether within primary or community care, or from specialist providers. What material is available is often prone to problems of definition and under-estimation.

Expressed Need (Chapter 7)

- The assessment of expressed need confirmed the applicability of a tiered model of care. At Tier 1 service users identified the need for information, social support and access to services which maintain activities of everyday living both for the individual and their families / significant supporters. For people who were more severely affected, the degree of expressed need was higher and should include specific help with personal care areas and everyday daily activities. The need for support for informal carers was also highlighted.

- The most commonly expressed need at Tier 2 was for better understanding and acceptance from primary care staff, particularly general practitioners. There was some degree of recognition by some service users that the absence of a specific diagnostic test could cause delays in the overall diagnostic process. Service users and providers also highlighted the need for better information. However, they also highlighted the need for a better understanding on the part of the primary care team on what advice to offer people diagnosed with ME-CFS regarding self-management and the need to provide care geared towards providing alleviation of unpleasant or worrying symptoms. It was felt that this need should be met in a structured way using some form of protocol or patient pathway. Care management was identified by both service users and service providers as a means of providing access to community health services to meet specific symptomatic needs and ongoing management within a long-term condition model of care. The need for mechanisms to aid the regular review of care, particularly for those with severe and prolonged disability, and, on-going, home-based assessment for the severely affected were also noted. Anticipatory care for people with ME-CFS was also highlighted, particularly in the context of possible co-morbidities that could emerge over time.

- At Tier 3 both service users and providers were clear in their view that there was a need for specialist services. Such a service needed to have a strong medical lead and provide access to the types of therapeutic advice that is usually found within neurological or rehabilitation services. The need for any specialist service to exist within the context of wider networks which encompassed other disciplines and services was expressed. Significant differences between service users and service providers only started to emerge when the *organisation* of Tier 3 services was considered. However, both service providers and service users made specific mention of the need for developing managed clinical networks (MCNs) for ME-CFS.
- Above all, service users and providers want services, irrespective of tier, that have positive attitudes towards ME-CFS and sufficient knowledge of the condition.

The Comparative Analysis: Evidence from Clinical Guidelines (Chapter 8)

- Current clinical guidelines are primarily based on “clinical consensus” or “best practice” statements which originate amongst those with expertise in the field. There is international consensus that referral and specialist input may be necessary. As such, they tend to focus on the need for effective diagnosis and the trial of symptomatic treatments where clinically indicated. This issue is dealt with more fully within the Scottish Good Practice Statement on ME-CFS: http://www.show.scot.nhs.uk/GoodPracticeStatementonME-CFSforGeneralPractitioners.

The Comparative Analysis: Evidence for Effective Service Models (Chapter 9)

- Despite detailed online and manual searches, evidence relating to detailed service models from other countries was elusive and a specific service model was not identified. In the absence of alternative models the Scottish, generic long term conditions model of care and the English CFS/ME Coordinating Centre model were considered as examples of practice that is coherent, transparent and underpinned by national guidelines.

- Given the long and contentious history which underpins the recognition of ME-CFS, it is perhaps not surprising that there is no clear, internationally recognised model of care which can be adopted in Scotland. Indeed the original model for a tiered service set out in the Scottish CMO's Short Life Working Group Report seems to be as good a starting point as any for Scotland.

Current Service Provision (Chapter 10)

- A further analysis of the Action for ME Scoping Study data on current service provision was undertaken and updated where possible. The re-analysis confirmed that the recommendations of the CMO's Short Life Working Group have not been implemented widely. This implies that little has changed, that needs are still present and that current service provision at all levels is inadequate.

A Proposed Model of Care for Scotland (Chapter 11)

- The health care needs assessment sets out a recommended series of actions to implement a tiered model of care for Scotland. Where possible, the recommendations for implementing the model draw on existing experiences and expertise across Scotland.

- Services will have to be able to meet the needs of people across the full severity spectrum of ME-CFS. The needs of the most severely affected, especially those who are unable to travel to be in receipt of care must be met. Care must be patient centred with care offered in a range of ways, including domiciliary care.

- The key developments in improving care within Tier 1 of a ME-CFS Service are:
  - providing supporting information to help ME-CFS patients and healthcare professionals understand the condition;
  - implementing care pathways to improve access to diagnosis; and
  - improving access to social and supportive care.

- The key developments in improving care within Tier 2 of a ME-CFS Service are based on the inclusion of ME-CFS as one of the chronic illnesses managed under the long term conditions arrangements. Amongst a number of developments this will include:
  - active case management within the primary care team and access to community health and social care services;
  - implementation of anticipatory care and the use of appropriately developed and quality assured self-management programmes for people with ME-CFS; and
  - improved information to support primary care teams and service users and to address stigma.

- The key developments in improving care within Tier 3 of a ME-CFS Service are:
  - the establishment of medical consultant-led, multidisciplinary teams across Scotland bringing together the range of necessary therapeutic professionals to diagnose, assess and, where needed, treat and manage complex cases; and

  - the establishment of managed clinical networks across Scotland to provide the broad range of medical and therapeutic inputs locally and regionally and – on a national level – to ensure there is a sharing of knowledge, experience and expertise, to provide cross-cover arrangements and to allow for the development of a more focused approach to researching and evaluating treatment interventions.

Infrastructure Issues (Chapter 12)

- The work which has underpinned this needs assessment identified a number of infrastructure issues which should be addressed. These may be broadly characterised as the need to develop effective clinical standards for services, developing education and training for professional staff and widening the research base for ME-CFS.

1 Introduction

Any formal assessment of health care needs in relation to Myalgic Encephalomyelitis – or Encephalopathy – (ME) and the broader spectrum of conditions that have been grouped together under the term Chronic Fatigue Syndrome (CFS) is a challenging exercise. The growing evidence base for both ME and CFS highlights that they are likely to be part of a range of conditions with differing underlying causes that can present in similar ways. However, at this time it is not simple to distinguish clearly between the two, not least because there is no unambiguous diagnostic test (or tests) for either condition. As a result, diagnosis is usually based purely on the patient's symptoms using criteria-based diagnostic tools that have been developed using a blend of research evidence and expert consensus. No criteria-based diagnostic tool has become widely accepted and this fact has, of itself, become an area for controversy. Even the language used to describe the condition has been a source of distress. In the words of one of those people with ME who helped create this assessment:

"The perception that chronic tiredness – or chronic fatigue – is the key defining feature of ME is misconceived and has caused much confusion, to the detriment of patients. Everyone gets tired and experiences fatigue at some time. But no healthy person feels the sort of overwhelming exhaustion and malaise that ME produces. 'Fatigue' is a quite inadequate term to describe it". (Scrutiny Panel Member)

The cause – or causes – of ME remains the subject of ongoing research; however, it can be defined symptomatically by characteristic features that include the incapacitating exhaustion and general, debilitating illness that occurs even after minimal exertion, neurological signs, cognitive and visual impairment, pain and the very real disabilities that are associated with the condition. Historically in the UK, this has given rise commonly to linking ME with CFS under the identifier CFS/ME. However, there are some who argue it should more usefully and appropriately be described ME-CFS, to reflect “ME and CFS”. In the light of this, this health care needs assessment (HCNA) uses the acronym ME-CFS to reflect this distinction.

Despite there being a large and growing body of published peer-reviewed research, there is, as yet, no evidence for a definitive cause of ME-CFS. There are many competing lines of research into the potential aetiologies including neurological deficits, endocrine problems, immunological responses, inflammatory processes, and the role of infections and of genetic predispositions; however, many of these remain in the early stages of exploration due to the under-development of the research effort. It would be wrong, however, to see the absence of definitive research as simply meaning there is insufficient evidence from which to develop a working approach to the care of people with ME-CFS. For example, the existing evidence of physical illness has been used to allow the continued inclusion of ME within the World Health Organisation International Classification of Diseases (Tenth Revision) as a neurological disorder.

Clearly people with ME-CFS do present with a clinical illness in which profound post-exertional exhaustion and malaise is associated with a wide range of disabling symptoms that include gastric problems, dizziness, headaches, sleep disturbances, difficulties with concentration and with cognitive processes, and muscle weakness or pain amongst many others. Not only can there be a high degree of variation between different people presenting with ME-CFS, there can also be a large variation in an individual sufferer’s symptoms that can change in severity and effect over time. At present there is no “cure” for ME-CFS. A small group of individuals seem to make a steady, albeit slow recovery over time. For the majority there is a cycle of remission and relapse associated with ME-CFS that creates a particularly complex set of problems over time. Other patients experience little or no remission or improvement and this includes people who experience a continuous level of severe symptoms bringing extreme disability over many years without significant improvement. Finally, some deteriorate over time. Overall, the mix of physical and cognitive symptoms can be as disabling as multiple sclerosis, rheumatoid arthritis, congestive heart failure or other chronic conditions. As such ME-CFS places substantial pressures not only on the sufferer, but also their families and carers. It also creates a challenge to health care services to effectively identify people with ME-CFS effectively and offer them services which can alleviate their symptoms and improve their overall quality of life.

Because the underlying causes and progression of ME or of CFS are not yet fully understood, they present something of a challenge to undertaking a formal HCNA. There is a lack of epidemiological data on which to base population estimates and identify the full spectrum of severity. Whilst there are clinical guidelines in existence – of which the English National Institute of Health and Clinical Excellence (NICE) guidance is the most recent – the research on which many are based can be argued to be limited in scope, notably with regard to the capacity for some people with ME-CFS to participate (eg the most severely affected). In addition, the evidence for what is effective care is relatively small in comparison to similar guidelines for other conditions. It is clear that very little of the material currently published is generally accepted by all interested parties.

Given the wide range of clinical presentations which are possible for ME-CFS, and the relative lack of research evidence, this HCNA must be seen as a “work in progress”. It provides population estimates based on what epidemiological evidence is available, considers models of care and describes the views of key stakeholders on what can be done to meet needs. What it cannot do is expect to be the final word on the matter.

2 The General Principles of Health Care Needs Assessment

The objective of any Health Care Needs Assessment (HNCA) is to specify services and other activities which impinge on health care relating to a specific disease or diseases.[1] In general, the principal activities involved in HCNA are:

- an assessment of incidence and prevalence;
- an analysis of the effectiveness and/or cost-effectiveness of services; and
- establishing the existing service baseline to help guide service development and redesign.

From these three components, health care planners and commissioners, together with other stakeholders, can determine the policy direction they wish to pursue. There can also be other objectives in HNCA. These might include:

- improving access and the allocation of resources at local, regional and national levels;
- targeting resources at area(s) of highest need; and
- securing the active participation of key stakeholders and players in understanding the need for change and how it can be achieved.

Undertaking such work usually requires a collaborative approach bringing together people with the necessary knowledge base and those with the appropriate technical skills. Broadly speaking, this requires that there is an:

- *Epidemiological Needs Assessment:*
    - incidence and prevalence;
    - effectiveness and cost effectiveness of services; and
    - description of baseline services.

- *Corporate Needs Assessment:*
    - reporting the demands, wishes and alternative perspectives of interested parties, for example service users and their carers, and stakeholders including professional, political and public views.
- *Comparative Needs Assessment*
    - comparing and contrasting the services in the population under study with those provided elsewhere.

Taken together a HCNA should, insofar as there is appropriate data available, describe the capacity of the population to benefit from a service or intervention and to make suggestions as to how such benefits can be delivered. Health care need is not, however, the only important factor in planning and delivering health care. Consideration may be given to, for example, political direction, health care costs, legislation, competing NHS priorities, patient voices and public involvement, professional opinion, scarcity of resources or expertise and the existing pattern of services. Given the NHS is a public-funded institution, it is also important to recognise the importance of population perceptions and the impacts of political processes.

The specific approaches taken in this HCNA are set out in chapter 4.

3 Background and Scope

This HCNA focuses on the needs of adults with ME-CFS. This is in line with the remit agreed by the Scottish Public Health Network with the Scottish Government Health Department. This is not to under-value the needs of children with ME-CFS which were subject to a detailed review undertaken by the UK Royal College of Paediatrics and Child Health in 2004.[2]

The HCNA should be seen within the context of a number of documents that have, over the last six years, contributed towards establishing current policy in Scotland regarding ME-CFS care. These documents have helped to inform the scope for this health care need assessment. In addition, these documents exist within the wider context of policy relating to health and social care in Scotland.

In Scotland, the publication of the report of the Scottish Chief Medical Officer's (CMO) Short-Life Working Group in 2002 provided the first formal statement on the development of services for people suffering from CFS/ME.[3] This report was commissioned and reported after the work of the UK Chief Medical Officer's Independent Working Group on CFS/ME. The remit for the group was specifically set out *"not to revisit the work of the Independent Working Group but to consider what practical steps may be required to take forward its recommendations in Scotland taking into account current knowledge and understanding of CFS/ME."* The Scottish Working Group's report set out nine principal recommendations concerning the development, commissioning and implementation of services for people suffering CFS/ME (See Box 3.1). This report was circulated to NHS Boards under cover of HDL(2003)2.

Box 3.1

Principal Recommendations from the Report of the Scottish Short Life Working Group[3]

- The Short Life Working Group recommends to NHS Boards that local health needs assessment for CFS/ME should form part of ongoing NHS Board programmes. Such needs assessment will provide improved information on the impact of CFS/ME in NHS Board areas.
- On the basis of needs assessment, NHS Boards should develop forward plans, suitable for their local area, on how service provision for CFS/ME might be developed over the next 2 to 3 years.
- In developing plans for service development, partnership between NHS Boards and the Voluntary Sector has a central complementary role. This partnership is especially important in the areas of advocacy and needs assessment.
- Early diagnosis and recognition can make a difference to the outcome of this condition. This will involve appropriate evaluation, effective treatment and follow-up relevant to the severity of the disorder. Integrated community and social support through statutory and voluntary organisations are all necessary.
- The Group recommends a tiered approach to service development building upon care at primary care, specialist local support and where necessary, ability to refer complex cases to more specialised services.
- Care and management should be patient-centred, and delivered locally by multi-agency teams. Care and management plans should be negotiated with the patient and carer, and should involve partner agencies.
- Different care pathways and services need to be developed for groups with particular requirements such as children and young people; the severely affected; housebound and bed-bound patients; and those living in remote or rural areas. Where the patient group is small, such as in remote and rural areas, it may be necessary to develop services for CFS/ME integrated with the management of other chronic diseases, or to work in partnership with other NHS Boards.
- There should be provision for education and training of all professionals, and the development of necessary skills for treating the disorder. Professional supervision is essential to maintain the quality of care.
- Future developments in the care and management of this condition should be informed by ongoing research. As services are developed, opportunities should be taken to develop the necessary research within Scotland.

In 2007 the Scottish Parliament Cross Party Group on ME (CPG on ME) produced a Legacy Report[4], setting out its priorities for a reformed CPG to consult upon within the new Parliament. Those priorities which relate to this HCNA are shown in Box 3.2.

Box 3.2

A Summary of the Scottish Parliamentary Cross Party Group on ME Legacy Report [4]

Points made within the scope of the HCNA:

- A Centre of Excellence for ME – *The CPG on ME would like to see the setting up of a Centre of Excellence for ME which would provide co-ordination of epidemiology, research and strategic needs; dissemination of research and best clinical practice throughout the health service; and monitoring the clinical course of ME in patients.*
- Managed Clinical Network for ME – *Whilst recognising the development of local MCNs in parts of Scotland, the CPG would like to see the development of a Scotland-wide MCN for ME to ensure greater uniformity in access to and quality of appropriate services and dissemination of information. The CPG considers that an effective and dedicated MCN is contingent upon the establishment of a Centre of Excellence.*
- Clinical Guidelines – *The CPG would like to see the development of clinical guidelines that reflect the unique and distinctive clinical presentation of ME and take full account of the growing body of biomedical evidence regarding the basis of this illness. The CPG recommends that the NICE guideline not be adopted in Scotland and that the Canadian guideline be adopted in Scotland.*
- Consultation and Active Participation – *The CPG would like the Scottish Government, NHS Boards and other policy makers to have meaningful consultation with biomedical researchers, people with ME, patient groups and the CPG. Specifically, the CPG expects that the Assessment of Needs of people with ME will involve active and formal consultation and be based upon the unique clinical features and symptom pattern of ME. Clinical Guideline development and the work of a Managed Clinical Network should also be committed to full consultation with people with ME and their carers, including the severely affected and housebound.*
- Education / Training of Health Professionals – *The CPG highlights the need for improved education of all health professionals in the recognition, treatment and management of ME in both training and continuing professional development. There is also a need for raising awareness amongst professionals in education and social services.*

Points beyond the scope the HCNA:

- Biomedical Research – *The CPG would like Government funding of ME research to be prioritised and, if possible, ring-fenced. The CPG would wish the recommendations of the Gibson Report into the status of CFS/ME research be adopted in Scotland.*
- PACE Trial – *The CPG expressed considerable concern regarding the expense, effectiveness and scientific validity of this trial.*
- Welfare Benefits Guidelines – *The CPG would wish the Scottish Government to reject the DWP guidance on ME.*
- Building alliances, understanding and partnerships and raising awareness – *The CPG would seek to continue to work with a wide body of groups and individuals to have a stake in improving the lives of those living with ME.*

As part of the preparation for this HCNA, the Scottish Government Health Department funded an independent scoping study. This was undertaken by Action for ME. This study, published in December 2007, identified 21 areas which should be either a characteristic of, or be specifically considered by, the HCNA[5]. These are shown in Box 3.3. It should be noted that one recommendation – that of seeking to ensure that the recommendations from the HCNA be included in strategic planning and be funded – is beyond the broad scope of a needs assessment exercise.

Box 3.3

A Summary of the Main Recommendations from the Action for ME Scoping Study for the HCNA[5]

The HCNA should provide:

- a clear definition of ME-CFS;
- an outline of prevalence rates;
- a thorough audit of services;
- a view on the role of health professionals in outreach and in primary care, including for those most severely affected with ME-CFS;
- a view on the role of specialist-led multi-disciplinary teams;
- an assessment of treatment options without over-emphasising Cognitive Behavioural Therapy and Graded Exercise;
- a consideration of best practice models from services for other long-term & chronic conditions;
- a view on the role of Managed Clinical Networks and patient pathways;
- a view on the quality, availability and accessibility of information offered to patients;
- a view on the training of GPs and other health professionals;
- a consideration on developments in IT to support telehealth and telecare;
- a view on what can be applied from the Short Life Working Group report;
- a view on how to prioritise ME within NHS Boards; and
- a view on the role of and funding for research in Scotland.

The HCNA should seek to:

- use diagnostic criteria and definition of the illness as its starting point;
- look at international models;
- consider the use of the NICE guideline;
- engage with GPs and health professionals who “deprioritise” ME;
- reach the most severely affected as well as newly diagnosed;
- consult with patients and include their views/perspective; and
- take a patient perspective.

These reports also identify a number of wider policy areas that will need to be considered in undertaking the assessment. These include Better Health, Better Care, the Scottish Government's health and wellbeing action plan[6] which outlines national policy regarding:

- the provision of health care services;
- putting the patient at the centre of care;
- long term conditions management – including self-managed care (where appropriate);
- creating greater integration within community planning partnerships for health and social care; and
- the development of managed clinical networks.

In addition, the application of existing initiatives relating to single shared assessment, supporting carers, and advocacy services to this area are clear.

On the basis of these three reports the broad scope for the HCNA for ME-CFS was identified. This report therefore covers the following major elements:

- the definition of ME-CFS and recommended diagnostic criteria;
- an updated epidemiological statement;
- a mapping of existing services at local, regional and national level;
- recommendations on appropriate models of care which can be adopted by NHS Boards across Scotland; and
- recommendations regarding wider aspects of ME-CFS services to support developments.

It is recognised that this HCNA is a necessary first step in developing a coherent, consistent service throughout Scotland.

4 Approaches Used

The general approach used by the Scottish Public Health Network (ScotPHN) was described in Chapter 2. This highlighted that there is no single, formal approach to undertaking HCNA which must be used in all circumstances. Rather, any HCNA should be undertaken using methods which are appropriate to the types of need being established. Clearly the aim is to use data collection mechanisms that are valid and robust. In general this requires a mixture of qualitative and quantitative data collection techniques.

The specific approaches used for the ME-CFS health needs assessment are outlined below.

4.1 The Epidemiological Assessment:

The purpose of this element of the HCNA was to establish the descriptive epidemiology of ME-CFS. It is important to note that there is no robust, epidemiological study which directly relates to Scotland. Indeed, this is a recognised lack within the wider context of ME-CFS research.

To address this, a brief review of the research literature was undertaken to identify new UK or international studies, published since 2003 which could be used to inform an updated epidemiological statement. Unfortunately, little epidemiological research would seem to have been published during this period. As a result, the basis of the descriptive epidemiology presented here is an update on that previously published in the CMO's Short-Life Working Group in Scotland[3] augmented by data specially commissioned from the UK ME Observatory.

In addition to the literature review for epidemiological studies, data from more local data collection systems were identified and reviewed for robustness. Where appropriate, this material has been used to help refine the estimates derived. This has allowed some insight into the likely use of primary care and specialist assessment services. The data used was drawn from a number of sources. Where available, existing data from within Scotland were used to describe the prevalence and consequence of ME-CFS. These data have been used to make estimates of the number of people with ME-CFS and the potential service demands that could occur.

The descriptive epidemiology is presented in Chapter 6.

4.2 The Corporate Assessment

The purpose of the corporate need assessment is to identify the varying needs, demands, wishes and alternative perspectives of interested parties and stakeholders. This includes professional and service user views as well as political and public views (where appropriate).

4.2.1. Identifying ME-CFS Service User Needs

The HCNA used three main approaches to identifying the needs of those with ME-CFS. Firstly, to seek the views of people with ME-CFS, ScotPHN commissioned Action for ME to undertake an independent, qualitative assessment. This work was undertaken via a series of three focus groups attended by 28 people with ME-CFS who were able to travel to the chosen venues in Glasgow and in Fife. In addition to this, telephone interviews were undertaken with a small group (n=4) of those most severely affected by ME or CFS. Younger people affected by the illness were interviewed (n=6) by the Association of Young

People with ME (AYME). The questions used during the focus groups and for the interviews are shown in Box 4.1.

Box 4.1:

Questions used during Focus Groups and Telephone Interviews

1) In your experience, and in general, what were the main health needs when you first became ill?
2) What are your health needs when trying to manage your ME?
3) Thinking back to when you were first diagnosed, was there anything that helped you and might help other people in a similar situation?
4) Once diagnosed with ME has there been any services or treatments that helped or would have been helpful if available to you? and
5) What information would you have liked to have received when you first became ill, or since, to help you manage your condition?

Secondly, an open Stakeholder Day for service users and those stakeholder organisations representing them was organised. This took place on the 23 June 2008. Appendix 1 contains the names of all those organisations that were invited to send representatives and a list of attendees. In total, some 37 people attended this day which was facilitated by members of ScotPHN and the Project Group for the HCNA. The people who attended this day considered three main questions:

1) how should we define ME-CFS?
2) what services are needed by people with ME-CFS? and
3) what good practice is already there that we can build upon?

A note summarising the output from this Stakeholder Day is posted on the ScotPHN website (see: www.scotphn.net). Finally, details from the unpublished review of patients needs undertaken in Lothian – “Believe in ME”[7] – were incorporated, as were those from the Action for ME Scoping Study[5].

4.2.2 Identifying Service Providers' View of Needs

The description of the baseline services are drawn from individual returns by health board area which were collected by Action for ME for their Scoping Study[4]. In addition a workshop day was held to elicit the views of service providers. This workshop, which was held on the 9 June 2008, was attended by 15 professionals involved in service provision representing a broad range of clinical expertise in the NHS and from the Voluntary Sector. The attendees considered the same questions as asked of those attending the Stakeholders' Day. A list of those attending is contained within Appendix 1 and, as before, a note summarising the output from this service providers' workshop is posted on the ScotPHN website (See: www.scotphn.net).

The expressed needs of both service users and providers are described in chapter 7.

4.3 The Comparative Assessment

The final component of the HCNA – that of comparing and contrasting services provided elsewhere to inform the development of those for the population under study – has been undertaken using two approaches.

The first approach was to undertake a specific literature review to identify key themes in service provision. Details of this literature review are set out in Appendix 2. The second involved a member of the ScotPHN team undertaking a site visit to one of the fourteen Clinical Network and Co-ordination Centres established in England to develop and provide services for people with ME-CFS. The centre chosen was in Bristol and supported both NHS and voluntary sector services across the northern sector of the English south west. A description service provided from this centre is available from the ScotPHN website (see: www.scotphn.net) or at: http://www.nbt.nhs.uk/services/criticalcare/cfs-me/).

The findings are described in chapters 8 and 9.

5 Defining ME and CFS

5.1 Criteria-based definitions

Fatigue – even long lasting fatigue – is often a symptom of an underlying illness. When the fatigue is chronic and is linked with other symptoms in a specific pattern, then it is possible to describe chronic fatigue syndromes (CFS). However, this is a very loose term and may cover a number of possible illnesses in which chronic fatigue is a feature. These include myalgic encephalomyelitis (or encephalopathy), post-polio syndrome, post viral fatigue syndrome, and some forms of fibromyalgia.

Perhaps the only incontrovertible aspect of defining ME and CFS is that agreement is elusive. Whilst this is largely a consequence of historical debate, the continued failure to recognise the importance of the wider research base and the many, differing clinical opinions regarding aetiology all continue to exert a divisive influence on the subject.

The term ME was first used in the 1950s. Current use of the term ME conforms largely to the spirit of this early use of the term and includes the neurological aspects of the condition. In 1969 WHO classified ME as a neurological disease and this is still included in the ICD-10 code as an alternative term for postviral fatigue syndrome (G93.3) and is classed as a "Disorder of the Brain". There are those who argue that the inclusion of neurasthenia (increased fatigue associated with mental effort) in the ICD 10 classification (F48.0) provides an opportunity for some to maintain a psychiatric origin for CFS. However, this description of a fatigue syndrome can lead to misunderstanding. Neurasthenia is not specifically characterized by *chronic* fatigue and should not be confused with ME-CFS.

In 1988, the US Centers for Disease Control (CDC) developed a working case definition[8] which introduced the term Chronic Fatigue Syndrome to replace the term “chronic Epstein-Barr virus syndrome”. In the UK, psychiatrists developed their own criteria that omitted the minor symptoms of the CDC definition creating a less strict definition and effectively resulting in a more heterogeneous population. Published in 1991 and known as the Oxford criteria, this has been criticised continually for its focus on fatigue and comparative lack of consideration of other physical symptoms[9]. In 1994, CDC revised its definition of CFS to adopt a broader definition. This emphasised fatigue as the key symptom. This definition – called the ‘Fukuda’ definition after its lead author – has become the standard, international criteria for research purposes[10]. This was further revised in 2003 to clarify inherent ambiguities in the 1994 version.

The current, working definition of CFS/ME in Scotland would seem to be the 1994 version of the CDC definition. This was used by the CMO’s Short-term Working Group on CFS/ME in defining the disease[8]. This definition requires that a diagnosis of CFS/ME should only be made when persistent or relapsing fatigue is present for a minimum of six months, in combination with a range of other symptoms and signs. Whilst this definition commands wide international recognition, and provides a framework in which CFS/ME is determined across populations, it remains essentially a definition for the purposes of research. Using such an approach diagnosis is based on clinical assessment of a combination of symptoms having excluded other possible diagnoses. Rather than adopting this definition – or its 2003 revision – without challenge, this HCNA, has undertaken a review of all the main definitions of ME-CFS. This review has also taken into account the work of NICE in England and Wales which proposed a variant on the CDC definition and introduces a wider range of physical symptoms which may be experienced by people with ME-CFS not contained within the CDC definition. Table 5.1 provides a comparison of all the main diagnostic definitions.

The review undertaken for this HCNA highlighted two further sets of definitions/diagnostic criteria from the 1990s, one Australian[11] and one from the UK[12]. However, these are variants on a theme and would seem to add little to the broader definition set out by the CDC (1994) definition.

In Canada, in 2003, Carruthers et al published a clinical guideline based on the views of an expert panel seeking consensus around a clinical case definition[13]. This definition – and a subsequent Australian guideline for primary and community health care professionals[14] - seeks to provide a clinical case definition for ME and CFS which emphasises the neurological features of the condition and the post-exertion fatigue / malaise which more psychiatrically-based definitions under-emphasise (see box 5.1). The Carruthers et al guidance – usually referred to as the Canadian Guideline – was supported by the Westminster Parliamentary Group on Scientific Research into Myalgic Encephalomyelitis (The Gibson Report)[15] as well as the CPG of the Scottish Parliament on ME (Legacy Paper)[4].

However, whilst such an approach can help identify the clinical features of ME, it can also have the effect of excluding other forms of CFS which do not display the wider neurological features. The consequence of this may be to exclude those who still require investigation and sign-posting towards appropriate assessment and – where appropriate – intervention. This concern was reflected in the broader definition adopted by the NICE CFS/ME guidelines published in England and Wales in 2007[16]. The NICE definition also suggests that a degree of debilitation in maintaining ordinary, activities of everyday living be required for a diagnosis of CFS/ME to be made. Whichever definition – or definitions – is used, it is clear that they need to be capable of allowing all patients to access appropriate assessment without implying a specific treatment regime is required.

5.2 Corporate views on defining ME-CFS

The issue of definition was considered by the two workshop groups who were asked to reflect on which of the two main definitions – the Canadian Guideline definition and the NICE definition – was preferable. These were considered specifically in response to the recommendations of the Action for ME Scoping Study[5]. In addition the workshops were asked to consider the possibility of adopting – as is the case in other areas of clinical practice – both definitions: the Canadian definition as a definition of ME and the NICE definition as a definition of CFS.

Within the service providers' workshop there was a robust discussion on whether a two definition approach was feasible or desirable. Overall, it was agreed that any definition should steer towards a clinical requirement rather than research criteria and that inclusive assessment criteria need to be in place. It was felt that any definitional approach should help towards specialist investigation, where this was required, as well as help and support. Those who supported the Canadian approach emphasised the benefits of a pathogenic definition which steers away from a fatigue-based definition, though there were some who felt that such a symptom-based definition could be useful if a test of severity or energy index could be incorporated into the definition.

However, it was noted that as there was no positive test for ME, the majority of ME diagnoses and those of CFS are currently made through the exclusion of other illnesses. This could therefore have the unintended consequence of increasing delay to diagnosis for some individuals. It was also seen as a potential problem that, for individuals where chronic fatigue was the *only* symptom of concern, this could exclude people with ME from appropriate, further investigation. Overall, it was agreed that broader diagnostic criteria could allow access to generic services such as secondary care and advice as well as community options eg community support or physiotherapy.

Within the stakeholders' workshop there was almost a universal view that only a definition based on the diagnostic criteria set out in the Canadian guidelines should be adopted. This was felt to be preferable as it emphasised the neurological symptoms and provided a stronger definition of ME. Some felt that such a definition was sufficient to cover all types of CFS, but this was not generally accepted. There was some recognition that this could result in the exclusion of some people with CFS from accessing assessment and receiving appropriate services.

5.3 ME and CFS – definitions for Scotland

Overall, it was the view of both the Project Group and its Steering Group that the review of evidence for definition and the discussion at the two workshops did not achieve a consensus. A pragmatic solution is therefore proposed with separate definitions for ME and for CFS.

Recommendation 1:

It is recommended that the clinical, symptomatic definition of ME outlined in the Canadian Guideline be adopted in Scotland (See Box 5.1).

Recommendation 2:

It is further recommended that a symptomatic definition of CFS based on that proposed in the NICE guideline be adopted in Scotland (See Box 5.2).

It is hoped that this pragmatic approach will allow clinicians to adopt an approach to diagnosis that can ensure that all those individuals for whom CFS exists are identified as rapidly as possible and also allow for more focused assessment and review to confirm a diagnosis of ME, where appropriate. Diagnostic issues are more fully explored in the Scottish Good Practice Statement on ME-CFS (SGPS)[1]:

http://www.show.scot.nhs.uk/GoodPracticeStatementonME-CFSforGeneralPractitioners.

Box 5.1
CANADIAN CONSENSUS CRITERIA FOR USE IN DIAGNOSING ME[13]

A patient with ME/CFS will meet the criteria for fatigue, post-exertional malaise and/or fatigue, sleep dysfunction, and pain; have two or more neurological/cognitive manifestations and one or more symptoms from two of the categories of autonomic, neuroendocrine, and immune manifestations; and adhere to item 7.

1. Fatigue: the patient must have a significant degree of new onset, unexplained, persistent, or recurrent physical and mental fatigue that substantially reduces activity level.

2. Post-exertional malaise and/or fatigue: there is an inappropriate loss of physical and mental stamina, rapid muscular and cognitive fatigability, post exertional malaise and/or fatigue and/or pain and a tendency for other associated symptoms within the patient's cluster of symptoms to worsen. There is a pathologically slow recovery period - usually 24 hours or longer.

3. Sleep dysfunction: there is unrefreshed sleep or sleep quantity or rhythm disturbances such as reversed or chaotic diurnal sleep rhythms.

4. Pain: there is a significant degree of myalgia. Pain can be experienced in the muscles, and/or joints, and is often widespread and migratory in nature. Often there are significant headaches of new type, pattern or severity.

5. Neurological/cognitive manifestations: Two or more of the following difficulties should be present: confusion, impairment of concentration and short-term memory consolidation, disorientation, difficulty with information processing, categorizing and word retrieval, and perceptual and sensory disturbances – e.g. spatial instability and disorientation and inability to focus vision. Ataxia, muscle weakness and fasciculations are common. There may be overload phenomena: cognitive, sensory –

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[1] The Scottish Good Practice Statement on ME-CFS (SGPS) was finalised following completion of the HCNA consultation. The HCNA acknowledges that the SGPS is the recognised clinical guidance on the diagnostic approach to ME-CFS. This is important as the SGPS also addresses the concerns raised by the Scottish Neurosciences Council regarding the use of the Canadian Consensus Document as a diagnostic tool.

e.g. photophobia and hypersensitivity to noise - and/or emotional overload, which may lead to ‘crash’ periods and/or anxiety.

6. At least one symptom from two of the following categories:

   a. Autonomic manifestations: orthostatic intolerance - neurally mediated hypotension (NMH), postural orthostatic tachycardia syndrome (POTS), delayed postural hypotension; light-headedness; extreme pallor; nausea and irritable bowel syndrome; urinary frequency and bladder dysfunction; palpitations with or without cardiac arrhythmias; or exertional dyspnea.

   b. Neuroendocrine manifestations: loss of thermostatic stability – subnormal body temperature and marked diurnal fluctuation, sweating episodes, recurrent feelings of feverishness and cold extremities; intolerance of extremes of heat and cold; marked weight change - anorexia or abnormal appetite; loss of adaptability and worsening of symptoms with stress.

   c. Immune manifestations: tender lymph nodes, recurrent sore throat, recurrent flu-like symptoms, general malaise, new sensitivities to food, medications and/or chemicals.

7. The illness persists for at least six months: It usually has a distinct onset, although it may be gradual. Preliminary diagnosis may be possible earlier. Three months is appropriate for children.

   The symptoms must have started after, or have been significantly altered after, the onset of the illness. It is unlikely that a patient will suffer from all symptoms in criteria 5 & 6. The disturbances tend to form symptom clusters that may fluctuate and change over time.

   Children often have numerous prominent symptoms but their order of severity tends to vary from day to day. There is a small number of patients who have no pain or sleep dysfunction, but no other diagnosis fits except ME/CFS. A diagnosis of ME/CFS can be entertained when this group has an infectious illness type onset. Some patients have been unhealthy for other reasons prior to the onset of ME/CFS and lack detectable triggers at onset or have more gradual or insidious onset.

Box 5.2
NICE CRITERIA FOR USE IN DIAGNOSING CFS[16]

Healthcare professionals should consider the possibility of ME/CFS if a person has fatigue with all of the following features:

- new or had a specific onset (that is, it is not lifelong)
- persistent and/or recurrent
- unexplained by other conditions
- has resulted in a substantial reduction in activity level
- characterised by post-exertional malaise and/or fatigue (typically delayed, for example by at least 24 hours, with slow recovery over several days)

and one or more of the following symptoms:

- difficulty with sleeping, such as insomnia, hypersomnia, unrefreshing sleep, a disturbed sleep–wake cycle
- muscle and/or joint pain that is multi-site and without evidence of inflammation
- headaches
- painful lymph nodes without pathological enlargement
- sore throat
- cognitive dysfunction, such as difficulty thinking, inability to concentrate, impairment of short-term memory, and difficulties with word-finding, planning/organising thoughts and information processing
- physical or mental exertion makes symptoms worse
- general malaise or ‘flu-like’ symptoms
- dizziness and/or nausea
- palpitations in the absence of identified cardiac pathology.

A diagnosis should be made after other possible diagnoses have been excluded and the symptoms have persisted for:

- 4 months in an adult
- 3 months in a child or young person (the diagnosis should be made or confirmed by a paediatrician)

Table 5.1 Comparison of Diagnostic Criteria

Key criteria **Must be present**	US CDC 1988	Australian (Lloyd) 1990	UK Oxford 1991	US CDC 1994	UK London 1994	Canadian (Carruthers) 2003	Australia 2004 Management	UK NICE 2007
Fatigue	√	√	√	√	√	√	√	√
Definite/new onset – not life long	√		√	√		√		√
Severe & disabling fatigue affecting physical and mental functioning	√		√			√	√	
Should have been present for =>6months and for more than 50% of that time	√	√	√	√	√	√ (3m in children)		=>4m (3m in children)
Additional criteria related to fatigue				Plus =>4 symptoms required from following categories				
No improvement with bed rest	√			√				√
Post-exertional or exacerbated by minor exercise		√		√	√	√	√	√
Causes significant disruption of usual, daily activities		√		√				√
Not the result of exercise/exertion				√				√
Prolonged recovery	√				√	√		√

Other Criteria	Plus =>6 symptoms required from following categories inc 9 above)		Not required to be present					Plus =>1 symptoms required from following categories
Sleep disturbance [or unrefreshing sleep]	√		√	√		√	√	√
Myalgia	√		√	√		√	√	√
Pain [other than myalgia eg headaches; lymph nodes; throat]	√			√		√	√	√
[Migratory] arthralgias	√			√		√		√
						Additional symptoms – required in combination		
Neuropsychiatric dysfunction	√	√						
Cognitive dysfunction						√	√	√
New onset short term memory impairment		√		√		√		√
Mood disturbance			√					
Neurological disturbances					√	√		
Variable involvement of cardiac & other bodily systems					√			
Extended relapse course with tendency to chronicity					√			
Marked variability in course of a day					√			
Dizziness/nausea						√	√	√
Orthostatic intolerance						√	√	
Palpitations						√		√
Gastro-intestinal symptoms (e.g. irritable bowel)						√	√	

6 Epidemiology

The absence of a diagnostic test for ME-CFS linked to the differences highlighted in the clinical guidelines, has made describing the epidemiology of ME and of CFS problematic. This is an international problem which is yet to find an adequate resolution. For Scotland, the Chief Medical Officer's Short Life Working Group on CFS/ME concluded:

> *"The precise number of people in Scotland affected by CFS/ME is unknown at present. No published studies on the incidence and prevalence are yet available for the Scottish population though a number of local studies have been carried out in some NHS Board areas. In the absence of this, reliance must be placed on published information."*[2]

As noted above, the review of the epidemiological research literature published since 2003 has highlighted that there has been no study specific to the Scottish population undertaken. The establishment of a virtual ME Observatory in the UK may do much to change this, though at present there is no Scottish university participating in this initiative. The following sections therefore are estimates based on the best available evidence from international research studies or other sources applied to Scottish populations.

6.1 The Estimated Prevalence of ME-CFS in Scotland

Deriving accurate estimates of the prevalence of ME-CFS in Scotland is difficult. The lack of a suitable definition which can be used for epidemiological studies, linked to the issues around diagnostic accuracy, make any estimates subject to error. These effects have been carefully explored in the research literature[17] and could reflect as much as a five-fold variation in prevalence. The applicability of any studies undertaken outside of Scotland can also be a source of error. As a result, any estimates must be viewed with a degree of caution.

It should also be noted that none of these estimates is based upon the definitions which are recommended for adoption in the HCNA.

Both the Short Life Working Group report from 2003[2] and the more recent guidance from the National Institute of Health and Clinical Excellence (NICE) from 2007[12] cite the overall prevalence of ME-CFS in the adult population at between 2 per 1000 and 4 per 1000. This is an estimated range based on published research studies from across the world and considered to provide at least an estimate within the correct order of magnitude.[18] Of these, it is estimated that 1 in 4 people with ME-CFS will be severely affected[3] (i.e. house bound or bed bound).

Table 6.1 shows the estimated number of people in Scotland by NHS Board area with ME-CFS. The table also shows an impact of population change over the next 10 years with projections of the numbers of people with ME-CFS. In all cases these estimates are based on a 2008 population base.

Table 6.1; Estimated and Projected Number of People with ME-CFS for 2008, 2013, 2018. Adults aged 18y and over

Health Board	2008				2013				2018			
	Population	Prevalence			Population	Prevalence			Population	Prevalence		
		Lower Estimate (0.2%)	Mid Estimate (0.3%)	Higher Estimate (0.4%)		Lower Estimate (0.2%)	Mid Estimate (0.3%)	Higher Estimate (0.4%)		Lower Estimate (0.2%)	Mid Estimate (0.3%)	Higher Estimate (0.4%)
Ayrshire & Arran	292,850	586	879	1,171	293,715	587	881	1,175	293,608	587	881	1,174
Borders	89,448	179	268	358	92,679	185	278	371	95,569	191	287	382
Dumfries & Galloway	119,662	239	359	479	120,341	241	361	481	120,503	241	362	482
Fife	287,601	575	863	1,150	294,307	589	883	1,177	301,149	602	903	1,205
Forth Valley	227,909	456	684	912	234,124	468	702	936	240,548	481	722	962
Grampian	431,065	862	1,293	1,724	444,281	889	1,333	1,777	455,768	912	1,367	1,823
Greater Glasgow & Clyde	955,688	1,911	2,867	3,823	958,487	1,917	2,875	3,834	956,695	1,913	2,870	3,827
Highland	247,897	496	744	992	254,148	508	762	1,017	260,206	520	781	1,041
Lanarkshire	439,699	879	1,319	1,759	445,795	892	1,337	1,783	451,863	904	1,356	1,807
Lothian	658,684	1,317	1,976	2,635	686,179	1,372	2,059	2,745	712,065	1,424	2,136	2,848
Orkney	15,853	32	48	63	16,240	32	49	65	16,703	33	50	67
Shetland	17,069	34	51	68	17,102	34	51	68	16,963	34	51	68
Tayside	318,386	637	955	1,274	326,569	653	980	1,306	333,354	667	1,000	1,333
Western Isles	20,996	42	63	84	20,731	41	62	83	20,646	41	62	83
Scotland	4,122,807	8,246	12,368	16,491	4,204,697	8,409	12,614	16,819	4,275,639	8,551	12,827	17,103

Table 6.2; Estimated Number of Males and Females with ME-CFS for 2008 by Selected Age-groups. Adults aged 18y and over. Mid 2008 based.

Health Board	Males						Females					
	18-29	30-39	40-49	50-59	60+	18+	18-29	30-39	40-49	50-59	60+	18+
Ayrshire & Arran	8	22	59	74	32	195	33	111	293	354	183	974
Borders	<5	7	19	24	11	63	8	33	92	109	58	300
Dumfries & Galloway	<5	8	24	32	15	82	11	40	116	149	82	399
Fife	9	24	59	70	29	190	37	113	279	331	168	928
Forth Valley	7	19	49	55	22	152	28	98	232	262	126	745
Grampian	13	40	92	112	42	300	53	174	414	495	229	1,365
Greater Glasgow & Clyde	34	86	191	216	82	609	142	403	957	1,032	506	3,040
Highland	6	19	52	68	29	175	23	90	244	312	155	823
Lanarkshire	13	39	93	107	41	292	55	189	457	514	239	1,455
Lothian	23	64	131	145	56	419	105	297	628	684	326	2,041
Orkney	<5	<5	<5	<5	<5	11	<5	6	16	20	10	52
Shetland	<5	<5	<5	5	<5	12	<5	7	16	20	9	55
Tayside	10	24	61	77	35	206	40	115	302	365	198	1,021
Western Isles	<5	<5	<5	6	<5	15	<5	8	19	25	14	68

Data relating to the age and sex distribution of ME-CFS was published in 2007 in a study from the US[19]. This suggests that ME-CFS is up to four times more common amongst women than men and that people in their mid-life (aged 40 to 59) are more likely to suffer from the disease. Table 6.2 shows age and sex specific estimates based on this study, adjusted for the estimated 2008 population in Scotland. It should be noted that these estimates are somewhat crude as – in the absence of Scottish data on the sex-specific prevalence – this has been held constant at 4 per 1000 population for both males and females. This will have the effect of over-estimating the number of women in older age groups in Scotland due to the greater proportion of older Scottish women compared to the US population on which the estimates are based[18].

Data from the US Centers for Disease Control (CDC) suggest that only 1 in 5 US citizens with ME-CFS will have been formally diagnosed[20]. There is no comparable data from the UK on which to base an estimate of under-diagnosis.

6.2 Estimates of symptomatic presentation and disease course

There is very little epidemiological data available relating to the types of symptomatic presentations which are common in ME-CFS. The CDC suggests that, in addition to the symptoms that define a diagnosis of ME-CFS, between 20% and 50% of people with diagnosed ME-CFS will have additional presenting symptoms. These include abdominal pain, alcohol intolerance, bloating, chest pain, chronic cough, diarrhoea, dizziness, dry eyes or mouth, earaches, irregular heartbeat, jaw pain, morning stiffness, nausea, night sweats, psychological problems (depression, irritability, anxiety, panic attacks), shortness of breath, skin sensations, tingling sensations, and weight loss. Epidemiological evidence relating to the severity of ME-CFS presentations is also limited. The reported prevalence of severely affected individuals at 25% of all sufferers does not take into account that many more individuals may be severely affected at some stage in their illness.

The Canadian Guideline Expert Group, in reviewing a wider range of published research, suggests that recovery from ME-CFS to pre-illness levels of functioning occurs in under 10% of patients[13]. The degree to which improvement can occur – albeit to less than pre-illness levels of functioning – is open to debate. A recent study of patient experiences in the Edinburgh area provides some insight into the health status of people with ME-CFS. This study showed that those people with ME-CFS described the progress of their illness more in terms of learning coping strategies that helped improve their quality of life and general functioning, rather than using the term recovery[7].

6.3 Estimates of current service use

Identifying accurate data for service use by people with ME-CFS in Scotland presents difficulties. The potential for under diagnosis, linked to the difficulties noted above relating to diagnostic criteria, make any recorded episodes of care likely to be an underestimate. However, some limited data relating to GP attendances, specialist assessment services and elements of wider, social support have been identified.

6.3.1 GP attendance

Data from the Primary Team Information programme, formerly the Continuous Morbidity Register, which produces data relating to attendances (or consultations) in primary care has been analysed to estimate the number of people recorded with a diagnosis of CFS. An attendance is recoded for seeing any member of the practice team. This is shown in Table 6.3.

Table 6.3 Estimated number[2] of ME-CFS patients consulting in General Practice in Scotland[3] and annual consultation rates per 1,000 population by age group[4] in Scotland; Financial year ending 31 March[5]; by age group

	2003/04		2004/05		2005/06	
Age Group	**Estimated No. of Patients Consulting**	**Consultation Rates**	**Estimated No. of Patients Consulting**	**Consultation Rates**	**Estimated No. of Patients Consulting**	**Consultation Rates**
0-14	150	0.2	250	0.3	150	0.2
15-24	650	0.9	700	1.0	450	0.6
25-34	1,100	1.5	900	1.3	1,150	1.6
35-44	1,750	2.1	1,450	1.7	1,550	1.8
45-54	1,800	2.5	1,600	2.2	1,400	1.9
55-64	1,250	2.1	1,150	1.9	1,250	1.9
65-74	450	0.9	350	0.8	450	0.9
75+	150	0.4	100	0.2	100	0.3
All Ages[6]	7,350	1.4	6,500	1.2	6,450	1.2

1 - Defined by the Read Code 'F286' Chronic Fatigue Syndrome (ME).
2 - Estimates are rounded to the nearest 50.
3 - Includes contacts with a GP, practice nurse, district nurse, health visitor.
4 - Population source: Community Health Index (CHI) as at 30 September 2004, 2005 and 2006.
5 - Based on 45, 44 and 44 practices that submitted complete PTI data for the years ending March 2004, 2005 and 2006. Figures are standardised by gender and deprivation.
6 - Totals may not equal sum of parts due to rounding.

It is of interest that these data would confirm for Scotland the observed higher proportion of ME-CFS sufferers amongst those in their middle years, as identified in the US study. However, it is also possible that this observation is explained by under diagnosis in other age groups.

Care should also be taken with these data as the Read Code used (F286) is defined as chronic fatigue syndrome (including ME). Taken together, these may mean that these figures are likely to be an underestimate of GP attendances. If this is the case, then it is probable that implementation of the SGPS will have an impact on these figures.

6.3.2 Specialist assessment services

NHS Lothian maintains a specific, specialist diagnostic assessment and review clinic for people with ME-CFS. Access to this clinic is governed by a specific, ME-CFS protocol[21] that has been agreed with GPs and is routinely available to them via the local RefHelp system which facilitates GP referral to specialist services. This use of this protocol provides some degree of confidence that the data derived from this clinic is diagnostically consistent. In 2006, the most recent year for which we have full data suitable for comparison with the foregoing estimates, the ME-CFS clinic saw 180 new patients for assessment. During the year, there were a further 269 review patients seen, accounting for 441 out-patient attendances. This allows crude estimates of new assessments in the adult population of 2.8/10,000 and for review attendances of 6.8/10,000. This data has been used to estimate the number of "assessments" undertaken and review attendances that could be expected in each Health Board area. These estimates are shown in Table 6.4. However, this has been considered by the clinic to be an underestimate of the true number of individuals seen with ME-CFS who are being seen across the full range of clinical services in Lothian. Data collected during the first half of 2010 would suggest that there may be upwards of 196 new assessments and 525 review attendances. This would suggest crude estimates of 3.0/10,000 new assessments and 8.0/10,000 review attendances. The impact of these half-year estimates is also shown in Table 6.4. Whilst this is a potentially more accurate estimate of activity, it is known that there are a number of individuals referred to the clinic who do not attend.

Table 6.4 Estimated number of New ME-CFS Assessments and Review Attendances at a Specialist Assessment and Management Clinic for 2008, 2013, 2018. Adults aged 18y and over. Mid 2008 based

Health Board	2006 Activity Based Estimates						2010 Activity Based Estimates					
	2008		2013		2018		2008		2013		2018	
	New	Review	New	Review	New	Review	New	Review	New	Review	New	Review
Ayrshire & Arran	82	199	82	200	82	200	88	234	88	235	88	235
Borders	25	61	26	63	27	65	27	72	28	74	29	76
Dumfries & Galloway	34	81	34	82	34	82	36	96	36	96	36	96
Fife	81	196	82	200	84	205	86	230	88	235	90	241
Forth Valley	64	155	66	159	67	164	68	182	70	187	72	192
Grampian	121	293	124	302	128	310	129	345	133	355	137	365
Greater Glasgow & Clyde	268	650	268	652	268	651	287	765	288	767	287	765
Highland	69	169	71	173	73	177	74	198	76	203	78	208
Lanarkshire	123	299	125	303	127	307	132	352	134	357	136	361
Lothian	184	448	192	467	199	484	198	527	206	549	214	570
Orkney	4	11	5	11	5	11	5	13	5	13	5	13
Shetland	5	12	5	12	5	12	5	14	5	14	5	14
Tayside	89	217	91	222	93	227	96	255	98	261	100	267
Western Isles	6	14	6	14	6	14	6	17	6	17	6	17
Scotland	1,154	2,804	1,177	2,859	1,197	2,907	1,237	3,298	1,261	3,364	1,283	3,421

2006 Activity based estimates for new assessments estimated at 2.8/10000 and review attendances at 6.8/10000 adult population

2010 Activity based estimates for new assessments estimated at 3.0/10000 and review attendances at 8.0/10000 adult population

6.3.3 Benefits Agency data

Data relating to those individuals in receipt of Incapacity Benefit from the Benefits Agency and for whom ME or CFS is given as a reason for the incapacity have been identified. This data – which is only available for Scotland as a whole – is based on a 5% sample of all Department of Work and Pensions claims that are analysed on a quarterly basis. Quarterly data for the 5 year period August 2002 to May 2007 has been analysed. This suggests that in a three month period, there will be some 200 people in receipt of incapacity benefit with the given reason being ME and 400 with the given reason being CFS.

It is difficult to interpret this data further. Whilst it clearly indicates that these are individuals for whom their condition makes it impossible for them to work, it is not a true indicator of severity of the condition. Given the known problems around diagnosis, there will be individuals who are unaware that they may be eligible for this benefit. As eligibility for Incapacity Benefit includes a medical assessment, this may also be problematic for some in convincing assessors of the reality of the condition. As a result it is likely that this is an underestimate of those who are unable to work as a result of ME or CFS.

6.4 Conclusions

The absence of an accurate case-definition for ME-CFS has made describing the epidemiology of ME and of CFS problematic. This is an international problem which is yet to find an adequate resolution. A review of the epidemiological research literature published since 2003 has highlighted that no study specific to the Scottish population has been undertaken. Recent guidance cites the overall prevalence of ME-CFS in the adult population at between 2 per 1000 and 4 per 1000. This is an estimated range based on published research studies from across the world and considered to provide a “best estimate". Of these, it is estimated that 1 in 4 people with ME-CFS will be severely affected[3]. There is very little data on service use, whether within primary care or from specialist providers. What material is available is often prone to problems of definition.

Recommendation 3:

a) There is an urgent need for a sound epidemiological study of ME and CFS in Scotland; in which regard consideration should be given to including ME and CFS within the Scottish Health Survey.

b) Routine reporting of ME and CFS should be considered within the context of developing information systems for Long Term Conditions monitoring under the Quality and Outcomes Framework (QOF).

7 Service Users' and Service Providers' Views of Need

In this section, the views of service users and of health care professionals who provide services for people with ME-CFS in Scotland regarding health needs is described.

7.1 Qualitative Data Analysis

As noted in chapter 4, the views of service users, their carers, and service providers were identified using a number of methods. These included for service users and carers a workshop, three focus groups and telephone interviews. For the service providers a workshop was also held. In addition, a review of relevant documentary evidence was undertaken. This included the recently completed review undertaken in Lothian of service users' views[21].

These provided a wealth of material which has been subject to a thematic analysis to identify key expressions of need. To structure this analysis, the tiered approach outlined within the CMO's Short-Life Working Group was adopted[2]. This considers three tiers of service: Tier 1 relating to the general services needed by people in the community; Tier 2 concerning the provision of services within primary care; and Tier 3 describing specialist services/specialist expertise that is specific to ME-CFS. A slightly updated version that takes into account the current use of the term "long term conditions" rather than the older phrase "chronic disease management" is reproduced in Appendix 3.

The thematic analysis captured the expressed needs from three specific groups of people with ME-CFS. These groups were adult service users; adults who are most severely affected; and younger adults who are in transition from children's to adult care services. Given the very broad spectrum of symptoms that can be experienced by people with ME-

CFS, it is perhaps surprising that the views of need expressed by service users were broadly consistent. Whilst there are some differences between the three groups of service users, the themes which emerged from the data identified a series of specific, expressed needs.

It is also notable that these themes were also consistent with the views of the service providers, though it should be recognised that whilst the service users often expressed their needs in terms of what they wanted of services, the service providers expressed their views of need in terms of types of configuration of services or the outcomes of care.

7.2 Expressed Need at Tier 1

(Relating to the general services needed by people in the community)

At Tier 1, almost universally, for service users the need for information, social support and access to services which maintain activities of everyday living both for the individual and their families / significant supporters was identified. Specific needs identified included:

- information about the illness and the range of services available;
- access to employment support;
- disability support;
- benefits advice; and
- domestic support (eg meals on wheels, home help etc).

The need that was being characterised was broadly similar to that of individuals with community care needs or who have other long-term conditions. Whilst such services were clearly being described as having been available in some areas, this was not universal. Moreover, in the absence of a diagnosis of ME or CFS, access to these services and support can be more difficult as the route to these services via a Single Shared Assessment is not put in place. This requires effective Tier 2 responses. In some cases the access to such services was dependent on medical assessments provided outside the NHS.

For people who were more severely affected, the degree of expressed need was higher. For them, in addition to the areas already mentioned, Tier 1 support should include specific help with personal care areas and everyday daily activities. The need for support for informal carers was also highlighted. What was particularly notable in relation to this group was the relative lack of connection to social care or to voluntary sector support services (over and above ME charities or self-help groups).

For younger adults, the process of transition was clearly difficult. The involvement of education services in addition to social services was seen as an extra source of stress on carers. The needs described above were expressed in terms of not only getting appropriate access to support once a diagnosis had been made (notably in relation to home teaching arrangements for those more severely affected), but also how to negotiate the transfer to adult services. In many respects the needs expressed echoed those of younger adults with other disabilities who were transferring from specific paediatric services into more general, often diffuse adult services.

7.3 Expressed Need at Tier 2

(Concerning the provision of services within primary care)

The most commonly expressed need was for better understanding and acceptance from primary care staff: particularly general practitioners. As Comber and Simpson contend, there is a need to end the culture of disbelief in ME-CFS that pervades primary care[7]. A further commonly expressed need was for rapid and accurate diagnosis, especially as there is a risk of further deterioration. This was seen by service users as key to accessing specialist services and providing support. There was some degree of recognition by some service users that the absence of a specific diagnostic test could cause delays in the overall diagnostic process, though this was by no means universally accepted. In such circumstances, the need for keeping ME-CFS patients well informed was highlighted.

Service providers also highlighted the need for better information at the Tier 2 level. However, they highlighted the need for a better understanding on the part of the primary care team on what advice to offer people diagnosed with ME-CFS regarding self-management and the need to provide care geared towards alleviation of symptoms. For the service providers, it was felt that this need should be met in a structured way using some form of protocol or patient pathway.

The use of information as a means of managing expectations was highlighted by both service users and providers. Many service users expressed the view that honesty and transparency about what was known and what could be offered was very important in establishing a trusting relationship with the primary care team. In return, the service providers considered that people with ME-CFS should be helped to have a better understanding of the diagnostic process that the general practitioner is likely to follow and that referral to a specialist is likely to be based on clinical presentation and criteria which will guide the preliminary investigations that need to be undertaken. Again, the need for an explicit protocol or pathway that would assist understanding of these elements of the overall process was highlighted.

The need for ensuring appropriate access to supportive care services at Tier 1 was highlighted. It was also noted that this may need to be initiated at point of diagnosis to meet presenting care needs.

The gate-keeping role of general practitioner was mentioned by service users, with specific mention being made in relation to therapy services:

- physiotherapy;
- occupational therapy (and access to home-loan equipment to support special mobility and independence needs); and
- dietetic / nutritional advice.

In addition, access to specific services to address particular symptomatic problems was identified:

- acute and chronic pain management;
- complementary therapies (where available); and
- counselling or psychotherapy to deal with anxiety and low mood states.

At Tier 2 the need expressed was not necessarily for specialist intervention from specialist professionals. For example, it was noted that not every individual would need or want such input. The main concern expressed was to ensure that health professionals had the appropriate attitudes towards acceptance of ME-CFS and sufficient knowledge of the condition and its variable presentations. Where they did not have such appropriate attitudes or knowledge, the view was strongly expressed that referral to a knowledgeable specialist was likely to be required if only to avoid causing a worsening of the sufferer's condition or where the individual was severely affected.

The role of the general practitioner or primary care team member in providing a care management role was identified by both service users and service providers. For service providers this focused on creating a formalised approach to undertaking a core assessment of those with ME-CFS, providing access to community health services to meet specific symptomatic needs and ongoing management within a chronic disease / long-term condition model of care. Anticipatory care for people with ME-CFS was also highlighted, particularly in the context of possible co-morbidities that could emerge over time.

The need to establish mechanisms to review care provision on a regular basis was specifically mentioned by service users. This was especially important to those who were more severely affected. Ensuring ongoing, home-based assessment, care and review of those who were housebound or with very limited independence was noted several times. Mention was made of the importance of nursing staff in providing this type of ongoing care management. As for the service providers, there was mention made amongst the service users of the long term condition management approach to meet the needs of someone with chronic disease. For those younger people in school or in transition, the need to integrate health care into the overall package of care that could include additional learning support and social support was a matter of concern to some. In this regard, the degree to which paediatric services may be responsible for negotiating additional learning support was mentioned by service providers.

It is reassuring that service providers agreed with the view that these differences in the patterns of care could be best captured in different patient pathways. Such an approach was felt to facilitate care provision and its management.

7.4 Expressed Need at Tier 3

(Describing specialist services/specialist expertise that is specific to ME-CFS)

Both service users and providers were clear in their view that there remained a need for specialist services. Such a service needed to have a strong medical lead and provide access to the types of therapeutic advice that is usually found within neurological or rehabilitation services. Indeed, one service provider specifically drew attention to the potential for such similarities of approach. However, it was also recognised that no one service could provide a universal service covering all aspects of potential assessment and advice. The need for any specialist service to exist within the context of wider networks which encompassed other disciplines and services was expressed. This was particularly noted in relation to accessing paediatric specialists and to psychological support.

Whilst there was general acceptance of the need for specialist services, there was less consistency in describing what the composition of any such specialist service would be. The nearest to such a statement was that the “ideal” service should encompass what was already in place in Glasgow, Lothian, and in Fife. By implication, this would mean that the ideal service should provide specialist diagnoses and assessment (Lothian), aimed at creating a holistic care programme (Glasgow), that could be delivered by Tier 2 and Tier 1 community and primary health and social care services supported by appropriate nursing care management (Lothian, Fife) and self management programmes (Lothian). Such a model of care was described by both service users and service providers in terms similar to those used to capture the arrangements currently being developed for neurological long term conditions.

Significant differences between service users and service providers only started to emerge when the *organisation* of Tier 3 services was being debated. Some service users felt that only a highly specialist team, based within a regional or national facility could provide the necessary degree of care required. Some suggested that a national Centre of Excellence be established to provide a focus for treatment, research and the development of national qualifications in ME-CFS. Other service users felt that more regional specialist services were needed that could be used to facilitate diagnosis and care planning and support more local health and social care teams to provide ongoing care. Service users highlighted the potential problems associated with transport. Both availability of transport and its potential for worsening the condition of someone with ME or CFS were noted. Some commented that this could be seen as a reason for not going down the route of national or even regional services. However, others did not agree with this view.

Service providers drew attention to the fact that even specialist services developed at either a large health board or regional level would still have to work in partnership with local Tier 1 and 2 services. They also noted that such services would still need to have access to a much broader range of medical, therapeutic and supportive services to ensure that care plans could be implemented and people with ME-CFS treated and maintained. The specific issues around meeting needs in remote and rural areas – including the potential for using both telehealth and telecare as means of providing specialist advice and care on a regional or national basis – were noted.

Both service providers and service users made specific mention of the need for developing managed clinical networks (MCNs) for ME-CFS. Some wished to see a national network which could restate a national service framework, whilst others wanted a regional approach to developing MCNs.

7.5 Conclusions

The thematic analysis has provided a wealth of information concerning the expressed needs of service users and service providers. This analysis would suggest that there was a much higher degree of agreement than disagreement between people with ME-CFS and those health care professionals who participated. The overall approaches explored highlighted the importance of active case management, with specific mention made of the long term condition model being used within Scotland.

Recommendation 4:

It is recommended that to meet these expressed needs, Health Boards in Scotland should develop a specific tiered ME-CFS service that provides:

- ***rapid and accurate diagnosis and assessment;***
- ***supportive care and treatment of presenting symptoms; and***
- ***provides access to wider social and economic support.***

Recommendation 5:

It is recommended that the characteristics of such services would include a local management of care, provided by the primary care team at its heart and supported by a specialist team that can facilitate diagnosis and assessment, and plan care on both a clinic and outreach basis.

Above all, both service users and providers want services that have positive attitudes towards people with ME-CFS.

8 The Comparative Analysis – Evidence from Clinical Guidelines

In this chapter the findings from the literature review in relation to clinical guidelines are presented. This review included the major, international reports and guidelines from the past six years. For an overview of the international perspective a paper by the New Zealand Guidelines Group (NZGG) provided a clear comparison of approaches to ME-CFS across Australia, Canada, the US and the UK[22]. This was supported further by reference to a Health Technology Assessment review of evidence prepared by the Norwegian Knowledge Centre for the Health Services[23]. Reference was also made to the CMO's Short Life Working Group[3], the Action for ME Scoping Report[5], and an advisory report to the Minister of Health, Welfare & Sport in the Netherlands[24] and the NICE Guidelines for England and Wales[16].

8.1 The Analysis

A summary table detailing the key areas of consensus and difference across the guidance documents is included in Appendix 4. When reviewing this appendix, it should be noted that only the UK data, which is from the NICE Guidelines, contains explicit recommendations. Other guidance documents are more discursive or presented as a simplified matrix, making comparison difficult. Therefore the comparison can only suggest areas where there is broad agreement and the issues noted attract support at an international level, or where there is a lack of agreement.

It is important to note that none of the guidelines was accepted without criticism. Despite this, each of the documents mentioned has contributed to broader consensus on many of the key ME-CFS issues. Explicit, evidence-based, international consensus regarding service needs is outlined below.

8.2 Aetiology and Diagnosis

The NZGG note the aetiology of ME-CFS is unclear but that it is frequently found to follow an infection. Exacerbating factors included exertion, sleep disturbance and stress[17].

Diagnostic testing strategies were found to be influenced more by particular perspectives on ME-CFS rather than evidence of effectiveness[22]. International comparison showed differences in the range of tests recommended, but there was no universal agreement on routine testing. This issue is dealt with more fully within the Scottish Good Practice Statement on ME-CFS:

http://www.show.scot.nhs.uk/GoodPracticeStatementonME-CFSforGeneralPractitioners.

8.3 Specialist Referral

International consensus can also be noted in relation to referral, with each country acknowledging the possibility that referral may be necessary as part of the diagnostic workup and that specialist input may need to be multidisciplinary. The NICE Guidelines considered there to be no research evidence for the benefits of specialist referral. However, the consensus methodology employed by the guidelines development process resulted in a recommendation that decisions regarding specialist referral should be made in conjunction with the patient and should only be made with due consideration for that person's condition (eg duration, complexity and severity of symptoms and the presence of co-morbidity) and the local services available. It is noted that referral should be offered immediately to those with severe symptoms.[13] The CMO's Short Life Working Group emphasised the role of the specialist in pain management.[3]

8.4 Information and training

Evidence suggests the need for more and better information and/or training for healthcare workers, patients and carers particularly in relation to employment/education and welfare benefits. From the international perspective, ensuring there was a shared understanding of the condition was viewed as essential to the shared management of the condition[22].

8.5 Care Management & Treatment Options

There was international agreement that management of care and/or treatment should be individualised. The approach to care management used should acknowledge variation in the abilities of any one person with ME-CFS on a day-to-day basis. Overall the aim should be to provide symptomatic treatment and supportive care to increase the person's quality of life, without making their condition worse.

Reflecting a generally held view amongst patient support groups, Action for ME emphasised patient autonomy and the need for care and treatment goals to be set by the patient[5]. The NICE guidelines also emphasise that healthcare professionals and the person with ME-CFS (and their carers) should seek to establish a collaborative relationship that supports the process of diagnosis, assessment and care. Whilst it may be fair to comment that patient autonomy and choice *per se* is presumed to be occurring, the actual experience of patients is different[7]. Healthcare workers should bear in mind that people with ME-CFS – as for anyone receiving NHS care - have the right to refuse or withdraw from aspects of recommended care without affecting the remaining elements of the care plan. This should not be a barrier to care in the future.

8.5.1 Physical Activity and Exercise

Management of physical activity is not considered consistently across guidelines. For example, the NICE guidance advises that service providers should be clear about the lack of evidence in the literature it reviewed, for benefit or harm with regard to such interventions. Other guidelines discuss physical activity in less formal terms as part of a more generalised approach to activity management.

There was international agreement – particularly in the Canadian Guideline and the New Zealand Guidelines Group report – that too much exercise can exacerbate symptoms and that limited exercise should only be introduced with caution[13, 22]. Views on graded exercise programmes varied. Although it was acknowledged that a degree of exercise relating directly to each individual's level of energy and/or pain may be beneficial, the international summary included a cautionary note that graded exercise programmes had recently been identified as harmful. A Norwegian evidence review noted a lack of information on the effect of graded exercise therapy on depression or quality of life. The NICE guidelines recommend Graded Exercise Therapy (GET) for people with mild to moderate ME-CFS citing randomised control trial evidence of significant improvements in measures of fatigue and physical function[16]. Recommendations relating to exercise programmes are contentious with some reports noting patient preference for Pacing (seeking to balance periods of activity and rest) over GET. NICE do make recommendation regarding GET as an option relating to physical activity, but warn of insufficient evidence in relation to its use for children and those severely affected[16]. It should be noted that the PACE trial which specifically explores the effectiveness of pacing, is due to report in 2010.

In Scotland, a significant proportion of people with ME-CFS who participated in the Action for ME Scoping Study said that GET made them worse with roughly 75% of those who reported undertaking GET describing themselves as getting worse. It should be noted that some 57% of responders did not participate in GET[5]. Data reported from the 25% ME Group, which represents the most severely affected with ME, suggest that 82% of people with ME that they surveyed described themselves as having deteriorated having undertaken GET. This included some who maintained that they had only become severely affected after having taken part in GET. In contrast to the NICE position, the Action for ME Scoping Study reported that of those individuals who reported undertaking pacing (89% of those surveyed), only 3% reported themselves as having deteriorated and 68% that it had been helpful. However, care should be taken to avoid over-interpreting these data as there is no way of differentiating between ME or CFS patients who contributed, or of how severely affected they were when a physical activity programme was offered, or of how well they were able to comply with the activity regime suggested.

8.5.2 Cognitive Behavioural Therapy

There are highly differing views on the role of Cognitive Behavioural Therapy (CBT) in the management of ME-CFS within the clinical guidelines. Indeed, the NZGG specifically comment that this ranges from what they described as an “uncertain role” to seeing CBT as an integral part of management. Within Scotland, the CMO’s Short Life Working Group commented that whilst there was a role for CBT in the management of ME-CFS for some patients, “It is not a technique that could or should be recommended to every patient”[3].

Most recently the guidance issued by NICE set out specific recommendations for the use of CBT in ME-CFS[13]. The guideline developers clearly stated that CBT should be offered to people with mild to moderate ME-CFS and provided to those who *actively choose to accept the offer*. They also set out very specific standards for providing CBT. These are set out in Box 8.1.

Box 8.1

NICE Recommendations regarding the use of CBT[13]

- A course of CBT should be delivered only by a healthcare professional with appropriate training in CBT and experience in CFS/ME, under clinical supervision. The therapist should adhere closely to empirically grounded therapy protocols.

- CBT should be offered on a one-to-one basis if possible.

- CBT for a person with CFS/ME should be planned according to the usual principles of CBT, and should include:
  - Acknowledging and validating the person's symptoms and condition;
  - Explaining the CBT approach in CFS/ME, such as the relationship between thoughts, feelings, behaviours and symptoms, and the distinction between causal and perpetuating factors;
  - discussing the person's attitudes and expectation;
  - developing a supportive and collaborative therapeutic relationship; developing a shared formulation and understanding of factors that affect CFS/ME symptoms;
  - agreeing therapeutic goals;
  - tailoring treatment to the person's needs and level of functioning;
  - recording and analysing patterns of activity and rest, and thoughts, feelings and behaviours (self-monitoring);
  - establishing a stable and maintainable activity level (baseline) followed by a gradual and mutually agreed increase in activity;
  - challenging thoughts and expectations that may affect symptom improvement and outcomes;
  - addressing complex adjustment to diagnosis and acceptance of current functional limitations;
  - developing awareness of thoughts, expectations or beliefs and defining fatigue-related cognitions and behaviour;
  - identifying perpetuating factors that may maintain or exacerbate CFS/ME symptoms to increase the person's self-efficacy (sense of control over symptoms);
  - addressing any over-vigilance to symptoms and related checking or reassurance-seeking behaviours by providing physiological explanations of symptoms and using refocusing/distraction techniques;
  - problem solving using activity management and homework tasks to test out alternative thoughts or beliefs, such as undertaking pleasure and mastery tasks (tasks that are enjoyable and give a sense of accomplishment);
  - building on existing assertion and communication skills to set appropriate limits on activity;
  - managing sleep problems, for example by addressing any unhelpful beliefs about sleep, behavioural approaches to sleep disturbance, stress management, and/or relaxation training;
  - treating any associated or comorbid anxiety, depression or mood disorder according to NICE clinical guidelines on these conditions; and
  - offering information on managing setbacks/relapses.

- People with severe CFS/ME should be offered an individually tailored activity management programme… which may:.. draw on the principles of CBT.

However, it should also be noted that only 40% in the Action for ME Scoping Study had tried CBT. Of these, some 39% had found it helpful, 44% to have not had an effect and 17% had found it to be harmful[5].

8.5.3 Other Treatments

Generally speaking, the clinical guideline documents presume that presenting symptoms or factors which relate to specific, possible causative factors (eg infections or inflammatory processes) are treated. Whilst there is a limited evidence base for drug treatments, symptomatic approaches include the use of non-steroidal anti-inflammatory drugs, other analgesics, muscle relaxants and (low dose) tricyclic antidepressants (where appropriate). However, it should be noted no specific clinical trials of these drugs in relation to ME-CFS have been undertaken. It is also relevant to note that anecdotal patient evidence and part of the wider research evidence on ME-CFS suggests that drug sensitivities are reported to be common.

As disturbed sleep patterns can be a feature of ME and other forms of CFS, there was consensus that it may be necessary to refer patients to a sleep specialist in order to distinguish between ME-CFS and primary sleep disorder. In the case of ME-CFS related sleep difficulties, non-pharmaceutical approaches should be tried before medication to help establish more successful sleep patterns.

Referral for specialist pain assessment and chronic pain management is also considered to be an important component in specific treatments to be offered.

8.6 Conclusions

Whilst the analysis undertaken has highlighted what consensus and difference exists, it also shows that there is a relatively small evidence base on which to base clinical guidance. The current clinical guidelines are primarily based on “clinical consensus” or “best practice” statements which originate amongst those with expertise in the field. As such, they tend to focus on the need for effective diagnosis and the trial of symptomatic treatments where clinically indicated.

Whilst there is a growing body of research, much of it is concerned with exploring possible causes of ME-CFS and very little is concerned with research which applies what is already known to determining effective therapeutic strategies. Furthermore, much of the research that is being undertaken is of a quality that would be difficult to use in formal, systematic reviews of the effectiveness of ME-CFS treatment.

As a consequence of this it is hard to see how, at the present time, the suggestion made within the Action for ME Scoping Study that a formal Scottish Inter-Collegiate Guideline Network (SIGN) guideline be developed. It is probable that such an approach – using the best available evidence at present – would not be able to draw on a sufficient body of research to complete the type of robust, quality-assured, systematic analysis which characterises SIGN’s work. Where such an approach was attempted by NICE, it has been subject to a number of challenges from stakeholders and interested parties on the grounds of the research that was considered suitable for inclusion.

Recommendation 6:

At the present time there is insufficient research evidence on which to base a SIGN ME-CFS Guideline for Scotland. However, a clinical guideline which supports effective diagnosis, signposts people with ME-CFS towards appropriate medical and therapeutic assessment and service, and provides the basis for ongoing care management is desirable. It is suggested that this is in keeping with the Scottish Good Practice Statement on ME-CFS.

9 The Comparative Analysis – Evidence for Effective Service Models

The review of guidelines and reports was also used to consider what international models of care are in operation. Despite detailed online and manual searches, evidence which provide detailed models from other countries has been found to be elusive. Whilst a specific service model was not identified, information presented to the Cross-Party Group on ME from ME Research UK, suggests that:

> *"In terms of a model to aim for – the 'Canadian Consensus' document could provide a suitable resource for assessment and treatment – though the interventions suggested were mostly aimed at symptomatic relief rather than treatment of underlying cause. Whichever model is used, there is at present a lack of suitable experience among health service staff, and training would need to be given."*

In Scotland, models of care based on the long term conditions approach have been considered and a generic model is discussed below.

In England, the CFS/ME Service Investment Programme funded the development of new services from 2004. In the absence of alternative models these serve as examples of, if not best practice, then practice that is coherent, transparent and underpinned by national guidelines. One of these services (Bristol), selected following discussion with Action for ME, was reviewed for this part of the comparative analysis to illustrate the type of structure that such a model may adopt, without endorsing any specific staffing configuration. For example, it is notable that this service did not provide a medical lead as other such services in England have (eg the CFS Diagnostic and Management Service in Queens Hospital, Romford).

9.1 The Long-Term Conditions Model

The Scottish Government's health strategy for providing supportive care for those with long term conditions was set out in the document Better Health Better Care: Action Plan[6]; though it had already been initially identified as a necessary element of Scottish health policy in Building a Health Service Fit for the Future (The Kerr Report).[25]

These documents set out a vision of a future health care service where the balance of care was shifted towards care which was:

1. geared towards meeting the needs of people with long-term conditions;
2. embedded in communities;
3. provided by a team of professionals;
4. provided on a continuous basis;
5. able to provide integrated care to meet needs holistically;
6. focussed on preventative care;
7. supportive of the patient and was a partner in their care;
8. able to encourage and facilitate appropriate self care;
9. supportive of carers as partners; and
10. able to make best use of new, high tech solutions.

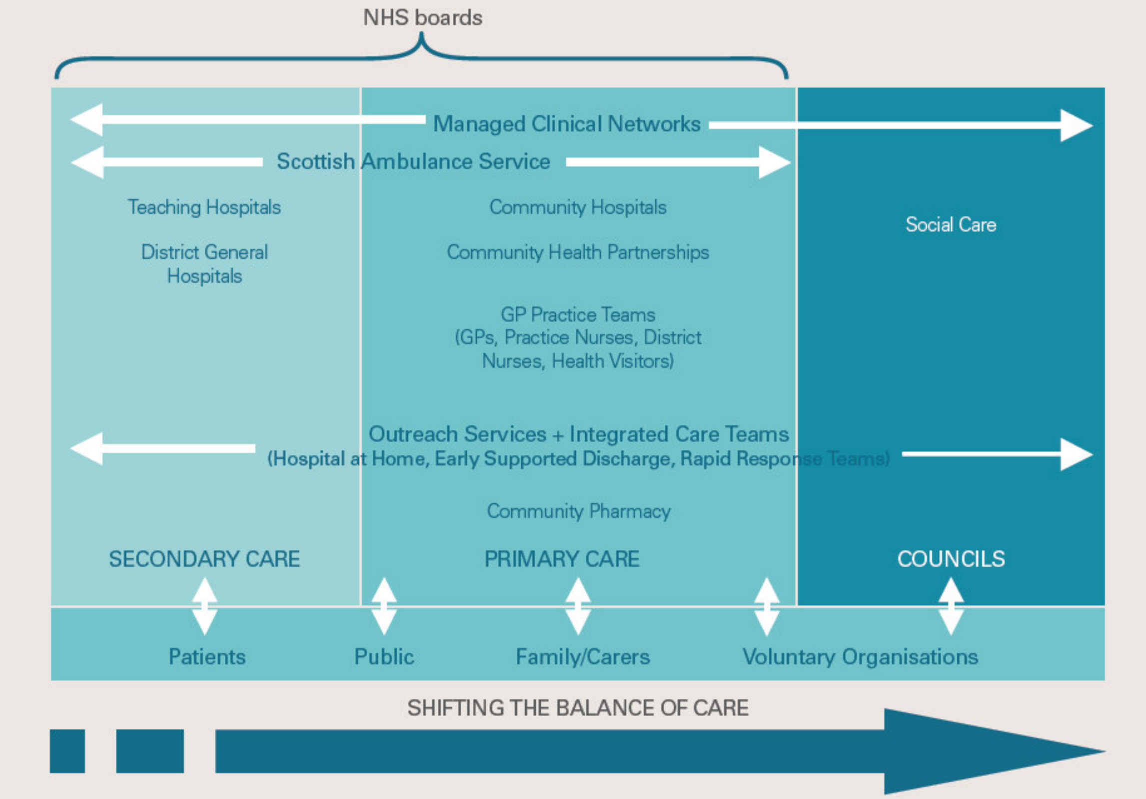
The overall approach outlined involved three "levels" of care:

1. supportive, self care / self management;
2. shared care – care that is provided by a range of professionals for those who need additional care or support to manage their conditions; and
3. intensive care or case management for that part of the population which have more complex or multiple needs.

In supporting the development of these approaches at Community Health Partnership and Health Board levels, the Scottish Government has provided continuing support for the Scottish Long Term Conditions Alliance and has established the Long Term Conditions Collaborative to work with the NHS in Scotland. Most recently, these approaches to a redesigned model of health care in Scotland have been reinforced within The Quality Healthcare Strategy for NHS Scotland[26] which has emphasised the priority the Scottish Government places on delivering actions to improve the management of long term conditions. Whilst this approach is at an early stage of implementation, a fact highlighted by Audit Scotland[27], there is already recognition of the usefulness of the approach in a range of situations. This is not least because – as figure 9.1 shows – the approach brings together a large range of services and organisations into the overall pattern of care for individuals.

Figure 9.1:

Summary of all the main parties involved in long-term condition care



Note: Different parts of the NHS as well as councils, voluntary organisations, patients and their families are all involved in long-term condition care. A shift in the balance of care to address changing healthcare needs will involve moving services and resources from secondary care into primary and social care, with more public and patient involvement.

Source: Audit Scotland 2007

As yet there has been no formal long term condition model of caring for ME-CFS put in place in Scotland. However, there is a high degree of similarity between the model of care set out in the Quality Healthcare Strategy for NHS Scotland[26] and the tiered model of service originally set out in the CMO's Short Life Working Group[3]. It is therefore feasible for such an approach to be developed and implemented within Scottish Health Boards.

Of particular benefit in so doing would be that such an approach would accommodate the differing needs for care of people across the full range of severities of ME-CFS. For example, those who are mildly, or moderately affected – once appropriately diagnosed could fit into a model of care that exists within the first two levels. Such an approach would have particular benefit as it would accommodate those who are most severely affected, as it provides a very specific way in which care or case management could ensure that their ongoing needs are being met.

9.2 The English Coordinating Centre Model

The Avon, Wiltshire & Somerset CFS/ME centre is one of 13 Clinical Network Coordinating Centres set up under the Department of Health funded CFS/ME Service Investment Programme 2004-2006. It co-ordinates three local adult multi-disciplinary teams (Bristol & Gloucester; Bath & Wiltshire; and Somerset) and a service for children and young people based in Bath. It operates 2 days per week. The model is one of a central coordinating team for the region, led by a Clinical Champion located in the Pain Management Clinic in Frenchay Hospital, providing a consultancy service to satellite teams. The Centre is participating in the MRC PACE trial. Overall the service providers consider that it conforms broadly to the NICE Guidelines[16].

It should be noted that – to date – there has been no formal evaluation of the effectiveness of the Coordinating Centre approach and the outcomes for people with ME-CFS that they have achieved.

The service offers:

- multi-disciplinary assessment (domiciliary and out-patient);
- consultation and advice in liaison with the Primary Health Care Team;
- direct clinical work (group or individual); and
- a multi-component rehabilitation package for symptom management or complex case management.

The service comprises the following staff:

Bristol based team		**Gloucester team**	
Team Leader/Clinical Champion	x 1	CBT Therapist	x 1
Assistant Psychologist	x 2	Specialist Physiotherapist	x 1
Specialist Occupational Therapist	x 2		
Specialist Physiotherapist	x 2		
Clinical Psychologist	x 2		
Counsellor	x 2		
Specialist Medical Advisor	x 1		
Administrator	x 2		

Referrals must be based on a diagnosis conforming to a standard protocol of symptoms and exclusions. The diagnosis should be supported by written results of prescribed tests. Referrals that do not include results of the required tests are referred back to the general practitioner. A considerable amount of time and energy has been given to developing understanding of ME-CFS amongst medical practitioners in the area and guiding them to ensure correct referral procedures. This has been emphasised as a key piece of work in encouraging acceptance of the illness amongst professionals, helping to streamline the referral process and supporting ongoing care.

The service considers that the range of practitioners is valued as each contributes towards a balanced approach to management and treatment. Of particular value is the medical adviser, a retired general practitioner who maintains strong links with general practitioners in the area. The team have exploited her good relations with colleagues to enhance the impact of outreach/education work with general practitioners. The team has seen an improvement in referrals since this work began. Other specialists within the team allow for distinct expert approaches with individual patients where required (eg physiotherapy where mobility needs to be addressed; occupational therapy to improve functioning for work or daily living).

In the case of patients with severe ME-CFS, the service liaises with community health services and social services where appropriate. Fewer services specifically for children and young people with ME-CFS exist. Understanding of ME-CFS amongst paediatricians is low. Liaison with referring paediatricians is addressing this issue. Time missed from school is used as a measure of severity of ME-CFS in children. Schools/education departments are crucial partners in supporting the patient and families/carers. Liaison with these bodies is an important part of improving understanding of ME-CFS and appropriate/timely referral.

The creation of the “Clinical Champion” helps to give weight to the team and its function. Recognition of the task of leading a Clinical Network has supported the work of the Centre.

The NICE Guidelines underpin the work, but the team admits to interpreting them broadly, in relation to the needs of their patients and the resources available. For example, advice on pacing is given routinely despite lack of evidence of benefit referred to in the Guidelines. This is because the prime aim of the service is to offer a supportive service that helps each patient identify a management strategy that suits their symptoms, their approach and their lifestyle. Many patients come to the service having researched their condition and are aware of pacing as an option.

Since 2005 the Centre has worked in partnership with Action for ME in operating a telephone helpline for people in the Bristol area, Somerset, Wiltshire, Bath & North East Somerset, Gloucestershire and Greater Manchester. The helpline offers information and support to those with questions relating to ME-CFS. This service has proved very popular and has attracted calls from people across the UK, not just the local area.

The Helpline allows expert advice to be given at the point of need and is viewed as being of significant benefit due to its immediacy. Use of the Helpline has been considerable. In the period beginning June 2006 to end of May 2007, 342 calls were logged to the Helpline, representing a 7.35% increase on the previous year. In 2006-7 most calls were from patients themselves (73.7%) with 20.2% from carers and 6% from professionals. The most common reasons for calling included: information on new NHS services (29.9%) and emotional support (25.4%). This represents a 9.2% increase on the previous year on those calling for emotional support[28].

9.3 Conclusions

Given the long and contentious history which underpins the recognition of ME-CFS, it is perhaps not surprising that there is no clear, internationally recognised model of care which can be adopted in Scotland. Indeed the original model for a tiered service set out in the CMO's Short Life Working Group Report[2] would seem to be as good a starting point as any for NHS Boards in Scotland.

Recommendation 7: ***(See also Recommendation 4)***

It is recommended that the tiered model for services proposed by the CMO's Short-Life Working Group be used as a basis for ME-CFS service development in Scotland.

Recommendation 8:

It is recommended that a dedicated helpline and website to provide information and support for people with ME-CFS and those who care for them be established in Scotland.

10 Current Service Provision

The current pattern of service provision across Scotland was assessed as part of the Action for ME Scoping Study[5]. This is summarised in appendix 5. The study concluded that:

> *"Wide variations in availability, accessibility and quality of care exist for patients with M.E .There is no agreed standard of care and treatment currently being met across Scotland. Services are sometimes being defined by local needs but this is by no means standard practice. Very few people with M.E. in Scotland are benefiting from specialist care and from the findings of this survey, none of the Health Boards felt that they were providing services for people with M.E. that were fully adequate."*

The study highlighted four specific services which could be construed as "specialist" services. These were the:

- ME-CFS Assessment Clinic in the Regional Infectious Diseases Unit in the Edinburgh Western General Hospital (NHS Lothian University Hospitals);
- Centre for Integrative Care at the Glasgow Homeopathic Hospital ;
- Clinical Nurse Specialist employed by NHS Fife; and
- Thistle Foundation Self-Management Course in Edinburgh, which is funded by Lothian NHS Board.

As part of this healthcare needs assessment a further analysis of the Action for ME Scoping Study data has been undertaken. This also sought to explore the degree to which the 13 Health Boards who responded were seeking to provide services across all three tiers of service outlined in the report of the CMO's Short-Life Working Group[3] (see Appendix 5). The fact that the expressed need (see Chapter 7) still echoes the recommendations of the CMO's Short Life Working Group implies that little has changed, that needs are still present and that current service provision at all levels is inadequate.

10.1 Services at Tier 1

Tier 1 service as outlined by the CMO's Short-Life Working Group focussed on the provision of information for people with ME-CFS, their carers and their family members, training and education for health care professionals and the development of local clinical guidelines. In terms of expressed need, Tier 1 should also meet social support needs and access to services which maintain activities of everyday living (see Chapter 7).

It is clear from the analysis of the Scoping Study that this has been not carried forward in a structured way. Information was only provided in eight Health Board areas of which only five provided written information, some provided self-help materials (2 Boards) and some directed people to web-based information (3 Boards). Only one area had developed their information with the aid of a local ME Support Group. Information provision was usually by GP, though in 2 Board areas medical consultants were the main information provider and in 1 Board it was the specialist nurse. Formal training was provided by only 3 Health Boards – one of which was on an "at-request" basis. Six Health Boards provided their staff with information about ME and other CFS, whilst a further three provided no information for staff.

Only one Health Board had developed local care pathways. This was the care pathway prepared by NHS Lothian for GP referral for specialist assessment by either adults or children's ME-CFS clinics.

No figures for the number of people with ME-CFS receiving general, social support services at Tier 1 were available. However, comments from the focus groups and the Action for ME scoping study suggest current provision at this level is sub-optimal and patients experience great problems accessing such services. This is largely because access to these services and support is usually dependant upon diagnosis.

10.2 Services at Tier 2

The CMO's Short-Life Working Group recommended that Health Boards make use of their chronic disease management approach to meet the initial needs of people with ME-CFS in such as way as they would receive care which was relevant and helpful and not likely to cause further harm. Health Boards in Scotland have now subsumed their chronic disease management approach within their model of care for those with a long term condition. This means that Primary and Community Care Services should seek to ensure that people with ME-CFS are able to receive:

- initial assessment and diagnosis against criteria and opportunities for confirmation of diagnosis (where required);
- information for people with ME-CFS and those supporting them; and
- referral to community health care services, social care and support services and complementary services to meet specific, symptomatic needs in a timeous fashion.

In many regards these actions would usually be subject to accepted care pathways. However, as already noted, only one Health Board had such a guideline and even that would only have related to the first bullet point (see 10.1 above and Chapter 8). As a consequence it is not surprising that the Action for ME Scoping Study found that over half of its respondents had had to wait over a year to receive a diagnosis and in 25% of cases that the wait had been over 2 years.

This long-term conditions "model" presumes that patients are routed into one of three broad streams of care:

1. low intensity support for individuals with access to self care approaches;
2. community care management by primary / community health care teams; and
3. intensive care management by primary / community health care teams with the support of appropriate specialists / multi-disciplinary teams.

In this model it should be noted that long-term conditions management presumes that there are systems in place to allow for active patient follow up and monitoring, the use of patient registers and review/recall systems that allow for care management. Currently, there is no specific system for ME-CFS in operation in Scotland. It should also be noted that, at this service tier, what is deemed “specialist” will probably relate to the type of care being offered and not be specific to the disease.

Broadly speaking, the further analysis showed that few NHS Boards in Scotland have formally attempted to address the needs of people with ME-CFS through existing services. Those that have done so have focussed on providing a Tier 2 service. In some cases the link to long term conditions management was specifically noted. Specific examples of how Health Boards were responding to need are shown in Table 10.1.

Table 10.1 Current Tier 2 Service in Scotland by Care Management Stream

Stream	**Service Responses (No. of Health Boards)**
Low intensity support with self-care approaches	• Self-management courses / services (3) • Voluntary sector support (7) • Social care assessment and support (8) • Employment assessment and support (7)
Community care management by primary / community health care teams	• Primary care / Family nursing (8) • Occupational Therapy (8) • Physiotherapy (not specified) • Dietetics (not specified)
Intensive care management by primary / community health care teams with the support of appropriate specialists / multi-disciplinary teams	• Chronic pain services (8) • Clinical psychology (not specified) • Skin integrity services (4)

However, even were there to be a full complement of Tier 2 services available for people with ME-CFS in all Health Boards, the lack of clear, clinical guidance would suggest that these streams of care management are unlikely to be offered in a systematic or equitable fashion. The lack of comprehensive training for front-line NHS staff might also suggest that any care offered may be less effective than desired. There is also a clear risk that those people with severe ME-CFS are less likely to be accessing necessary services. The views from service users and health care professionals described above would also support such a view (see Chapter 7).

10.3 Services at Tier 3

The model of care for Tier 3 proposed by the CMO's Short-Life Working Group envisaged the development of secondary care services that would provide clinical investigation and confirmation of diagnosis, assessment and support for the long-term condition management provided by Tier 2 services. The analysis of the Scoping Study data showed this had not occurred in the majority of Health Board areas.

The CMO's Short Life Working Group envisaged that Tier 3 services would:

- develop expertise in medical assessment and diagnosis;
- develop expertise in rehabilitation services;
- develop expertise in the management of difficult problems;
- have a role in the monitoring of the services; and
- have links to research, training and audit of services

In this regard, the Short Life Working Group was clearly intimating that developing services on a pattern we would now consider to be a managed clinical network would be required. As greater expertise was developed and confidence developed in both Tier 3 and Tier 2 services, it was suggested that the network could move towards the development of wider aspects of managed clinical networks in addressing areas such as the development of treatment modalities, management of more complex cases and developing research and information links.

Specific mention was made by the CMO's Short Life Working Group of the need to develop specific pathways for involving the Tier 2 community teams from health and social services, including rehabilitation and the access to appropriate everyday living support for those severely affected. This included developing protocols and facilities for respite care. A different care pathway to the adult services was identified as necessary for children and young people. This was identified as requiring the involvement of paediatric services at an early stage and other statutory and non-statutory agencies involved in education and children's social work.

The analysis of returns from the Action for ME Scoping Study did not demonstrate meaningful progress in developing Tier 3 Services across Scotland. Only NHS Lothian and NHS Greater Glasgow and Clyde had elements of a Tier 3 service. Both NHS Fife and NHS Dumfries and Galloway had sought links to "regional" Tier 3 services based in Edinburgh. However, these have not been formalised in any way.

NHS Lothian had established a Managed Clinical Network Development Group. This had developed care pathways for both adults and children as far as diagnostic and assessment services. However, these pathways did not consider treatment modalities and made referral to Tier 2 services as their main approach. The availability of the PACE Trial at the University of Edinburgh did provide some access to specialist medical management and rehabilitation services for those who entered the trial. For those who did not, onward referral for consultant care or specialist intervention was available to meet identified needs. In Greater Glasgow, the holistic approach adopted by the Homeopathic Hospital provided access to some of the wider therapeutic interventions. However, the approach taken by both Health Boards seems to have been that once a care plan was identified, it was expected that this would be delivered by Tier 2 services.

Overall, even where a Tier 3 service was in place, access to treatment modalities was poor. Waiting lists for accessing Tier 2 rehabilitation and therapeutic services were noted. This lack of capacity to offer existing therapies to people with ME-CFS at either Tier 2 or Tier 3 is of concern.

10.4 Conclusion

Whilst the overall model of care proposed by the CMO's Short Life Working Group remains valid[3] – indeed in many respects sits extremely well with the current national policies set out in The Quality Healthcare Strategy for NHS Scotland[26] – the implementation of the group's recommendations have been partial. The fact that expressed needs still echo the recommendations of the CMO's report of 2002 would imply little has changed and that current service provision remains inadequate.

At both Tier 1 and Tier 2, appropriate links to service delivery and development have been identified but not implemented. The presumption that existing services should be able to extend to accommodate the needs of people with ME-CFS without appropriate training and information does not seem to have resulted in any noticeable change in the experiences of people suffering from ME-CFS. At Tier 3, services have not been developed by the majority of health boards. Where these have been developed, services which do not have access to sufficient capacity to plan and initiate treatment under specialist supervision have been of limited success. Developing Tier 3 services in isolation from Tier 2 services that can provide rehabilitative care / long term condition management specific to individuals with ME-CFS has also been shown to be ineffective.

In other words, developing a specialist service at Tier 3 without having **both** specialist input at Tier 3 and the capacity for Tier 2 services to provide ongoing care appropriate to ME-CFS patients would be counter-productive.

11 A Proposed Model of Care for Scotland

Chapters 7 and 10 highlighted that the tiered model of care – whilst remaining fit for purpose in Scotland – has not been implemented in a robust way across Scottish Health Boards. The model ***could*** meet the expressed needs of people with ME-CFS provided that it was fully implemented and the necessary capacity issues in existing services addressed. Implementing the model ***could*** also be better supported if a number of specific, wider infrastructure issues are addressed.

In this chapter, the actions needed to implement a tiered model of care for Scotland are presented. Where possible, the recommendations for implementing the model will draw on existing experiences and expertise across Scotland. In the next chapter, the wider, infrastructure issues are considered.

11.1 General Service Characteristics

The infrastructure of any service is, of course, only part of the solution. The values which underpin the services that are delivered are also critical. The needs of people with ME-CFS, whether in terms of the specific, presenting symptoms, how they vary over time, or the severity of how the individual is affected, means that services must be flexible and responsive in meeting need. There is a clear link here to the requirement placed on the NHS to develop patient-centred care and improve the overall patient experience. As one of the focus groups put it, the general approach should be one of treating what is treatable and supporting that which is not.

The services should strive to offer diagnosis, treatment and care that is tailored to the individual's needs and clinical presentation. This will mean offering care in a range of settings, including domiciliary care as well as health care settings. The service will also have to be able to meet the needs of people with ME-CFS across the full severity spectrum. The needs of the most severely affected, especially those who are unable to travel to be in receipt of care, must be met.

Clearly, ensuring there is sufficient capacity in the service to meet needs will be an issue. However, it will also be necessary to ensure that services are provided in a way that all people with ME-CFS can benefit.

11.2 Service Implementation at Tier 1

The key developments in improving care within Tier 1 of a ME-CFS Service are:

- providing supporting information to help ME-CFS patients and healthcare professionals understand the condition;
- implementing care pathways to improve access to diagnosis; and
- improving access to social and supportive care.

Such developments need to have the capacity to operate on a clinic or outreach basis.

11.2.1 Providing supporting information about ME-CFS

The most pressing need at Tier 1 is for information for people with ME-CFS, those who care for them and for health professionals. Such information needs to consider not only the nature of the condition, but also the care and support that is available and how people can access help.

It is clear from the very many differing opinions about what underlies ME-CFS, and how those with these conditions should be cared for, that we should expect little initial agreement about what might be included in such information.

It therefore seems appropriate to seek to develop a core information set across Scotland. This should be developed within a clear framework that brings together service users, providers and the independent sector, to provide information for patients, their carers and for health care professionals. This material should use the materials developed by organisations such as Action for ME, the 25% Group and the ME Association as well as the Primary Care Information Pack developed by NHS Lothian. The development of a national, core information set will provide a template against which any local adaptation of the information to meet local circumstances can take place.

Recommendation 9:

A broadly constituted stakeholder group should be established to:

(a) ***create a national, core information set which can be used for people with ME-CFS and their carers;***

(b) ***create a national, core information set which can be used for health and social care professionals; and***

(c) ***explore appropriate ways of making such information widely available.***

11.2.2 Implementation of care pathways

The need for local care pathways is outlined in Chapter 7 above. The generic pathway described in the SGPS should be adapted for local use in an open and transparent manner.

Recommendation 10:

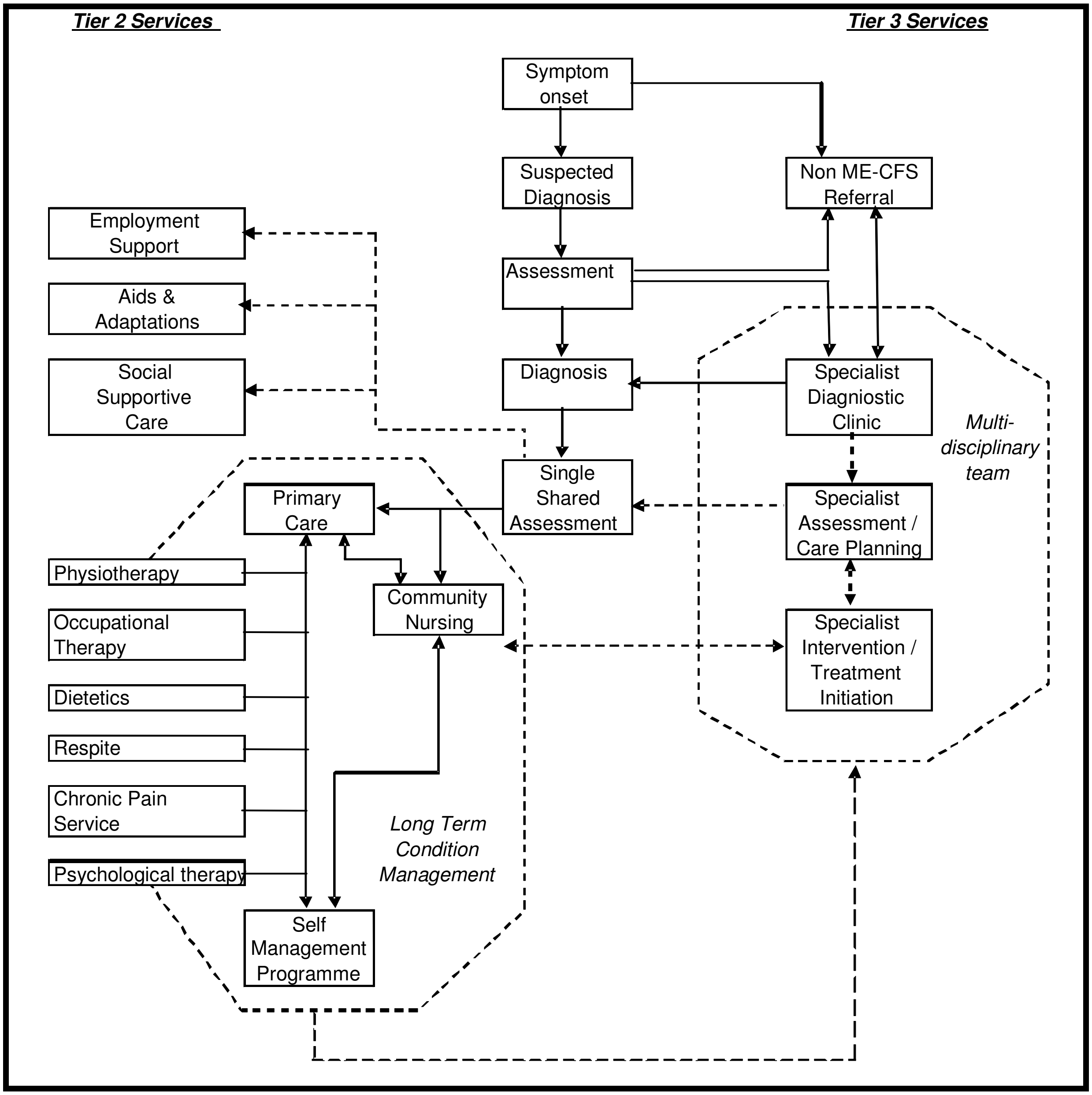
NHS Boards in Scotland should develop formal, care pathways for the diagnosis, assessment and management of people with ME-CFS as outlined in the report of the CMO's Short Life Working Group. These local pathways should be compatible with the Scottish Good Practice Statement on ME-CFS.

The training of health care staff, recognised as a Tier 1 service issue, is considered in Chapter 12 below.

11.3 Service Implementation at Tier 2

Health Boards in Scotland should ensure that ME-CFS is clearly identified as an area which is being managed under their Long Term Conditions arrangements. A general model for Long Term Conditions was described in Chapter 9. The relationship between the elements of Tier 2 services and Tier 3 services are explored in a generic pathway (created during the Stakeholder's Group meeting):

Generic Tier 2 and Tier 3 ME-CFS Pathway



Recommendation 11:

NHS Boards in Scotland should formally identify ME-CFS within their Long Term Conditions Plan or Strategy. Management of ME-CFS should be carried out in line with local arrangements for other long term conditions, where appropriate.

11.3.1 Accessing the Primary Care Team

At the heart of the Tier 2 service is the general practitioner or primary care team member in providing assessment of those presenting with symptoms suggestive of ME-CFS, providing access to community health services to meet specific (symptomatic) needs and ongoing management within the long term condition model of care. As an indicator of expected need, the analysis undertaken in Chapter 6 suggests that for a GP practice of around 2,500 patients, it could be expected that a member of the primary care team would be consulted on at least three occasions by a person with ME-CFS in a given year.

However, as described above (see Chapter 7), the most commonly described need was for better understanding of, and acknowledgement of the condition ME-CFS from primary care professionals; particularly general practitioners. As the Tier 2 service will usually provide the first port of call for virtually all people with ME-CFS, early experiences which promote trust and acceptance will help manage expectations, reduce frustration and allow for better management of the condition. Having a local care pathway will help not only to promote better understanding and acceptance from primary care professionals, but also to improve speed of diagnosis and delivery of care. It will also help to inform and guide ME-CFS patients about the overall process being followed to reach an appropriate diagnosis and excluding other possible causes of the presenting symptoms.

11.3.2 Community Health and Care Provision

A key element in the management of people with ME-CFS must be the provision of supportive care. In this regard, having clear, referral criteria for therapy services will be needed in relation to:

- physiotherapy and occupational therapy;
- access to home-loan equipment to support special mobility and independence needs;
- dietetic / nutritional advice; and
- social care services (via single shared assessment).

In addition access to specific services to address particular symptomatic problems were identified. For example:

- pain management; and
- psychological therapies to deal with anxiety and low mood states.

Ideally, any services should be delivered by those with a sound understanding of ME-CFS.

Recommendation 12:

When developing local approaches to Long Term Conditions Management, NHS Boards should ensure that:

(a) ***assessment and review mechanisms are in place for people with ME-CFS, including domiciliary assessments /review where needed; and***

(b) ***appropriate referral mechanisms for people with ME-CFS to receive appropriate supportive therapies are in place; and***

(c) ***appropriate referral mechanisms for people with ME-CFS to access services that can meet specific, symptomatic needs are in place.***

Chapter 10 highlighted that there was a need not only to make use of the rehabilitation services and the wider range of community and specialist services to support people with ME-CFS, but also to ensure that such services had sufficient capacity to provide appropriate care.

Recommendation 13:

When developing local approaches to Long Term Conditions Management, NHS Boards should ensure that both rehabilitation services and specialist, symptom-specific services have sufficient capacity to support people with ME or CFS in addition to the many other people with long term conditions for whom they will be providing care.

For those younger people in transition, there is a need to develop specific transition pathways as part of local clinical guidance. This will help ensure effective transfer of care to adult services.

Recommendation 14:

Local arrangements for transition to adulthood should be extended to cover the needs of young people with ME-CFS. These arrangements should be included in local care pathways.

11.3.3 Self-Management Programmes

The place for self-management in helping people with long term conditions is clear. That it has a role in relation to ME-CFS is also supported by the service providers and service users. Specialist service providers were concerned to ensure that advice given by the primary care team to people diagnosed with ME-CFS regarding self-management should be of a high standard and not likely to exacerbate symptoms. Similarly, whilst the move by some Health Boards to develop self-management programmes should be extended across Scotland, such programmes should not put already vulnerable people at increased risk. Self-management programmes for people with ME-CFS must therefore be subject to effective quality assurance and regulation. This is critical for both programmes directly provided and managed by the NHS, as well as those provided in the Third Sector or by independent sector providers.

Recommendation 15:

NHS Boards in Scotland should develop, or facilitate the development of, Self-Management Programmes to support people with ME-CFS.

These programmes should be subject to appropriate quality assurance:

(a) for the NHS, such quality assurance should be provided by NHS Quality Improvement Scotland; and

(b) for the independent or third sectors, guidance on quality assurance should be developed on a wide, partnership basis.

Recommendation 16:

Consideration should be given to developing an appropriate regulatory framework for the provision of Self-Management Programmes by independent or voluntary sector providers as for independent healthcare providers.

Tier 2 services – notably the primary care team – will have a major role in ensuring that the mechanisms established to manage long term conditions are applied in the case of people with a diagnosis of ME-CFS. This will mean that they actively review care provision on a regular basis. This will be especially so in relation to those who are severely affected.

11.4 Service Implementation at Tier 3

Chapters 7 and 9 identified that there was general agreement from service users and service providers that there was a need for specialist, Tier 3 services. However, there was a lesser degree of agreement about what would be the constituent parts of such a specialist service or how it should be organised. As Chapter 8 highlighted, apart from the experiences drawn from services in England and Wales, the international comparative analysis undertaken for this HCNA was also unable to draw any conclusions on what such a service should offer or how it could be organised. The following description of a specialist service is therefore based on a synthesis of the available evidence and expressed need. It is expressed in terms of what is likely to be needed across Scotland as a whole, rather than attempting to specify at either regional or board level what would be needed. As such it should be seen as a “best-fit” to guide NHS Boards – individually or regionally – to use in formulating their responses. It is not a statement of “best practice” which is to be followed unthinkingly.

11.4.1 The Multidisciplinary Team

At the heart of the Tier 3 Service should be a multidisciplinary team (MDT). The MDT could not provide a universal service covering all aspects of ME-CFS care. It would be responsible for:

- specialist diagnoses and assessment;
- initiation, establishment and monitoring of specific management interventions;
- care planning for more complex cases;
- supporting the community and primary health and social care services providing long term conditions management within Tier 2 services;
- providing education and training for health and social care professionals working with Tier 2 services; and

- providing advice to those developing and delivering self management programmes.

It is acknowledged these would be provided on both a clinic and outreach basis as required.

Whilst there was variation in opinions of what should constitute the MDT, there is sufficient consensus that it should include:

- Lead medical consultant – with a background in a suitable medical specialty;
- Other medical staffing;
- Clinical nurse specialist with a background in a suitable nursing field;
- Physiotherapist;
- Occupational Therapist;
- Dietician; and
- Clinical Psychologist.

It was, however, stressed by all concerned that all staff within the MDT should have experience in dealing with ME-CFS, including severe cases of ME-CFS. The MDT also needs to be sufficiently resourced to be able to manage people with ME-CFS using new technologies (eg telehealth and telecare), as well as being able to provide domiciliary care.

11.4.1.1 Lead Consultant and Medical Staff Requirements

If the data from Chapter 6 relating to specialist diagnostic and assessment use from NHS Lothian are used as a basis for specific medical staffing levels, it can be estimated for Scotland as a whole that – at current levels of use – by 2013 there could be between 1,177 and 1,261 new assessments and 2,859 and 3,364 review attendances. In the light of the activity data from NHS Lothian, it is estimated that provision of 4 dedicated clinical sessions for an ME-CFS clinic per week, based on the availability of clinical sessions for 40 weeks of the year. This estimate does not take into account the potential increase in ME-CFS patients being transferred to a ME-CFS clinic diverted from general clinical sessions provided across a range other specialities. This has been estimated to equate to a further 2 clinical sessions per week. Assuming that this is a fair reflection of clinical activity necessary across Scotland, this would equate to roughly 9 sessions of consultant and other clinical time per 1,000,000 adult population per week across Scotland dedicated to ME-CFS diagnosis, assessment and follow up and care. Overall, this suggests that roughly 37 clinical (consultant and other medical) sessions would be required weekly for the whole adult population.

After allowing for the necessary components within any medical consultant's job description, the consultant staff will have a series of wider responsibilities which need to be taken into account. These include the wider liaison role for the broader range of specialist services, encompassing other disciplines and services, notably in relation to paediatric services and involvement in the teaching and training of NHS and other staff, . Given also the pattern of the population split across Scotland which requires coverage across the more remote and rural areas, it may be more realistic to suggest that there is a need for a minimum of 3 whole time equivalent consultant posts across NHS Scotland (assuming a 10 planned activity contract).

In addition to the consultant staffing, there is clearly a need for additional medical sessions to provide ongoing care. This is a task that appropriately trained associate specialists, specialty doctors and sessional GPs, working under the clinical leadership of a consultant, would be well suited to undertake, depending on local needs and circumstances. In this context, a preliminary level of twelve clinical sessions across is likely to be required, though this is almost certain to expand for a period of time. In the longer term, the impact of better education and development amongst health care professionals generally will result in achieving a sustainable level of staffing across Tier 2 and 3 services. The indicative costs for a lead consultant and additional medical staffing, within the context of a wider MDT, are shown in Appendix 6.

At present there is no higher level, specialist training for specialists in ME-CFS. As a result, it is to be expected that any consultant level appointment is likely to be made from an existing specialist discipline which provides a basis from which the necessary clinical skills could be developed. In this regard clinical neurology, rehabilitation medicine, or infectious diseases are potential specialties. However, there are also likely to be physicians from other areas of speciality practice with an interest in ME-CFS who could be suitable. It may be that in the short term it proves difficult to appoint whole time consultants to such posts. In this circumstance, consideration may need to be given to allocating consultant planned activities within the job plans of existing consultants. Whatever the specialty of the consultant, it must be expected that there will need to be a period of time where training and development will be needed to allow them to become effective specialists in the area of ME-CFS.

Recommendation 17:

a) Consideration should be given as to how best to facilitate the development of consultant posts for ME-CFS at NHS Health Board or NHS Regional Planning Group level across Scotland. These consultants should lead multidisciplinary teams to provide services at Tier 3.

b) Consideration should be given as to how best to provide an appropriate skill-mix in medical provision as part of the multidisciplinary teams to provide services at Tier 3.

11.4.1.2 Tier 3 MDT Healthcare Professionals

The review of available guidelines on ME-CFS showed them to be remarkably silent on indicative workforce requirements. This includes NICE whose implementation advice to support its Clinical Guideline recommends local action planning to support implementation[29]. Similarly, the Department of Health guidance which supported the development of the Clinical Network Co-ordinating Centres in England, does not provide any insights[30].

As with medical consultants, there is little hard data on which to base a specific requirement for healthcare professional staffing within the Tier 3 MDT. Clearly, if there is a need for at least three whole time equivalent medical consultants, then one can presume they would each lead an MDT comprising the staff described above. This would be broadly in line with the approach taken within the English Clinical Network Co-ordinating Centres.

However, this may not be a sufficient level of workforce to provide the service elements described above and a more structured approach to determining the necessary Tier 3 MDT workforce should be undertaken either by individual Health Boards or NHS Regional Planning Groups. This could usefully follow the recent guidance published by Skills for Health – Workforce Planning Team relating to the development of MDTs for neurological long term conditions[31]. Indicative costs for staffing an MDT are provided in Appendix 6.

Recommendation 18:

NHS Health Boards / NHS Regional Planning Groups should prioritise the development of consultant led services, supported by a Tier 3 Multidisciplinary Team for ME-CFS. Consideration should be given to a more detailed workforce plan in the medium term.

11.4.1.3 Initial Development of MDTs

The review of existing services across Scotland highlighted that there were no areas which had sufficient, appropriately trained or experienced staff that would be able to immediately establish such MDTs. Therefore care should be taken to recognise that all staff appointed to such MDTs will require a period of training (see section 12.2) to allow them to develop and provide a Tier 3 service. Time will also be needed to help develop local services working at Tier 2 to be able to work with a Tier 3 service.

Recommendation 19:

In establishing MDTs, the NHS Boards or NHS Regional Planning Groups should:

(a) ***ensure that once staff are appointed, an appropriate period of staff training is funded to allow an effective service to be established; and***

(b) ***ensure that MDTs have a suitable lead in time to develop effective collaborative working arrangements with local services at Tier 2.***

11.4.2 Managed Clinical Networking

The work of a Tier 3 MDT presumes that there is both local and regional managed clinical networking. At the Scottish level, the existence of at least three such MDTs should provide the basis for a national ME-CFS network which can ensure there is a sharing of knowledge, experience and expertise, provide cross-cover arrangements and allow for the development of a more focused approach to researching and evaluating treatment interventions in collaboration with the wider research community. Such a network could also provide a way of accessing UK wide and international research and clinical practice. The ability for networks to rapidly report possible adverse reactions, notably in relation to care or treatment regimes is also important in the context of ME-CFS.

Within each of the regional areas, local networks to Health Board services will be necessary to ensure that the care pathways are well specified and that patient referrals are as smooth and timely as possible. Whilst many of the basic arrangements for a network are already in place across Scotland, experience from other clinical networks shows that developing "managed" clinical networks takes time and appropriate resourcing. This is also borne out by the observation that whilst the CMO's Short-Life Working Group had recommended the development of such collaborative working in 2002, only two areas in Scotland have got as far as even formalising a "development" process of local MCNs.

Recommendation 20:

NHS Health Boards and NHS Regional Planning Groups should develop managed clinical networks in order to ensure that there are effective clinical services to meet the health care needs of people with ME-CFS.

Recommendation 21:

NHS Health Boards and NHS Regional Planning Groups, working with key stakeholders, should decide how best to ensure the development of such clinical networks for ME-CFS both regionally and across Scotland.

Recommendation 22:

NHS Health Boards should ensure that services which operate at Tier 2 for ME-CFS should have the opportunity and capacity to participate in the development and operation of the clinical networks at regional and national level.

11.5 Conclusions

Implementation of the model of service outlined in this chapter would go some way towards addressing the relative lack of progress that characterised NHS Scotland’s response to the report of the CMO’S Short-Life Working Group. It will also be supported by a number of actions which would seek to develop the wider infrastructure in which the NHS in Scotland operates. These are considered in the next chapter.

12 Infrastructure Issues

The work which has underpinned this need assessment identified a number of infrastructure issues which should be addressed. These may be broadly characterised as the need to develop effective clinical standards, developing education and training for professional staff and widening the research base for ME-CFS.

12.1 Developing standards for clinical practice

Effective health care delivery needs clear standards for clinical practice. Clearly standards of clinical practice will be supported by the type of national guideline for ME-CFS recommended in Chapter 8. However, as with other long-term conditions, services for people with ME-CFS will come from a range of both specialist and generalist services working at local, regional and national levels operating within network structures.

As a consequence, there is a need for explicit service standards for all services involved in caring for people with ME-CFS, irrespective of whether they are Tier 2 or Tier 3 services.

Recommendation 23:

NHS Quality Improvement Scotland should work with all interested parties to develop service standards for ME-CFS services in Scotland. Consideration should also be given to developing specific standards for clinical networks as part of this development.

12.2 Developing education and training

The need for better education and training of healthcare staff was a constant theme throughout the needs assessment, albeit that there was a lack of agreement on what the contents of such education and training should be. Some indication of what the content *could* be, may be inferred from the type of outcomes that were being envisaged for the education and training were it to be made available.

Across all healthcare professionals, there was an expectation from service users that education and training from the earliest stage of education would increase the acceptance of ME-CFS as a real, physical disease. It was also felt that such education and training would lead, amongst professionals, to a culture more accepting of people with ME-CFS, and ultimately lead to better therapeutic relationships.

Recommendation 24:

NHS Education Scotland should work with independent ME-CFS organisations to develop solutions to ME-CFS issues which would be included within education packages. These should be fed into undergraduate, foundation and professional training of health care staff across Scotland.

Similar outcomes were envisaged through the provision of education and training for existing health care staff across Scotland. However, given the very wide-ranging approaches to health care professional development, achieving this aim is very complex. Some of the service providers drew attention to the approach which had been taken by charities and independent providers in the palliative care sector in appointing appropriately experienced general practitioners as facilitators to help develop local education and training for primary care teams to meet what has become known as the “Gold Standards Framework” which promotes effective clinical practice within local clinical networks. This approach has been highly successful, especially when linked to the educational role of specialist Tier 3 services in supporting primary and community heath services.

Recommendation 25:

The Third Sector and Independent Sector agencies that work with and for people with ME-CFS should explore how best they can develop educational support for health care providers modelled on the approaches of similar agencies.

12.3 Using and Widening the Research Base

There was an overwhelming call for undertaking more research into the causes and treatment of ME-CFS from service users. Indeed, there were some users who felt that they would prefer to have no service development at all to meet existing needs and that all resources should be focussed on research. There was a broad range of possible research areas identified, with the strongly expressed view that it should not be undertaken in relation to psychiatry. Many – arguably all – of the areas for further research which were suggested reflected an underlying certainty that such research should focus on identifying the cause or treatment for ME-CFS.

Both service users and service providers considered that the current strategic direction for research into ME-CFS was insufficient and ill-focussed. Clearly, there is a need to revisit the overall strategy for research in Scotland and examine how the research base for ME and CFS can be broadened.

Irrespective of this, there was a general view expressed that the wide, existing research base should be disseminated to allow better understanding by health care professionals and service users.

Recommendation 26:

The existing research strategy in Scotland in relation to ME and CFS research should be reviewed by the Chief Scientist's Office and a new strategy developed, aimed at broadening the evidence base for ME-CFS. To ensure effective communication of the existing, diverse evidence base, consideration could be given to developing a Centre for Research Excellence and Dissemination.

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