

Objective rehabilitation parameters in chronic fatigue syndrome Jan b Eyskens

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The chronic fatigue syndrome: studies exploring gait automaticity and trunk-arm endurance as objective rehabilitation parameters

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*Good tests kill flawed theories;
we remain alive to guess again*

Sir Karl Popper (1902 - 1994)



Chapter 1

In search of a rational rehabilitation of chronic fatigued patients

Chapter 1

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Preface

Once in a while, everyone feels tired for a period of time. What if this feeling continues, especially if no underlying cause can be identified? How do we best approach patients' complaints?

In the early eighties, more and more patients presented themselves complaining of generalized pain and a persistent fatigue in physiotherapy clinics. No low-tech test existed to individualize exercise programs. In 1921 Muscio questioned if a fatigue test was even possible.

This thesis focuses on finding low-tech parameters that could help during the assessment and follow-up of patients with the chronic fatigue syndrome (CFS) fulfilling Fukuda et al.'s criteria.

At the start of my research on the subject, the personal computer was just born and PubMed was not yet available. What helped was reading, carefully listening to patients and watching them standing, walking, moving. Their fatigue seemed to be provoked by remaining upright for a prolonged time. On the other hand, lying down seemed to reduce the fatigue that patients were complaining of. The major aim of the 2000 pilot study was to test whether patients with CFS differed from healthy sedentary controls in their ability to hold their body up against gravity, to make (forceful) movements and to move around in space.

This pilot study - 'In search of a rational rehabilitation of chronic fatigue patients' - pointed to the fact that indeed the musculature of fatigued patients supposed to hold their bodies up against gravity was relatively weaker than the musculature to move, to make (forceful) movements and to move freely about in space.

Later, an already validated test (Timed Loaded Standing) was used to evaluate combined trunk and arm endurance confirming this dysfunction. A following research investigated automaticity of gait. Differences in quality of gait between CFS patients and non-disabled controls were found.

My interest as a physiotherapist, looking for data to adapt the graded exercise therapy approach to each individual patient, was aroused

by the scores of self report questionnaires related to the patient's complaints, namely fatigue and one's ability to function during the day. Only trunk-arm endurance proved to be statistically significantly related to the 'physical functioning' subscale of the 'Short Form Health Survey'. Although apparently a dysfunction in patients with CFS, the degree of lack of automaticity of gait seemed not to be related to the individual level of their complaints.

Since the etiology of CFS is still unknown although different biomarkers were studied, a 'gold standard' to assess and fine-tune diagnosis and rehabilitation results is still an important issue.

Concepts used in this thesis

Defining the Chronic Fatigue Syndrome

When to use the words tiredness, exhaustion or fatigue

Tiredness and exhaustion have to be distinguished from fatigue. A workable vocabulary related to the concept 'fatigue' has been written in 2007 by Trendall et al. [1]. Tiredness can be defined as "A normal lessening of strength and energy. Dispelled by rest, sleep or nourishment" [2]. Exhaustion can be seen as "the body's energy reserves exceed its ability to replenish these reserves forcing the body to stop functioning" [2] but also as a "totally decompensating and overwhelming state of illness" [3]. Fatigue is supposed to be normal when "related to exertion. It is rapid in onset, short in duration and relieved by a good night's sleep and/or nourishment". Normal fatigue 'invites' the organism to avoid further strain and stress [2] [4] [5].

Fatigue is one of the most frequent complaints in first line medical practices [6]. It is important to consider that treatment of fatigue has a better prognosis when started in the first six months [7]. Sometimes fatigue is related to a medical treatment [8] or can be understood as a specific physical problem, as for example in chronic obstructive pulmonary disease [1]. A tool was developed to help the assessment of fatigue in these pulmonary patients based on qualitative research

methods. Four themes emerged out of this research: patients' interest in surroundings; their physical capacity; their personality and mental state; and their physical appearance [1].

Chronic fatigue as a term

When fatigue as a complaint lasts for more than six months and when it lacks medical causes, it is said to have become chronic [9] [10]. After a thorough investigation regarding at least underlying neurological, somnologic, psychiatric or internal pathology, a 'diagnosis by exclusion' can be made to label the patient's complaint: chronic fatigue syndrome (CFS). The word 'syndrome', pointing to a "combination of symptoms and signs which have been observed to occur together so frequently and to be so distinctive that they constitute a recognizable clinical picture" is useful in medical communication and research [11].

A following step consists of describing specific and uniform criteria to define the illness as is done in case definitions [12]. Following such a route is important for interpersonal communication and scientific purposes. Regarding communication, not only the patient him/herself, but all healthcare providers (physicians, physiotherapists, psychologists and others) will benefit from defining criteria as sharply as possible to describe a patient having fatigue as a primary complaint lasting for more than six months. This could help to fine-tune the assessment of this fatigue at the start of the treatment, but also when evaluating therapy and/or rehabilitation programs. Well-defined case definitions would not only help trans-disciplinary and medico-legal interactions and insurance procedures, but also every contact between each care provider and their patient [13].

In regard of research, precise case definitions are needed to allow researchers to hypothesize as focused as possible towards an underlying cause [14]. This is important since CFS patients - seen from terms and situations as tiredness and exhaustion - have normal or near-normal aerobic capacities when compared to a sedentary population and that their complaints are not detected by various physical tests [15] [16] [17] [18] [19].

For that reason, the following descriptions are not stringent enough

to define populations for scientific research: “Induced by the disease process or treatment, rather than activity. It usually has an insidious onset, is cumulative and persists over time. It is rarely dissipated by sleep, although it may temporarily be eased by rest, a change in activity or diversion” [2]. The same applies for “a decreased capacity to perform physical and mental work, overwhelming exhaustion, lack of energy and tiredness, and a combination of these” [20] [1].

At least twenty sets of diagnostic criteria exist for CFS [21] [22]. I will present an overview of the most used case definitions in a chronological way. This is only possible when also mentioning ‘myalgic encephalomyelitis’ (ME) and “systemic exertion intolerance disease” (SEID) as these terms were used in parallel regarding fatigue as a patient’s principal complaint.

Different case definitions regarding CFS and ME

Ramsay published a first case definition in 1988 [23] using the term ‘myalgic encephalomyelitis’ (ME) due to the resemblance of symptoms with those of patients suffering from polio [23] [24] [25] [26].

The same year, Lloyd et al. published a paper also using ME as a term [27], but two years later they used the label CFS [28]. From then on, the word CFS was mostly used to describe and detach patients’ fatigue complaints to outline diagnostic criteria when they experience their fatigue as severe, persisting or relapsed for at least six months. Their fatigue also has to be characterized as new or definite onset, not alleviated by rest, and when it represents the reason for having to diminish patients’ daily life activities [21] [22].

Also in 1988, Holmes et al. set up the first widely applied set of criteria for CFS in order to replace another term: ‘chronic Epstein-Barr virus syndrome’ [29] [30] [31] [32] [33].

The Holmes et al. case definition used major and minor criteria that patients had to fulfill in order to be labeled as ‘CFS’ [34] [35] [36] [37]. To them, CFS as a term could be used when a combination of non-specific symptoms, including severe fatigue, weakness, malaise, subjective fever, sore throat, painful lymph nodes, decreased memory, confusion, depression, decreased ability to concentrate on tasks, and various oth-

er complaints were presented [29]. This research group also pointed to a remarkable absence of objective physical or laboratory abnormalities [29].

In 1991, Sharpe et al. developed the so called Oxford criteria [38] [39], stating that the diagnosis of CFS could be attributed to patients with severe and disabling fatigue as their principal symptom and when fatigue was affecting their physical and mental functioning. Other symptoms may also be present: myalgia, mood swings and sleep disturbances. Psychiatric co-morbidities preclude the use of CFS as a label [38].

A few years later, in 1994, Fukuda et al. described a revision of the 1988 criteria by Holmes et al. in their 'Center for Disease Control & Prevention criteria' [40]. A thorough medical history, physical and mental status examination, and laboratory tests had to be used checking for patients' conditions requiring treatment. Complaints of medically unexplained fatigue, after clinical evaluation, needed to be explained as persistent or relapsing, that is of new or definite onset, not the result of ongoing exertion, not substantially alleviated by rest and results in substantial reduction in previous levels of occupational, educational, social, or personal activities. In addition, four out of eight other criteria have to be met: substantial impairment in short-term memory or concentration; sore throat; tender lymph nodes; muscle pain; multi-joint pain without swelling or redness; headaches of a new type or severely unrefreshing sleep; and post-exertion malaise lasting more than 24 hours. These criteria list various symptoms to exclude a number of patients and included a note on the use of laboratory tests.

The prevalence of Fukuda et al.'s criteria, the ones used in this thesis, depends on the type of population that was surveyed. In a general adult population, CFS ranged from 0.007% to 2.8% and from 0.006% to 3.0% in a general practice [41]. It is more common in women, in members of minority groups and when lower educated [42] [43]. Age range peaks are observed between 29 and 35 years of age. CFS is less common in children [44]. Another study stated that the pooled prevalence for self-reporting assessment was 3.28% and 0.76% for clinical assessment [45]. The same study also points to the fact that patients,

physicians, health insurance companies, and other stakeholders should be cautious of prevalence determined by self-reporting of symptoms.

In 2003, Carruthers et al. proposed the 'Canadian Clinical Case Definition' [44]. According to their criteria, patients must also show neurological, neuro-cognitive, neuroendocrine, dysautonomic, and immune manifestations [46]. The article states: "Patients who develop ME/CFS often lose the natural antidepressant effect of exercise, feeling worse after exercise rather than better". This is remarkable, given the fact that graded exercise therapy is an evidence-based treatment for CFS and the guideline for physiotherapists [43] [47] [48]. The question how to exercise has also been asked explicitly [49].

Several pathophysiological pathways in various fields have been investigated and significant heterogeneity characterizes even fatigued people meeting the diagnostic criteria for CFS. Afari et al. provide a good overview [41].

In 2012, a Flemish Dutch organization (Vlaams-Nederlandse Onderzoekersgroep-Chronische Vermoeidheid, VNO-CHROVER) thought it was important to use a clear definition since fatigue as a complaint was frequently reported in primary and secondary care but also caused insurance related problems [13]. They agreed on the definition: "Chronische vermoeidheid is het zelfgerapporteerde lichamelijk en/of mentaal onwel bevinden, langer dan zes maanden aanhoudend en zich uitdrukend in uitputting, als gevolg waarvan iemand lichamelijk en/of mentaal niet kan functioneren op het door hem/haar gewenste niveau." This can be translated as follows: "Chronic fatigue is the self-reported physical and/or mental feeling of being unwell, persisting for more than six months, expressing one's exhaustion, as a result of which someone can not physically and/or mentally function at his/her desired level." [13] The article states that, since objective tests are not available, fatigue can only be measured using self-report questionnaires [13].

Their definition stresses the fact that the uttered fatigue should be felt as an unpleasant physical or mental feeling lasting more than six months and influencing patient's daily life functioning. It clearly states: the patient is not able to do what he/she wants to do. This can be related to a physical level (sport, household, physical exercise), a mental

level (attention, concentration, memory) or to both levels [13].

In 2015, a committee of the 'American Institute of Medicine', recommends that the complaints/disorder should be renamed as "systemic exertion intolerance disease" (SEID) and advises to replace 'ME' but also 'CFS' as labels stating that it is not a psychiatric or psychological illness [39]. Their report concludes that "a thorough history, physical examination, and targeted work-up are necessary and often sufficient for diagnosis", this to allow large percentages of undiagnosed patients to get an accurate diagnosis and receive appropriate care [50]. The report reintroduces the phrase 'disease' to substitute the concept of 'syndrome' [39], although both phrases have different meanings and connotations [51].

The need for objectivity during assessments

Whatever case definition is used, one problem when assessing chronic fatigue remains the same: the lack of objective physical biomarkers and/or signs that could be related to the individual patient's degree of fatigue [52].

Descriptive case-definitions based on symptoms and invalidation, without the character of a clear nosological or diagnostic entitlement, pose a permanent diagnostic problem in patients complaining about chronic fatigue. Still, patients should be classified according to their severity in order to optimally guide therapy, to predict prognosis and to estimate medico-legal and insurance questions [21] [53].

Specific qualitative and quantitative aspects of an exercise program have to be underpinned by randomized controlled trials using case definition as precise as possible, to minimize the risk of bias [54].

The only way to assess the subjective complaint of fatigue is the use of self-report questionnaires. Their outcome makes it possible to keep track of the degree of the fatigue of the individual patient at intake and in follow-ups [55] [56].

When asked for the precise functions that provoke their fatigue, most patients with CFS answer "having to be upright for a prolonged time" [57].

Asked for the ones that reduce fatigue, the answer is often "lying

down". During their intake, most patients are sitting in a slumped position. This made us think that the individual degree of fatigue could eventually be objectified measuring patient's global functioning.

This thesis will explore the search for two low-tech physical tests that could eventually be used during assessment and follow up of patients with chronic fatigue fulfilling Fukuda et al.'s criteria.

Self-report questionnaires related to fatigue as a subjective complaint

Physicians have only subjective informative parameters, namely the patient's story and complaints regarding his/her fatigue, eventually underpinned by results obtained from self-report questionnaires to label the complaints to one of the case definitions [58].

Often one or more of the following questionnaires are used [59]: the multidimensional fatigue inventory [60], the "Checklist Individual Strength" (CIS), a scale with a good reliability measuring fatigue and related aspects during the last two weeks [61] [62], and the MOS 36-item short form health survey (SF-36), a scale widely used for measuring health and functional limitations or health-related quality of life [63] [64].

In our research [65], we used the CIS's total score and its subscale 'fatigue' [61] [62] and two subscales of the SF-36: 'physical functioning' and 'vitality' as both subscales are expected to express specifically CFS patients' functional impairment [63] [64].

These 'subjective procedures' pose practical problems for physicians, health care organizations and the medico-legal system [66]. Therefore, different objective tests have been developed to help to quantify the severity of subjective complaints during assessments [67] [68].

Possibly, this could lead to differentiating sub-groups, to individualization of the degree of patients' fatigue and to distinguishing parallel disabilities. It could also help the assessment and follow-up of the patients and different policies aimed to activate patients fulfilling the criteria [69].

Rehabilitation: regaining the functional abilities in despite of prolonged fatigue

The World Health Organization defines rehabilitating people suffering disabilities as “a process aimed at enabling them to reach and maintain their optimal physical, sensory, intellectual, psychological and social functional levels”. Rehabilitation also involves adopting another way of living, consisting of choosing functions atypical for the life before the accident or disease.

The available knowledge to set up a rehabilitation program is based on recommendations obtained using randomized controlled trials and, if possible, on a meta-analysis of various studies and it is subject to certain barriers [70].

A rehabilitation program for an individual patient has to consider two distinct classifications in parallel: the International Statistical Classification of Diseases and Related Health Problems (ICD) and the International Classification of Functioning, Disability and Health (ICF).

The ICD is designed as a health care classification system. It provides a system of diagnostic codes to classify diseases but also the indications, symptoms, complaints, the social circumstances, and related external causes. A first version (ICD-6), published in 1949, was suitable for reporting morbidity. After numerous fixes and improvements, a new version (ICD-11) is expected by 2018. The system is also used to structure insurance and reimbursement modalities. Presently CFS patients fulfilling Fukuda et al.'s criteria are listed using 'Code R53.82' in the ICD-10 version.

Second, the ICF classification is used to describe and measure health and disability and takes into account contexts and environmental factors of patients and are written out for patients with CFS [71]. The ICF criteria combine the information regarding the body and its functioning and relate this information to the patients' possibilities to participate in his/her personal environment. In respect of rehabilitation, the entire sphere of CFS requires the classification of all relevant functions, including the contexts of patients, within the ICF framework [72].

Using both ICF and ICD criteria offers an interactive and dynamic way

of thinking during the assessment, paying attention to the continued evaluation and lastly the rehabilitation of patients with CFS. A way to apply both classifications has been proposed as 'the field model', a tool that can be used during the intake and assessment of chronic patients, especially in the case of fatigue treated in a general medical practice [73] since it can help patients and health care providers alike [74] [75].

Fatigue as a symptom is also commonly seen in physiotherapy clinics where it is still a challenge to apply the guideline of graded exercise therapy (GET) for the rehabilitation of patients with CFS [76] [77].

It is based on a 1997 randomized controlled trial by Fulcher and White showing results regarding fatigue, functional capacity, and fitness, obtained one year after a twelve-week program consisting of graded aerobic exercises in patients who had complaints for a mean of 2,7 years. The alternative treatment was a 'flexibility' program [78].

As a conclusion, these findings support the use of appropriately prescribed graded aerobic exercise in the management of patients with the chronic fatigue syndrome.

Trunk and arm endurance

Due to the functional problems and complaints of older osteoporotic women (chronic back tiredness and fatigue) Shipp et al. used the 'timed loaded standing (TLS)' test, a measure of combined trunk and arm endurance [79].

TLS measures the time a person can stand while holding a two-pound dumbbell in each hand with the arms at 90 degrees of shoulder flexion and the elbows extended. Important aspect in relation to this thesis is that statistically significant correlations were found between TLS, gait velocity, a six minute walk distance, and the MOS-36 subscale Physical Function [79].

Since provocative activities in osteoporotic patients were similar to those in patients with chronic fatigue (back tiredness when standing, working with the arms in front of the body, sitting to rest, planning rest periods), I used the TLS test to look for a similar weakness in CFS patients [80].

Walking

Gait is defined as a person's manner of walking, that is, the act of traveling by foot. While walking, proprioceptive senses are generated including the senses of position and movement of our limbs and trunk, next to the sense of effort and force needed [81].

A task or function is supposed to be automated if an additional task is not influencing the first one [82] [83] [84].

Loss of automaticity of gait has been described in old people in relation to falling over and has implications for the rehabilitation [85] [86]. In patients suffering from Parkinson's disease, loss of automaticity of gait worsens their ability to perform secondary tasks [87]. Also in Parkinson's disease, deficits in attentional set shifting, dual tasking and response inhibition is related to the saturation of frontal processing mechanisms due to a loss of automatic motor control [88]. In stroke patients gait automaticity may be restored over time to a functional level [89].

It is proposed to discourage smart-phone use while walking since it affects the pedestrian's awareness of critical roadside events; when the walker is feeling unstable and feeling insecure, there will be consequences during dual tasking; although it will not be thought of as causing the consequences except perhaps after a few falls [90] [91].

In clinical settings the "Stops walking when talking" (SWWT) is a conventional test conceived by Lundin and Olsson [92] [93].

During visits to hospitals in Scandinavia I saw physicians walking in hospital corridors with hospitalized patients nearing the end of their rehabilitation. When the physician perceives loss of automaticity of gait when asking the patient to talk about his/her home situation, the patient was referred to an intermediate solution, for instance a rehabilitation center [92] [94].

Research questions

Observations in a pilot study

A pilot study was set up in 1998 to eventually improve the assessment of patients with CFS [95]. The idea of this pilot study emerged from observing how patients with CFS performed their exercises. Already at the start of their movements, an inefficient habit could be observed. In a vertical position, during either sitting or standing, it seemed as if they were pulling their head down - pulling their head to their neck - as the first procedure in reshaping their body. We even noticed that when lying on their back, their head was pulled in their necks - downwards - before engaging in the movement.

The pilot study's major aim was to test whether patients with CFS differed from patients with Spasmophilia as well as patients with Fibromyalgia syndrome in their ability to hold their body up against gravity,

Pilot study populations

Clinical populations based on referrals by general practitioners were used. The criteria were not the same as is customary for scientific research. In eventual follow-up research, each individual's functional abilities in daily life and eventual traumatic experiences during their lifespan could better be quantified.

The populations consisted of patients with a medical diagnosis of Chronic Fatigue Syndrome (CFS) [40], Fibromyalgia Syndrome (FMS) [96] or Spasmophilia Syndrome (SPS) [97], the latter being the control group.

The three groups were selected from the patient's files of the Department of General Internal Diseases at the University Hospital of Antwerp, UZA, and invited for a physical examination. Objectifying the patient's functional status was done by means of anamnesis and clinical examination, executed by the referring medical doctor (G. M.), aimed to define the degree of dysfunction.

261 patients were invited by postal mail, of which 241 indeed received their letter of invitation (51 CFS, 57 FMS and 153 SPS patients).

99 patients responded positively (38%). Finally, 92 patients (34%) participated in the study: 19 of the 51 CFS patients (37 %), 14 of the 57 FMS group (24%) and 55 of the 153 SPS group (35%). The average age among groups was similar. For CFS mean age 40,6 yr, (range 23–61 yr), for FMS mean age 40,9 yr, (range 18–60 yr) and for SPS mean age 41,0 yr, (range 19–66 yr). Prior examinations indicated that the general cardiac condition did not differ from that of a normal population, nor was there a difference between the three groups of testees.

All physical measurements were done by the same clinical researcher.

Methods used in the pilot study

Intensity of fatigue

By a visual analogue scale (VAS) [98], participants were asked to compare the actual intensity of their fatigue.

Measurement of pain thresholds

Pain thresholds at eighteen Tender Points (topographically defined by the American College of Rheumatology) [99] were measured by using a ‘pain threshold meter’ (Wagner Pain Test™ Model FPK Algometer Figure 1.1) so that not only the number of positive points, but also the



Figure 1.1. Algometer, used to measure pain thresholds of Tender and Trigger Points in test subjects with myofascial pain and fibromyalgia expressed in Kg/cm². Equipped with a pressure distributing surface, it can be used to measure the amount of force performed by the test subject and is expressed in Kilograms. The arrow in the figures below indicate where and in which direction the device was placed to measure the resistance of the 1RM (resistance maximale) force exerted by the test subject. Note: Strength was measured through making use of a Wagner Pain Test™ Model FPK Algometer with a pressure pad.

Myofascial Pain Index (MPI) could be calculated according to the 1990 guidelines of the American College of Rheumatologists [99] [100] [101].

Psychological parameters

A Tampa Scale for kinesiophobia [102] was used to evaluate differences in fear of movement.

Morphological parameter

The possibility of the presence of short humeri being a provocative factor in myofascial pain as stated by Travell & Simons was checked [103]. Travell and Simons referred to the book of reference, 'Human-scale 1/2/3', being an important tool for everyone who designs furniture for the human body. Dr. Travell prescribed her patients with short upper arms, amongst them the late President J.F. Kennedy, to elevate the armrests of their office chairs and even to use older style rocking chairs [104].

Functional parameters

The parameters used for the evaluation of muscle strength of the arms in an upright standing position were: the power to push each arm upwards/downwards from a standing position (arms stretched forward by 90°, figures 1.2 and 1.3), exo- and endorotations of both arms (elbows flexed, figures 1.4 and 1.5).

Given the fact that the project was a pilot investigation to observe differences between the three populations tested, no normal values were used. Today, it is clear that the absence of a healthy control population in the pilot project is a major shortage [105].

Results of the pilot study

Fatigue

Fatigue intensity was similar in the groups that were studied. The CFS group reported more fatigue in the past; actual fatigue was most severe in the FMS group [40] [96].

By splitting up groups according to the length of their upper arms, we found a correlation between 'fatigue now' on a VAS scale and short humeri as a specific morphological characteristic particularly in the CFS group.

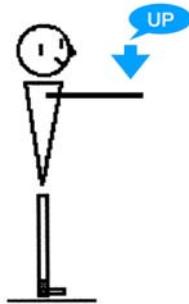


Figure 1.2. Testing the strength to push each arm upward from a standing position.

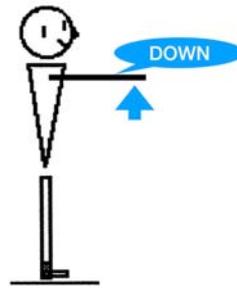


Figure 1.3. Testing the strength to push each arm downwards from a standing position.



Figure 1.4. Exo-rotation of the upper arm, elbow flexed.



Figure 1.5. Endo-rotation of the arms, elbow flexed.

Pain

Both FMS and CFS groups met the ACR criteria regarding the number of Tender Points (scoring 11 or more points reported to be painful with a pressure less or equal than 4 Kg/cm² measured at 18 topographically defined places. The SPS group scored slightly lower.

However, MPI (myofascial pain index) data as measured (the mean value measured on these 18 topographic points) were, as could be expected, lowest (low values pointing to more sensitivity and thus felt as more painful for the same pressure) in the FMS group, followed by the CFS group, and highest in the SPS group [97].

Measuring MPI values may therefore be more useful to evaluate the difference between CFS and FMS patients compared to only counting the number of Tender Points.

A recent article by McManimen and Jason [105] implies that when patients with CFS also meet the criteria of fibromyalgia as a secondary diagnosis, this situation appears to amplify their complaints of post-exertion malaise and worsens patients' physical functioning compared to those meeting only CFS criteria [106] [107].

Kinesiophobia

The Tampa Scale for kinesiophobia (having a cut off value of 36) proved only for FMS to be significantly positive. However, another phenomenon arises in relation with this scale. Tampa scores, spread out versus patients' ages, demonstrate that the fear of moving increases with age in all three groups (Figure 1.6). In other words: the chance that patients will revert to move more spontaneously decreases with the duration of their symptoms. One can therefore expect a further deconditioning if patients are not motivated to move in a structured manner from the onset of the complaint.

Muscle strength

The strength to lift each arm upwards from a standing position is weaker in CFS and FMS groups compared to a non-diseased population. We looked at the relationship between reported values of fatigue and measured values for 'lifting the arms'. The weaker the strength to lift the arms, the greater the complaints of fatigue (Figure 1.7). This is compatible with the provoking activities and postures as reported by these patients.

In order to exclude generalized muscle weakness [108], bilateral measurements of shoulder exo- and endorotation (elbows flexed) as well as the value of pushing each arm caudally (from a stretched forward position at 90% elevation) were taken.

We obtained the ratio 'posture/movement apparatus' by dividing the average value of lifting both arms by the average of the six other measurements: arm caudally, endo- and exorotation left and right shoulders. This ratio was lowest in the CFS group.

Reported intensity of fatigue (measured using a VAS scale) was higher in CFS patients when the above-described ratio was lower, confirming a selective weakness of 'posture apparatus' muscles (Figure 1.8). As far as we know, this line of reasoning and the calculation of the ratio of

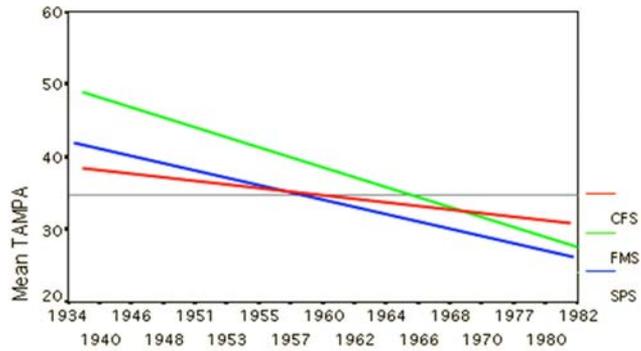


Figure 1.6. The Tampa Scale for kinesiophobia versus date of birth of CFS, FMS and SPS patients.

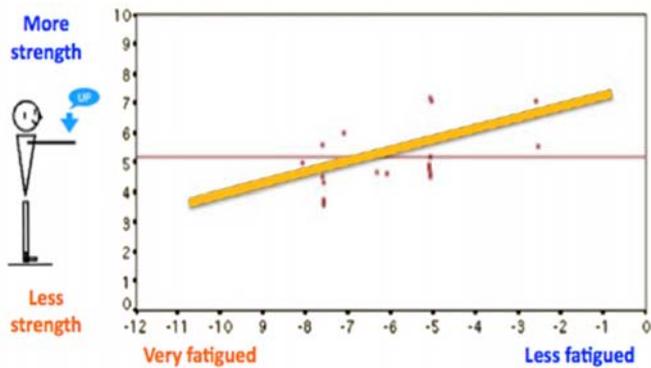


Figure 1.7. The strength to lift each arm up (forward stretched by 90° to the body) compared to 'fatigue now' obtained using a visual analogue scale.

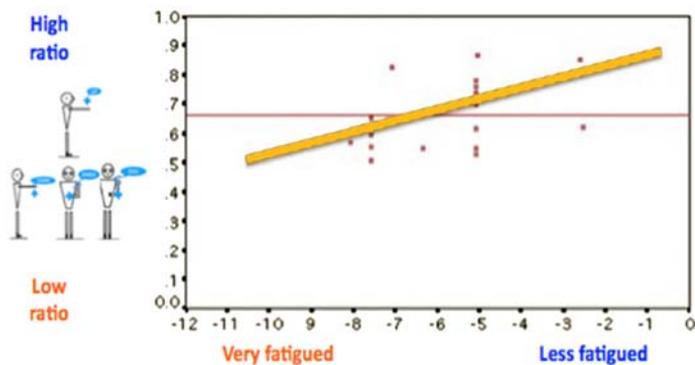


Figure 1.8. the ratio 'posture/movement apparatus' - obtained by dividing the average value of lifting both arms by the average of six other measurements: arm caudally, endo- and exorotation left and right shoulders - opposite to the reported fatigue on a visual analogue scale. This ratio was lowest in the CFS group.

the 'posture/movement apparatus' are original and not yet introduced in the medical literature. This finding can be important, because it can serve to assess, to retest and to compare results of a rehabilitation approach.

The start of a search for low tech tests

This pilot study revealed that CFS patients included show a reduced possibility to load their anti-gravitational means used when standing as a specific weakness that asked for more substantiated research [103] [118]. Since the results were compatible with the provocative (standing and sitting down) and reducing (lying down or slumped sitting) factors when asked for at intake in patients with these complaints and fulfilling Fukuda et al's. criteria [40], this was the start of a search for low tech tests.

Three research questions

Since the pilot study [103] pointed towards a deficiency in postural musculature of CFS patients, two main research questions were formulated.

In order to test whether CFS patients differ from other populations in holding themselves upright, either while sitting or standing, my first research question is: "Do patients with CFS have a deficiency in holding themselves upright against gravity?"

To find an answer to that question, an already validated test was used, Shipp et al.'s "Timed loaded standing: a measure of combined trunk and arm endurance suitable for people with vertebral osteoporosis" [78].

My second research question originated as a result of responses given by CFS patients when asked what (for them) constitutes a provocative or reducing factor while walking. Often their answer was that walking at the seaside or in a quiet environment was not that bad, whereas walking in busy shopping streets was very difficult and provoked their typical fatigue, sometimes for a time span of several days after the walk.

This led to the second research question: "Does automaticity of gait

in a population of patients with CFS differ from that of a healthy control population?”

In order to be able to answer this question, I adapted Lundin and Olson's 'Stops Walking While Talking' test (SWWT) [92] to a 'Stops Walking with Eyes Closed with secondary Cognitive Task' (SWECCCT) test with only a few minor changes [109].

Both research questions resulted in an objectivation of the dysfunction: TLS and SWECCCT show to be impaired in CFS patients compared to a population without CFS.

While two physical functions (trunk-arm endurance, automaticity of gait) indicate to be reduced in a population of patients with CFS fulfilling Fukuda et al.'s criteria, personal data of subjects in this population obtained through self-report questionnaires ('Checklist Individual Strength' [61] [62] and the 'Short Form Health Survey') were available [63] [64]. For the measurement of self-reported fatigue levels, the CIS' total score and its subscale 'fatigue' were used and for self-reported physical functioning and vitality two SF-36 subscales ('physical functioning' and 'vitality') were evaluated.

These data provided the opportunity to formulate the following research questions:

- 1) Does a relationship exist (and if so, in what way) between CFS patients' trunk-arm endurance and self-reported levels of fatigue, physical functioning and vitality?
- 2) Does a relationship exist (and if so, in what way) between CFS patients' automaticity of gait and self-reported levels of fatigue, physical functioning and vitality?

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