The University of Rome "Foro Italico"
The German Sport University, Cologne
The University of Southern Denmark, Odense
The Norwegian School of Sport Sciences, Oslo
The University of Vienna

European Master in Health and Physical Activity

"Self-Efficacy as an assessment tool in a multidisciplinary rehabilitation treatment for young CFS/ME patients"

A longitudinal pilot intervention study

Anita S Monsen

Student
Anita Skimten Monsen

Marit Sørensen

Supervisor
Marit Sørensen

Anders Farholm

Co-Supervisor
Anders Farholm

Academic Year: 2013 - 2015

Abstract

Introduction: Myalgic encephalomyelits (ME), also known as Chronic Fatigue Syndrome (CFS) is characterized by unexplained, long-lasting, disabling fatigue resulting in severe impairment in daily functioning. Capability to resume a normal life-style including social activities, schooling, sports and other activities, are essential for young CFS/ME patients for physical-and psychological development. The aim of the present thesis is to examine self-efficacy for being physically active as an assessment tool in treatments for young CFS/ME patients, and if self-efficacy can influence participation in PA. This research is the first to include both objectively measured PA and self-efficacy in a longitudinal design for young CFS/ME patients. **Method:** Data was collected from a pilot study conducted at a rehabilitation institution in Norway. Self-efficacy scale (SES) and PA was measured at 3time points (TPs); before (TP1), during (TP2), and 12 weeks after receiving a 4-week multidisciplinary treatment (TP3). The treatment included graded activity treatment (GAT), and development of individual activity plans as a coping strategy to resume a balance between activity, rest and a normal sleep pattern. **Results:** 24 participants (age 17, 83 SD ± 2) were included for analyses. Results indicated increased self-efficacy for being physically active during the treatment (nearly significant, $p=0.06$), but were not maintained 12 weeks after. Neither change in upright activity nor MVPA between the TPs were significant in the mean time of the total sample. However, participants who increased upright activity during the treatment had a decrease 12 weeks after ($p<0.05$), while participants who decreased upright activity during treatment maintained their activity 12 weeks after ($p>0.05$). There were a negative relationship between PA and SES during the treatment. This can be explained by mastery in coping strategies involving reduction in activity to avoid symptoms. **Conclusion:** Feasibility of design and method are considered as good, giving opportunity to investigate changes self-efficacy and PA over time in a safe environment. Because of low statistical power this results should be used with caution. However, assessment of self-efficacy in treatment for CFS/ME is informative and important to understand PA behavior, but further investigation is needed.

Forord

Jeg fikk interesse for CFS/ME under mitt arbeid som idrettspedagog på CatoSenteret. Jeg hadde hørt om CFS/ME tidligere, men ikke i forbindelse med utdannelsen. Jeg kunne ikke forstå hvorfor disse pasientene var pasienter. Eller hvorfor de kunne være aktive den ene dagen, men ikke den neste? Det var frustrerende og ikke kunne forstå, så mer kunnskap måtte til! Søken etter kunnskap ble vanskeligere enn forventet. Det var mange teorier og konklusjoner, men få svar….

Mitt felt er fysisk aktivitet og helse. For meg er det helt nødvendig å være i bevegelse, både for helsen, men også for humøret og "godfølelsen". Jeg tar det forgitt at jeg kan delta i de aktivitetene jeg ønsker, eller kunne løpe til toget når jeg er sent ute. Dette er noe en CFS/ME pasient må planlegge, tilrettelegge og vurdere for og unngå å bli sengeliggende, eller alvorlig syk. Jeg ønsker å kunne bidra, støtte og forstå disse pasientene, og har derfor forsøkt å skrive en oppgave som reflekterer dette. Jeg håper jeg har bidratt med noe fornuftig, som kan være med på å videreutvikle forståelse for denne sykdommen.

Jeg vil først og fremst takke veilederen min Anders, som gjorde en vanskelig jobb litt lettere! Statistikk og SPSS…. Ja… hele PC'n hadde vel flydd ut av vinduet hadde det ikke vært for deg. Jeg vil også takke CatoSenteret for samarbeid og datainnsamling, og håper på videre forskning og spennende resultater! Jeg vil også takke deltakeren for at de har bidratt med spennende resultater og svart på det samme spørreskjemaet 3 ganger, og snart en 4 gang!

Nå er det på tide å bli sosial igjen! Takk til kjæreste, familie og venner som har hatt forståelse for at dette har tatt tid og oppmerksomhet!

og PL sier "it's time to move"!

Anita Skimten Monsen

Abbreviations

ATP	Adaptive Pacing Therapy
CBT	Cognitive behavioral therapy
CFS	Chronic Fatigue Syndrome
CS	Catosenteret (rehabilitation institusion)
DLW	Doubly labeled water
ESES	Exercise Self-Efficacy Scale
GAT	Graded Activity Therapy
GET	Graded Exercise Therapy
HRQL	Health Related Quality of Life
ME	Myalgic encephalomyelitis
Min	Minutes
PA	Physical activity
PEM	Post-exertion malaise
PNI	Psycho-neuro-immunological
PSES	Physical self-efficacy scale
PL	Polar Loop
SCT	Social Cognitive Theory
SES	Self-Efficacy Scale
QOL	Quality of life

List of Figures

List of tables

Table of content

1. Introduction

Myalgic encephalomyelits (ME), also known as Chronic Fatigue Syndrome (CFS) is characterized by unexplained, long-lasting, disabling fatigue resulting in severe impairment in daily functioning (Prins, van der Meer & Bleijenberg, 2006). Recently, there has been increased interest in the field of CFS/ME which has caused a growing body of the literature on the topic. However, more knowledge is needed to understand the pathophysiology of CFS/ME and what kind of treatments that may improve the chance of recovery.

A challenge in the search of new knowledge in CFS/ME is the variety of case definitions and diagnostic criteria, causing heterogeneity in the sample of patients. There is no cure for CFS/ME, but treatments may help relieve the symptoms. The National Institute for Health and Care Excellence (NICE) recommends individual programme of treatment with the aim to maintain, and if possible increase emotional and physical abilities, as well as managing the physical and emotional effects of the symptoms (NICE, 2007). Maintenance of daily functioning and physical activity (PA) is important for health, and especially for young patients who are in a critical period of the physical and psychological development. Research has shown that inactivity, drop-out of school and other social activities is a consequence of CFS/ME (Sulheim et al., 2014).CFS/ME patients do not choose to be inactive. Inactivity is natural consequences of the complexity of their illness and should be taken seriously.

Post Exertional Malaise (PEM) is one of the core symptoms of CFS/ME. PEM is worsening of some, or all symptoms after physical or mental activity. A study which tested PEM using exercise testing on two consecutive days to examine the performance of CFS/ME patients and controls, found a major decrease in performance by the CFS/ME patient in the second test, while controls had no decrease in performance (VanNess, Snell & Stevens, 2007). Patients do not want to experience PEM, and therefore they may reduce or avoid PA. Individual treatment and follow-up is essential for most CFS/ME patients where they learn coping strategies and improve daily functioning. Commonly used treatments for young CFS/ME patients are; *Cognitive Behavioral Therapy* (CBT), *Graded Exercise therapy* (GET), *Graded Activity Therapy* (GAT) and *Adaptive Pacing Therapy (ATP)* also known as "*Pacing*". PA is a major part, and target in the treatment. Development of a baseline of activity can be beneficial to avoid worsening of symptoms also on days they feel particularly bad. Strategies to better cope with their illness is important for the capability to participate in preferred or important activities such as schooling are important and often part the treatment.

Effects and acceptance of the commonly used treatments are widely discussed in the literature. GET is experienced from some patients to worsen their symptoms (Francis, 2014), but there are little research supporting that GET is harmful (Larun, Burberg, Odgaard-Jensen and Price, 2015). ATP seems to be more accepted, but has shown small effect of improvements. An overall conclusion is that there are little consensus about CFS/ME. A multidisciplinary treatment that is tailor-made for each individual may be the most beneficial, but more research is needed. Heterogeneity in the patient groups which are included for research is common, probably because of the variety in use of diagnostic criteria. CFS/ME is therefore poorly understood and misunderstandings might occur. Hence, close follow-up and cooperation between patients and health care therapists are necessary for improvements and quality in treatments.

Change in behavior is often necessary for the patients to better cope with their illness, commonly restriction of some activities for capability to perform others. The behavior change is in the aim to find a balance between different activities, such as school and social activities, in combination with rest to avoid worsening of symptoms. Coping strategies for this illness are essential for daily functioning and a “normal” life style, including school and social activities. Assessment of the patient’s thoughts and beliefs about their capability to perform a behavior, also known as self-efficacy, might be informative in treatments. Self-efficacy as an assessment tool in young CFS/ME patients might provide understanding of the patient’s behavior, what to aim for and which goals to set during, or after a treatment period. In addition, self-efficacy may also predict future behavior (McAuley, Mailey, Szabo & Gothe 2013). Efficacy beliefs influence how people feel, think, and motivate themselves, as well as how they behave (Bandura, 1997). Self-efficacy is one of the most investigated psychological factors of behavior (McAuley et al., 2013). According to Bandura (1997), a highly efficacious individual will seek more challenging tasks and better cope with pain and setbacks than individuals with low self-efficacy. Bandura (1997) also claims that most behavior is shaped and initiated in thought, and that cognitive constructions act as a behavioral guide. Researchers have shown the importance of self-efficacy for being physically active both for patients and healthy individuals (McAuley et al., 2013).

According to performed literature search, no previous research has investigated the role of self-efficacy for being physically active in CFS/ME patients. This thesis is therefore covering a gap in the literature. The aim of this thesis was to investigate the role of self-efficacy for being physically active, in treatment for young CFS/ME patients (15-25 years of age). The main research question was if self-efficacy could influence participation in PA for young CFS/ME patients, and is the first to include both objectively measured PA and self-efficacy in a longitudinal design for young CFS/ME patients. Data was collected in cooperation with an ongoing pilot study. Data collection was performed in cooperation with an ongoing pilot-study conducted at *Catosenteret* (CS), a rehabilitation institution in Norway. Self-efficacy scale (SES) and PA was measured at 3 time points (TPs); before, during, and 12 weeks after receiving a 4-week multidisciplinary treatment programme, tailor made for young CFS/ME patients. The treatment included GAT, and development of individual activity plans which were developed as part of the coping strategies to resume a balance between activity, rest and a normal sleep pattern. The pilot study is conducted by CS in the aim to investigate improvements and recovery from the treatment. A longitudinal design was chosen to investigate a variety of outcome variables on the same participants over a period of time. Before describing method and findings relevant literature on the topic will be presented.

2. Theory

In this chapter the relevant literature in CFS/ME will be presented. This complicated and serious illness provides many challenges for research in this topic, which is reflected in the literature. CFS/ME is an umbrella term that includes both ME and CFS. For a long period of time, clinicians and researchers have developed separate case definitions and diagnostic criteria for both CFS and ME, although the names describe conditions with similar symptoms and unknown etiology. The variability in definition and diagnostic criteria is confusing and can cause misunderstandings about the illness. In the World Health Organization's (WHO) *International Classification of Disease (ICD),* Tenth Revision, which will be published in October 2015, both CFS and ME will be probably be coded identically and classified as disorder of the nervous system (ICD G93.3) (IOM, 2015). Importantly, "fatigue syndrome" is classified under mental and behavioral disorders (ICD F48.0). A variety of names have been proposed for this condition. Patients, clinicians, and researchers have still not agreed on which term to use. CFS is reported to be stigmatizing and that the term "fatigue" is not the defining characteristic of the condition and therefore leads to misconception. The name ME is criticized, because there is a lack of evidence for encephalomyelitis (brain inflammation). In a new report from the Institute of Medicine (IOM) published in February this year (2015) they suggest to rename the illness as "Systemic exertion intolerance disease" (SEID). The name SEID is developed from what they claim are the main characteristics, and what the patient experience (IOM, 2015). They also suggest in the report that a new code should be assigned to this disorder in the ICD that is not linked to chronic fatigue or neurasthenia (IOM, 2015).

If CFS/ME is going to be renamed is not yet decided. In this thesis the combined term is used to cover both conditions. This is in context to The Norwegian Directorate of Health, the subordinate agency of the Norwegian Ministry of Health and Care Services. CFS/ME is also used despite if other terms are used in the literature that is mentioned.

2.1 Epidemiology of CFS/ME

Epidemiological methods can give information about distribution of illnesses and allow investigators to make inference about risk factors as well as factors causing it (Jason, Porter & Rademaker, 2011). Association between risk factors can lead to better understanding of etiology, even if the cause is not known. However, if there is bias in the sampling methods it might lead to over- or underestimates of the true prevalence rate and might impede further research (Jason et al., 2011).

2.1.1 Prevalence

Early epidemiological studies in CFS/ME, estimated that about 20 000 adults in the United States (US) had the condition (Reyes et al., 1997). For a long period CFS/ME was thought to be a rare condition and little effort was done in developing of new knowledge (Jason et al., 2011). The low estimate from Reyes and colleagues might have been caused from selection bias or the methodology used, because only a few years later the prevalence was shown to be much higher. Jason and colleagues (found a prevalence of CFS/ME to be about 800 000 adults in the US (Jason et al., 1999). Today the true prevalence is still not known. Somewhere between 800 000 and 2.5 million Americans are estimated to have CFS/ME (Jason et al., 1999; IOM, 2015).

There is no epidemiological research from Norway, but projections from international studies indicate a total number between 10 000 and 20 000 CFS/ME patients with different severity (Helsedirektoratet, 2014_a). Pediatric CFS/ME is less common than in adults. It is suggested to be at least 300 children and adolescents with CFS/ME in Norway that is below 18 years (Helsedirektoratet, 2014_a). However, clinical experience indicate that far more than 300 children and adolescents are affected (Wyller, 2008). IOM (2015) have found estimates with numbers varying from 0.03 to 1.29 percent in pediatric CFS/ME. Differences in suggested prevalence may be because of different methodology and application of CFS/ME definitions, or underreporting of CFS/ME. Currently there is no exact estimate of pediatric CFS/ME, but we know the illness is highly prevalent and has a major impact on the ones that develop the illness. CFS/ME can be obtained in all ages, but is rarely reported in children younger than 10 years and elderly over 70 years. The average age of onset is 33 years (Davies & Crawley, 2008; IOM, 2015) and is also seen more commonly in females than in males (Knight, Scheinberg & Harvey, (2013).

2.1.2 Etiology and pathology

The etiology or cause of CFS/ME is not completely understood (Agency for healthcare Research and Quality [AHRQ] 2014; IOM, 2015). There are many possible precipitating factors such as viral infections, glandular fever or influenza virus, as well as exhaustion, mental stress, depression or traumatic events (Helsedirektoratet 2014_a; IOM 2015). There are lot of disagreement in the literature related to cause, development and maintenance of CFS/ME. In pediatric CFS/ME it is commonly reported to be developed after an acute flu-like or mononucleosis-like illness, but also a gradually onset of CFS/ME may occur.

Symptoms of CFS/ME are sometimes associated with an active virus infection. Therefore several studies have investigated if CFS/ME is caused by a virus. Epstein-Barr virus (EBV) which may cause infectious mononucleosis (also called mono) and other illnesses is one of the viruses along with other herpes viruses that have been found in some CFS/ME patients (Straus et al., 1985; Sairenji, Yamanishi, Tachibana, Bertoni & Kurata, 1995; Chia, 2005). Low circulating cortisol levels are also observed in CFS/ME patients and research has reported abnormalities of the hypothalamus-pituitary-adrenal (HPA) axis (Cleare, 2003). Other hypothesis of the HPA axis has been investigated such as hypersensitivity, but it is more likely to be a complex change in the HPA axis associated with CFS/ME rather than a single uniform change (Cleare, 2003).

Abnormalities of immune function such as reduced number of T-cells, reduced levels of cytokines, altered function of natural killer cells (NK-Cells), and altered levels of immunoglobulin IgG have been reported, but with inconsistent results (Sanders & Korf 2008). Psychiatric and psychosocial hypotheses are also suggested to be both the cause of CFS/ME and a maintaining factor. High personal standards of performance and perfectionism, over activity and work holism can be important predisposing factors for CFS/ME (Nijs, Meeus, Oosterwijck, Ickmans, Eupen & Koz, 2013) There is also research supporting a genetic disposition of CFS/ME but the mechanisms are still unknown (AHRQ, 2014; IOM, 2015). The genes that might be involved appear to have a role in the HPA axis functioning and the sympathetic nervous system. The importance of viral, endocrine, immune, psychological and other factors are investigated in several studies but there is still no consensus and agreement in whether this is causing CFS/ME, or is a consequence of CFS/ME. Various theories have been developed, but currently there is not enough evidence and adequate results for any of the proposed theories to be completely accepted in the literature. Multiple trigger mechanisms have been proposed, including infections, stress and trauma (Prins, 2006) as well as an autoimmune etiology (Fluge et al., 2011). The patients who suffer from this illness are often seriously ill, causing the patient to be house- or in worst case bed-bound. However, the cause and maintenance of the illness remains unknown. However, biological symptoms may affect psychological and social factors and vice versa. The human body and mind is in connection whatever that might cause or maintain any illness. A common way to describe a phenomenon in the biomedicine is cause-effect relationship. Illnesses are commonly far more complex, rather than simply one cause – effect. There might be multiple interacting causes and contributing factors.

2.1.3 The complexity

CFS/ME is as mentioned an illness that is poorly understood. Based on all the theories around the topic, CFS/ME seems to be caused by many different factors, and maintained by others. To use evidence-based approaches is not an easy task in the treatment of CFS/ME when there is little consensus. Experiences from the patients and specialists, in addition to the results from research, are all of importance when building an tailor made treatment for CFS/ME patients. The *Biopsychosocial* model can be used as a clinical guide to understand the patient's subjective experiences as an essential contributor to diagnose, health outcomes, and human care (Borrell-Carriò, Suchman & Epstein, 2004).The biopsychosocial model states that biological, psychological and social factors all plays a significant role in the context of the illness as well as maintenance, and is commonly used to understand CFS/ME. Models and research field can be used to understand the complexity of an illness and to build tailor-made treatments based on the literature.

Another model used to understand the complexity of CFS/ME is *Psychoneuroimmunology* (PNI). PNI is a field that studies the interactions between the central nervous system, the endocrine system and the immune system (Maier, Watkins, Fleshner 1994; Zachariae, 2009). Behavior and stress may impact these interaction processes and cause implication for health. There are two main pathways of bi-directional interaction between the brain and the immune system, the autonomic nervous system and the HPA axis. The immune system is affected both directly and indirectly through chemical transmissions messengers released by nerve cells, endocrine organs, and immune cells. In addition, the immune system may release cytokines which influence the nervous system. Stress or change in homeostasis may cause change in function of the immune system. A hypothesis is that an infection of the vagus nerve causes release of proinflammatory cytokines and other substances which lead to a pathological increased version of normal sickness behavior (VanElzakker 2013).

The variety in theories of what causes CFS/ME, what maintains it, and how the disease is understood is complicated and confusing. This is also reflected in the diagnosing procedures of the disease.

2.2 Diagnostic criteria and case definitions

A major problem in current research and literature is the variety in the use of diagnostic criteria and case definition for CFS/ME. Diagnostic criteria provide guidance to clinicians on the specific signs, symptoms or test results that indicate the presence of a disease (Coggon, Martyn, Palmer & Evanoff, 2005). Case definitions are specific types of diagnostic criteria listed to define an illness or condition (Center for Disease Control and Prevention [CDC] 2013). Case definitions establish the presence of disease-specific pathology through examination or testing. Case definitions are often assessed in terms of sensitivity, the ability to identify patients with a disease correctly, and specificity or the ability to exclude patients that do not have the disease (Coggon, Martyn, Palmer & Evanoff, 2005). The pathology of CFS/ME remains unknown and there is no diagnostic test for the disorder. Therefore assessment of sensitivity and specificity for the case definitions developed is not conducted precisely. Most of the diagnostic criteria for CFS/ME were developed through the consensus of experts. Currently about 20 sets of diagnostic criteria or case definitions for CFS/ME exists (Brurberg, Fønhus, Larun & Flottorp, 2014) and none of them serves as a gold standard. The Norwegian Directorate of Health, has published a National guideline for CFS/ME (Helsedirektoratet 2014$_a$). In the National guideline it is recommended to use *The Center for disease control and prevention* (CDC) diagnostic criteria from 1994 (Fukuda, 1994) or the *Canadian Consensus criteria* (CCC) (Carruthers et al., 2003). In pediatric CFS/ME it is recommended to use the diagnostic criteria developed by Jason et al., (2006). Further in this chapter I will go through some of the essential elements in some of the most common used diagnostic criteria.

The Fukuda definition (Fukuda, 1994) is the one most cited for research purpose (Bruberg, Fønhus, Larun , Flottorp & Malterud, 2014) as well as commonly used in clinical practice for both adults and in pediatric CFS/ME (IOM, 2015). The Fukuda definition requires prolonged or chronic fatigue that persists or relapses for over 6 months. In addition, four or more of the following minor symptoms must be present;

- *Impaired memory or concentration*
- *Sore throat*
- *Tender cervical or axillary lymph nodes*
- *Muscle pain*
- *Multi-joint pain*
- *New headaches*
- *Unrefreshing sleep*
- *Post-exertion malaise*

The Fukuda definition is criticized to be overly inclusive and to lead to heterogeneity among the patients (IOM, 2015; Bruberg et al., 2014). Another critique is that it does not require what some consider core symptoms of CFS/ME such as PEM and neurocognitive symptoms. Kennedy, Abbot, Spence, Underwood and Belch (2004) found significant differences in simple clinical measures and outcome measures between groups of patients who all fulfill the Fukuda definition. They also concluded that their response to treatment may vary (Kennedy et al., 2004). The CCC was developed by Carruthers and colleagues (2003) and published as a working case definition. The CCC includes more symptoms than the Fukuda definition to better identify patients with a unique combination of symptoms. It requires symptoms to be present over six months from six following symptom categories;

- *Fatigue, including substantial reduction in activity level*
- *PEM, and/or post-external fatigue*
- *Sleep dysfunction*
- *Pain*
- *Neurologic/cognitive manifestation*
- *Autonomic, neuroendocrine, or immune manifestation*

The above mentioned diagnose criteria are just two out of 20 that are developed in the attempt for better consensus. To just mention some of the others, the national Institute for Health and Clinical Exellence (NICE) developed clinical guidelines in 2007 (NICE, 2007), and The International Consensus Criteria for ME (ME-ICC) developed new criteria in 2011(Carruthers et al., 2011). Importantly, all testing and evaluation in the diagnosing procedure is based upon exclusion of other potential causes for symptoms. Most of the diagnosis criteria were originally developed for adults, yet they are often used to diagnose children and adolescents (IOM, 2015). A concern is that children and adolescents may not be able to compare premorbid or morbid physical functioning in the same way as adults (IOM, 2015). The International Association for CFS/ME (IACFS/ME) published a case definition developed in specific for pediatric CFS/ME in 2006 (Jason et al., 2006). The pediatric definition requires 3 months of clinically evaluated, unexplained, persistent or relapsing fatigue and cardinal symptoms of CFS/ME (PEM, unrefreshing sleep, pain, and neuroendocrine, or immune manifestation). The main difference is the shorter period of time to diagnose the children and adolescents. In addition, assessment of leisure, social and educational activities is suggested to detect a decrease in their functionality (Jason et al., 2006).

The most recent proposal of diagnostic criteria is developed from the IOM committee on Diagnosis Criteria for ME/CFS (IOM, 2015). The IOM diagnostic criteria is developed on the basis of comprehensive literature review and input from patient advocacy, and research communities. The criteria are claimed to focus on the central symptoms of the disease (IOM, 2015). The IOM (2015) diagnosis requires that the patient have the following three symptoms;

1. *A substantial reduction or impairment in the ability to engage in pre-illness levels of occupational, educational, social, or personal activities, that persist for more than 6 months and is accompanied by fatigue, which is often profound, is of new or defined onset (not lifelong), is not the result of ongoing excessive exertion, and not substantially alleviated by rest.*
2. *Post-external malaise*
3. *Unrefreshing sleep*

In addition, *Cognitive impairment* and/or *Orthostatic intolerance* is also required. The variety in use of diagnostic criteria and inclusion criteria in research are a major problem. Heterogeneity in the patient groups that is investigated may cause inconsistent results and difficulties in making any conclusions. In a systematic review, Brurberg and colleagues (2014) identified 38 studies on comparison and evaluation of the existing diagnosis criteria for CFS/ME. Most of the studies had serious limitations, and there was no rigorous assessment of the reproducibility of feasibility of case definitions (Brurberg et al., 2014). Coggon and colleagues (2005) support that the variety of diagnostic criteria and case definitions has caused heterogeneity and is a major challenge when doing research in both adults and pediatric CFS/ME. The new suggestion of diagnostic criteria for CFS/ME from IOM (2015) is claimed to facilitate diagnosis and care for the patients with CFS/ME and might be the beginning of a more consistent and broad dissemination. Good research methodology is needed to develop new and adequate knowledge in the field of CFS/ME. Consensus and agreement in the diagnosis criteria is essential in the ability to compare methods and important findings. Importantly, feasibility of the research method should be considered in concern of the patient's ability to participate. It might be necessary to divide the patient group into severity degree such as mild, moderate and severe degree. This will be described later in the chapter, first I will go through some of the main symptoms.

2.3 Symptoms of CFS/ME

2.3.1 Daily activities

The IOM (2015) describes five core symptoms of CFS/ME, first; *Reduction or impairment in ability to carry out normal daily activities, accompanied by profound fatigue*. Fatigue is universal in pediatric CFS/ME, usually in combination with other symptoms which lead to markedly functional impairment (Davies & Crawley 2008; Knight et al., 2013; Nijhof et al., 2011). Most CFS/ME patients experience to be bed- or house-bound at some period of their illness, and in severe cases it might last for longer periods. Symptoms can persist for years, and most patients never regain their pre-disease level of health or functioning (IOM, 2015). Research has found that even though some patient with CFS/ME improves from their illness they remain symptomatic and may not return to their premorbid level of functioning (Jason, Barker & Brown, 2013). Dr. Bell and Bell (2010) suggests that the best indicator of relative health for CFS/ME patients is upright activity performed each day. In other words, the importance of actually doing something that is important for the patient. The adolescents and young patient's capability to resume school, social and family activities is essential for QOL and normal development.

2.3.2 Post-Exertional Malaise

The second core symptom mentioned by IOM is PEM. PEM is considered to be a central feature of CFS/ME, and is worsening of some or all symptoms that occurs after physical, cognitive or emotional exertion and leads to reduction in functional ability (Carruters et al., 2003; IOM 2015). Activities such as walking up a flight of stairs, or as minimal as using a computer or phone might be triggers, as well as emotional distress, infections, standing or sitting up for an extended period, decreased sleep quantity/quality or infections. PEM might occur immediately following a trigger, but also hours or days after. It is reported worsening of symptoms in a range from 30 minutes to after 10 days (IOM, 2015). The duration of PEM is also unpredictable and can vary from hours, days, weeks and even months (Nijs et al., 2010). A common characteristic of PEM is delayed ability to return to prior levels of physical capacity after exertion (IOM, 2015).

Black and McCully (2005) compared CFS/ME patients with sedentary controls in their ability to increase daily activity in a daily walking program. In the first 4-10 days the patients were capable to reach the prescribed activity goal each day. After this period of time, the daily walking and the total activity counts decreased, while the control group maintained their daily walking and the total activity goals. The study support that the CFS/ME patients develop exercise intolerance, and inability to sustain activity levels that are over the individuals limitations. The result is associated with pronounced worsening of symptoms (Black & McCully, 2005). Several clinical exercise test studies have shown that CFS/ME patients suffer from PEM, and recover very slowly in some cases (Keller, Pryor & Giloteaux 2014; Snell, Stevens, Davenport & Van Ness, 2013). The capacity to recover from exertion is decreased, while the "recovery time" is prolonged. This delayed lack of recovery in patients with CFS/ME is demonstrated by performing two cardiopulmonary exercise tests separated by 24 hour. CFS/ME patients were unable to reproduce most physiological measures at both (Keller et al., 2014). Another study showed no significant differences between CFS/ME and control group after the first cardiopulmonary exercise testing. However, the second test showed significantly lower values for both oxygen consumption and workload (Snell, Stevens & Davenport, 2013). Importantly, repeated cardiopulmonary exercise testing can be a clinical indicator for diagnosing CFS/ME (Keller et al., 2014), but this kind of test carries substantial risk for these patients as it may worsen their condition (Nijs et al., 2010; IOM, 2015).

2.3.3 Unrefreshing sleep

Unrefreshing sleep is common in CFS/ME patient and is the third core symptom mentioned by IOM (2015). The term unrefreshing sleep is used to describe a light sleep that even after many hours does not relieve fatigue. They may wake up exhausted and with a feeling as if they had not slept at all. Sleep disorders may be on top of unrefreshing sleep for CFS/ME patients, the consequences goes beyond mere tiredness. In young patients sleep disturbance is an often reported symptom (Bell, 1995). Development of a normal sleep-awake-cycle should be part of the treatments for CFS/ME.

2.3.4 Cognitive Impairment and Orthostatic intolerance

The two last core symptoms mentioned by IOM (2015) are; *Cognitive impairment* and *orthostatic intolerance (OI)*.Cognitive impairments include mental fatigue, memory problems and loss of concentration (Helsedirektoratet, 2014_a; IOM, 2015).This might be problematic in relation to school and other social activities and cause high impact on the patients daily functioning. OI is worsening of symptoms when the person stands upright and improves when lying down. Upright position is for most people an easy task, for those who suffer from OI, standing upright can become extremely challenging. Upright position cause increased heart rate, low blood pressure, dizziness, nauseous and fatigue. The complex, effective and rapid and neurologic compensation to maintain blood pressure and consciousness is failing.

2.3.5 Severity and Prognosis

The above mentioned symptoms are suggested to be the core symptoms in CFS/ME (IOM, 2015). However, symptoms and functional impairments is individual and vary among the patients (Helsedirektoratet 2014_a; IOM 2015). When a patient extends his/her tolerance limit they often experience a relapse or worsening of symptoms, which may occur immediately or after hours, or days. Restitution period may be 24 hours or more, in worst case weeks or months. Symptoms and impairments of the illness may change regularly. One day the patient's may feel good and can attend school or other activities, while the next day they might be house-bound. The change in severity is complicated, and difficult to cope with. Especially young patients struggle to find a balance between PA and rest (RCPCH, 2004). They tend to overdo activity when they feel better which often lead to worsening of symptoms and functional impairments the next day/days.

CFS/ME can be divided in mild, moderate, severe and very severe degrees (Helsedirektoratet 2014_a). Patients of mild degree indicate that the activity level is reduced by at least 50% compared to before the illness occurred. The patients can take care of themselves, manage personal hygiene, light house work etc. Moderate forms of CFS/ME indicate impaired mobility which limits all types of daily activities and might cause them to be housebound. Patients with severe degree of CFS/ME are spending most of the day lying down/resting, and might be using a wheelchair. They might also need help for personal hygiene. Patients with very severe degree are bedridden (Helsedirektoratet 2014_a). They may also be very sensible for sensations from light and sound, and may have problems with food intake. Importantly, CFS/ME patients may be capable to mobilize strength to perform daily tasks and activities that are beyond their tolerance limits. It is important to stay beneath this limit to avoid relapse, but at the same time it is also important for the patient to maintain the activity level that they master. The prognosis of CFS/ME also vary, some may suffer from CFS/ME a short period while others may suffer their whole life. An early diagnose is important so that treatment strategies as soon as possible, both medications to reduce symptoms, and coping strategies to improve daily functioning.

2.4 CFS/ME impact on Daily activities and Functioning

CFS/ME and especially PEM can be a major disability factor for daily activities and routines. Previous studies support that the disability of CFS/ME patients is not only related to the fatigue but also additional symptoms, such as cognitive impairment, hypersensitivity and OI (Wyller & Helland, 2013). Jason, Porter and Shelleby (2010) have reported that children and adolescents have symptoms related to their daily life routine, such as loss of friends, trouble in school and drop out of social activities.

2.4.1 Education and Social development

CFS/ME has major impact on schooling and social development (Wyller, 2008; Nijs et al., 2011). Impairment caused by their illness lead to drop out of school, loss of contact with friends and reduction in ability to perform daily tasks which involve PA (Wyller, 2008). Crawley and Sterne (2009) found that in a sample of 211 children with CFS/ME, 60 percent attended school only two days per week. In addition, reduced school attendance among children with CFS/ME was associated with poor physical function (Crawley & Sterne 2009). Several studies show that CFS/ME cause restrictions and reductions of activities as well as a decline in school performance (Smith, Mitchell, Corey, Gold, McCauley, Glover &Tenover, 1991; Carter, Edwards & Kroneberger, Michalczyl & Marshall, 1995). Dowsett and Colby (1997) found that 42 percent of all medically certified long-term sickness related absence was due to CFS/ME (Dowsett & Colby, 1997). A Norwegian study found that 75 percent described their impairment as being house-bound most of the time and they could only leave home with careful planning (Bringsli, Gilje & Getz, 2014). Another study did a comparison of QOL characteristics in 25 children with CFS/ME and healthy controls and found that CFS/ME patients had profoundly reduced QOL (Kennedy, Underwood & Belch 2010). The CFS/ME patients scored lower than controls on the global health item of the Child Health Questionnaire, as well as most other items such as mental health, self-esteem, general health perceptions and physical functioning (Kennedy et al., 2010).

Daily activities, responsibilities, social interactions, schooling can be important indicators of disability and impairment (Schweitzer, Kelly, Foran, Terry & Whiting, 1995). To address PA level in CFS/ME patients as well as to learn coping strategies to maintain daily functioning should be part of treatment and follow-up for these patients. As we see, PA, or the fact of actually doing something (upright activity) can be indicators of the patient's daily functioning (Schweitzer et al., 1995; Bell & Bell 2010)

2.4.2 Physical activity and Functioning

PA is defined as all movement that is produced by contraction of the skeletal muscle and that substantially increases energy expenditure (Caspersen, Powell, & Christenson, 1985). PA is part of everyday life and is classified as behavior. Walking, gardening, cleaning, as well as sport and exercise are all examples of PA. There exists many forms of PA and include both occupational and leisure-time PA. Objectively measured PA in young CFS/ME patients are not conducted, but results from surveys and questionnaires show a major reduction in PA and daily functioning. Torres-Harding and colleagues (2006) measured PA and functioning with the Child Health Questionnaire and the Child behavior checklist. They found that children and adolescents with CFS/ME had difficulties concerning PA, including limitations in physical and psychosocial functioning and a negative impact on the ability to engage in everyday activities (Torres-Harding, Jordan, Jason & Arias, 2006). A systematic review from Nijs and colleges (2011) support the notion that patients with CFS/ME are physically inactive and often perform less PA in comparison to healthy sedentary controls (Nijs et al., 2011). However, what we need to remember is that CFS/ME patients do not choose to be inactive. Inactivity is a consequence of CFS/ME. Once the illness is established, the everyday situation changes dramatically. The impact of the CFS/ME makes it impossible or at least extremely difficult to continue their daily activities and routines. Carefully planning, and the priority of responsibilities and important activities becomes a major part of their day.

CFS/ME patients have shown to perform specific activities less frequently because they expect it to result in high fatigue levels (Nijs et al., 2011). PEM is probably the main reason for patients with CFS/ME to avoid PA and CFS/ME clearly impede the patient's ability to function on a regular basis both physically and cognitively (Nijs et al., 2011; Taylor, O'Brien, Kielhofner, Lee, Katz & Mears, 2010). PA should be encouraged for CFS/ME patients (Helsedirektoratet 2014$_a$), but the activity need to be graded to the individual tolerance limits. A good start for creating a baseline for daily activity is to cooperate with the patient. Especially because the knowledge in this field is poor, the health care therapists should listen to the patient when they provide guidance in coping strategies and when they encourage activities. In some cases it might be necessary to reduce the PA level, while others might need to increase PA. The severity of the illness differs individually in a high degree. Movement therapy based on carefully limited workloads has shown promising results for CFS/ME patients. Exercise therapy has also shown improvements for some patients, but this is probably for mild to moderate conditions of CFS/ME (Prins et al., 2006). The term exercise is

PA that is a planned, structured and repetitive bodily movement performed to improve or maintain one or more components of physical fitness (Caspersen, 1985). CFS/ME patients should not be advised to undertake vigorous exercise (NICE, 2007). PA is a broader term and includes daily activities, and low intensity. This might be a more achievable and an accepted term for CFS/ME patients rather than exercise.

Unfortunately, there is a lack of research concerning PA recommendations for CFS/ME patients. What we know is that PA can increase cognitive and psychological function (Tremblay, Colley, Saunders, Healy & Owen, 2010). PA is also associated with improvement in physical function, fatigue and mood disorder in chronic illnesses such as cancer, cardiac heart failure, and in particular multiple sclerosis and fibromyalgia which both are associated with debilitating fatigue (Nijs et al., 2013). There is much more to PA than only energy expenditure or physical fitness. PA is behavior, which is necessary for developmental, educational and health outcomes (Smith & Biddle 2008). PA promotes healthy development of musculoskeletal tissues, cardiovascular system, neuromuscular awareness (i.e. coordination and movement control), and maintenance of a healthy bodyweight (Andersen, Schnohr, Schroll & Hein, 2000). In addition, there is also good documentation for psychological benefits and the social development (WHO, 2010; Andersen et al., 2000). The process of physical growth, biological maturation, and behavioral development has shown positive association with PA, and is also important for CFS/ME patients. A study from Gordon & Lubitz, (2009) found that a 4-week intensive graded exercise programme for adolescents with CFS/ME increased their ability to resume a normal life-style, including sports and schooling commitments. The recommendations for children and adolescents is to be in at least 60 minutes of PA in moderate- to vigorous intensity (MVPA) every day to reduce the risk of disease, and improve physical and mental health (WHO, 2010). Probably, this is not intended for CFS/ME patients, where especially vigorous intensities should be avoided (Helsedirektoratet 2014$_a$; NICE, 2007).

Daily tasks and activities need to be carefully planned for the ability to perform them without experiencing relapse or worsening of symptoms. If one that suffers from CFS/ME fail to stay beneath their tolerance limit, days or even weeks of recovery may be necessary. This may reduce the perceived capability for being physically active and could lead to avoidance of PA in general which also include schooling and socializing, which is important for development. Children and adolescents are in a developmentally vulnerable period of life. Impairments of

CFS/ME might affect self-concept, body image, autonomy, socialization and cause academic problems (Gordon & Lubitz, 2009). Failing to cope with their illness and not being able to socialize or attend school can be extremely challenging for these patients both physically and mentally. To target these changes in the patient's thoughts of being physically active could be done with self-efficacy as an assessment tool. However, this is not investigated. I will later describe the role self-efficacy might have when in treatments for CFSME patients, but first the common treatments mentioned in the literature will be presented.

2.5 Treatments for CFS/ME

So far, no documented curative treatment of CFS/ME exists (Helsedirektoratet 2014_a). Treatment and management of CFS/ME include strategies so that the patient can better cope with his/her condition. Strategies and/or medication may reduce symptoms and improve daily function and quality of life. Instructions of self-management e.g., pacing, realistic goals, physical self-awareness, basic rights, understanding emotions, exercise and relaxation could benefit the patients (Helsedirektoratet, 2014_a). PA, not necessarily as exercise, but daily activities and function should be addressed and incorporated in the treatments for pediatric CFS/ME. It is recommended to establish a baseline of activity that the patient can manage even on a bad day (Royal College of Pediatrics and Child Health [RCPCH] 2004). A diary can be helpful to evaluate sleep wake cycles, diet, activity, school attendance, social contact and emotion. Fatigue Scales can also be informative when establishing a baseline.

Once a baseline is established, CFS/ME patients should get advice about balancing the amount of activity carried out each day. Activities can be physical, mental or social but the same amount should be reached each day, neither too much nor too little. The PA level to aim for in a treatment should depend on the individuals abilities and his/her tolerance limits. The amount established as an activity baseline should also be encouraged to accomplish on days they feel particularly bad. Importantly, they should not increase the activity on days they are feeling better (RCPCH, 2004). When the patient manages to achieves the activity baseline consistently, the amount of activity can be gradually increased, in addition to decreased rest. Small steps should be taken in a close cooperation and follow-up together with health care therapists. Assessment tools to target activity level, perceived ability, diaries or similar might be helpful for both the patient and the health care therapist. The patient should also be advised about a healthy diet, especially if there are signs of poor diet and lack of appetite (RCPCH, 2004). PA should also promote enjoyment and perceived competence. Especially for young patients' mastery experience, joy and social aspects is important. In the literature of CFS/ME the benefits, as well as harms, of commonly offered treatments are also subject for disagreement and little consensus.

The three most common treatment strategies discussed in the literature concerning PA for CFS/ME patients are CBT, GET and *Pacing* (Helsedirektoratet 2014$_a$; IOM, 2015; NICE 2007, RCPCH, 2004). There is an intense debate about the possible harms or benefits of the treatments. However, there is also little consensus in what exactly separate them, and my impression is that strategies used in reality differs from the one explained in research. Although CBT, GET and Pacing are three separate treatments there is a considerable overlap in what they are and what they are trying to achieve. There is a lack of standardized definitions for these terms, therefore when evaluating research studies it is important to be aware precisely what the intervention includes, not only by what it is called. Further in this chapter the most common treatments that are mentioned will be presented in addition to their issues and critiques. Importantly, medical treatments used in CFS/ME are not presented in this thesis.

2.5.1 Cognitive Behavior Therapy

CBT has the aim to help patients adjust and feel more in control of their illness and symptom. The cognitive element and the behavioral element are two major components in CBT (Prins et al., 2006; RCPCH 2004). The cognitive element focuses on the identification of thoughts and how they are modified, as well as beliefs and assumptions which might shape how the patient understands CFS/ME. The behavioral element aims to gradually and consistently introduce a change in behavior, such as PA. The CBT treatment is tailored to the needs of the patient and can include different techniques of monitoring.

There are few published randomized control trials (RCT) of CBT performed in children and adolescents. One that is published by Stulemeijer, de Jong, Fiselier, Hoogveld & Bleijenberg (2005) reported significantly greater decrease in fatigue severity, functional impairment and increase in school attendance in the intervention group compared with the control group. They concluded that CBT is an effective treatment for CFS/ME in adolescents. Prins and colleagues (2001) published a multicentre RCT performed on adult participants, where they concluded that CBT was more effective than other common treatments such as GET and pacing. This was also supported by a systematic review performed by Price, Mitchell, Tidy & Hunot

(2008). Lasting benefits of CBT is also shown in a 5-year follow-up (Deale, Husain, Chalder &Wessely, 2001).

2.5.2 Graded exercise therapy

GET is a structured and supervised program of exercise and is also referred to as *exercise therapy*. It is based on the patient's current level of ability and limitations. The intensity and duration of exercise will in the beginning be at a very low level and increases very slowly depending on progress. GET is based on a model of de-conditioning and avoidance of activity which both are thought to maintain fatigue and disability. The aim of GET is to increase fitness and stamina and to reduce physical deconditioning.

In 2004 a review published in The Cochrane Collaboration aimed to investigate the relative effectiveness of exercise therapy and control treatments for CFS/ME (Edmonds, McGuire & Price, 2004). They included only RCTs with patients having a clinical diagnosis of CFS/ME and of any age, in total five studies were included. They found that those receiving exercise therapies were less fatigued than the control participants and that physical functioning was significantly improved. Importantly, there were more dropouts in the exercise therapy group. In addition an earlier systematic review included three of the same RCT's and reported an overall beneficial effect from exercise therapy (Whiting, Bagnall, Sowden, Cornell, Mulrow & Ramirez, 2001). These reviews (Edmonds, 2004; Whiting, 2001) support that some patients may benefit from exercise therapy, and did not find any evidence supporting that exercise therapy may worsen outcomes on average. Importantly, exercise therapy may be less acceptable in other patients groups with more severe conditions of CFS/ME. The high dropout rate may be an indication that some of the participants were not able to participate. Recently an update of above mentioned review (Edmonds, 2004), were published by Larun and colleagues (2015). The review involved adults diagnosed with CFS/ME (CDC-1994 or Oxford criteria). In total 1518 participants were included in the review. The authors concluded that patients with CFS/ME may generally benefit and feel less fatigued following exercise therapy (Larun et al., 2015). A positive effect was observed in sleep pattern, physical function and self-perceived general health. No evidence supported that the exercise therapy was harmful, and serious side effects were rare in all groups. However, limited information made it difficult for the authors give any conclusion about the safety of exercise therapy (Larun et al., 2015). Importantly, they included participants who were able to participate in exercise therapy. This does not show the whole picture of CFS/ME. In addition most studies are performed with an adult sample therefore more studies should be performed in children and adolescents.

2.5.3 Adaptive Pacing Therapy (APT)

APT assume that CFS/ME is an organic disease and aims to carefully match activity levels to the amount of energy available for each patient and not go over the individual tolerance for PA (Nijs et al., 2013). Therapists work with the patients to help monitor activity and symptoms, aiming to improve QOL and create the best conditions for a natural recovery (White et al., 2011). This is a popular treatment and more accepted by patients than CBT and GET. Surveys by patients' organizations in the UK have reported that CBT and GET sometimes can be harmful and therefore "pacing" is preferred by patients and specialist health care (The ME Association, 2010).

2.5.4 Research and Recommendation

The PACE trial (Pacing, graded Activity and Cognitive behavior therapy: a randomized Evaluation) was the first in the world to test and compare the effectiveness of the four main treatments currently available for CFS/ME patients (White et al., 2007). The trial is based in Edinburg, Oxford and three London hospitals. Participants are randomly allocated to one of the treatment; Adaptive pacing, CBT, GET and Specialist medical care, and then given a 12-month program. The specialist medical care includes information about the illness, general advice about managing the illness and prescription of medication. Standardized details about treatments provided can be read elsewhere (White et al., 2007). The patients included are diagnosed with the oxford criteria for CFS/ME, all participants under 18 years were excluded.

In total, 641 patients from the PACE trial were included by White and colleagues (2011) for investigation of effectiveness and safety of all four treatments. Findings from the PACE trial suggest that individually delivered CBT and GET, when added to medical care are more effective than, and as safe as pacing. Therefore they recommend that patients attending secondary care with chronic fatigue syndrome should be offered individual CBT or GET in addition to medical care (White et al., 2011). Recently, Chalder, Goldsmith, White, Sharpe and Pickles (2015) investigated the same trial trough a secondary mediation analysis. They found that fear avoidance beliefs were the strongest mediator for both CBT and GET. Changes in both beliefs and behavior mediated the effects of CBT and even more for GET. The authors support a treatment model in which both beliefs and behavior play an important part (Chalder et al., 2015).

Most studies including the PACE trial is done on adults, therefore no conclusion can be made concerning younger patients. Knight and colleagues (2013) critically investigated interventions used for management of CFS/ME in children and adolescents in a systematic review. The majority addressed behavioral interventions such as multidisciplinary, psychological and exercise interventions. Strongest evidence was found for interventions using CBT, and weaker evidence for multidisciplinary rehabilitation. Little evidence exists on the maintenance of intervention effects (Knight et al., 2013). In addition, there is little evidence that the above mentioned treatments are beneficial for patients with severe degree of CFS/ME. However, GET or CBT is recommended for patients with mild or moderate degree (NICE, 2007; Helsedirektoratet, 2014_a).

2.5.5 Challenges and recovery

CFS/ME patients face many challenges and have not always been treated as well as they should neither by society or the health care. Some may feel that their symptoms have been downplayed and that earlier negative experience of previous treatment is ignored. The severity and degree of CFS/ME varies, as well as the individuals functional capabilities. Heterogeneity may be the reason why some are not taking CFS/ME seriously. A major problem is simply the fact that they do not know enough about this disease, which naturally lead to misunderstandings for patients, therapists and others. CFS/ME need more clarification and increased interest in research to better understand the complexity of this disease.

There is still no consensus about the final goal in CFS/ME treatments. How recovery should or could be defined is also discussed. Should recovery be defined as *the total absence of symptoms and a return to normal functioning*? Most patients' goal settings include coping strategies and daily function which is close to "normal". They carefully divide their work capacity into certain activities that is important for them and exclude others of less importance. There is no laboratory tests or goal-standard to establish the absence (or presence) of CFS/ME. Dr David S. Bell, which is one of the world's leading experts on CFS/ME, has suggested upright activity to be the most reliable indicator of prognosis and recovery. Upright activity includes time spent in actually doing something, like school, walking, cleaning or cooking (Bell & Bell, 2010). In other words, to have the capability to do more daily activities that is important to the person.

GET is not necessarily the most preferable treatment among patients. Speaking of personal experience the term exercise is obviously not the right term to use concerning a disease where

PEM is the main symptom. PA or activities may be more reasonable terms, which can be described as upright activity. Bodily movement is essential for human functioning and QOL. Final goals in treatments of CFS/ME is not agreed, but educating the patient to avoid relapse and worsening of symptoms caused by unfavorable activity patterns seems reasonable and helpful for the patients daily functioning. Graded activity therapy (GAT) is suggested as a term which better describes activity adaption as a treatment for CFS/ME patients. Activity adaption requires good practical arrangements, daily goals and activity plans. Both too high and too low levels of activity are associated with negative consequences. In mild to moderate degree of CFS/ME, activity adaption and PA might be beneficial and improve the condition of the patients (Larun et al., 2015).

Longitudinal designs may give more information about recovery and change in the outcome variables of interests. Houdenhove (2009) suggest that research concerning treatments for CFS/ME patients should be shifted towards investigation of effectiveness of treatments employed by clinicians in “real life” setting. Common treatment strategies, including CBT and GET do not use similar strategies and standardization of treatments and the result could be difficult to compare. Houdenhove suggests that design and evaluation of tailor-made treatments are more preferable in research of CFS/ME patients. Rehabilitation institutions should formulate a treatment philosophy that is based on current evidence, and involve both psychological and physiological aspects (Houdenhove, 2009). Each patient needs individual care and follow-up. Quality care needs assessment and cooperation with the patient. The individual patient probably knows allot more of his/her version of CFS/ME than what is found in the literature or explained by others. Listening to the patient and to assess progress is important when the knowledge about CFS/ME is poor. There are many challenges that follow a disease like CFS/ME and the preserved capability of performing any activity or daily tasks is not necessarily the strongest among the young patients. The ability to define a goal, persevere, and see oneself capable is essential in the coping strategies for CFS/ME no matter what kind of treatment approach.

2.5.6 What is missing?

In general people tend to only attempt challenges or tasks they believe they can accomplish Bandura (1977). In treatments, where coping strategies include creating a baseline for PA, both occupational and leisure time activities, it could be necessary to target self-efficacy for being physically active. Strength of self-efficacy for being physically active could influence participation in PA, as well as predict behavior. Treatments for CFS/ME commonly aim to change behavior and to develop coping strategies but the patients self-efficacy is not assessed as far as I know. Self-efficacy can indicate a person's belief to succeed in a behavior or to achieve a goal. It can be all kind of goals such as attending school three days a week, increase everyday activity, or to wake up at nine every morning. The belief in own ability to succeed in a particular situation is described as determinants of how people think, behave and feel (Bandura, 1994). Self-efficacy might have an important impact on many factors like motivation, psychological states and behavior. Results from the PACE trial showed that changes in both beliefs and behavior mediated the effects of CBT and GET (Chadler et al., 2015). In addition, increased perceptions of personal capabilities and mastery experience might be factors that influence improvements in treatment of CFS/ME (Prins et al., 2006). An individual's self-efficacy is found to play a major role in how goals, tasks, health behavior and challenges are approached. Hence, self-efficacy might be important to assess in treatments for CFS/ME patients where goal setting and behavior change are a main parts. In daily situations which may contain many challenges a strong self-efficacy could benefit the patients in coping strategies. Self-efficacy as an assessment tool to assess each patient's perceived capability to achieve important goals, tasks or behavior might be beneficial in commonly used treatments for CFS/ME patients. As mentioned above, the health therapist should listen and develop coping strategies together with the patient. Assessing the patient's thoughts of capability to perform a specific behavior or activity before, during and after received treatment can improve the quality of the treatment as well as follow-up procedures. Self-efficacy might also be informative when establishing a baseline for activity level and when behavior change is part of the goal setting. In the aim to understand the patient, and to give them a sense of being understood, assessment tools could help finding out about the patient's expectations and concerns.

For some years ago, Kerns, Weinberg & Rosenberg (1998) found that a stronger sense of self-efficacy was suggested to moderate responses to CFS/ME and related events. Self-efficacy was also suggested to have an important role in the appraisal of the dimensions of somatic symptoms, disability and distress (Findley et al., 1998). This is apparently the only study covering self-efficacy in this population. However, Taylor and colleagues support the need of an occupation-focused rehabilitation program that focuses on changing young people's attitude about their own performance (Taylor et al., 2010). In addition, PA can enhance self-esteem in children and adolescents as well as positive judgment of the global and physical self

(Smith & Biddle 2008), an indication of this can be self-efficacy. Another interesting point of view is self-efficacy's role as a predictor of PA.

2.6 Self-efficacy Theory

The psychologist Albert Bandura at Stanford University introduced the concept of perceived self-efficacy in the context of cognitive behavior modification in 1977 and self-efficacy represents the key construct in Social Cognitive Theory (SCT) (Bandura, 1977; 1997). Self-efficacy is defined as people's judgment of their capabilities to organize and execute a specific behavior or action (Bandura, 1977) and the self-efficacy theory is built on the belief that people generally only attempt challenges or tasks they believe they can accomplish, and they will not try if they believe they will fail. People that have a strong sense of self-efficacy might have the belief of accomplishment in many situations. A strong sense of self-efficacy and the belief in accomplishment rather than failure reduces stress and risk of depression (Bandura, 1997). It is proven that a strong sense of personal efficacy is related to better health, higher achievement, and more social integration. People with a low self-efficacy may doubt their ability to accomplish specific situations or tasks, and may see them as threats. Small barriers might lead to avoidance and preventing them from being successful. They lose faith in their capabilities and might give up easily.

Behavior change is facilitated by an individual sense of control (Bandura, 1997). High self-efficacy reflects an optimistic point of view of own capability to deal with stress, tasks or specific behavior. It mirrors a sense of control over ones environment and ability to master challenges. Conversely, a person with low sense of self-efficacy often has pessimistic thoughts about own accomplishment, and is associated with depression, anxiety, and helplessness. Self-efficacy affects how a person interprets situations here and now, and it also affects how people visualize and construct future situations (Bandura, 1997). In other words perceived ability in the task may enhance or impede the motivation to act or perform a behavior. Self-efficacy affects intention to behavior change. Effort to reach a goal and persistence to continue striving despite barriers or setbacks that may weaken the motivation to continue are influenced by self-efficacy (Schwarzer & Fuchs 1995). Interestingly, both beliefs and behavior are shown to mediate the positive outcome effects from commonly used treatments for CFS/ME patients (Chandler et al., 2015). However, the role of self-efficacy in CFS/ME patients is not investigated.

The challenges CFS/ME patients experience both due to symptoms and the uncertainty of their illness. This may affect their self-efficacy for being physically active or to reach goals or change behavior due to better coping strategies. Self-efficacy expectations can be subject to external and internal influences and are therefore an ideal target for manipulation and intervention strategies (McAuley et al., 2013). Originally, the self-efficacy theory was proposed to account for the different results achieved by diverse methods used in clinical psychology for the treatment of anxiety (McAuley,1992) Since then it has been expanded and applied to other domains of psychological functioning, including health behaviour and PA as well as in sport and exercise.

2.6.1 Clinical use of self-efficacy

Self-efficacy is shown to be important also in a clinical context. A review done by Kopershoek, van der Bijl & Hafsteinsdóttir (2011) present an overview of the literature focusing on the influence of self-efficacy and self-efficacy enhancing interventions on mobility, activities of daily living, depression and QOL in stroke patients. Their results supports the self-efficacy theory in that patient’s with high self-efficacy functioned better in everyday activities than patients with low a self-efficacy. Self-efficacy was also negatively associated with depression and the review highlighted the importance to further emphasize the role of self-efficacy in the care for stroke patients (Korpershoek et al., 2011). Another review found similar results in patients with cardiovascular disease (Katch & Mead 2010). They investigated the role of self-efficacy in cardiovascular disease self-management and found that self-efficacy was an important component in the treatment. Another recent study done by Patterson, Meyer, Beaujean & Bowden (2014), used SCT to understand PA among dialysis patients where they found a significant relationship between PA and self-efficacy.

According to above mentioned research self-efficacy is proven to have an important role for improvement outcomes and PA in clinical use. Self-efficacy as an assessment tool can provide important information about subjective feeling of accomplishment, and be an important determinant for behavior, motivation and performance.

2.6.2 How to change Self-efficacy beliefs?

Self-efficacy is highly changeable, and the strength may vary due to different variables such as mood, environment or encouragement. Bandura identifies four main ways in which self-efficacy can be affected; *mastery experience, social modeling, verbal persuasion and physical/emotional state* (Bandura, 1997; McAlister, Perry & Parcel, 2008). The figure 2.1 is retrieved from Bandura (1997) and illustrates the four ways which affect the strength of self-efficacy and thereby behavior.

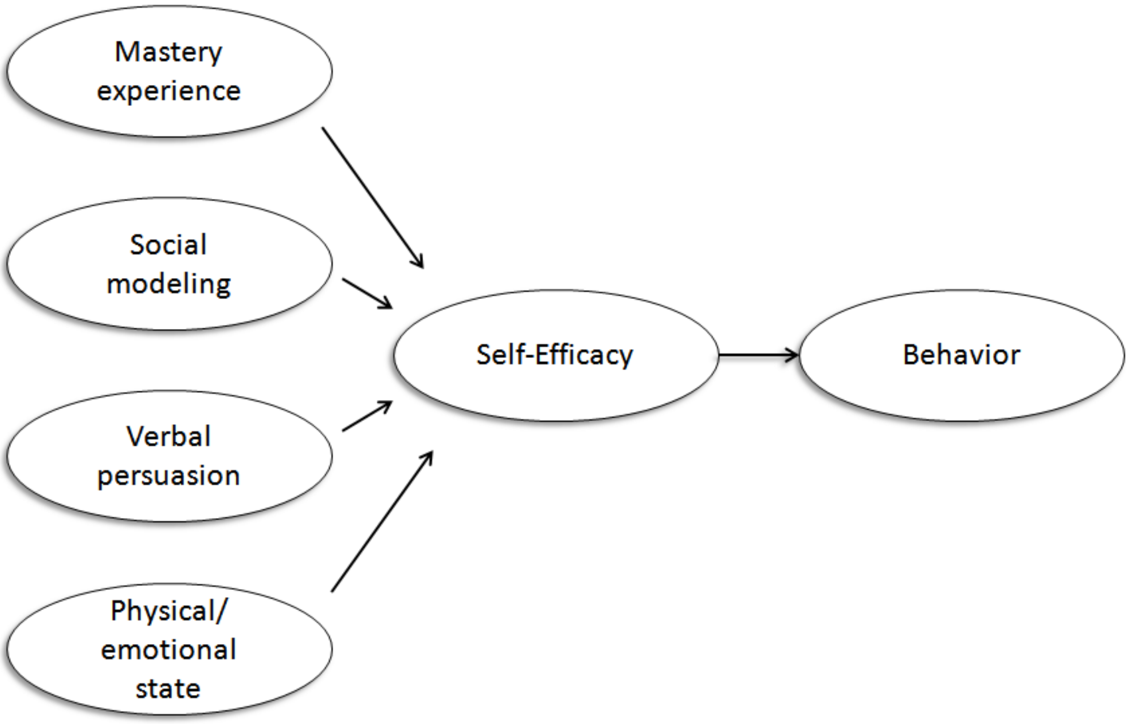


Figure 2.1: *Illustration of sources which affect Self-Efficacy and thereby behavior, collected from Bandura, 1997*

Mastery experience is suggested to be the strongest source of self-efficacy (McAlister et al., 2008). Prior success or accomplishment in a task or behavior may develop into a positive belief in the capability to engage in that or similar behaviors/tasks. The individual's history of successful experience develops confidence and typically increases self-efficacy. However, many factors might influence how experiences are cognitively processed. A person might still doubt own capabilities in a behavior although he/she achieved success at an earlier stage in the same behavior. In the same way that success might increase self-efficacy failure or negative experience may decrease it. CFS/ME patients might have prior experience of failure and limitations in function due to their illness and PEM might have been a consequence. According to these statements these negative experiences CFS/ME patients might experience could cause a decrease in self-efficacy in everyday activities such as schooling, social activities or sports. *Social modeling/vicarious experience*s is another way to affect self-efficacy and include that by watching others comparable to ourselves being successful in a behavior or task we can learn and increase belief in one owns ability to performe the same behavior (McAlister et al., 2008). If prior experience is missing or when people doubt their ability of success, self-efficacy might be developed trough vicarious experience. Rehabilitation approaches are often organized in groups where the patients get opportunity to learn from each other.

Verbal persuasion from others can also affect self-efficacy. Encouragement from other people such as “you can do it!” may increase the confidence and thereby effort in the specific behavior (McAlister et al., 2008). Support and feedback from social groups might be helpful to increase self-efficacy, but the compliments and encouragement has to be realistic for the individual receiving it. Positive encouragement or feedback can increase self-efficacy, and negative feedback might weaken it. However, the verbal persuasion does not contribute as much as the individual’s own experience or mastery experience. Verbal persuasion is also only a short term effect and need to be linked with actual successes. *Physical and emotional states* are also factors that influence self-efficacy (McAlister et al., 2008). Positive emotions such as happiness and excitement might increase self-efficacy, while negative states like depression or anxiety might weaken it. The emotional states can influence even though it is not related to the current task. The daily situations for a CFS/ME patient might be challenging, depression, pain and stress response are common symptoms in CFS/ME. Hence, this might affect the self-efficacy in many tasks they could have been capable of doing on a good day.

2.6.3 Self-Efficacy for being physically active

The activities an individual choose to pursue, the degree of effort they expend in pursuit of their goals, and the levels of persistence in face of failures, setbacks and difficulties are theorized to be influenced from efficacy expectations (McAuley et al., 2013). Self-efficacy as a predictor and a mediator is well investigated for both patients and healthy individuals (McAuley et al., 2013). Harris, Owen, Victor, Adams & Cook (2008) examined objective PA measures and a broad range of health, psychological and anthropometric variables. They found that lower accelerometer step-count was independently predicted by a low exercise self-efficacy. Self-efficacy is also demonstrated to be a powerful correlate in the prediction of long-term maintenance of PA. McAuley (1993) and Bandura (1989) reported that under challenging circumstances higher cognitive control systems, such as self-efficacy, significantly influenced behavior. Hankonen, Absetz, Ghisletta, Renner & Uutela (2010) completed 3-mounth lifestyle interventions for overweight adults. They found that changes in self-efficacy across the intervention period positively predicted exercise behavior change.

Similar results were found in a 12 month counseling intervention for older men where self-efficacy was significantly associated with changes in PA both directly and indirectly (Hall et al., 2010).

McAuley et al., (2011) suggest that self-efficacy ratings during an intervention are more informative than baseline measures. After the patients are exposed to a new behavior or task they can better draw decisions on recent experiences to make a more accurate judgment on if they will be successful in the future. This is supported by Wilbur, Vassalo, Chandler, McDevitt &Miller (2005) which found that levels of self-efficacy at baseline did not predict exercise adherence during the maintenance phase. Importantly, individuals who had improvements in self-efficacy during the intervention period had better results in the maintenance phase (Wilbur et al., 2005). This means that changes that occur during rehabilitation programs can influence future behavior in a stronger degree. In-patient rehabilitation treatments commonly last only for a short period of time. After finishing the treatment it might be challenging to continue target behavior when they arrive home to their familiar environment (and possibly old habits). According to Wilbur et al. (2005) a stronger sense of self-efficacy beliefs for a behavior that is developed during the treatment period might give higher probability for the patients to continue performing the same behavior at home. In fact, commonly used treatments often emphasize the ways which affect self-efficacy. For instant rehabilitation treatments often aim for mastery experience in PA, and coping strategies aiming to improve daily functioning and mastery. Groups are developed for socialization and to share experiences as well as doing activities in groups might emphasize vicarious experience. Encouragement from the health therapists or the others in the group is verbal persuasion and is another way to affect self-efficacy. Increased self-efficacy might already be a big part in treatments, but not specifically targeted.

2.6.3 Why self-efficacy as an assessment tool?

As mentioned self-efficacy is proved to be an important mediator and predictor of PA, and is widely used in clinical treatments in a variety of patients (McAuley et al., 2013; Harris et al., 2008; Hankonen, Absetz, Ghisletta, Renner & Uutela, 2010). Previous research findings suggest that interventions involving PA should also integrate strategies to improve self-efficacy in order to maximize improvements in disability outcomes and improve QOL(McAuley et al., 2013). Targeting self-efficacy in interventions enhances behavior change not only directly, but also indirectly through other social cognitive constructs (McAuley et al., 2013).

PA is often a main part treatments for CFS/ME patients, therefore their beliefs in own capability to perform PA could be important to assess before, during and after a treatment approach. The patients' self-efficacy of performing daily activities might give information for developing a baseline in the treatments, as well as predict future behavior. Self-efficacy scales as an assessment tool in young CFS/ME patients is not investigated, despite of proven importance in other patient groups (McAuley, 1993; 1989; McAuley et al., 2007; 2013). Increased self-efficacy of performing PA might stimulate enhanced skills to cope with daily stressors and increase feelings of accomplishment. As Bandura (1997) has stated, individuals who believe they are capable of accomplishing specific tasks are more likely to try harder to achieve their goals and persist longer when they encounter barriers. Building strategies for self-efficacy in specific behavior such as PA, school attendance or social activities can increase efficiency of treatment. Hence, self-efficacy should be assessed.

2.6.4 Suggested model

To better illustrate how self-efficacy and CFS/ME may influence each I developed a model (Figure 2.2) which is based on the previous model shown in this chapter (Figure 2.1) created by Bandura. The model illustrates how the four ways which affect self-efficacy may be influenced by CFS/ME and thereby changes in self-efficacy beliefs for being in PA. According to Bandura (1997); Mastery experience, social modeling, verbal persuasion and physiological/emotional state affect self-efficacy, self-efficacy again influence actual behavior which in this case is PA. PA may also affect the symptoms of CFS/ME negatively and lead to decreased self-efficacy beliefs.

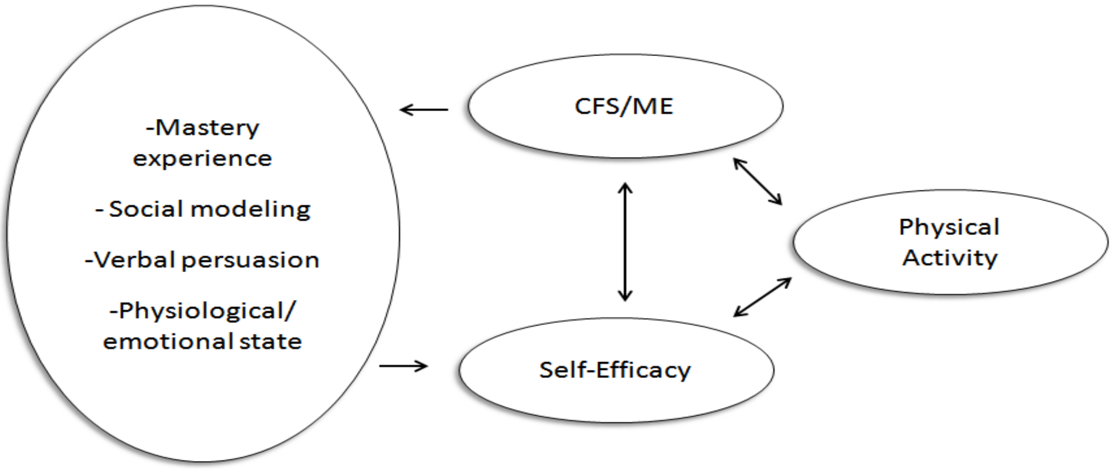


__Figure 2.2:__ The model illustrating the hypothesis that CFS/ME affects self-efficacy both directly and through the four main sources identified by Bandura (1997). CFS/ME and Physical activity (PA) is also moderating each

other. Worsening of symptoms after PA may decrease self-efficacy beliefs while graded amount of PA and improvement of symptoms will increase self-efficacy and again promote PA.

CFS/ME and self-efficacy also affect each other which both affect PA. In addition PA will possibly affect both self-efficacy and CFS/ME, this relationship may be both negative if too much or too little, but positive if adapted the person's capabilities. Different degrees of self-efficacy might be important to target in treatments including PA. The patient's self-efficacy may be helpful for both the therapist and the patients in order to plan and reach the right amount of PA. How strong self-efficacy is in young CFS/ME patients and how self efficacy can be used as an effective tool is not investigated in previous research. This master thesis has the aim to test feasibility of self-efficacy as an assessment tool in a rehabilitation treatment for young CFS/ME patients. The model is used for development of current hypothesis, if self-efficacy is affected during the treatment, and if the strength of self-efficacy influences PA.

2.7 Research challenges and assessment

In order to develop a research method assessing the importance of self-efficacy as an assessment tool for being physically active, both PA and self-efficacy need to be measured. Either of them is a simple manner to measure because of the complexity of both terms. Further common methods used in research involving PA and self-efficacy wil be presented.

2.7.1 How to measure physical activity

PA is a complex term and difficult to standardize, it consists of several movement patterns and involves many challenges concerning the measurement tool (Thomas et al., 2005). Subjective methods such as questionnaires and surveys are commonly used in research with many participants. Self-assessment questionnaires are appropriate for large epidemiological studies because they are low in cost and easy to administrate. However self-assessment is shown to have restrictions in information and several limitations (Helsedirektoratet, 2009). Smaller studies that require higher accuracy may use advanced techniques such as DLW or indirect calorimetry which is more precise, but also difficult to assess and high in cost. Few research trials can afford these advanced techniques, therefore other devices which can give an objective measure of PA are preferred. Accelerometer has become a standard method for assessing physical activity in field-based research (Helsedirektoratet 2009; Welk, Schaben & Morrow, 2004). Accelerometers provide an objective indicator of PA and are easy for the participants to use compared to questionnaires and activity log.

ActiGraph is a commonly used accelerometer in science and is validated against DLW which is the gold standard measuring energy expenditure (EE) (Ekelund et al., 2001). However, ActiGraph is criticized for not being able to measure activities with lower intensity as accurate as moderate or high intensity. A validation study of PA monitors for assessing lower intensity activities show that accelerometer-based activity monitors (ActiGraph GT3X and ActivPal) did not perform as well as more advanced pattern-recognition monitors. The multi-sensor monitors appear to have advantages compared to the standard accelerometry-based monitors (Calabró, Lee, Saint-Maurice, Too & Welk 2014). It is also shown that the placement of the device is important for the results, and that outcome results from wrist worn devices commonly indicate a higher PA level because they cover more upper body movements than hip worn (Hildebrand, Van Hees, Hansen &Ekelund 2014).

Recently advances in technology have led to the emergence of new consumer-based activity monitors designed for everyday use. Many new activity monitors have been released into the market such as Fitbit Flex, Nike Fuelband, Jawbone UP, Polar Loop etc. However, the reliability and validity should be assessed before used in research. A study aimed to examine the validity of energy expenditure estimates from a variety of consumer-based, physical activity monitors under free-living conditions (Lee, Kim & Welk 2014). 60 healthy males and females wore eight different types of activity monitors simultaneously while completing a 69 minutesprotocol. It included both monitors worn on the hip and the wrist. They found that the estimates from the Body Media FIT, Fitbit Zip, and Nike fuel Band where each within the 10 % equivalent zone around indirect calorimetry estimate (Lee et al., 2014). This indicates that the new developed devices available at the market also are accurate and reliable. To decide which PA monitors that is appropriate in different cases or population is not an easy task. Liao, Benzo & Ries (2014) suggest that new technology devices may be appropriate in self-motivated exercise programs, thus clinical trials and validation studies are needed (Liao et al., 2014). Other benefits of the recently developed accelerometer devices are that they are lightweight, easy to wear and have improved accuracy of PA monitoring. Because these devices also include real-time feedback, they may help to improve participant self-motivation strategies.

Which method to use for a particular purpose require considerations of several factors such as the research question, the population, availability, principal investigator and staff training and budget. Many factors need to be considered when choosing an activity monitor to assess PA in young CFS/ME patients. It should not be an impact to wear the activity monitor, it should cover all upright activity as well as it should be valid and reliable.

2.7.2 How to measure self-efficacy

There is no all-purpose measure of perceived self-efficacy (Bandura 2006). Self-efficacy reflects judgment of capability for a specific behavior under specific circumstances, therefore the measurement of self-efficacy need to be appropriate and specific for the purpose. A researcher must develop a self-efficacy scale for the specific population for investigation that is tailored to the particular domain of functioning. Perceived efficacy plays a key role in human functioning because it affects behavior directly and by its impact on other determinants such as goals and expectation, perception of impediments and opportunities in the social environment (Bandura, 2006).

Self-efficacy measurements can be labeled in three domains; Level, strength and generality (Bandura, 1997; 2006). Self-efficacy beliefs can be designated in terms of levels by the number of activities individual judge themselves capable of performing above a selected cutoff value of efficacy strength. However, measuring levels will lose the predictive information from self-efficacy (Bandura, 2006). Efficacy strength incorporates efficacy level and is generally a more sensitive and informative measure than efficacy level. Strength refers to how strongly people believe they are capable of performing a behavior or action and can indicate perseverance of the individual. Individuals with weak efficacy beliefs are easily counteracted by difficulties, whereas individuals with strong efficacy beliefs will preserve their efforts despite difficulties and obstacles. Importantly, strength of perceived self-efficacy is not necessarily linearly related to choice of behavior (Bandura, 1977), but the stronger the sense of self-efficacy, the greater perseverance and likelihood that the chosen activity will be performed successfully. Generality can vary across types of activities, situation, and type of individuals toward whom the behavior is directed. Assessment linked to activity domains, situational contexts, and social aspects reveal the patterning and degree of generality of beliefs in efficacy (Bandura, 2006). Perceived self-efficacy is a major determinant of intention and a judgment of capability to execute given types of performances. Efficacy items should accurately reflect the construct. Self-efficacy is concerned with perceived capability. Therefore the phrase “can do” is a better judgment of capability than “will do”, which is a statement of intention (Bandura, 2006). In addition a wide range of scores should be available to adequately capture strength.

Efficacy beliefs influence whether people think optimistically or pessimistically and influence the course of action people choose to pursue, the challenges and goals they set for themselves and their commitment to them etc. Meta analyses confirms the influential role of perceived self-efficacy in human self-development, adaption, and change (Moritz, Feltz, Fahrbach, & Mack, 2000; Stajkovic & Luthans, 1998). In the measurement of perceived self-efficacy to follow a health-promoting exercise or PA routine, individuals judge how well they can get themselves to follow the routine under various impediments such as tiredness, depression, pain or if they have more interesting things to do. The identified challenges for the specific population must be built into the efficacy items (Bandura, 2006)

There have been developed many different questionnaires and scales to measure self-efficacy beliefs for children, adolescents and adults, as well as in different groups of patients. However, no questionnaire or scale is made to measure self-efficacy specified on adolescents with CFS/ME. Existing tools that is developed are either too specific to other diseases, not suitable or to general. In a cross-sectional study from Saunders et al. (1997), they validated The self-efficacy scale that is taken from social cognitive theory (Bandura, 1986; Reynolds et al., 1990; Sallis et al., 1992). It includes confidence in ability to be physically active. They found internal consistently and test-retest reliabilities that was adequate and provide support for the use of the self-efficacy scale to measure psychosocial determinants of physical activity in preadolescent children. A PhD thesis from Sæbu (2011), conducted at the Norwegian School of Sport Sciences, they investigated PA and motivation in young adults with a physical disability and assessed self-efficacy (Sæbu, 2011). The items in the questionnaire used is translated to Norwegian and collected from the ESES – exercise Self-Efficacy Scale. ESES is validated in a sample of 368 individuals with spinal cord injury (Kroll, Kehn, Ho & Groah, 2007). Findings indicate that the ESES is a reliable instrument with high internal consistency and scale integrity (Kroll et al., 2007). This questionnaire might is also be suitable for children and adolescents with CFS/ME, but it is necessary with small modifications to more precisely target adolescents with CFS/ME

2.8 Ethics

A well-designed study is needed to be sure that a sample such as young CFS/ME patients has the ability to participate without being exposed for harm. Research interventions for this population are challenging, especially because of vide diagnose criteria that cause heterogeneity in the patient group. There are still much in the field of CFS/ME which is not completely understood and cause challenges for a researcher. Ethical concerns are especially important for this population. As a researcher, there is a responsibility to consider the ethical issues involved in own research (World medical association [WMA] 2013). Ethical judgment is necessary in order to carry out a research study. Different ethical principles are made concerning different research areas, for instant medicine, health or exercise. The choice of method and participants decide which principle to follow. Young CFS/ME patients are considered as a vulnerable group (WMA, 2013). When doing research on vulnerable groups, it is necessary to be aware of the professional guidelines of considerations.

In Norway, researcher must conduct according to the principles of the Declaration of Helsinki and apply to the *Research ethics committee* (REC) and/or Norwegian Social Science Data Services (NSD) before starting the research (Ruyter, 2009). REC or NSD will always have the final decision if the research is acceptable and not harmful for the participants. The most essential task of the researcher is not to harm the participant and it is seen as the bedrock of medical ethics. Research and literature give us adequate knowledge when conducting an intervention, but what when adequate knowledge is deficient? The research field of CFS/ME is complicated and confusing. However, the complexity of CFS/ME needs more investigation to secure quality treatments which improve the patients daily functioning. The inadequate evidence in safe methods, due to both research and treatments should be an ethical concern before conducting research on young CFS/ME patients. When the knowledge is not adequate enough, the clinical experience of therapists and doctors can be helpful in conducting a research project that is not harmful for patients. In order to investigate young CFS/ME patient's activity level and self-efficacy for being physically active without taking any risk of harm, this master thesis collected data in cooperation with a rehabilitation institution conducting a larger pilot study. Investigate self-efficacy as an assessment tool during an evidence-based treatment, tailor-made for young CFS/ME patients by a specialist team is conducted. Observation of patients over time might give important information about improvements and effects from treatments and in the same time not harmful for the patients. However, the feasibility of the design and method is an important part of this master thesis.

REC and NSD controls the research that is performed. They will decide if the research is necessary, beneficial and not harmful for the participant. When the participants include children and adolescents, it is some challenges we need to address. First, the study method must be appropriate for these participants, and the consent should be given to both the adolescents and the parents. Participants have rights to be informed about what they are participating in, and the reason for the investigation (Harris & Atkinson, 2009). Informed consent is obligatory and should contain aims, method, sources of findings, conflicts of interests, anonymity etc (WMA, 2013). The parental consent is required, but a parent cannot consent on behalf of the participant. The adolescents should also get full information about the research, volunteered participation and that they can withdraw at any time. It is the researcher's responsibility to provide information in the sense that is understood by the adolescents. Ethical guidelines should overweight the research itself (Harris & Atkinson 2009). CFS/ME is an illness where little or very little exposure can cause worsening of symptoms and this should be considered when deciding the research method and design.

3. Research question and Hypotheses

The aim of the present thesis was to investigate self-efficacy for being physically active as an assessment tool in treatments for young CFS/ME patients. The complicated procedure in development of a main research question was not an easy task. Many directions and possibilities were suggested, but with a lack of theory this was a complicated procedure. The main research question is;

"Can self-efficacy influence participation in physical activity for young CFS/ME patients?"

The suggested model (Figure 2.2) was used during the research to investigate patterns in PA and self-efficacy, and how it might affect each other. In addition there were also investigated if there were any relationship between the variables. The hypotheses are expected results of a study or experiment (Thomas et al., 2005). The hypotheses are used to create structure in the research. An essential feature of a hypothesis is that it is testable. In statistical hypothesis testing two hypotheses are compared, these are called null hypothesis and the alternative hypothesis.

3.1 Operationalization

Self-efficacy is the person's judgment of their capability to perform a behavior or activity. In this thesis, the behavior of performance is PA. PA is any bodily movement produces by the skeletal muscles which increase energy expenditure. Young CFS/ME patients are in current study a term which includes patients between 15-25 years of age which are diagnosed with CFS/ME in specialist healthcare services. For children and adolescents the Jason (2006) pediatric criteria are recommended for use in Norway. Importantly, for adults the Fukuda definition (Fukuda, 1994), or the CCC (Carruthers et al., 2003) are recommended (Helsedirektorated 2014$_a$). Exact which diagnostic criteria that have been used are not known. Only patients with mild to moderate degree of CFS/ME are included in present thesis.

3.2 Statistical Hypothesis testing

3.2.1 Hypothesis 1

Perceived self-efficacy for being physically active, increase after the participants receive treatment at CS.

- Hypothesis (H0): There is no change in perceived self-efficacy after treatment at CS.

3.2.2 Hypothesis 2

PA increases after the participants receive treatment at CS.

- Hypothesis (H0): There is no change in PA after treatment at CS.

3.2.2 Hypothesis 3

There is a positive correlation between perceived self-efficacy and PA.

- Hypothesis (H0): There is no correlation between self-efficacy and physical activity level

4. Method

Empirical research describes data that is based on objective observations. The use of the empirical method can provide important knowledge but it is necessary to be aware of possible limitation. In this chapter I will describe the choice of design and method used for answering the research question and hypothesis. In addition, the possible limitations and threats to the validity of the thesis will be described.

4.1 The Overall Pilot-Study

This thesis has taken part in an ongoing pilot-study investigating recovery and improvements in young CFS/ME patients after receiving a 4-week inpatient multidisciplinary CFS/ME treatment. The pilot-study is conducted in a real life setting at CS, a rehabilitation institution in Norway with a longitudinal design. CS started the 4-week CFS/ME treatment programme for children and adolescents diagnosed with CFS/ME in 2008. Recently there has been an increased interest in the field of CFS/ME, but there is still a lack of adequate knowledge in many areas. One area where there is little consensus among researchers is which treatments provide the best results, and what kind of result are aimed for. Do the patients improve or recover, is it harmful or safe? More understanding and research are needed in order to help patients in the best possible manner. There exists little research about treatments, improvements and recovery, especially, in pediatrics and young CFS/ME patients. Hence, CS is now conducting a pilot-study that aims to answer the following question;

> *"Do children and adolescents with CFS/ME achieve prolonged recovery by attending a tailor-made multidisciplinary rehabilitation treatment at Catosenteret?"*

The pilot-study is conducted to evaluate feasibility of design and method chosen to investigate treatment outcomes and improvements in the patients after the 4-week rehabilitation treatment. A longitudinal design is chosen to measure long-term changes up to 1 year after the rehabilitation treatment. The pilot-study gives the opportunity to conduct a larger, control trial when evaluation of design and method are reached.

4.1.1 A 4-week Multidisciplinary Treatment

The rehabilitation institution is organized around a multidisciplinary treatment team. The treatment team works together with the patient to achieve the best possible level of functioning, increase degree of coping and improved QOL. The philosophy at CS is built upon concepts and beliefs associated with the biopsychosocial model, whereas biological, psychological and social factors play a significant role in CFS/ME. CS believe there is a link between the state of mind and the individual's health, but they also emphasize biological conditions based on PNI. They emphasize the theory that there is an ongoing immune response perhaps due to viral infection also supported by VanElzakker (2013). Based on the biopsychosocial model and PNI, CS has developed a tailor-made treatment programme for young CFS/ME patients. The treatment facilitates self-determined forms of motivation. They encourage effort and provide opportunities for activities and sport. Both occupational and leisure time PA is a main part of the treatment. CS has chosen to use the term GAT in their treatment rather than the term GET. GAT is more related to "activity management" described in the guidance from NICE (2007). Different activities concerning the individual limitations are selected, adapted and graded in the therapy to promote health and well-being. GAT is a goal-oriented and person-centered approach tailored to the needs of the person with CFS/ME. The therapy includes establishing a baseline, where specific activities need to be increased or decreased in relation to the individual's own limits and goal settings. Short bouts of PA with low intensity and guided systematic rest periods throughout the day are encouraged. The patients have the opportunity to take part in different sport and activities such as horseback riding, climbing, kayaking, hiking etc. But the main aim is the ability to resume a normal life-style with a balance between rest and PA.

All the CFS/ME patients develop an individual activity plan together with the treatment team. The activity plan is a tool in order to achieve control and balance between their occupational and leisure time PA. The activity plan is a fundamental part of the treatment at CS which the patients also bring with them for further development when they return home. When completing the 4-week rehabilitation treatment there is arranged cooperation with an external therapist from the home town. The activity plan is for the patients to bring with them back home for further development and adaption to their life-style.

The treatment at CS is individualized, but the patient is also part of a group. The CFS/ME group normally consists of 6-9 patients which. The patient group provides an opportunity to meet others in the same situation, and can be used for group conversations and to share experiences. The group gives the patient an opportunity to socialize, since some of the patients may have been house-bound for a longer period. The group treatment is important for the development of social skills. Coaching and guidance of coping strategies are performed at an individual level but also in the group. Patients undergo treatment primarily in the daytime, in addition to two evenings during the week. Schooling and education are offered at CS. Every morning during the week (except weekends) the patients meet up in lessons, with the possibility for individual adjustments.

The rehabilitation stay is developed for young CFS/ME patients between 10 and 25 years of age with a mild to moderate degree of CFS/ME. Their functional level should indicate ability for participation in GAT. Before attending CS, the patients must undergo extensive diagnostic assessment in primary and/or specialists services, where the pediatric criteria from Jason (2006) are mostly used. The application process has to go through the Regional Health Authority for South-Eastern Norway (RHA). If the patient is under 16 years the parents must be present, however the parents or siblings are encouraged to participate during the treatment for all the patients.

4.1.2 Data collection

The baseline data collection started in September 2014 and has included 4 groups. The 4 groups which were included attended CS for 4 weeks in the time period September 2014 – January 2015. Assessment tools that are included in the pilot-study for a quantitative data collection are the *Adolescent Sleep Wake Scale*, *self-efficacy scale (SES)*, *Chalder Fatigue Questionnaire*, *Coop Wonca charts* and *Activity diary*. The data collection is illustrated in *Figure 4.2*. Baseline measurements were collected one week before the participants attended CS for treatment (TP1). In the last week of the treatment at CS, the next data collection was performed (TP2). Then a follow-up 12 weeks after received treatment (TP3), and 1 year after received treatment (TP4). Since the pilot-study is an ongoing project the data material at TP4 is not yet collected and not used in this master thesis. The final report from the pilot study will be published in January/February 2016. This master thesis took part of the pilot-study from the beginning and prepared the SES as an assessment tool and outcome variable of self-efficacy for being physically active. The SES as an assessment tool is investigated, in addition to objectively measured PA in this thesis. Examination of change over time as well as feasibility of method and design with regards to a vulnerable patient group is performed.

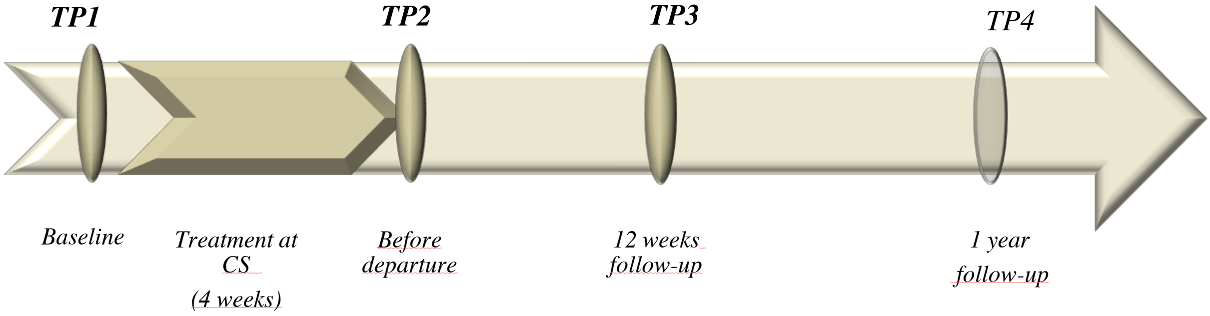


Figure 4.2: *Timeline which shows the data collection time points (TP) from baseline (TP1), last week at CS (TP2), 12 weeks after received treatment (TP3) and 1 year follow-up (TP4). TP4 is not yet conducted and will not be used in this thesis. This is therefore illustrated with a more transparent marker on the arrow.*

4.2 Purpose and design

Objectively measured PA is not performed in previous research in young CFS/ME patients, nor has self-efficacy. Hence, the purpose of current master thesis was to test feasibility of SES for being physically active as part of the pilot-study, and if self-efficacy can influence participation in PA for young CFS/ME patients. Design of the research is a longitudinal pilot intervention study. The longitudinal design allows data collection from the same participants over a longer time period and is a type of observation study (Thomas et al., 2005). Because of time limitations, this thesis only includes the data collection until 12 weeks after the treatment at CS. There are some methodological issues that need to be considered due to a longitudinal design alongside the time required. I will quickly go through some issues regarding current design.

One of the most common issues is *unrepresentative scores*, called outliers and occur in all research, but are particularly problematic in longitudinal research (Thomas et al., 2005). Outliers may occur when there is lack of attention or motivation to do the task, or if there are other distractions. Unrepresentative scores can be an issue in current design. The participants may have been distracted when they performed the questionnaires at home (TP1 and TP3). Another concern might be if the participants had a bad day with symptoms such as headache, muscle pain and fatigue and spent less attention to answer the questionnaire. Bad mood or lack of motivation to answer the questions can also cause unrepresentative scores. Concerning the PA measurements, unrepresentative scores because of attention or motivation are less likely since the participants wear the device on the wrist with little effort. However, they might have taken it off causing less wear time. Another concern due to unrepresentative scores in current research is the "Hawthorne effect". The participants might change their PA behavior as they know they are being observed. The Hawthorne effect is investigated in previous studies and increased activity pattern is observed the first day but seems to come back to normal following days (Mattocks, et al., 2008). To control for this effect the first day of recordings is not used in the analysis. Unclear semantics can also be an issue, meaning that words used to explain a task need to be appropriate and understandable for the participants attending the research (Thomas et al. 2005). Identical instruction for all participants is a standard rule, but in some cases it might be necessary for more explanation for children than for adolescents. Since the participants include children, adolescents and young adults the information of the task was explained in a simple manner so that all participants could understand. In addition the task in the questionnaire for self-efficacy was given with practical examples. Written information in how to use and charge the device to measure PA was given at all TPs (*Appendix 5).* The project leader also followed up all participants by phone calls before and after the data collection took place.

A longitudinal design with more than one data collection may increase the possibility of dropouts (Thomas et al., 2005). When investigating patients or clinical trials, health problems might occur during the intervention or study period and should be considered before starting the research project. The participants in this sample have CFS/ME of mild to moderate degree, but dropouts caused by health problems or other factors might occur. Since the thesis also includes only 3 TPs the risk of dropout is reduced. A recent study from Dougall and colleagues (2014) investigated dropout rates from CFS/ME patients included in the PACE trial (White et al., 2011). Negative health events affecting eyes, nose and throat were reported by 54% of the participants. 46% of participants reported health problems attributed to CFS/ME, and a small number had gastrointestinal, psychiatric/psychological and musculoskeletal health events (Dougall et al., 2014). There were no serious health related events associated with either CBT or GET, but "pacing" was associated with more frequent deterioration in physical functioning. The authors also stated that if the patient had more symptoms associated with CFS/ME, as a more severe degree it predicted health problems in general and a diagnosis of depressive disorder at baseline predicted increased health problems (Dougall et al., 2014).

Lack of reliability is when the participants do not have reliable performance and is especially an issue when testing younger children. This should not be an issue in this thesis, where there is no specific testing of performance. PA is measured objectively during 8 days. However, if they change behavior due to the test there will be lack of reliability. The participants were asked to act/perform normally during the measurement period. They were also told about the importance of them to act as normally as possible to provide accurate information of their situation and future treatments.

4.3 Ethics and Procedure

The regional Committee for medical Research Ethics and the Norwegian Social science Data Service approved both the pilot-study *(Appendix 2*) and this master thesis *(Appendix 3)*. Young patients with CFS/ME who were offered a rehabilitation stay at CS during the autumn of 2014 were invited to participate in the research. The ones that accepted the invitation were given informed consent and additional information about treatment, the pilot-study and the master thesis *(Appendix 1)*. The participants gave written consent before taking part in the research, and informed about their right to withdraw from any part of the data collection at any time. The assessment tools used in this thesis included the SES which is a paper-and-pencil questionnaire administration of self-efficacy *(Appendix 4),* and objectively measured PA from 3 time points (TP) see *Figure 4.2*. All data materials used in the current study are de-identified. The name and other characteristics serving to identify a person have been removed or replaced with a number. Indirect personal information is categorized into broad categories such as age interval etc. rather than exact age of the participants. The project leader, who has the responsibility for correct and proper compliance of the legislative provisions on data security and internal control, is the only one who can identify individuals through the separate list of names and other information.

4.4 Measurements

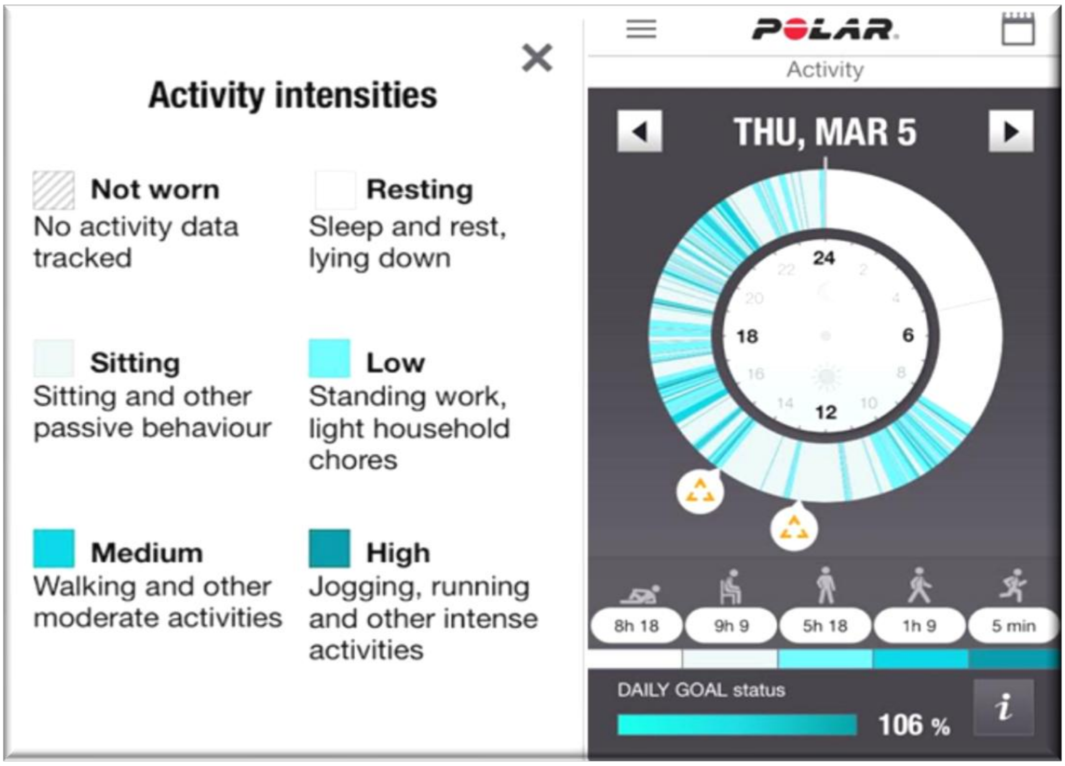
4.4.1 Self-efficacy

Bandura (2006) has developed guidelines for constructing efficacy measures. These include domain specification, gradation of challenge, content relevance, response scaling, phrasing of items, item analyzing, minimizing response bias and validation. I will further go through some of those pertinent for this thesis. Self-efficacy items are collected from the *Exercise Self-Efficacy Scale* (ESES), which was used in a PhD thesis conducted at the Norwegian school of sport science (Sæbu, 2011). They investigated PA and motivation in young adults with a physical disability and translated the self-efficacy scale to Norwegian. An example of an item is “I am confident that I can be physically active no matter how tired I feel”. Response where given on a ten point scale ranging from *Not true* (1) *to always true (10)*. In this thesis small changes of the items were done to more precisely target young CFS/ME patients (*Appendix 4*). In total 14 questions were chosen with the aim of covering self-efficacy for being physically active.

Efficacy beliefs that are specific to particular domains of functioning are stronger predictors of behavior than global expectations (Bandura 2006). I have chosen to use PA in all items and some barriers concerning CFS/ME that might affect PA behavior. The items are built upon their confidence in their capability of performing PA in everyday life. A compressive assessment of all that is needed to succeed will produce a more predictive efficacy than just one aspect of the behavior (Bandura, 2006). Therefore multiple items (n 14) rather than a single item are used in current study. Ensuring that efficacy measures and performance/behavior measures are concordant is necessary (Bandura, 2006). Small corrections of chosen items where done to more precisely target young CFS/ME patients and their barriers to PA behavior. Strength of self-efficacy was measured on a ten point scale ranging from *Not true* (1) *to always true (10)* and all questions started with “*I am confident that I can…*” The items reflected physical, psychological and practical barriers for being physically active. The highest possible sum score at SES is 140 and lowest is 14, the higher score the stronger the self-efficacy and vice versa.

4.4.2 Physical Activity

PolarLoop (PL) which is one of the newly-developed activity trackers available on the market was used to assess PA both as “*upright activity*” and moderate to vigorous activity (MVPA). PL is a small lightweight device that is placed on the wrist and it captures movements in three dimensions. Polar has used an unknown method concerning executions to transfer the movement frequency into activity intensity and analyzed in 30 seconds epochs using a 1-5 minute time window (Virtanen, 2014). Intensity of activities is calculated into metabolic equivalents (MET’s) taking into account the age, height and weight of the individuals. Performed activity is categorized into six activity levels: *Non-wear, Sleep/rest, Sitting, Low, Medium and High* (*See Figure 4.3)*. MET values in the different activity levels are set to low (1.8-3.5 MET’s), medium (3.5-6 METs) and high (>6MET’s) more details is found in the Polar report written by Virtanen (2014). In addition amount and type of movements are registered and transferred to estimation of steps in 60 seconds time epochs, but steps as an outcome variable is not used in this thesis. PL includes the internet service *Polar Flow*. All data analysis is uploaded from PL to created profiles for all participants on Polar Flow. Data is displayed as total minutes performed in PA or inactivity divided into the given activity intensities (*Figure 4.3)*.



Activity intensities

Not worn No activity data tracked

Resting Sleep and rest, lying down

Sitting Sitting and other passive behaviour

Low Standing work, light household chores

Medium Walking and other moderate activities

High Jogging, running and other intense activities

Figure. 4.3: Picture from the website Polar Flow after uploading activity from Polar Loop. The figure shows the given activity intensities from polar, and the information given from recorded activities at the following internet service PolarFlow.

The participants used PL consecutively for 8 days at all TPs. One day is a 24 hour recording, including nighttime and sleep. If there were days without recordings, they were excluded from the analysis. In addition, the first and the last day of recorded data were excluded to control for bias. The outcome variable used in this thesis is minutes/day in upright activity (*Low, Medium* and *High). According* to Bell and Bell (2010) upright activity is a reasonable and informative variable concerning CFS/ME. MVPA are commonly used in the literature, and provide information possible to compare with the PA guidelines (WHO, 2010). Hence, this variable is also included in analysis. MVPA include minutes performed in *Medium* and *High* activity intensities given from PL *(Figure 4.3)*. Importantly, counts/minutes are not available in the recording given from PL therefore the possibility to compare current results with other objectively measured PA is limited. Wear time of PL was analyzed as such. How many hours of wear time the participants used PL per day. This was conducted to control for little wear time or empty days of recording.

4.5 Validity and Reliability

The validity and reliability in the research method and the assessment tools are essential for the findings and the researcher's conclusion. Quality of the research depends upon the validity of the scientific method, and the ability to scientifically answer the questions of interest. Validity is defined as the degree to which a test or instrument measures what it is supposed to measure (Thomas, Nelson & Silverman, 2005). An integral part of validity is reliability and is understood as consistency or repeatability of a measure. The results from a test can be reliable and not valid, but never valid and not reliable. Cook & Campbell (1979) has identified four important threats to validity when doing research. This is s*tatistical-, internal-, construct- , and external validity*. The threats to validity and issues in current method are further described.

4.5.1 Statistical validity

Probability statements are often used to describe the confidence in the statistical findings. The p-value is commonly used to say if the results are statistically significant. If the P-value is <0.05 the difference or relationship of this size or result would be expected less than 5 times in 100 (Thomas et al., 2005). Level of probability is often sat before the research and is called alpha (α). In behavioral research this is normally 0.5 or 0.1 (Thomas et al., 2005). But might also be higher concerning the aim of the research. Alpha is used to control for type I error. There are two type of error a researcher may make, type I and type II error.

- *Type I error is to reject the null hypothesis when the null hypothesis is true*
- *Type II error is retain the null hypothesis when the null hypothesis is false*

The chance of error still exists but with α they are specified. Type II error is controlled by beta (β) and has a relationship with α. When α is sat increasingly smaller, β becomes larger. Therefore it is necessary to decide which type of error that is most important to avoid. Is it more important to avoid concluding that self-efficacy is an important assessment tool when self-efficacy is not an important assessment tool for young CFS/ME patients when it in reality is (type II).This is not easy to decide, when wanting to avoid both types of error.

In addition to reporting the significance of the results the "meaningfulness" of the difference between two means can be estimated. This can be done by giving the effect size (ES), which is the standardized value that is the difference between the means divided by the standard deviation also called delta, $ES = (M_1 - M_2)/SD$ (Cohen, 1969). M_1 is the mean of one group, while M_2 is the mean of a second group, which SD to use is discussed in the literature but commonly a pooled SD is suggested (Thomas et al., 2005). The ES can then be compared to guidelines for behavioral research suggested by Cohen (1969).

- *< 0.2 is small*
- *0.5 is moderate*
- *> 0,8 is large*

Power is another concept in statistic, the greater the power the more likelihood for detecting a real difference or relationship (Thomas et al., 2005). The size of the sample is extremely influential on power (Thomas et al., 2005). If number of participants increase the power increase. It is possible to estimate the sample size needed to reach a specific power and to identify an important effect. To estimate the sample size a pilot or previous research must have been conducted. There is no previous research in current field of interest and the ES is not known. However, an estimation guess is performed out of interest.

Alpha could be set to .05 to avoid type I error. Cohen (1988) suggest that in behavioral research the seriousness of type I to type II error should be in a 4-to-1 ratio. Beta is therefore .20 which control for type II error. Power is 1 – beta, which gives 0.8 and is commonly accepted as an appropriate power in behavioral research (Thomas et al., 2005). A large sample size in current population is difficult to gather, as well as dropouts need to be considered so 20 participants in current design are reasonable. If a moderate ES (0.5) is decided 20 participants with an alpha of .05 and a moderate ES of 0.5 would give a statistical power of .30 according to tables given in Thomas et al., (2005). Current study would therefore have a 30 percent chance of detecting a real difference. This was expected, a small sample group affect the power. Expected ES are normally based on previous research, and current design does not include any control group, estimation of the prevalence of young CFS/ME patients is also poor. Estimation of sample size to detect a real difference is therefore not making much sense. When using a dependent t-test (repetitive measurements of the same group of participants) the effect could be given in percent, but also estimation of the ES. Subtracting the pretest from the posttest and divide it on the pretest SD would give an estimation of the ES and meaningfulness of the results and can be used for future research. Since self-efficacy as an assessment tool as well as objectively measured PA is not investigated in earlier studies the feasibility of the research is a main part of this thesis, and the overall pilot-study.

4.5.2 Internal validity

Internal validity is about the ability to prove causality, if the conclusion made accurately reflect the variables investigated, and not something else. This is especially important in experimental research where elimination of the possibility that anything else than the independent variable caused the change in the dependent variable. A longitudinal design has the aim to investigate the change in variables in the same group of people over more than two TPs. Considerations about other extraneous factors which can influence the results are important for the conclusion. History is one of the threats to internal validity identified by Campbell & Stanley (1963) and is important in longitudinal designs as changes can occur between the TPs. The treatment for young CFS/ME patients at CS is a multidisciplinary individualized approach, which is a real life setting. This made it difficult to standardize the intervention and external factors. In longitudinal designs it is nearly impossible to stop external factors such as illness, personal characteristics, psychological mood or societal influences. On the other hand, when investigating real life situation these external factors will always be part of the participant's situation to some extent. In this thesis the measurement periods are only some weeks apart and the improvements or effects are most likely to be because of the multidisciplinary treatment at CS. However, exactly what causes the improvements or changes are not known for sure. Maturation is another threat important in longitudinal design, but since current study only includes the first three TPs in the pilot-sturdy, maturation should not be a big concern. *Dropout* as discussed earlier might be a caused by worsening of symptoms or the need of treatment elsewhere. Other threats to internal validity identified by Campbell & Stanley (1963) that might be important to consider in this thesis is *instrumentation*.

Instrumentation threats to the internal validity are caused by the assessment tools. The preferred assessment tools can be validated against some criterion. Criterion validity tells how well a test or device correlate with a measure that is already validated or accepted in research (Thomas et al., 2005). Measures that are used as criterion are often considered as the gold standard in its field. For instant DLW is considered as a gold standard for metabolic rate and for energy expenditure. These gold standards are often difficult to assess, require a laboratory and are expensive. Hence, more easily administered assessment tools are preferred, especially in field research. The preferred assessment tool can be correlated against the gold standard to secure validity and reliability. Evaluation of the assessment tools is part of the pilot-study but more as in the procedure and design. Criterion validity should be acceptable in the SES sins it is prepared from the ESES which is an already used questionnaire in the literature. The ESES is validated in a sample of 368 individuals with spinal cord injury (Kroll et al., 2007) and they concluded the ESES is a reliable instrument with high internal consistency and scale integrity. In addition the SES was tested for internal consistency at baseline with good results (*see table 5.1*). Translation was performed by a Norwegian PhD research team (Sæbu, 2011)

Concerning PL the validity of the device is not adequately performed as far as I know. PL is a relatively new developed device not used in research but the method Polar uses is evaluated. The "Polar method" is compared to indirect calorimetry which shows less than 0.5 MET mean square error (MSE) in activities like sitting, reading, using PC, dusting etc. In activities like mopping, moving items, vacuuming the MSE was 0.5-0.75 MET and in activities such as football, jogging, running and mowing the lawn the MSE was 1.0-2.0 MET (Virtanen, 2014). The method used by PL to calculate MET into kcal has been developed from indirect calorimetry as a gold standard in short performances (Brugniaux et al., 2010), and DLW technique in longer performance (Kinnuen, Tanskanen, Kyröläinen & Westerterp, 2012). The correlation coefficient was 0.86-0.89 regarding the total energy expenditure (Virtanen, 2014; Kinnuen et al., 2012).In addition a pilot-test was performed before using PL in this thesis. PL was validated against *direct observation* as a gold standard in everyday PA. The pilot test included 5 participants which performed 10 minutes blocks in each activity level described by PL (fig. 4.3). The conclusion was that PL is a valid assessment tool with a mean relative difference at only 2 percent, but more validation studies are needed.

Repeated testing is also identified by Campbell & Stanley (1963) to be a threat for the internal validity. The participant's behavior or intellectual growth in the field might be caused by repeated testing. Therefore the results might not apply to untested patients. Possible effect might occur from the measurements. Participants may change their answers or behavior due to the testing. The feeling of self-efficacy is an individual measure where there is no right or wrong answer. However, the participants could get an impression of what a socially acceptable answer might be. Influences from the other participants or the health therapist might also occur during the treatment. In experimental research many of the threats to internal validity can be controlled by randomization of participants, placebo or double-blind setups. However, when a control group is not conducted, the randomization process and placebo method have less meaning. Randomly selection is not weighted in this design for practical and ethical reasons. Random assignment can rarely be used in real world settings (Thomas et al.

2005). Since this thesis is based on real world setting it was not possible to conduct the randomization process, since the participants applied for the treatment.

This thesis is part of a larger longitudinal design which includes a multidisciplinary treatment where many external and internal factors might influence improvements. Observation of effects, and health related and functional improvements in the CFS/ME patients after learning coping strategies for their illness as well as pattern of interest are more actual in current design. Feasibility of procedures, assessment tools, design and method are the main aim and not necessarily about cause and effect. Studies are never perfect, completely free from threats to the validity, but should aim to reduce the threats as much as possible. When dealing with patients where the etiology and pathology of their illness is unknown and the effects of commonly used treatments are widely discussed, observation research of real life situations might be a place to start. Observation and a humanistic point of view could be beneficial before conducting larger RCTs.

4.5.3 Construct validity

Self-efficacy refers to an individual's beliefs in their capability to complete tasks, reach goals and/or execute behavior, and plays a critical role in how we think, feel and behave. In psychology, a construct is a trait or ability that is based on theories. A construct exist in theory and has been observed to exist in practice. Construct validity is the experimental demonstration that a test measures the construct it claims to be measuring. It refers to whether the operational definition of a variable actually reflects the true theoretical meaning of the concept. In this thesis the concern would be whether the SES matches up with the theoretical concept of self-efficacy and if PL actually measure PA. Evaluation of the measurement tool is essential for establishing construct validity. Correlation can be used to evaluate how well the measurement tool agrees with other previously validated tests that measure the same construct. This is basically the same issue which concerns instrumentation threats. Construct validity can be a threat to validity in this thesis. If misunderstanding has occurred when answering the SES, or if the participants answer what they believe is the favorable answer. The items in SES are validated in previous literature. However, small modifications are done to more precisely target young CFS/ME patients but this should not have affected the construct validity. In addition, to define the construct as precisely as possible also called operationalization is of importance for construct validity. Precise explanation of the terms was given before the participants answered the questionnaire. Concerning PL's ability to measure adequately measure PA and divide it into the activity intensities was pilot tested before current research with good results. However, more validation studies are needed.

4.5.4 External validity

External validity refers to the ability to generalize the results of other populations or situations. External validity is threatened in four ways identified by Campbell & Stanly (1963). *Reactive or interactive effects of testing*, this means that the pretest may affect the results of the treatment. This should not be a big issue in current results since there is no favorable answer to the questions asked in SES and answers are given as perceived capability. Concerning PA, some participants may want to achieve the same or higher amount of PA at posttest and therefore change their normal behavior pattern. *Interaction of selection bias and experimental treatment*, that if a group is selected on some characteristics, the treatment may work only on these participants. This may be a threat for the ability to generalize the results in current study because the specific sample group. *Reactive effects of experimental arrangements* means that, the treatment only may benefit the participants in an exact setting that is used in the research (e.g., in a laboratory) and not in the real world. The treatment at CS is individualized and may be only transferable to other individuals in the same setting. *Multiple-treatment interference* is when participants receive more than one treatment, the previous or additional treatment may influence. If the participants received any treatment elsewhere, they were excluded from the analysis. However, the treatment at CS is a multidisciplinary approach where individualized treatment strategies are performed. Current sample size is very specific and small, therefore the results from current sample is not necessarily possible to generalize in the population. However, other young CFS/ME patients may apply to the current results.

4.5.5 Reliability

Reliability is the degree to which a measurement tool produces stable and consistent results. Standardized procedures of the measurement, the information given and data collection are important for the reliability. A correlation coefficient can be used to assess the degree of reliability. The coefficient of reliability range from 0 to 1 and reflects the degree to which the measurements are free of error variance. The closer the coefficient is to 1 the less error the score reflects. Test-retest reliability refers to the test's consistency among different administrations. This is not performed in current study because of the design is including an intervention. However, the internal consistency of the items in SES is tested. Cronbach's alpha is a coefficient of reliability, or consistency and is commonly used in psychometrics such as self-efficacy. In current master thesis, Conbach's alpha was used to measure the internal consistency of the items in the SES. Since all of the items (n14) in the SES measures self-efficacy the internal consistency should be high. A commonly accepted rule of describing internal consistency is that 0.6 -0.7 is acceptable, 0.7 – 0.9 is good, and >0.9 is excellent. The Conbach's alpha results for SES are listed in *Table 4.1*. The alpha was over 0.8 at TP1 which indicates a good internal consistency, in addition at TP2 and TP3 the Conbach's alpha was over 0.9 which is excellent. When looking at the item total statistics (*data not shown*) it did not seem that removal of any of the 14 items would give a higher internal consistency.

4.5.6 Other issues or advantages

Because longitudinal studies involve repeated measures the management of the data is sometimes complicated. Data for each individual participant have to be linked, and dropouts during the research are problematic causing loss of data material. Loss of participants can affect any research but is particularly more likely in longitudinal designs. Even the whole sample group can be lost if the research lasts for a longer period, especially when dealing with patients of any kind. Attrition or dropouts is a risk that must be taken in the ability to conduct a longitudinal design. However as chosen in this thesis a shorter research period might reduce the risk of dropouts but still give a descriptive insight of the results. Comparing the patients with their "earlier selves" allows detailed information about improvements and outcome variables and might be a big step of developing knowledge in CFS/ME and to more appropriate understands about the young CFS/ME patient.

4.6 Statistical analysis

Statistical Package for the Social Sciences (SPSS) version 21 was used for the statistical analysis. In addition to SPSS, Microsoft Office Excel 2010 was used to make charts and tables. The total mean for the sum score was calculated for analysis, in addition to the mean score at each question. SD is used to measure how much variation it is in the distribution, low SD means values are close to the mean, while high SD indicate that the values are more spread out. Because of a high variation in the score, median (the middle value) and range (minimum-maximum) is also given for most analysis. Is the effect or relationship of interest reliable, is it significant? And how strong is the effect or relationship of interest? Statistics are used to describe data, determine relationships among outcome variables and to test for differences among groups. A common procedure in research is hypotheses testing, where the statistical test will tells whether to reject or retain the null hypothesis (H0). H0 hypothesis are usually the opposite of the statement of interest and commonly takes form as "no difference".

Correlation is a statistical technique used to determine the relationship between two or more variables, for instant self-efficacy and PA. The degree of relationship between these variables is of interest. If self-efficacy increases, does PA increase? Another interest could be if self-efficacy could act as a predictor for the level of PA. The coefficient of correlation is a quantitative value of the relationship which range from -1 to +1. A perfect positive correlation as a value of +1, a perfect negative correlation has a value of -1, while 0 is no correlation at all. Importantly, cause - effect is established by theory, logic, and the total experimental situation (Thomas et al., 2005). Hypotheses testing and correlation in this thesis is conducted to establish if there is any change in the variables over time, and if there is any relationship between the variables. However, patterns of change and possible improvements, in addition to feasibility of method and design are emphasized in this thesis rather than only cause-effect established by statistical significance. Researchers often expect that evidence or the results should support the alternative hypothesis (the one of interest). Hypotheses can be one- or two tailed, two tailed statistical test can be used when the effect can go in both directions such as in this thesis; there is no change in self-efficacy. Rejection of this H0 hypothesis can be caused by both a negative or positive change in self-efficacy, and a two-tailed test should be used. To decide which statistical tests to use, the sample characteristics and distribution should be investigated.

4.6.1 Distribution of the data

Normal distribution assumptions for statistical analysis need to be considered in order to decide whether to use nonparametric or parametric statistical tests (Thomas et al., 2005). A normal distribution means that the outcome variables take place around the mean, and tails off symmetrically away from the mean (Gaussian distribution/bell curve). The mean, median, and mode present the center of the distribution where ± 1 SD from the means include 68% of the scores, ± 2 SD from the mean include 95 % while ± 3 SD includes 99% of the scores (Thomas et al., 2005). The distributions mean, skewness and kurtosis coefficients should be calculated in order to determine distribution of the data. Skewness describes the direction of the hump of the distribution curve, and the direction of the tail. Kurtosis is a description of the vertical characteristics of the curve showing the data distribution (Thomas et al., 2005). It is usually required that normal distributed data has values of skewness and kurtosis within +/-2 (Hair, Anderson, Tatham & Black, 1995). However, there is also literature which accepts values of skewness within +/- 3 and kurtosis within +/-10 as normal distributed data (Kline, 2005). If the skewness and kurtosis exceed the recommended values it may decrease the reliability of the results and one should consider removing some of the extreme values. The skewness is more important to secure reliability (Thomas et al., 2005). If the data do not meet the assumptions required to perform parametric tests, it is necessary to use nonparametric statistical tests (Thomas et al., 2005). In the current data, analysis of the skewness and kurtosis were performed at all TPs listed in *Table 4.1*.

Table 4.1: *Results from the Shapiro-Wilk test (p-value), Skewness and kurtosis performed on all data set used for analysis. In addition the Conbach's alpha (α) is given for the self-efficacy scale.*

	Shapiro-Wilk test	Skewness	Kurtosis	Cronbach's α
SES TP1	.04*	-.24	-1.37	.86
SES TP2	.82	-.19	-.58	.91
SES TP3	.51	-.45	-.85	.92
Upright Activity TP1	.05*	1.13	1.00	-
Upright Activity TP2	.61	-.29	-.09	-
Upright Activity TP3	.59	-.17	-.90	-
MVPA TP1	.01*	1.62	3.37	-
MVPA TP2	.01*	1.39	2.20	-
MVPA TP3	.03*	.77	-.79	-

Note: * p<.05

The skewness and kurtosis was within +/-2 at all TPs for the SES data. For upright activity and MVPA the skewness was within +/-2, but the kurtosis is >3.The skewness is within +/-2 in both upright activity and MVPA, but the kurtosis is >3. The skewness which is the one that has a high impact on reliability is within the recommended values of Hair and colleagues, and the kurtosis is within the recommended values from Kline (2005). The *Shapiro-Wilk test* was used to test if the data were normally distributed (Shapiro & Wilk, 1965; Razali & Wah, 2011) listed in *Table 4.1*.The H0 hypothesis in this test for of normality, is that the data are normally distributed. When the test is significant (p<.05) it means the data is not normally distributed and the Ho is rejected. In addition a visual inspection of the histogram and normal Q-Q plots was performed. The Shapiro-Wilk shows that both SES and upright activity at TP1 are significant, meaning they are not normally distributed. Significant values are also seen in MVPA at all TPs. This is also seen with a visual inspection of the histogram and normal Q-Q plots. A small sample size reduces the probability for normally distributed data in addition to outliers. There did not seem to be any outliers in the data set, but values are spread out from the mean. Due to the data sets which were not normally distributed and a small sample size nonparametric statistic is used in this thesis. Nonparametric statistics make no assumptions about the probability distributions of the variables and are used for the statistical analysis.

In the hypotheses testing, the differences between the TPs were tested with t-test for dependent sample, using the nonparametric alternative *Wilcoxon's matched pairs test*. To test if there were any relationships between self-efficacy and PA variables the *Spearman R* (Siegel & Castellan, 1988) were used given as correlation coefficient and if it was significance. The total mean for the sum score was calculated for analysis, in addition to the mean score at each question. SD is used to measure how much variation it is in the distribution, low SD means values are close to the mean, while high SD indicate that the values are more spread out. Because of a high variation in the score, median (the middle value) and range (minimum-maximum) is also given for most analysis.

5. Results

In this chapter the results will be presented. First, it is shown an overview of the descriptive statistic, and the important characteristics. Secondly, results from the *Wilcoxon Signed Rank Test* are presented showing change over different TPs in both SES and PA, findings concerning research questions and hypothesis will be presented.

5.1 Descriptive Statistic

In total, 25 participants accepted the invitation to partake in the study. Unfortunately, one participant had to drop out during TP2 because of psychological problems, and treatment elsewhere. Therefore, 24 participants were included for analysis in current study. There were more females (n = 19) than males (n =5) and the age of the participants ranged from 15-24 years of age (mean 17, 83 SD ± 2). All 24 participants completed the SES at all three TPs. The score in SES is given as the sum of all answers ranging from 1-10 (n14), giving a maximum score at 140. The mean (±SD) and the median (max/min) for the participants are listed in *Table 5.1* for all TPs. The mean score in SES is relatively high at TP1 but increases at TP2 which indicates a stronger self-efficacy. There is a decrease in the mean score from TP2 to TP3 indicating a weaker self-efficacy 12 weeks after the participants received treatment. The PA recordings were divided in upright activity and MVPA. The participants wear time of PL was calculated. At TP1 there was two participants with unreliable data, one had recordings only during the weekend, while the other did not have recordings at all of unknown reasons. After excluding the unreliable data, 22 participants were included for further analysis. From visual inspection and calculations there were not much difference between the week days and the weekend. Therefore conclusion was made to use 3 days from the week and 1 day for the weekend was used for the mean analysis (Tue, Wed, Fri and Sat). The same days were used at all TPs. The included participants for the PA analysis, fulfilled to have more than 4 hours of wear time at TP1 *(Appendices 7)*. At TP2 and TP3 there were more days with less wear time and some of the participants had days with only 4 hours. In the interest of keeping as many participants as possible for analysis who recordings during the days decided were included. All the 22 participants were also included for TP2, but at TP3 there was in total 4 more participants whom had to be excluded from the analysis, in total 18 participants were included for analysis at TP3. At TP1 the participant's mean time in upright activity was, 4 hours and 25 minutes, where 35 minutes (±26) was in MVPA. At TP2 the mean time spent in upright activity was 4 hours and 33 minutes, where 38 minutes (±23) was spent in MVPA. At TP3 mean time in upright activity was 4 hours, where 32 minutes (±24) was in MVPA.

Table 5.1: *Central tendency of SES, Upright activity and MVPA at all TPs given in mean ± SD and median (min-max). Wear time of polar loop are listed as hours.*

	Mean (Score± SD)	Median (Range)	Wear Time (Hours)
SES			
TP1 (n=24)	92 ± 1.50	102 (60 – 128)	---
TP2 (n=24)	99 ±1.55	101 (56 – 136)	---
TP3 (n=24)	95 ±1.68	97 (50 – 131)	---
Upright activity			
TP1 (n=22)	265 ± 90	258 (154-499)	23 (SD ± 2.20)
TP2 (n=22)	273 ± 76	283 (104-422)	22 (SD ± 4.12)
TP3 (n=18)	240 ± 80	231 (99-359)	22 (SD ± 4.27)
MVPA			
TP1 (n=22)	35 ± 26	29 (6-118)	23 (SD ± 2.20)
TP2 (n=22)	38 ± 23	35 (11-104)	22 (SD ± 4.12)
TP3 (n=18)	32 ± 24	21 (3-81)	22 (SD ± 4.27)

Note: The highest achievable score in SES is 140. Each question (n14) has a score which range from 1-10, higher score indicate a greater self-efficacy in the domain of physical activity (PA)

5.2 Change in self-efficacy (Hypothesis 1)

The hypothesis *"Perceived self-efficacy fort being physically active increase, after the participants receive treatment at CS"* was tested in the purpose to accept or reject the null hypothesis (*no change*). To measure if there was any significant change between the different TPs the *Wilcoxon Signed Rank Test* for related samples was used. The analysis from both the individual questions and in total are listed in *Table 5*.2. The analysis shows that there is nearly a significant change in SES from TP1 to TP2. The mean score at TP2 had a relative change at 8.16 percent compared to TP1. This indicates that the participants seemed to increase their self-efficacy beliefs in domain of PA after received treatment at CS.

Table 5.2: The mean score with standard deviation (SD) and change between the time points (TPs) for all questions (Q1-14 in the self-efficacy scale (SES). In addition the total SES mean and changes (p-value) are given.

	Mean score			**Change**		
	TP1 (n24) Mean ± SD	TP2 (n24) Mean ± SD	TP3 (n24) Mean ± SD	TP1-TP2 P-Value	TP1-TP3 P-Value	TP2-TP3 P-Value
Q1	4.92 ± 2.89	5.58 ± 2.41	5.38 ± 2.60	.28	.25	.78
Q2	6.50 ± 2.32	6.50 ± 2.59	6.79 ± 2.11	1	.60	.61
Q3	6.67 ± 2.20	7.17 ± 2.39	7.33 ± 2.41	.32	.32	.71
Q4	8.38 ± 1.91	8.46 ± 1.84	8.63 ± 1.79	.85	.85	.73
Q5	7.25 ± 2.58	6.88 ± 2.42	7.13 ± 2.49	.54	.79	.75
Q6	8.13 ± 2.56	8.33 ± 2.30	7.92 ± 2.43	.43	.72	.26
Q7	8.17 ± 2.44	8.21 ± 1.89	7.79 ± 2.15	.83	.38	.36
Q8	5.96 ± 2.46	7.13 ± 2.13	6.63 ± 2.02	.04*	.49	.10
Q9	6.75 ± 2.77	7.75 ± 2.01	7.58 ± 2.26	.03*	.04*	.52
Q10	5.88 ± 2.46	6.92 ± 2.29	5.96 ± 2.53	.02*	.67	.04*
Q11	6.96 ± 2.55	7.38 ± 2.39	7.08 ± 2.62	.51	.57	.84
Q12	6.46 ± 2.52	6.92 ± 2.23	6.42 ± 2.76	.36	.92	.28
Q13	4.00 ± 2.57	5.08 ± 2.32	4.92 ± 2.59	.01*	.10	.71
Q14	5.88 ± 2.68	7.08 ± 2.32	5.46 ± 2.73	.02*	.59	.01*
Sum score	**92 ± 1.50**	**99 ± 1.55**	**95 ± 1.68**	**.06**	**.49**	**.26**

Note: SES scores range from 1-10, higher score indicate a greater self-efficacy in domain of physical activity (PA).* P < .05

The difference in the individual questions between the different TPs, question 8, 9, 13 and 14 had a significant positive change (p < .05) from TP1 to TP2. There was one question (Q5) which seemed to have a negative change but this was not significant. From TP1 to TP3 there was only a relative change at 3.40 percent, showing no significance. This can be explained with a small decrease (not significant) in the mean score from TP2 to TP3, with a relative change -4.40 percent. It was only Q14 who had a significant negative from TP2 to TP3. *Figure 5.3* illustrate the mean score for all questions in the SES in graphs from all TPs.

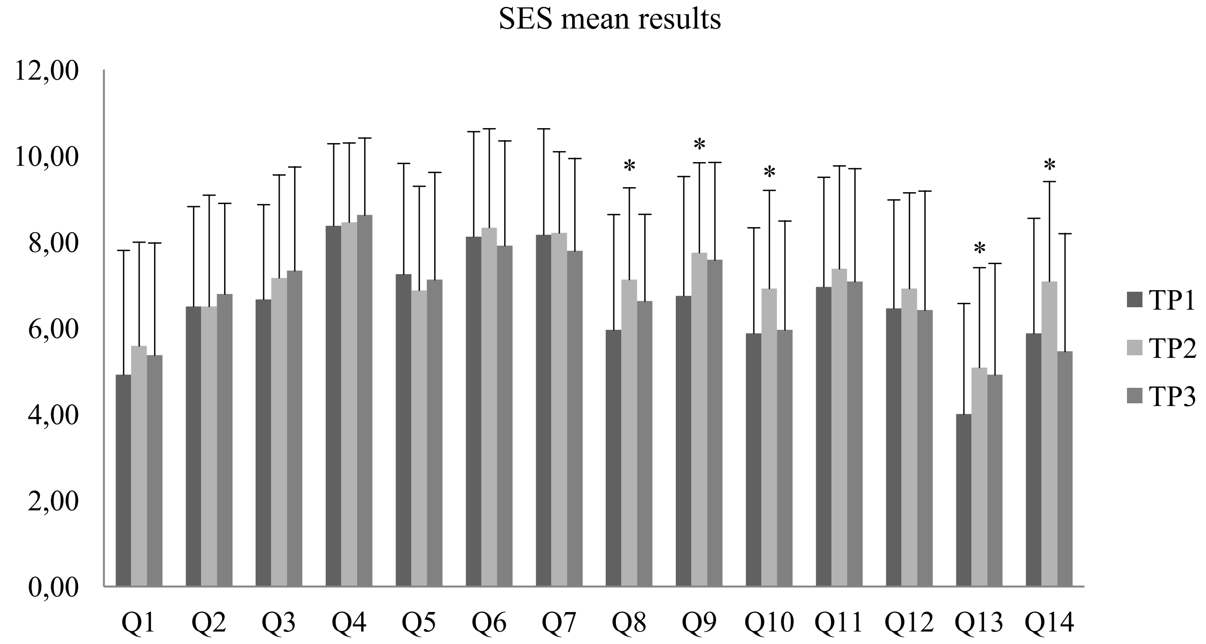


Figure 5.3*:* Mean scores for the questions in SES (n14), there is significant changes from TP1 to TP2 in question 8, 9, 10, 13 and 14. In addition there is a small decrease from TP2 to TP3.

Note: The p-value is only given for significant changes at TP2 compared to TP1 *P<.05

The results from the *Wilcon Signed Rank Test* show a tendency towards a positive change from TP1 to TP2. According to these results the alternative hypothesis is supported and H0 hypothesis rejected. The perceived self-efficacy for being physically active increase after the participants receives treatment at CS. However, self-efficacy for being physically active seemed to decrease 12 weeks after received treatment.

5.3 Change in Physical Activity (Hypothesis 2)

The mean time spent in upright activity and MVPA is illustrated in *Figure 5.5*. There are only shown small changes between the TPs with no significance. A high SD indicates that the data are spread out over a wide range from the mean. In both upright activity and MVPA, the median might give a better description of the central tendency.

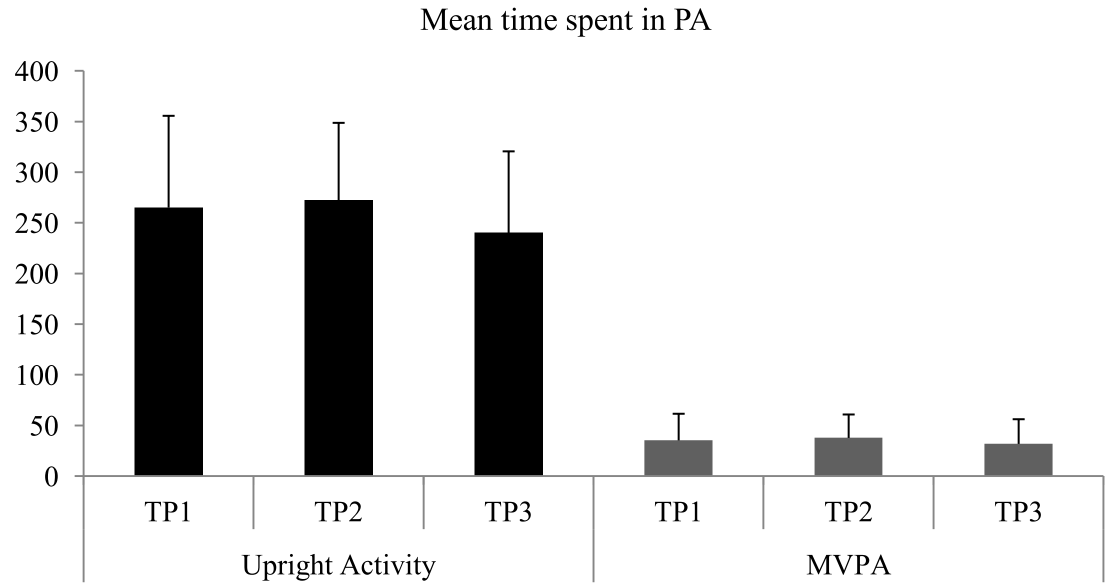


Figure: 5.5: Mean time in spent in Upright activity and moderate to vigorous physical activity (MVPA) given in minutes for all TPs.

The time spent in upright activity and MVPA given as the median describe a larger change between the TPs than the mean, but the changes are still small. A visual inspection of the differences between the days showed minimal differences shown in *Figure 5.6*. There are also minimal changes in upright activity in the weekend and weekdays, and therefore no single analysis from the weekend is performed. The participants which was excluded from the TP3 analysis, but fulfilled TP1 and TP2 (n 4) are shown in Figure 5.7.

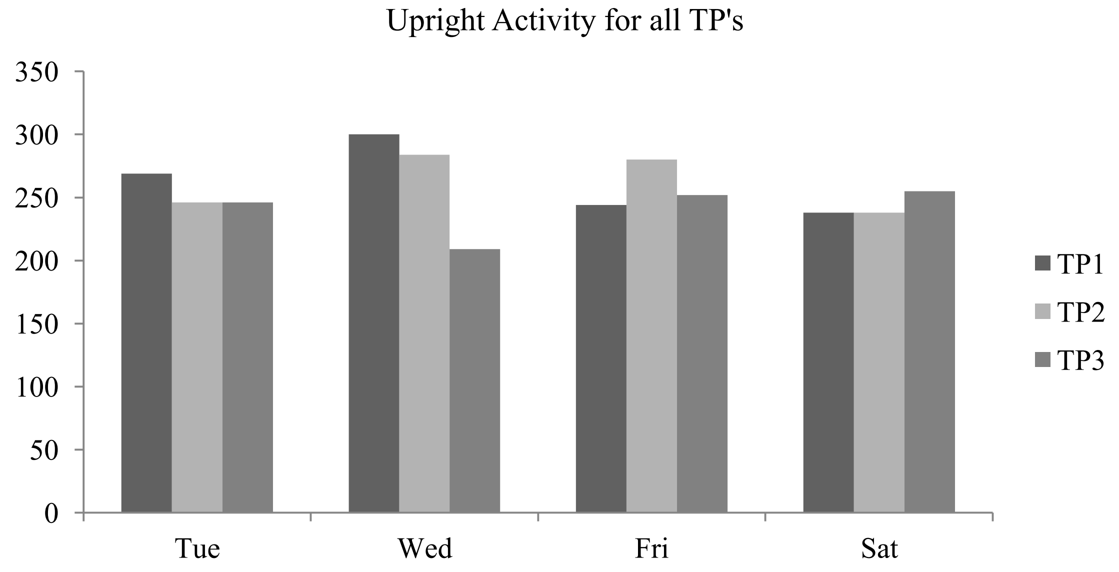


Figure 5.6: *Illustrate the differences in mean time spent in upright activity for the days included for analysis*

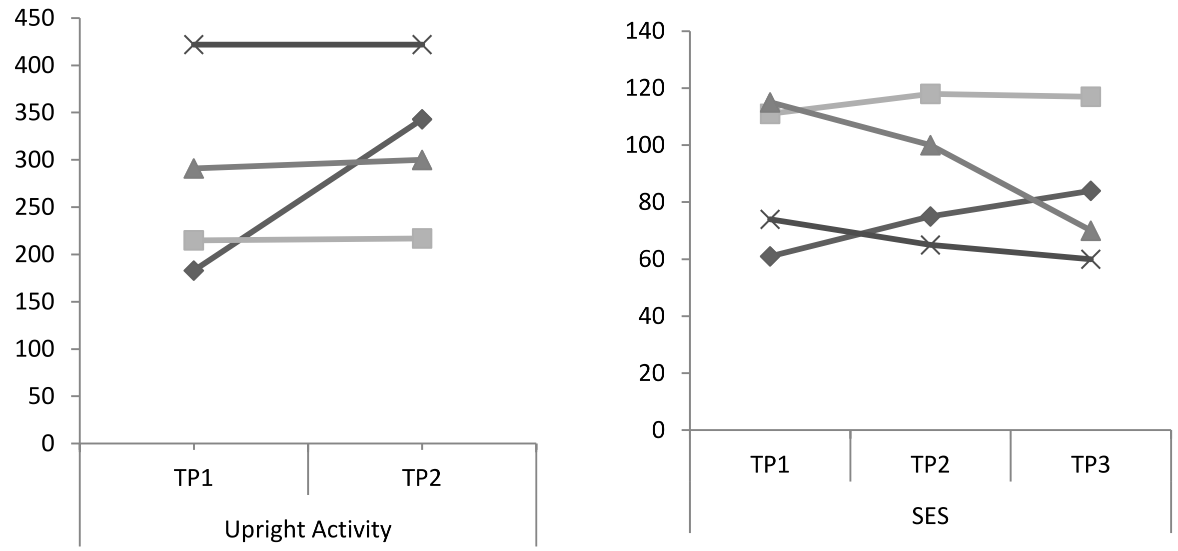


Figure 5.7: *Illustrates the patterns in upright activity and self-efficacy for being physically active (SES) for the participants (n4) who were excluded from the physical activity analysis at time point 3 (TP3).*

Figure 5.7 shows the patterns of the four participants in both upright activity (left) and SES score. Three of the participants have little change in upright activity from TP1 to TP2, while one has an increase of 160 minutes from TP1 to TP2. When looking at the SES, the one who increase in upright activity also increases in SES. There is also one of the participants who decreases allot in SES from TP2 to TP3.

Table 5.3: *The mean change in minutes spent in upright activity and moderate to vigorous physical activity (MVPA) with the following p-value.*

Change in Physical Activity

	TP1-TP2 (n22) *P-value*	**TP1- TP3** (n18) *P-value*	**TP2-TP3** (n18) *P-value*
Upright Activity	.90	.28	.31
MVPA	.45	.35	.62

*P<.05

The hypothesis *"PA increases after the participants receive treatment at CS"* was analyzed by comparing the differences between the TPs in both upright activity and MVPA using t-test for related sample (*Wilcoxon Signed Rank Test*). The results from the analysis are listed in *Table 5.3* showing no significant change in either upright activity or MVPA. Hence, according to these results H0 hypothesis is retained. There are no significant changes in PA between the TPs.

5.3.1 Further analysis in PA

The data in upright activity and MVPA has a very large SD and it might be outliers or extreme values in both directions causing no changes between the TPs. For further analysis the individual changes in upright activity was investigated and illustrated in *Figure 5.8*. There was approximately half of the sample group who had a negative change from TP1 to TP2. This indicates that these participants decreased their activity level at CS compared to their activity level at TP1. There were also participants who increased their activity level at CS compared to TP1.

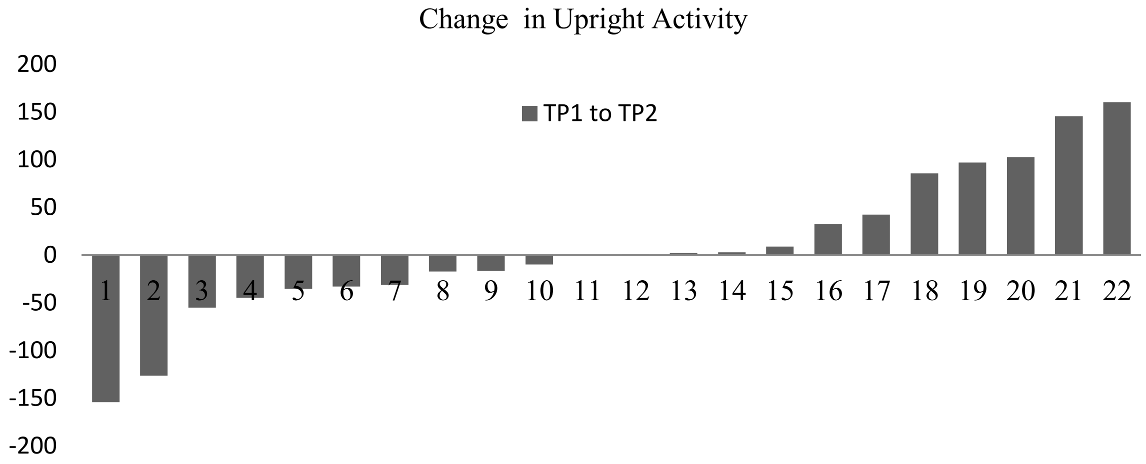


Figure 5.8: *Illustrate the mean change in minutes from TP1 to TP2 in the total sample group (n22).*

The sample group was divided in two, respectively those who decreased and those who increased their time spent in upright activity from TP1 to TP2. The central tendencies for the two groups are listed in *Table 5*.4. The group which decreased their time spent in upright activity at TP2 had a mean reduction of 47 minutes which gives a relative decrease at 15.6 percent. The group which increased their time spent in upright activity at TP2 had a mean increase of 61 minutes giving a relative increase of 26.5 percent. There are no significant changes from TP1 to TP3 or from TP2 to TP3 for the group which decreased their time spent in upright activity, but the group which increased had a significant negative change from TP2 to TP3. In summary the participants who decreased their time spent in upright activity at TP2 are generally a more active group than the ones who increased at TP2.The same pattern is seen in the SES. The group which decreased their upright activity at TP2 had a stronger self-efficacy than the group which increased at TP2. However, there is no significant change in the SES score between the different TPs.

Table 5.4: *Present the central tendency for the group who decreased their time spent in upright activity and those who increased their time spent in upright activity.*

	Decreased at TP2			Increased at TP2		
	No.	*Mean ± SD*	*Median (Range)*	*No.*	*Mean ± SD*	*Median (Range)*
Upright Activity						
TP1	11	301 ± 105	271 (159-499)	11	230 ± 57	215 (154-324)
TP2	11	254 ± 89	252 (104-422)	11	291 ± 60	311 (186-368)
TP3	10	252 ± 83	246 (99-359)	8	226 ± 80	232 (111-329)
SES						
TP1	11	96 ± 22	103 (60-128)	11	87 ± 20	80 (61-115)
TP2	11	105 ± 22	105 (65-136)	11	94 ± 23	99 (56-133)
TP3	11	102 ± 24	104 (60-131)	11	90 ± 21	91 (50-117)

	Change between TPs	Change between Tps
Upright Activity	(p-value)	(p-value)
TP1→TP2	.005**	.003**
TP1→TP3	.241	.779
TP2→TP3	.386	.050*
SES		
TP1→TP2	.213	.213
TP1→TP3	.859	.359
TP2→TP3	.355	.423

Note: **.Correlation is significant at the .01 level*. Correlation is significant at the 0.05 level

The *Figure 5.9* illustrate that the group who increased time spent in upright activity at TP2, decrease at TP3 ($p < .050$). The group who decreased upright activity at TP2 maintained the same activity level at TP3 showing no significant change from TP2 to TP3. When dividing the participants into two groups depending on whether they decreased or increased their upright activity at CS there are significant changes and H0 hypothesis rejected. However, the alternative hypothesis "*PA increases after the participants receive treatment at CS"* is only partly supported. There is not necessarily an increase in PA after received treatment. Some participants also seemed to decrease their level of PA.

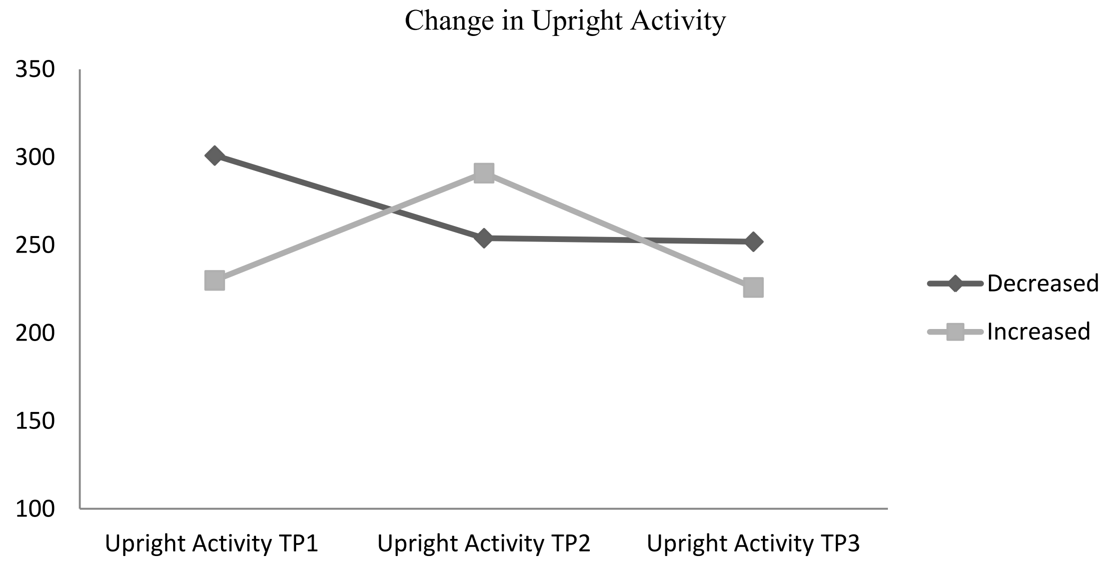


Figure 5.9*:* Illustrate the pattern of the mean changes between the TPs divided in two groups, respectively those who decreased and those who increased time spent in upright activity.

5.4 Correlation between Physical Activity and Self-Efficacy (Hypothesis 3)

The *Spearman rank correlation* was used to investigate the relationship between the participants score in self-efficacy and time spent in PA. Spearman's correlation coefficient (denoted as r_s) measure the strength of association between the variables and is listed as a matrix in *Table 5.5*. The Spearman's correlation coefficient shows little or no relationship between SES and PA at TP1. There was a negative correlation between SES and upright activity at TP2, which was statistically significant. The negative relationship was also seen between SES and MVPA at TP2. In addition, there was a negative correlation between SES scores at TP3 and MVPA at TP2.According to these results H0 hypothesis rejected. However, the results do not support the alternative hypothesis "*There is a positive correlation between perceived self-efficacy and PA".* The relationship between self-efficacy and PA tend to have a decreasing trend according to the Spearman rank correlation analysis.

Table 5.5: *Spearman rank correlation matrix for 9 variables in mean scores*

	1	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**
1. SES TP1(n24)	-								
2. SES TP2 (n24)	.624**	-							
3. SES TP3 (n24)	.500*	.731**	-						
4. Upright Activity TP1 (n22)	-.032	-.187	-.041	-					
5. Upright Activity TP2 (n22)	-.282	**-.572****	-.338	.540**	-				
6. Upright Activity TP3 (n18)	-.148	-.331	-.119	.659**	.451	-			
7. MVPA TP1(n22)	.098	-.176	.024	.615**	.434*	.441	-		
8. MVPA TP2 (n22)	-.283	**-.699****	**-.495***	.352	.827**	.393	.457*	-	
9. MVPA TP3 (n18)	-.045	-.059	.253	.306	.383	.547*	.498*	.450	-

Note: **.Correlation is significant at the .01 level*. Correlation is significant at the 0.05 level

The negative Spearman correlation coefficient corresponds to a decreasing monotonic trend between self-efficacy and upright activity at TP2 plotted in *Figure 5.10*. The same decreasing monotonic trend is also seen for SES and MVPA at TP2 *Figure 5.11*. The last statistically significant correlation coefficient between SES at TP3 and MVPA at TP2 is plotted in *Figure 5*.12

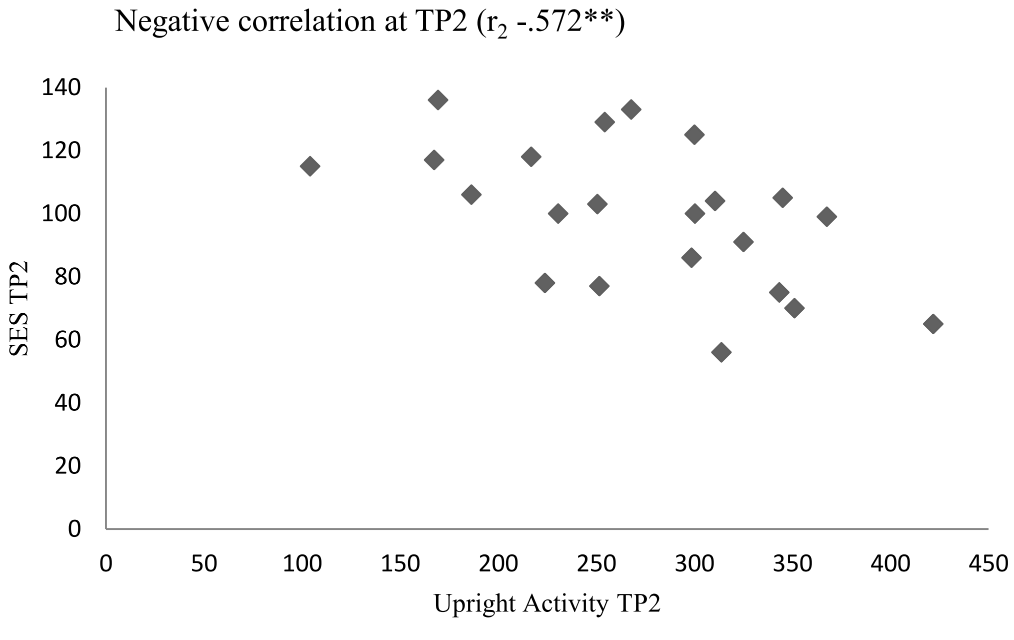


Figure 5.10 The negative Spearman correlation coefficient (r_2 = -.572**) corresponds to a decreasing monotonic trend between self-efficacy scale (SES) and minutes spent in upright activity for the participants (n22) at TP2. Note: **.Correlation is significant at the .01 level*. Correlation is significant at the 0.05 level

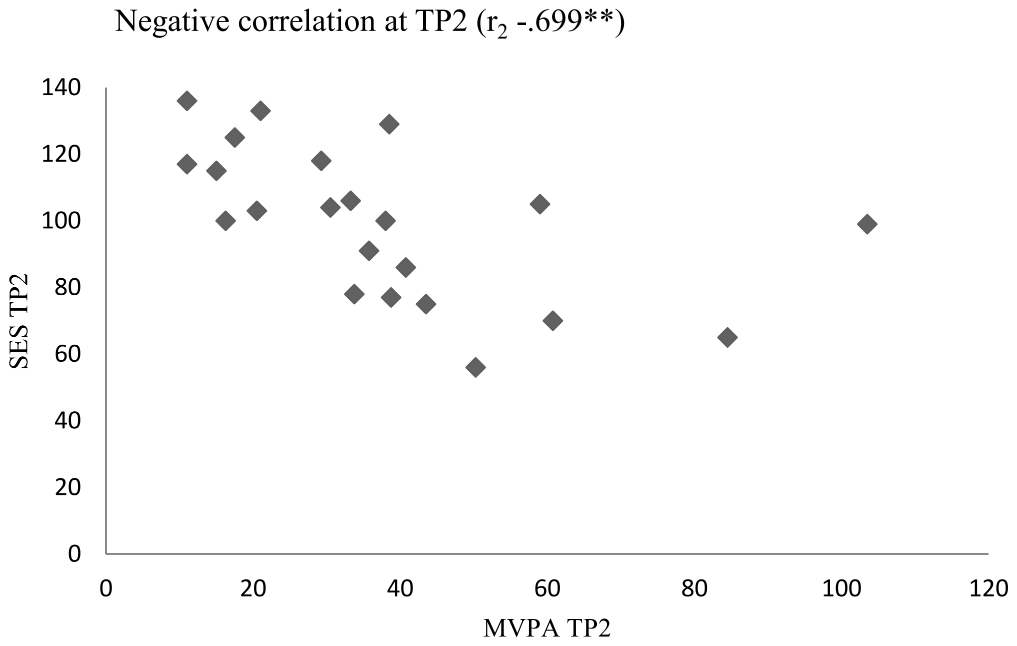


Figure 5.11 *The negative Spearman correlation coefficient (r_2 = -.699**) corresponds to a decreasing monotonic trend between self-efficacy scale (SES) and minutes spent in moderate to vigorous physical activity(MVPA) participants (n22) at TP2.*

Note: **.Correlation is significant at the .01 level*. Correlation is significant at the 0.05 level

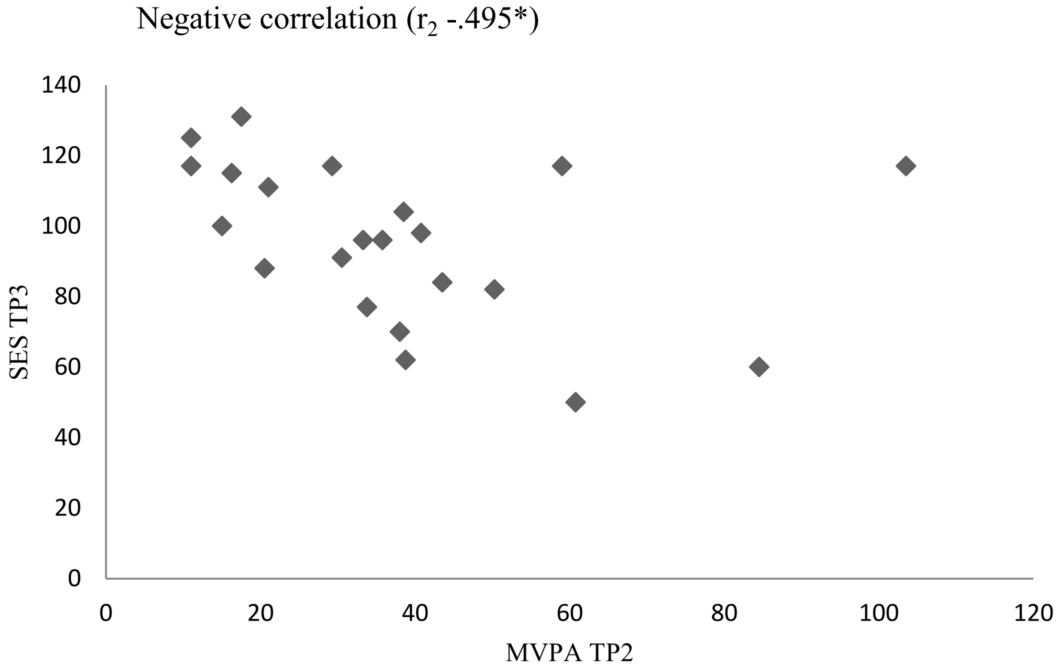


Figure 5.12 *The negative Spearman correlation coefficient (r_2 = -.495**) corresponds to a decreasing monotonic trend between self-efficacy scale (SES) at TP3 and minutes spent in moderate to vigorous physical activity(MVPA) at TP3for the participants (n22) at TP2.*

Note: **.Correlation is significant at the .01 level*. Correlation is significant at the 0.05 level

The *Spearman rank correlation* was also used to investigate the relationship between the change between the TPs in self-efficacy and time spent in PA. The results are presented in a matrix *(Table 5.6)*. According to the Spearman's correlations there was no relationship between the change in variables for self-efficacy and PA.

Table 5.6*: Spearman rank correlation matrix denoted as r_s for 9 outcome variables in change between the time points (TPs).*

	1	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**
1. SES TP1 to TP2(n24)	-								
2. SES TP1 to TP (n24)	.407*	-							
3. SES TP2 to TP3(n24)	-.319	.697**	-						
4. Upright Activity TP1 to TP2(n22)	-.093	.077	.017	-					
5. Upright Activity TP1 to TP3(n18)	-.129	-.211	-.188	.317	-				
6. Upright Activity TP2 to TP3(n18)	.082	-.179	-.153	-.620**	.484*	-			
7. MVPA TP1 to TP2(n22	-.282	-.126	-.016	.841**	.389	-.451	-		
8. MVPA TP1 to TP3(n18)	.011	.092	.034	.399	.519*	.036	.564*	-	
9. MVPA TP2 to TP3 (n18)	.264	.106	.070	-.562*	.015	.577*	-.548*	.240	-

Note: **.Correlation is significant at the .01 level (2-tailed), *. Correlation is significant at the 0.05 level (2-tailed)

6. Discussion

The purpose of this thesis was to investigate the role of self-efficacy for being physically active in young CFS/ME patients, and if assessment of self-efficacy before, during and after could provide important information for treatments and follow-up of young CFS/ME patients. Any relationship between self-efficacy and PA was also investigated. A longitudinal pilot intervention study was chosen for the purpose of investigating the research question and hypothesis, and was conducted at a rehabilitation institution in Norway. The intervention was a multidisciplinary rehabilitation treatment, tailor-made for young CFS/ME patients and lasted 4 weeks. GAT was a central part of the treatment and individual activity plans were developed. The activity plans were adjusted continuously during the treatment to establish an activity baseline in cooperation with health care therapists. In addition, the activity plans were also planned to use as a coping tool for when they returned home. SES and objectively measured PA were measured at all TPs. The three TPs included for data collection in this thesis made it possible to compare and investigate changes in the mean and median scores in SES, and the time spent in PA before, during and after the treatment at CS. Unfortunately, no control group was included in the current design, because of practical reasons and research limitations. However, this thesis is one of the first conducted in the field and can contribute with important knowledge that can be used in future studies.

CFS/ME is a complicated and serious illness, and research on this topic is complicated with many challenges in both design and method. However, more research is necessary for the ability to understand this complex illness, and how to help the patients to better cope with everyday activities and their daily situations. Investigation in a “real life” setting gave the opportunity to observe how a multidisciplinary treatment affects self-efficacy and PA and if there are any relationship between these variables. The current design is a safe and informative way to investigate and develop new knowledge on a vulnerable sample group. Since data is collected from a pilot study and the sample size is small, the power in the statistics is low. Hence, patterns that might indicate change and improvements and feasibility are emphasized rather than only statistical significance. In addition a qualitative perspective or a humanistic point of view is incorporated in the discussion of the results. Eichberg (2014) suggests that movement and health studies are placed between quantitative and qualitative methods and address the tension between the quest for evidence and the comparative method. The field of knowledge needs cross-disciplinary connections and overlaps of the qualitative and quantitative method to understand human life (Eichberg, 2014).The results from this pilot study can be used to understand patterns of an important psychological construct for behavior in CFS/ME patients and how it is related to their activity level. The results will now be discussed systematically according to the previous chapter with the aim to answer the research question; *"Can self-efficacy influence participation in physical activity for young CFS/ME patients?"* Further discussion will include strengths and limitations of the design and method, and if self-efficacy is an informative assessment tool in treatment for CFS/ME patients.

6.1 Descriptive statistic and Central tendency

There were more girls than boys in the sample group of 24 participants. This is not surprising as epidemiological studies show that CFS/ME is more common in females than in males (Knight et al., 2013). The inclusion of participants with a combination of adolescents and young adults can be problematic, since the participant under the age of 18 might have been diagnosed from the pediatric criteria (Jason et al., 2006), while the participants over the age of 18 might have been diagnosed with the Fukuda definition (Fukuda, 1994) or the CCC (Carruthers et al., 2003). This might have led to heterogeneity in the sample group. Heterogeneity in the sample group is a common issue in the research of CFS/ME and difficult to avoid (IOM, 2015; Bruberg et al., 2014; Kennedy (2004). There are currently about 20 sets of diagnostic criteria or case definitions and none of them are used as a gold standard (Bruberg, 2014).The participants in this sample group were not randomly selected because of ethical reasons and investigation in a real life situation where the participants had applied for treatment at the rehabilitation institution.

The central tendency of scores in SES, and minutes in upright activity and MVPA are given as both mean ± SD, and median (min– max). Median can in many situations give, a better description of the central tendency when data is not normally distributed (Thomas et al., 2005). SES data were normally distributed at TP2 and TP3, but not at TP1 (Shapiro-Wilk $P<0.02$). However the skewness and kurtosis were within +/-2 which are often required values for a normal distributed data (Hair et al., 1995). Most of the data were symmetrical, but a SES had a small negative skew, especially at TP1, with a long tail to the left. The PA data were more positive skewed with the tail going to the right, except upright activity at TP2 and TP3 which had small negative skew. Both upright activity and MVPA are used as variables to describe PA. According to the Shapiro-Wilk test of normality, PA was not normally distributed at any of the TPs in MVPA, and not in upright activity at TP1. Due to the small sample size and lack of normality in some of the TPs, nonparametric statistical tests were chosen. With nonparametric statistic, no assumptions are made about the distribution of the values. Nonparametric tests are criticized to be less powerful (ability to reject a H0 HYPOTHESISwhen it is false), but there are little agreement in the literature about this (Thomas et al., 2005). The values from the Shapiro-Wilk test, skewness and kurtosis of the data are listed in *Table 4.1*.

6.2 Self-efficacy

The main findings in self-efficacy were a nearly significant change at TP1 compared to TP2 (p = .063). Probably, this would have been significant with a larger sample size. There was a decreasing pattern in the SES score from TP2 to TP3, but this was not significant. The SES scores from all TPs are illustrated in *Figure 5.3*. The main pattern from the results indicates an increase from TP1 to TP2, and a decrease from TP2 to TP3. The increase from TP1 to TP2 was significant in question 8, 9, 10, 13 and 14, and the decrease from TP2 to TP3 was significant in question 10 and 14. The questions can be translated to;

I am sure that ...

- *Q8; ...I can perform planned exercise/activity goals that I have made*
- *Q9; ...I can be physically active although I have CFS/ME*
- *Q10; ...I can be physically active most days*
- *Q13; ...I can manage to bring my friends to be physically active most days*
- *Q14;... I can be more physically active*

The complete questionnaire is found in *Appendix 4*, in Norwegian (translation of the questionnaire is not performed). The increase in these questions can be explained as a stronger self-efficacy in capability to success in planned goals, and that CFS/ME as a barrier for being in PA is reduced. Question 13, might be associated to more social aspects which might have been affected trough socialization in the treatment groups. The participants also seem to get a stronger perceived capability to be physically active as reported from question 10 and 14. According to Bell and Bell (2010) upright activity is the best indicator of relative health for CFS/ME patients. If the participants in this sample group feel a stronger capability to be in PA this could be an indicator for their health and improvements after treatment.

The self-efficacy might have been affected by sense of capability, or as stated by (Bandura 1997) mastery experience. The other main ways to affect self-efficacy suggested by Bandura is probably also affecting the increase and effects from the treatment at CS (see Figure 2.1). The treatment at CS emphasize mastery experience, by encouraging PA at the patient's level, preferred sports/activities and teaches the participants how to cope with their illness in a best possible manner. When they manage to perform activities within their limits they might get a feeling of mastery which affects self-efficacy for being physically active. The three other ways to affect self-efficacy mentioned by Bandura might also have been emphasized through socialization in the group, support and care. However, self-efficacy was an unknown term for the specialist team before it was included as part of this thesis. The treatment the participants receive is not built to emphasize self-efficacy, but the coping strategies used at CS still seemed to affect the participant's self-efficacy for being physically active. If self-efficacy could be emphasized in the treatment as known term and its possible effects, it could be used to better understand the patients, and what they need of follow-up.

The improvements in self-efficacy seen at TP2 are not sustained at TP3. This indicates a decrease in self-efficacy for being physically active 12 weeks after the treatment. Hence, the effects in increased self-efficacy did not seem to be long-lasting effects. One exception is question 9, which still shows a positive change at TP3 compared to TP1. The participants still feel capable of being physically active despite their illness, also 12 weeks after receiving treatment. The decrease from TP2 to TP3 in self-efficacy for being physically active is only statistically significant in question 10 and 14 which cover perceived capability to be in PA in a more direct manner than the other questions. If the improvements were from the treatment only is unknown, to control for this bias a control group could be included in future research.

Even though there are some interesting patterns seen in the results, they do not cause a rejection of the H0 hypothesis and indicate no change in self-efficacy between the three TPs. Just out of curiosity, a paired samples t-test for normally distributed data was performed on the total scores comparing all the TPs. This test yields a significant positive change from TP1 to TP2 ($p = 0.04$). As mentioned the nonparametric tests are criticized to be less powerful. So the ability to reject H0 hypothesis when it is false is reduced in a nonparametric test (Thomas et al., 2005). The SES at TP1 was, however, not normally distributed. Hence the H0 hypothesis is retained. Because of the low power, the changes in SES should be understood as patterns and possible changes. Further investigations are needed to conclude the importance of self-efficacy in treatments and to possibly reject H0 hypothesis.

6.2 Physical activity

The main findings of objectively measured PA indicated no change of statistical difference in the mean time spent in either upright activity or MVPA between the TPs (*Table 5.3)*. Hence, the H0 hypothesis is retained. This indicates that there is no change in PA after treatment at CS. However, with a visual inspection of the central tendency, the pattern in upright activity seemed to be similar to the pattern seen in self-efficacy (*Table 5.1*). There is a small increase during the treatment at CS, and a small decrease 12 weeks after the participants received treatment. The mean time spent in upright activity was approximately 4 ½ hour at TP1, where 35 of those minutes were spent in MVPA. At TP2 the mean time spent in upright activity increased by 8 minutes. When using median, the increase from TP1 to TP2 was larger (25 minutes). This indicates that the participants tend to increase their time spent in upright activity during the treatment.12 weeks after received treatment the upright activity had a mean decreases by 33 minutes. The median shows a larger decrease by 52 minutes *(Table 5.2)*.A decrease in upright activity can be interpreted as failure in keeping the same amount of activity, which might have been developed as an activity baseline. However, the mean changes in both upright activity and MVPA are small with no statistical significance.

There was little change in upright activity between the days during the measurement period (8 days).The same activity pattern, with little change between days is also seen in the days chosen for analyses (*Figure 5.6*). An important consideration in the data from PA is the large range between the minimum and maximum time score, and large SD. Hence, investigation of individual changes from TP1 to TP2 was conducted. The Individual changes in minutes from TP1 to TP2 are shown in Figure 5.8. There were some participants who decreased upright activity, and some who increased upright activity during the treatment. Further analysis was performed and listed in Table 5.4. The changes from TP1 to TP2 were statistically significant in both groups ($p<0.005$). A possible explanation could be standardization of the treatment at CS. However, then the mean of the two groups would have been similar. The treatment at CS is individualized, and can also be shown in the large range of minimum and maximum score (Table 5.4). The mean patterns of upright activity for the two groups are illustrated in *Figure 5.9*. The group who decreased upright activity during the treatment seemed to maintain the same level, also 12 weeks after treatment, while the group who increased had a significant decrease 12 weeks after ($p = 0.05$). According to these results it is possible that the participants, who increased upright activity at CS, exceeded their limits. The activity level at CS might have been too high due to their individual limitations. Therefore, they were not capable of maintaining this level of upright activity when they returned home. Additionally, they had a higher activity level at CS compared to the other group. The reason for the decrease 12 weeks after rehabilitation is not known, and cannot be explained by current results. According to the literature, PEM is a central feature of CFS/ME and might occur immediately after a trigger, but also days after (IOM, 2015). The duration of PEM is also unpredictable and might last for a long period of time. One explanation of the decrease at TP3 for this group could be PEM. However, their activity level was approximately the same as they had at baseline (*Table 5.4*). When looking at the mean and the median they had a lower SES score than the ones who maintained their activity level at TP3. There were no significant changes in SES for neither of the groups.

Reduction in upright activity 12 weeks after received treatment could be interpreted as no improvements. If the participant failed to maintain the activity which was sat as baseline because of increased symptoms they need to adjust the activity plan for the ability of coping. Another explanation of reduction could be lack of motivation. Question 4 in the SES target perceived ability to be physically active without help from a health care therapist or similar, the mean score are relatively high at all TPs, and TP3 is slightly higher than at TP1 and TP2 (*Table 5.2*). Hence, lack of help from the therapist does not seem to be a barrier for being in PA. However, this is the mean score in question 4 from all participants. Further investigation could be conducted for each question in the SES for the participants who decreased in upright activity at TP3. There is a possibility that some of the questions may have a lower score according to barriers.

If the participants failed to follow the coping strategies they learned at CS. A closer follow up after the treatment might be necessary. There was also a pattern of a small decrease in SES score (not significant). Failure in coping strategies learned at CS might be indicated by a decrease in SES (failure instead of mastery experience might cause a lower self-efficacy in the behavior or task). According to the suggested model (*Figure 2.2)*, PA and CFS/ME can affect self efficacy, both negatively and positively in addition to the four main ways stated by Bandura (1997). It all affects each other but are probably most affected by mastery experience which is according to the self-efficacy theory the strongest source. If the participants fail in coping strategies, or to maintain their activity level on bad days their self-efficacy might decrease. If their self-efficacy decreases they are less likely to continue following the developed activity plan. However, these are just assumptions, further investigation is needed. Further, it might seem that closer follow-up after a 4-weeks treatment programme is necessary. There were also many participants who had little wear time of PL at TP3. Some were also missing whole days of recording. Reasons for this are not known. These participants were excluded from the analysis when using TP3, but maintained in TP1 and TP2 analysis.

Figure 5.7 represent the four participants who fulfilled wear time of PL at TP1 and TP2. Three of the participants approximately the same amount of upright activity at TP1 and TP2. Interestingly, the last participant had a decrease of 160 minutes from TP1 to TP2. This could be instrumentation bias, or unrepresentative score from TP1 or TP2. However, if the score is representative, it could have been that this participant had bad days, or was house-bound before attending CS. If looking at the self-efficacy score this participant increased also allot from TP1 to TP3. The patterns here are extremely interesting and could be important to analyze as part of a treatment programme. One could speculate allot from this data, so as an additional assessment tool such as activity log could have been informative. The type of activity or why the activity is performed (or not performed) is not given from objectively measured PA and is considered as a limitation. However, when adding an activity log one could get additionally information which can provide necessary information to understand the participants, and what kind of follow-up that is needed. Is it "just" lack of motivation, or is it because of the illness?

According to the analysis conducted when separating the sample into two groups the H0 hypothesis was rejected. However, according to present results there is not necessarily an increase of the PA level during the treatment at CS, decrease of the activity level were also present in this results (*Figure 5.8*). The exact reason occurrence of decrease or increase is not known, but could be coping strategies for their illness. Further investigation of this should be conducted but with the use of activity diary or other tools to discover the reason, a combination of qualitative techniques could also be beneficial. The participants who decreased their activity level during the treatment at CS managed to maintain the same activity level at TP3, this might be indication of improvements, and that they master coping strategies. However, the possibility of instrumentation bias could also have affected these results and should be considered. The fact that it might be subgroups in the sample or heterogeneity, causing differences in the severity degree of the illness is also common in this research field and should also be further investigated (IOM, 2015). For the objectively measured PA it would have been interesting to compare the PA level of the CFS/ME patients with a healthy peer group. The PL is not used in previous literature. In fact, no objectively measured PA is performed on CFS/ME patients. Hence, no comparisons can be made in this thesis. In future research, a control of both a healthy peer group and a peer group which is not receiving any treatment could be added to strengthen the design. So far self-efficacy and PA measurement have been discussed and compared with each other, but what is the statistical relationship between the variables?

6.3 Correlation

The main findings in the correlation analysis were a negative relationship between self-efficacy and upright activity at TP2 (see *table 5.5)*. This means that if the participant has a high self-efficacy the level of PA are low, and conversely, the lower self-efficacy the more physically active. This is contrary to most research on self-efficacy and PA which report a positive relationship (McAuley et al., 2013; Harris et al., 2008; Hankonen et al., 2010; Bowden, 2014). According to Bandura, strength of perceived self-efficacy is not necessarily linearly related to choice of behavior (Bandura, 1977), but the stronger the sense of self-efficacy, the greater perseverance and likelihood that the chosen activity will be performed successfully.

This negative correlation can be described from the results listed in *Table 5.4*. The participants, who had a relatively high level of upright activity at TP1 (5 hours), spent less time in upright activity at CS. In addition they maintained their activity level from TP2 to TP3. This negative relationship between the variables can indicate that they managed to change their activity level towards a more beneficial level due to their symptoms. Hence, mastery experience in PA level increased or maintained their self-efficacy score in the SES. This is just an assumption and further research is needed to either approve or disapprove the explanation. Another possible explanation is that the group with a higher activity level at TP1 knew they were able to be more PA active, but at CS they followed a restricted pattern of PA and had to reduce the level of PA. They have previous mastery experience in a higher level of PA, therefore a high score in SES despite a low level of PA.

Individuals with weak self- efficacy beliefs are easily discouraged by difficulties, whereas individuals with strong self-efficacy beliefs will preserve their efforts despite difficulties and obstacles (Bandura, 1977). The SES that was developed for current research was aimed towards the perceived ability to increase the level of PA. In the beginning of this research a part of the SES preparations, there was an expectation that CFS/ME patients in most cases were inactive because of their symptoms, and that the intention in most treatments was to carefully increase the activity level. In some circumstances this may be correct. However, the opposite might also be a way of coping with the illness. This pattern was seen for some of the participants in this sample group (*Figure 5.9*). It is all about finding a balance between preferred activity (hobby, social activities etc.), necessary activity (school attendance etc.) and rest. The patients need to find a level of PA they also can maintain on bad days (NICE, 2007; Helsedirektoratet 2014$_a$). Restrictions of some activities might also be beneficial for CFS/ME patients. A stronger relationship is seen between self-efficacy and MVPA at TP2 (*Table 5.5).* A possible explanation is that the patients who had a higher activity level (and higher intensity activities) at baseline, learned to better cope with their illness by reduction of activities which triggered PEM or other symptoms.

The hypothesis "*There is a positive correlation between perceived self-efficacy and PA*" is not supported. However, according to the findings there is a negative relationship between the variables causing H0 hypothesis to be rejected. This is commonly called negative results, when the results are in contrary to the hypothesis of interest. This brings me to the area of publication bias, or the "*Negativland*" stated by Laws (2013). Laws (2013) address this relevant issue in psychology. He states that negative findings are commonly downplayed or failed to be mentioned. Overuse of phrases such as no significant trends, or worse, reframe the results as positive are commonly seen in psychology. The term HARking has become common in the research. HARking is when the researchers build their hypothesis after the results are known, and when they are aligned to fit. Denial of the existence of negative results and the importance of replication are common whether we want or not and this trend need to be changed (Laws, 2013). This is probably not an issue only concerning psychology, but also in other areas of research. The research field of CFS/ME or any research field may be postponed for publication bias. Published studies might also be affected by outcome reporting bias, or analysis reporting bias. There are less likely that the researcher publishes his/hers results if they are in conflict regarding their hypothesis (Law, 2013). Publication bias in reviews commonly results in selective publication of those with positive findings. If this is common, there will be a major problem in the concern of searching for knowledge from the literature to develop evidence-based treatments for young CFS/ME patients. Therefore all findings should be reported, both negative and positive, as well as no findings at all.

There is an urgent need for more knowledge on the topic of CFS/ME. Both negative and positive findings should be presented to explain the knowledge base. In this thesis there are some results which are contrary to the literature. Self-efficacy seemed to have a negative relationship with PA, this is opposite of the hypothesis, but are still interesting results and important to report. As discussed above it might even be a result of improvement. When the participants reduced their activity level they managed to be active without trigging PEM, also on bad days. They experience mastery and thereby affecting self-efficacy for being physically active, which is according to the suggested model (*Figure 2.2)* and Bandura (1997). Mastery experience increases self-efficacy, even though the level of PA decreases to a lower level than at baseline illustrated in *Figure 5.9*. This group of participants which decreased their level of PA, did probably find a level possible to maintain. The mean time spent in upright activity was 252 minutes, which is about 4 hours. Since there is no control group of healthy individuals using PL, is not possible to say whether this is a low level or not for CFS/ME patients.

There is commonly a high focus on increasing the level of PA to improve health. Young CFS/ME patients might overdo their limits in the aim to improve. PEM is unpredictable and can vary from hours, days, weeks and months (Nijs et al., 2010) and the common characteristic is delayed ability to return to prior levels of physical capacity after exertion (IOM, 2015). Avoidance of activities which trigger PEM is essential for daily functioning. Too much sedentary behavior is not good for any individual's health (WHO, 2010) but too much of PA whether it is occupational or leisure time activity could be counterproductive. Coping strategies are all about finding a balance between activities, diet, sleep and rest, and to be aware of limitations. If 4 hours of upright activity is the participants limit it is much better to stay within this limit, rather than extend it and be bed-bound for a longer period of time.

The reason why some of the participants had days without wear time is unknown, one reason could be that they were bedbound/house bound and therefore felt it unnecessary to wear the PL. The importance of participants wearing the PL also during bad days should be highlighted in future research, in addition to better follow-up at TP3. At TP3 4 participants had to be excluded because of too many days of non-wear, the reason for this is not known. However, some of the participants might have lost their motivation to perform the given task. TP3 was the third time they performed the procedure, and the loss of "excitement" might have been a reason. In the other hand the Hawthorne effect might have been reduced at TP2 and TP3, compared to TP1. Concerning objectively measured PA an important bias to consider when analysis the results is the change in behavior as the participants know they are being observed (Wickstrom & Bendix, 2000). Instrumentation, or construct validity can as mentioned in the method chapter, also be a source of bias in present results. The further discussion will focus on strength and limitation in this thesis before the summary and conclusion are made.

6.3 Strengths and Limitations

The research field of CFS/ME is as mentioned a complicated area, especially research on young patients. Adequate knowledge is necessary for the possibility to offer safe and effective treatments for individuals which suffer from this illness. CS is one of few rehabilitation institutions specialized in the field of CFS/ME where PA is a main part of the coping strategies. The opportunity to be part of the pilot study to investigate patterns in self-efficacy for being physically active as well as the level of PA at 3 TPs is a major strength of this master thesis. The longitudinal design gave the opportunity to investigate the same participants over time and their changes in the outcome variables. It is discussed if it could be called a longitudinal design when the research last less than a year (Thomas et al., 2005). This thesis included data collection at baseline, during an intervention/treatment and 12 weeks after. Because of time restrictions in a master thesis, the follow-up after one year (*Figure 4.2*) was not possible to include in this thesis. However, if interested, the pilot study is an ongoing project where the one year follow-up will be conducted at a later TP for further investigation (*Appendix 6*).

Longitudinal design with more than one data collection may increase the possibility of dropouts (Thomas et al., 2005). In present pilot only one participant had to drop out because of psychological reasons. All other participants completed all measurements (to some degree) at all TPs. Hence, the feasibility of the design is a good way of investigating CFS/ME patients without a high rate of dropouts. Hovedenhove (2009) suggests that investigations the effectiveness in treatments for CFS/ME patients should be conducted in real life situations. This thesis supports this method of investigation on CFS/ME patients, as well as a combination of quantitative and qualitative research. This method is a safe, as well as informative way to investigate improvements from the treatment and how self-efficacy might work as an assessment tool. In some situations it is necessary to conduct quantitative and statistical significant results, but sometimes it might be interesting to just ask the patients or to investigate trends or patterns, even though they are not statistically significant. Both methods have their strengths, but they also have limitations which is important to consider when applying research into practice.

Another important strength in this thesis is that it covers a gap in the literature. Assessment and investigation of self-efficacy for being physically active are commonly used in both healthy individuals and clinical situations (McAuley et al., 2013; Kopershoek et al., 2011; Katch &Mead, 2010; Patterson et al., 2014), but as far as I know not in CFS/ME patients. The

PACE trial was also investigated by Chalder and colleagues (2015) in a mediator analysis and support a treatment model in which both beliefs and behavior play an important part. The PACE trial is conducted on adults, but the important mediators for improvements are likely to be similar in younger patients.

This thesis the first to include both objectively measured PA and SES in a longitudinal design for young CFS/ME patients. Normally, the ActiGraph has been used in the literature in several cases and populations. A limitation in ActiGraph is its ability to measure lower intensity (Calabró et al., 2014), and that pattern-recognition monitors perform better in lower intensity activities. To capture low intensity activities is essential when investigating CFS/ME patients. The PL was chosen for use in the pilot-study both because of its popularity on the market, and the ability to measure everyday activity, sleep pattern, steps. It is also water proof and easy to use. Evaluation of PL is also part of the pilot-study at CS, and they consider continuing using PL as a tool in future treatment for young CFS/ME patients. PL seem to be a valid measure of upright activity for CFS/ME patients, but in future research a validation study comparing PL against a goal standard should be performed to ensure instrumental validity.

The use of SES to assess self-efficacy seems to be effective and helpful concerning change of perceived capability for being physically active. A multidisciplinary 4-week of treatment which include GAT seem to improve perceived self-efficacy for young CFS/ME patients and may play an important role when creating a baseline for activity in treatment, as well an indication of improvement in daily functioning. If activity adaption and PA is beneficial for all patients cannot be concluded, but it seems to be beneficial for many patients which suffer from mild to moderate degree (Larun et al., 2015). According to results from Gordon and Lubitz, (2009) a 4-week graded exercise programme for adolescents with CFS/ME gave positive results in the participant's capability to resume a normal life-style, including sport and schooling. This thesis also show improvements after a 4-week treatment programme for young CFS/ME patients, but the improvements does not seem to be maintained. According to this thesis, GAT seems to improve perceived self-efficacy during the treatment period, but scores show a tendency to fall back to the same level they had before the treatment at CS when retesting after 12 weeks. As mentioned in the method, there will also be a follow-up one year after received treatment, which will give more important information of improvements. However, if self-efficacy decreases back to the original level already 12 weeks after received treatment, it might also be hypothesized that self-efficacy stays at the original level also after one year. Importantly, we learn from our mistakes, meaning, if they manage to use the activity plan and stay within their limitations they might achieve success. Or better, they might recover. The results from the TP3 however, indicate that closer follow-up is needed after a 4-weeks rehabilitation treatment.

I will further discuss some more strength and limitations in this design and measurements according the ones mentioned in the method.

6.3.1 Statistical validity and the sample group

Two basic aspects of statistical conclusions are validity-statistical power and ES (Thomas, et al 2005). A concrete estimation of the sample size needed in this thesis to avoid type I and type II error when doing hypothesis testing was not possible because of the lack of previous research. The estimation of power in this research showed low power. The size of the sample is extremely influential on power, and as the number of participants increase, the power increase. The sample size in this thesis was 25, one participant dropped out from the research because of psychological issues. In addition two were excluded from the PA analysis because of missing data at TP1. At TP3 four participants more were excluded from the data because of missing wear time of PL. The small sample size in this thesis leads to low power of the results. A sample size of 22 participants gives statistical power of 0.30 with a moderate ES (0.5), meaning a 30% chance of detecting real difference. This means that pilot study has low power and this should be considered when drawing conclusions. In a pilot study evaluation of the feasibility of design and method chosen to investigate the treatment outcomes and improvements of important variables is the main aim and not necessarily cause and effect (Thomas et al., 2005). Importantly, causation is not determined from any statistics or correlation, but rather by theory, logic and the total experimental situation.

Randomization to control threats to internal validity is difficult in a “real life” setting (Thomas et al. 2005). To ask the participants to receive another treatment than they applied for or no treatment at all is not possible due to ethical considerations. The process of gathering enough participants for this kind of design and method is not a simple manner. Therefore a control condition was not applied in the current design. Further investigation on self-efficacy and PA level should be conducted with a larger sample size for the ability to increase validity-statistical power. A design which allows randomization and control condition should also be considered.

6.3.2 Assessment of Self-efficacy

Strength incorporates efficacy level and is a sensitive and informative measurement of self-efficacy (Bandura, 2006). PA was explained as all leisure- and occupational activities with examples to assure that the participants understood the term PA. Efficacy beliefs influence whether people think optimistically or pessimistically about a specific behavior. Present results show that young patients with CFS/ME have more optimistic thoughts about being physically active after the rehabilitation stay. However, twelve weeks after the rehabilitation treatment the optimistic thoughts seemed to decrease.

Questionnaires are practical and easy to administer, however some biases may occur which threaten validity and reliability. Under or over reporting might be a bias in this sample group. However, the questions are about how the person's perceived capability is for being physically active. Self-efficacy is individual, and the answers are probably not more socially correct in any direction. The definition of PA was carefully explained including examples to avoid misunderstandings. Importantly, PA is a broad term (Caspersen, Powell & Christenson, 1985) and can be understood differently among the participants .The questions in SES are collected from the ESES which is validated (Kroll et al., 2007) and suggested to be a reliable assessment tool. However, small changes were done in order to adjust the scale to context of CFS/ME, this might have influenced both reliability and validity of the scale. Conbach's alpha was used to test the internal consistency of the scale (see table 5.1). The alpha was above 0.80 at all TPs which is regarded as good, and 0.90 which is regarded as excellent. When looking at the" item total statistics" which shows if removal of any items would not give a higher Conbach's alpha, it did not seem that removal of any of the questions would give a higher internal consistency. Although the SES seems to be a valid and informative assessment tool, as mentioned above, some adjustments should be added to more precisely target the coping strategies used in treatment and not only capability to increase PA or ability to be in PA.

A factor that may influence response to a questionnaire is the length and complexity. Participants are more likely to complete a short simple questionnaire rather than a long complex one which takes longer time to complete (Thomas et al., 2005). This is considered as strength in this thesis where only 14 items were used. 14 questions seemed to be acceptable and did not cause any impact on the participants, the SES was completed by all 24 participants at all 3 TPs.

6.3.3 Assessment of Physical activity

Inactivity can be defined as sitting or lying down behavior which results in energy expenditure less than 1.5 MET. CFS/ME patients are probably spending more time in inactivity than healthy individuals due to their symptoms. Upright activity is used in prior research from Bell and Bell (2009) aiming to address recovery. When measuring upright activity or PA at baseline, change can be an indication of recovery. Upright activity *(Low, medium* and *high intensity PA)* was in this thesis presented as PA measured during a day, this seemed to be a more accurate outcome variable rather than only MVPA or low intensity activities.

The outcome variable called upright activity in this thesis seemed to be a reasonable and good way to measure PA in CFS/ME patients rather than only MVPA, which is commonly used in PA research (WHO, 2010; Kolle et al, 2010). All activity which is performed in an upright position is included as PA and the remaining time during a day is probably inactivity. CFS/ME patients are generally not capable of being in high intensity activities without worsening of symptoms, therefore the "low" intensity category was also included. Upright activity is suggested to be the best indicator or relative health (Bell & Bell, 2010) and seems to be the most reliable indicator of prognosis and recovery. When a patients is getting better it also means that they are capable of doing activities they could not do before. *Hours of Activity scale* was measured by Bell and Bell (2010). It ask the subject to estimate average total number of hours of upright activity within an average 24 hour day.Healthy subjects seems to be more than 12 hours of upright activity in a day, severe CFS/ME patients experience 2 or less hours per day, while moderate CFS/ME experience 3-7 hours and mild 8-10 hours (Bell & Bell, 2010) The mean hours spent in PA, or upright activity from the participants in this sample group was about 4 ½ hour. Importantly, this is not comparable because of the above results is from a survey that used self-report measurement scale, while this thesis used objectively measured PA.

MVPA is as mentioned, a commonly used in the literature. In a nationally representative cohort of children and adolescents, Kolle and colleagues (2010) demonstrated with objectively measured PA that the mean time in MVPA for 15-year-olds girls was 62 minutes, and 68 minutes for 15-year-olds boys, the MVPA was higher in 9 year olds. WHO (2010) guidelines for PA recommend 60 minutes in MVPA for children and adolescents. For the possibility to compare results MVPA was also measured in this thesis. However, a limitation in this thesis is the use of relatively new activity tracker, which is not validated in the literature. This may be a source of measurement error causing bias in the results. Another concern is the method used by Polar to translate counts/minutes, into minutes in the different activity intensities. Counts/ minutes are also commonly presented in the literature (Kolle et al, 2010) and give the possibility to compare results if cut off points are known. Commonly, epoch length is shorter when measuring children and adolescents (10, 30 or 60 seconds) and MVPA are sat to all activity > 2000 counts/minute, which represent >3 METs and a walking pace >4 km/hour (Kolle et al., 2010; Ekelund et al., 2004; Andersen et al., 2006). For PL this cut off points are not known .Virtanen (2014) explain the method used for calculation, but not in detail, which make it difficult to compare these results with other objectively measured PA studies where ActiGraph or other activity trackers are used.

The choice of using PL to measure PA was taken because it is easy to use both for the participant and the research leader who was not familiar with the use of ActiGraph or other accelerometers commonly used in the literature. PL is used as a watch and can be used also when sleeping (comfortable to use) and it is waterproof. PL does not cause any physical or psychological impact for the participants, which was a concern when choosing an activity tracker in this sample group. Importantly, the PL was pilot tested by the author of this thesis before use, and seems to measure PA with only 2 percent difference from observed activity. PL provides information about the activity pattern in young CFS/ME patients, and is easy to use. An issue is short battery time (5 days), which might cause underestimation of the activity level. The charging of PL takes only some minutes, but the participants might forget to take it on immediately after completed charging. That the participants might take it off, for any reason is also a limitation that is difficult to control. Investigation of wear time was conducted before analyzing the data *(Appendix 7)*. The mean wear time at TP1 were 23 (± 2.20) hours (*Table 5.1)*. However, at TP3 there might be necessary to highlight the importance of wear time. Another concern is that at some days some of the participants had more than 24 hours. This might be a limitation of PL. Hence, validation studies are necessary. Another possible explanation could be mistakes performed when plotting the data. The data was given as SPSS file, mistakes might have occurred when plotting data given from PL.

Future studies should consider an objective method which can give more precise information rather than questionnaires or PA diaries only. However, an activity tracker does not give any information considering type or purpose of the activity. This might be important information therefore activity diaries could be used in addition. Participants in this sample group filled out activity diaries but this is not used in this thesis. Combination of the two methods should be used for further investigation.

In addition to the concerns of the reliability and validity of PL there was also necessary to consider what method that is preferable in the population of interest and the number of days required to estimate habitual PA. The number of monitoring days required for children and adolescents ranges from 4 to 9 days (Trost, Mclver & Pate 2005). In the current study 8 days of monitoring was performed with PL. However, the first day and last day was excluded because of a non wear data was higher in these two days. This was probably because there was not specified a time when to start using PL the first day *(Appendix 5)*. They were only asked to use PL consecutive for one week. In addition some may have taken it off before the measurement period was finished. Therefore to avoid measurement error from the first and last day they were not included in the analysis. When investigating wear time of PL *(Appendix 7)* there were some days without wear time (0 minutes). To avoid excluding participants from an already low sample size, the analysis included 4 days (3 weekdays and Saturday) of recording, instead of 6 days. The exclusion criteria in wear time was low (<4 hours) to keep as many participants as possible. This should be considered, for future research and highlighted in the information sheet for the participants. Previous studies have shown that adolescents were more physically active during weekdays than weekends (Kolle, Steene-Johannessen, Andersen & Andresen, 2010). In this thesis there were minimal changes (*Figure 5.6*). Therefore, none of the analyses separating weekdays from the days in the weekend are shown.

6.3.4 Pilot study

The purpose of conducting a pilot-study is to examine the feasibility of an approach or intervention that is intended to use in a larger study (Leon, Davis & Kraemer, 2011). The lack of good scientific methods in investigation of CFS/ME patients is a remaining problem (IOM, 2015). Therefore CS decided to conduct a pilot-study to be able to later perform a larger study in the best manner. Pilot-studies represent a fundamental phase of the research process and are important work to reduce methodological faults and lack of validity. Pilot-studies are used to evaluate recruitment, randomization, measurements, new methods and implementation of the intervention. This was exactly the purpose of the pilot project at CS, whereas evaluation of research method, intervention and measurements is a big part of the investigation. Since this thesis collected data from the pilot-study and the research field of CFS/ME has many challenges and little agreement in preferred inclusion criteria's, method, treatment etc. The statistical powers in these studies are typically low. Importantly, this thesis should not be used to evaluate significant changes, but rather to find interesting patterns in self-efficacy and PA before and after the treatment intervention. Self-efficacy is not proved to be important for young CFS/ME patients according to these results but there seem to be a change which should be investigated more deeply.

6.3.5 Selection bias and Generalization

Selection bias can be a threat to both internal and external validity (Campbell & Stanley, 1963). Participants in this sample group were not randomly selected. This was because the data collection was done in a real life setting and on a vulnerable population group. Participants in this sample group had to go through an application process before receiving rehabilitation at CS, regardless of their participation in the pilot study. Randomization or randomly selecting participants within the present design would have been unethical.

External validity refers to the generalization of results and may be a limitation in this thesis. CFS/ME patients are a specific population. Selection bias occurs when participants that is included in the research differ in a meaningful way from those who not (Thomas et al., 2005). The participants in current study is of mild to moderate severity of CFS/ME and may differ allot from those who have a more severe degree of this illness. Diagnose criteria might also be an issue concerning generalization. Which diagnostic criteria that are used are not known in this thesis and is a limitation for generalization and validity of the results. This is information which is protected for ethical reasons and was not included in the application procedure to NSD. This is important information both for generalization and for ability to compare the results with other research findings. Hence, this should be included in future research. However, the Jason (2006) pediatric criteria are recommended for use in Norway (Helsedirektoratet 2014_a) and are the most common for diagnosing children and adolescents with CFS/ME. Importantly, some of the patients were older and might have been diagnosed by the Fukuda definition (Fukuda, 1994) or CCC (Carruthers et al., 2003) which also is recommended for use in Norway (Helsedirektoratet 2014_a). Importantly, the results observed in this thesis may not necessarily be generalizable to those with severe or very severe degree of CFS/ME. Use of different diagnostic criteria also makes it difficult to generalize results from this sample group to other CFS/ME patients.

6.4 Summary

CFS/ME is a research field which needs more attention and more adequate knowledge to understand the complexity of this illness. Current literature is in many situations confusing with little consensus, causing misunderstandings of the illness. There exist over 20 sets of diagnostic criteria's or case definitions, and none serve as a gold standard (Bruberg et al., 2014). This cause heterogeneity in the patient group and highlight the importance to report degree of severity. Heterogeneity in patients and the individual symptoms are problematic when doing research, causing variation in effects and conclusions. CFS/ME and following symptoms such as PEM, has a major impact on young patients daily functioning and could impede them to have a "normal" life-style, including educational and social activities. Daily PA is important for young patients who are in a critical period of physical and psychological development. However, many CFS/ME patients experience to be bed- or house-bound some period of their illness, and in severe cases, might last for longer periods (IOM, 2015). Research has also found that some patients, which might improve from their illness, remain symptomatic and do not return to premorbid level of functioning (Jason et al 2013).

The possibility to resume schooling, socialization, sports, and other activities are essential for QOL (Kennedy et al., 2010; Schweitzer et al., 1995). Treatments such as GET and CBT seem to be beneficial for improvements and coping strategies in adults (Chalder et al., 2015). However, little evidence exists for efficiency of treatments in younger CFS/ME patients. There is also a lack in the literature of research covering self-efficacy in the domain of PA as an important psychological determinant for behavior and coping strategies. Self-efficacy as an assessment tool could improve individual treatment strategies, in the aim to better understand each individual perceived capability for being physically active. The strength of self-efficacy for being physically active might also influence actual behavior. The time spent in actually doing something can be measured as upright activity suggested by Bell and Bell (2010), and could be a reliable indicator of prognosis and recovery.

This thesis is the first to include both objectively measured PA and self-efficacy in a longitudinal design for young CFS/ME patients. Data is collected from a pilot study conducted at CS (a rehabilitation institution, in Norway). Investigation in a real life setting with a specialist treatment team provided a safe and informative method for investigation of self-efficacy and PA in a vulnerable patient group. SES was prepared in the aim to target perceived capability for being physically active in young CFS/ME patients. In order to investigate if self-efficacy could influence participation in PA, PL was used to measure upright activity and MVPA. The data was collected at three TPs; before, during and 12 weeks after the participants received a 4-week multidisciplinary treatment, tailor made for young CFS/ME patients. The treatment included GAT, and individual activity plans which were developed as part of the coping strategies to resume a balance between activity, rest and a normal sleep pattern.

Self-efficacy for being physically active seemed to have a positive change during the treatment, but did not seem to be long lasting, however further follow-up will be investigated at TP4 (one year after received treatment). Because of the data were not normally distributed, both mean and median were used to describe the central tendency, and nonparametric statistical analysis were used. Time spent in upright activity did not seem to change between the TPs. This could be because of heterogeneity in the patient group. Further investigation was conducted where the group was divided into two subgroups. Those who decreased and those who increased in upright activity during treatment were analyzed separately. Results showed that participants who increased activity at CS, had a decrease 12 weeks after treatment ($p<0.05$). The participants who decreased their activity at CS maintained their activity level 12 weeks after, with no change ($p= 0.42$).This indicates that some of the participants needed to reduce activity level for the capability to maintain their baseline of PA without worsening of symptoms or relapse. A balance between rest, schooling, social activities and a normal sleep pattern are essential in the treatment at CS. To find a baseline of activity which is possible to maintain also on days they feel particularly bad is important to avoid relapse.

In the correlation analyses there was a significant negative relationship between PA and SES during the treatment. This is in contrary to most research on self-efficacy form being physically active (McAuley et al., 2013; Harris et al., 2008; Hankonen et al., 2010; Bowden, 2014). A possible explanation could be that some of the participants reduced their upright activity and experienced mastery in the aim of coping with their illness, and to avoid PEM or relapse. Hence, a stronger self-efficacy for being physically active was correlated with a reduced PA level. This could be an indication of improvement for these patients.

Importantly, this thesis is a pilot with low statistical power. Statistically significances are reported in some of the analysis, but because of low statistical power and a high possibility for type I error (rejecting H0 hypothesis when it is true), this results should be used with caution. However, the result should also highlight the role of self-efficacy in treatments for CFS/ME patients, as well as the importance of individualization in coping strategies, especially in PA. Causation is not determined from any statistics or correlation, but rather by theory, logic and the total experimental situation. When applying research into practice a more humanistic point of view is also necessary. CFS/ME is as mentioned a complex and poorly understood illness, there is therefore necessary to understand and respond adequately to patients suffering from this illness, and to give them a sense of being understood. Evidence-based treatments are important, but practical experience is essential.

6.4.2 Conclusion

This thesis has provided interesting results in the field CFS/ME. SES has been used to measure self-efficacy for being physically active. SES can be informative for development of individualized coping strategies, as well as for behavior. Upright activity can be used as an indication of improvements, or provide information about the amount of time spent in all kinds of PA in an upright position. Feasibility of the design and method are considered as good, giving opportunity to investigate changes self-efficacy and PA over time in a safe environment. However, some adjustment should be added to the SES to better target the actual coping strategies for each individual, rather than only capability to increase PA. Validation of PL is needed to control for construct validity and instrumentation bias. In addition, a larger sample size and estimation of ES are needed to increase statistical power.

To answer the research question; *"Can self-efficacy influence participation in physical activity for young CFS/ME patients?"* probably yes, but more research is needed.

7. References

Agency for Healthcare Research and Quality [AHRQ]. (2014). *Diagnosis and Treatment of Myalgic Encephalomyelitis/Chronic Fatigue Syndrome*. AHRQ Publication No. 15-E001-EF. Available from: http://effectivehealthcare.ahrq.gov/ehc/products/586/2004/chronic-fatigue-report-141209.pdf

Andersen L.B., Schnohr P., Schroll., & Hein H.O. (2000). *All-cause mortality associated with physical activity during leisure time, work, sports and cycling to work*. Arch Intern Med. 2000 Jun 12;160(11):1621-8.

Andersen L.B, Harro M., Sardinha L.B., Froberg K., Ekelund U., Brage S., Anderssen S.A. (2006).*Physical activity and clustered cardiovascular risk in children: a cross-sectional study* (The European Youth Heart Study). Lancet 368(9532): 299–304.

Bandura A. (1977). *Self-efficacy: Towards a unifying theory of behavioral change*. Stanford University; Physiological review 1977. Vol. 84, No 2, 191-215.

Bandura A. (1986). *Social foundations of thought and action: A social cognitive theory*. Englewood Cliffs, NJ: Prentice- Hall, Inc

Bandura A (1989). *Human agency in social cognitive theory*. American Psychologist, 44(9), 1175-1184.

Bandura A. (1997). *Self-Efficacy, The exercise of control*. New York: W.H. Freeman and Company.

Bandura A. (2006) *Guide for constructing self-efficacy scales*. In Pajares F. & Urdan T (Eds.) *Self-efficacy beliefs of adolescents* (pp.307-337). Information Age Publising. USA

Bell D. S. & Bell D.E (2010). *Definition of recovery in Chronic Fatigue Syndrome*. Journal of LiMe. 2010;4:23-27. Available from: http://www.investinme.org/Documents/Journals/Journal%20of%20IiME%20Vol%204%20Issue%201.pdf

Bell D. S. 1995. *Chronic fatigue syndrome in children and adolescents: A review*. *Focus & Opinion Pediatrics* 1(5):412-420.

Beverly D.W (2005). *Chronic fatigue syndrome in children*. Current Paediatrics vol.15, 246-252. Available from http://ac.els-cdn.com/S0957583905000102/1-s2.0-S0957583905000102-main.pdf?_tid=10cbeedc-4965-11e4-8185-00000aacb35e&acdnat=1412166095_2571d3be89bb00895c42ceeca74906be

Black C. D., & McCully K.K. (2005). *Time course of exercise induced alterations in daily activity in chronic fatigue syndrome*. Dynamic Medicine 4(10).

Borrell-Carriò F., Suchman A.L & Epstein R.M. (2004). *The Biopsycosocial Model 25 Years Later:Principles, Practice, and Scientific Inquiry*. Ann Fam Med. Nov;2(6): 576-582

Bringsli G.J., Gilje A. & Getz B.K. (2014). *The Norwegian ME Association National Survey: Abridged English verson*. Oslo, Norway: The Norwegian ME Association.

Brugniaux J.V., Nieva A., Pulkkinen I., Laukkanen R.M., Richalet J.P. & Pichon A.P. (2010). Polar Activity Watch 200: a new device to accurately assess energy expenditure. Br J Sports Med;44(4):245-9.

Brurberg, K. G., Fønhus M.S., Larun L., Flottorp S. & Malterud K. (2014). *Case definitions for chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME): A systematic review*. BMJ Open 4(2):1-12

Calabró M.A., Lee J.M, Saint-Maurice P.F., Yoo H. & Welk G.J. (2014). *Validity of physical activity monitors for assessing lower intensity activity in adults*.Int J Behav Nut Phys Act: Sep 28;11(1):119.

Campbell D.T., & Stanly J.C. (1963). *Experimental and quasi-experimental designs for research*. Chicago: Rand McNally.

Carruthers B.M., Jain A.K., De Meirleir K.L., Peterson D.L, Klimas N.G., Lerner A.M … van de Sande M.I (2003) *Myalgic encephalomyelitis / chronic fatigue syndrome: clinical working case definition, diagnostic and treatment protocols*. J. Chronic Fatigue Syndr. Vol 11, No 1 p. 7-115.

Carruthers, B.M., van de Sande, M.I., de Meirleir, K.L., Klimas, N.G., Broderick, G., Mitchell, T … Stevens, M.L. (2011). *Myalgic encephalomyelitis: international consensus criteria. Journal of Internal Medicine 270* (4), 327–338

Carter B.D., Edwards J.F., Kronenberger W.G., Michalczyk L. & Marshall G.S. (1995) *Case control study of chronic fatigue in pediatric patients*. Pediatrics. 1995 Feb;95(2):179-86.

Caspersen C.J., Powell K.E. & Christenson G.M. (1985). *Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research*. Public Health Rep. 1985 Mar-Apr; 100 (2): 126-31.

Center for Disease Control and Prevention [CDC] (2013). *National Notifiable Diseases Surveillance System* (NNDSS): Case definitions.

Chalder T., Goldsmith K.A., White P.D., Sharpe M., Pickles A.R. (2015) *Rehabilitative therapies for chronic fatigue syndrome: a secondary mediation analysis of the PACE trial*. Lancet Psychiatry 2015; 2: 141–52

Chia J.K.S (2005). *The role of enterovirus in chronic fatigue syndrome*. Endocrinol Metab 15;55-59

Cleare A.J (2003). *The neuroendocrinology of chronic fatigue syndrome*. Endocrine Rev 24: 236-252.

Coggon, D., C. Martyn, K. T. Palmer, & B. Evanoff (2005). *Assessing case definitions in the absence of a diagnostic gold standard*. International Journal of Epidemiology 34(4):949-952.

Cohen J. (1969). *Statistical power analysis for the behavioral sciences*. New York: Academic Press

Cohen J (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). New York: Academic Press

Cook T.D., & Campbell D.T. (1979). Quasi-experimentation: Design & analysis issues for field settings. Boston: Houghton Miffilin Company

Crawley E. & Sterne J.A. (2009). *Association between school absence and physical function in paediatric chronic fatigue syndrome/myalgic encephalopathy*. Arch Dis Child. 2009 Oct;94 (10):752-6.

Davies S. & Crawley E (2008). *Chronic fatigue syndrome in children aged 11 years and younger*. Arch Dis Child. 93(5):419-21.

Deale A., Husain K., Chadler T. & Wessely S. (2001). *Long-term outcome of cognitive behaviour theraphy versus relaxation therapy for chronic fatigue syndrome: a 5-year follow-up study*. Am J Psychiatry. Dec;158(12):2038-42

Dowsett E.G. & Colby J. (1997). *Chronic fatigue syndrome in children. Journal was wrong to critizise study in schoolchildren*. BMJ 1997 Oct 11;315(7113):949.

Edmonds M., McGuire H. & Price J.R. (2004) *Exercise therapy for chronic fatigue syndrome*. Cochrane Database of Systematic Reviews, Issue 3. Art. No.: CD003200. DOI: 10.1002/14651858.CD003200.pub2.

Eichberg H (2014). *Explanation or Understanding? Movement Studies Between Natural Sciences and Cultural Studies*. University of Southern Denmark, Denmark. |In Physical Culture and Sport. Studies and Research. Volume 63, Issue 1, Pages 5–21

Ekelund U., Sjöström M., Yngve A., Poortvliet E., Nilsson A., Froberg K., Wedderkopp N & Westerterp K.R. (2001). *Physical activity assessed by activity monitor and doubly labeled water in children*. Med Sci Sports Exerc; 33:275-81.

Ekelund U., Sardinha L.B, Anderssen S.A, Harro M., Franks P.W, Brage S., … & Froberg K. (2004). *Associations between objectively assessed physical activity and indicators of body fatness in 9- to 10-y-old European children: a population-based study from 4 distinct regions in Europe (the EuropeanYouth Heart Study)*. Am J Clin Nutr: 80(3): 584–590.

Findley J.C., Kerns R., Weinberg L.D. & Rosenberg R. (1998). *Self-Efficacy as a psychological moderator of chronic fatigue syndrome*. Journal of behavioral medicine Vol. 21, No 4, 1998.

Fluge O, Bruland O, Risa K, Storstein A, Kristoffersen EK, Sapkota D, Næss H, Dahl O, Nyland H, Mella O (2011). *Benefit from B-lymphocyte depletion using the anti-CD20 antibody rituximab in chronic fatigue syndrome. A double-blind and placebo-controlled study. PLoS One:6:e26358*

Fukuda K., Straus S.E, Hickie I., Sharpe M.C., Dobbins J.G., & Komaroff A. *The chronic fatigue syndrome: a comprehensive approach to its definition and study*. Annals of Internal Medicine 1994; 121: 953-959. Available at: http://www.ncf-net.org/patents/pdf/Fukuda_Definition.pdf

Clare Francis, Action for M.E (2014) *M.E. Time to deliver – initial findings of action for M.E.'s 20014 survey* Action for M.E. Available at: http://www.actionforme.org.uk/Resources/Action%20for%20ME/Documents/get-involved/time-to-deliver-updated.pdf

Hair J.E., Anderson R.E., Tatham R.L., & Black W.C (1995). *Multivariate data analysis with readings*.Englecwood Cliffs New Jersey: Prentice-Hall

Hall K.S., Crowley G.M., McConnel E.S., Bosworth H.B., Sloane R., Ekelund C.C & Moerey M.C. (2010). *Change in goal ratings as a mediating variable between self-efficacy and physical activity*. Ann Behav Med ; 39-(3):267-73.

Hankonen N., Absetz P, Ghisletta P., Renner B. & Uutela A (2010). *Gender differences in social cognitive determinants of exercise adoption*. Psychol Health 25;(1):55-69.

Harris D.J & Atkinson G. (2009). *International journal of sport Medicine - Ethical Standards in Sport and* Exercise Sciense Research. Int J Sports Medicine 30: 701-702

Harris T.J., *Owen* C.G., *Victor* C.R., *Adams* R. & *Cook* D.G. *(2008*). *What factors are associated with physical activity in older people, assessed objectively by accelerometry?* Br J Sports Med. *2009* Jun;43(6):442-50. doi: 10.1136/bjsm.2008.048033.

Helsedirektoratet (2009) *Fysisk aktivitet blant voksne og eldre i Norge. Resultater fra en kartlegging i 2008 og 2009*. Helsedirektorated; Oslo

Helsedirektoratet (2014$_b$). *Kunnskapsgrunnlag fysisk aktivitet – innspill til departementets videre arbeid for økt fysisk aktivitet og redusert inaktivitet i befolkningen*. Utgitt 02/2014 IS-2167. Available from: http://helsedirektoratet.no/publikasjoner/kunnskapsgrunnlag-fysisk-aktivitet/Publikasjoner/IS-2167.pdf

Helsedirektoratet [Norwegian Directorate of Health] (2014$_a$). *Nasjonal veileder – pasienter med CFS/ME – Utredning diagnostistikk, behandling, pleie og omsorg*. Utgitt 02/2014, IS-1944.

Hildebrand M., Van Hees V.T., Hansen B.H. & Ekelund U. (2014). Age-Group *Comparability of Raw Accelerometer Output from Wrist- and Hip-Worn Monitors. Published on Feb. 5, 2014*

Houdenhove B.V (2009). *Treatment of chronic fatigue syndrome:How to find a "new equilibrium?* Patient education and Counseling. Vol 77, issu 2. pp. 153-154.

Institute of Medicine [IOM] (2015). Beyond Myalgic Encephalomyelitis/Chronic Fatigue Syndrome: Redefining an Illness. Washington, DC: The National Academies Press.

Jason L.A., Barker K. & Brown A. (2013). *Pediatric myalgic Encephalomyelitis/Chronic Fatigue Syndrome*. Rev Health Care; 3 (4): 257-270

Jason L.A., Bell D.S., Rowe K., Van Hoof E.L.S., Jordan K., Lapp C., … De Meirleir K (2006) *A pediatric case definition for myalgic encephalomyelitis and chronic fatigue syndrome*. *J Chronic Fatigue Syndr; 13:1e44*.

Jason L.A., Porter N, Shelleby E, Till L., Bell D.S., Lapp C.W., … De Meireleir K (2010). *Severe versus Moderate criteria for the new pediatric case definition for ME/CFS*. Child Psychiatry Hum Dev; 40(4):609-20. doi: 10.1007/s10578-009-0147-8

Jason L.A., Porter N. & Rademaker A (2011). *Epidemiologic approaches to community-based research*. In: Jason L.A., Glenwick Dc., Editors Methodological approaches to community-based research. Washington, DC: American Psychological Association

Jason L.A., Richman J.A., Rademaker A.W., Jordan K.M., Piloplys A.V., Taylor R.R., … Piloplys S. (1999). *A community-based study of chronic fatigue syndrome*. Arch Intern Med; 159 (18): 2129-37.

Katch H. & Mead H (2010). *The role of self-efficacy in cardiovascular disease self-management: a review of effective programs*. Patient Intelligence, 2:33-44

Keller B. A., Pryor J.L., & Giloteaux L. (2014). *Inability of myalgic encephalomyelitis/ chronic fatigue syndrome patients to reproduce VO2 peak indicates functional impairment*. J Transl Med; Apr 23;12:104.

Kennedy G., Abbot N.C., Spence V., Underwood C., & Belch J.J. (2004). *The specificity of the CDC-1994 criteria for chronic fatigue syndrome: comparison of health status in three groups of patients who fulfill the criteria*. *Annals of epinemiology 14(2): 95-100*

Kennedy G., Underwood C. & Belch J.J.F (2010). *Physical an Functional Impact of Chronic Fatigue Syndrome/Myalgic Encephalomyelitis in Childhood*. Pediatrics 125 (6): e1324-e1330

Kinnunen H., Tanskanen M., Kyröläinen H., Westerterp K.R. (2012). *Wrist-worn accelerometers in assessment of energy expenditure during intensive training*. Physiol Meas;33(11):1841-54. doi: 10.1088/0967-3334/33/11/1841.

Kline R.B. (2005). *Principles and practices of structural equation modeling*. 2nd edition. New York: The Gulford Press.

Knight S.J., Scheinberg A. & Harvey A.R. (2013). *Interventions in pediatric chronic fatigue syndrome/myalgic encephalomyelitis: a systematic review*.J Adolesc Healt. 53 (2): 154-65

Kolle, Steene-Johannessen, Andersen & Andersen (2010). *Objectively assessed physical activity and aerobic fitness in a population-based sample of Norwegian 9- and 15-year-olds*. Scand J Med Sci Sports 20: e41-e47.

Kopershoek C., van der Bijl J. & Hafsteinsdóttir T.B. (2011). *Self-efficacy and its influence on recovery of patients with stroke: a systematic review*. J Adv Nurs. Sep;67(9); 1876-94

Kroll T., Kehn M., Ho P-S & Groah S. (2007). *The SCI Exercise Self-Efficacy Scale (ESES): development and psychometric properties*. Int J Behav Nut Phys Act 2007; 4: 34.

Larun L., Burberg K.G., Odgaard-Jensen J. & Price J.R (2015). *Exercise theraphy for chronic fatigue syndrome*.Cochrane Database Syst Rev. 10;2:CD003200

Laws R.K., (2013). *Negativland – a home for all findings in Psycology*.Laws BMC Psycology 2013, 1:2.

Lee J-M., Kim Y & Welk G.J (2014) *Validity of Consumer-Based Physical Activity Monitors*. Med. Sci. Sports Exerc., Vol.46,No.9, pp. 1840–1848, 2014

Leon A.C., Davis L.L. & Kraemer H.C (2011). *The role of Pilot Studies in Clinical Research*. J Psychiatr Res. May: 45(5): 626-629.

Liao S-Y., Benzo R., Ries A.L. & Soler, X. (2014). Physical *Activity Monitoring in Patients with Chronic Obstructive Pulmonary Disease*. Journal of the COPD Foundation. Vol 1. Nr 2

Maier S.F., Watkins L.R. & Fleshner M. (1994) *Psychoneuroimmunology: The interface between behavior, brain, and immunity*.Am Psychol, 49 (12) pp. 1004–1017

Mattocks C., Ness A., Leary S., Tilling K., Blair S.N., Shield J. … Riddoch C. (2008) Use *of accelerometers in a large field based study of children: protocols, design issues and effects on precision*. J Phys Act Health;5 Suppl 1:S98-111.

McAlister, A., Perry, C.L. & Parcel, G.S. (2008). *How individuals, environment and health behaviors interact: Social cognitive theory*. In: K. Glanz, B. K. Rimer & K. Viswanath (Eds)), Health behavior and health education: theory research and practice (4th ed.). (p.169-188). San Francisco: Josey-Bass.

McAuley E., Morris K.S., Doerksen S.E., Motl R.W., Liang H., White S.M., Wójcicki T.R. & Rosengren K (2007). *Effects of change in physical activity on physical function limitations in older women: mediating roles of physical function performance and self-efficacy*. J Am Geriatr Soc.;55(12):1967-73.

McAuley, E. (1992). Self-referent thought in sport and physical activity. In T. S. Horn (Ed.), Advances in Sport Psychology, (pp. 101-118). Champaign, IL: Human Kinetics

McAuley, E., Mailey, E. L., Szabo, A. N., & Gothe, N. (2013). *Physical activity and the personal agency: Self-efficacy as a determinant, consequence, and mediator*. | In Martin Ginis, K. A. (Ed.), *Handbook of Physical Activity and Mental Health*. London: Routledge

McAuley. E. (1993). *Self-efficacy and the maintenance of exercise participation in older adults, journal of behavioural medesine*. 16 (1). 103-113

Moritz, S. E., Feltz, D. L., Fahrbach, K. R., & Mack, D. E. (2000). The relation of self-efficacy measures to sport performance: A meta-analytic review. Research Quarterly for Exercise and Sport, 71, 280-294.

National Institute for Health and Care Exelence [NICE]. (2007).*Chronic fatigue syndrome/myalgic encephalomyelitis (or encephalopathy) Diagnosis and management of CFS/ME in adults and children*. London: NICE clinical guideline 53. Available from:

http://www.nice.org.uk/guidance/cg53/resources/guidance-chronic-fatigue-syndromemyalgic-encephalomyelitis-or-encephalopathy-pdf

Nijhof, S. L., K. Maijer, G. Bleijenberg, C. Uiterwaal, J. L. L. Kimpen, and E. M. van de Putte. 2011. Adolescent chronic fatigue syndrome: Prevalence, incidence, and morbidity. Pediatrics 127(5):E1169-E1175

Nijs J., Meeus M., Oosterwijck J., Ickmans K., Eupen I.V & Kos D. (2013). *Tired of being sedentary. Physical activity as a treatment goal in patients with chronic fatigue syndrome*. In Martin Ginis, K. A. (Ed.), Handbook of Physical Activity and Mental Health. London: Routledge

Nijs J., Aelbrecht S., Meeus M., Van Oosterwijck J., Zinzen E. & Clarys P. (2011). *Tired of being inactive: a systematic literature review of physical activity, physiological exercise capacity and muscle strength in patients with chronic fatigue syndrome*. Disabil Rehabil;33(17-18):1493-1500.

Nijs, J., Van Oosterwijck J., Meeus M., Lambrecht L., Metzger K., Fremont M. & Paul L. (2010) *Unravelling the nature of postexertional malaise in myalgic encephalomyelitis/ chronic fatigue syndrome: The role of elastase, complement C4a and interleukin-1beta*. Journal of Internal Medicine 267(4):418-435.

Patterson M.S., Meyer U.M.R., Beaujean A.A. & Bowden R.G (2014). *Using the social cognitive theory to understand physical activity among dialysis patients*. Rehabil Psychol. Aug;59(3):278-88.

Price J.R., Mitchell E., Tidy E. & Hunot V (2008) *Cognitive behaviour therapy for chronic fatigue syndrome in adults*. Cochrane Database Syst Rev 2008 Jul 16;(3): CD001027

Prins J.B., Bleijenberg G., Bazelmans E., Elving L.D., de Boo T.M., Severens J.L., … van der Meer J.W. (2001). *Cognitive behaviour therapy for chronic fatigue syndrome: a multicentre randomised controlled trial*. Lancet. 2001 Mar 17;357(9259):841-7.

Prins J.B., van der Meer J.W & Bleijenberg G (2006). *Chronic fatigue syndrome*. Lancet 2006 Jan 28;367(9507):346-55

Razali N. M., & Wah Y.B (2011). Power comparison of Shapiro-Wilk, Komogorov-Smirnov, Lillefors and Anderson-Darling tests. *Journal of Statistical modeling and analytics, 2(1), 21-33*.

Reyes, M., Gary, Jr., H.E., Dobbins, J.G., Randall, B., Steele, L., Fukuda, K., ... Reeves M.D. (1997). Surveillance for chronic fatigue syndrome – Four U.S. cities, September, 1989 through August, 1993. Morbidity and Mortality Weekly Report Surveillance Summaries, 46, 1-13.

Reynolds K.D., Killen J.D., Bryson S.W., Maron D.J., Taylor C.B., MacCoby N. & Farquhar J.W. (1990). *Psychosocial predictors of physical activity in adolescents* Prev Med, 19. pp. 541–551

Royal College of Pediatrics and Child Health (RCPCH) (2004). Evidence Based Guideline for the Management of CFS/ME (Chronic Fatigue Syndrome/Myalgic Encephalopathy) in Children and Young People

Ruyter, K W. *Medisin og helsefag [Medicine and Healthsubjects]* (Sist oppdatert: 06. oktober 2009). The Norwegian National Committees for Research Ethics [Online]. Retrived 3. Oct 2014 from: http://www.etikkom.no/FBIB/Introduksjon/Innforing-i-forskningsetikk/Medisin-og-helsefag/

Sæbu, M (2011). *Physical activity and motivation in young adults with physical disability: a multidimentional study base don a cross-sectional survey and an intervention-study*. Doctoral thesis. Norwegian School of Sport Science, Oslo. Available from: http://hdl.handle.net/11250/171307

Sairenji T., Yamanishi K., Tachibana Y., Bertoni G. & Kurata T. (1995). *Antibody responses to Epstein-Barr virus, human herpesvirus 6 and human herpesvirus 7 in patients with chronic fatigue syndrome*. Intervirol 38:269273.

Sallis J.F., Simons-Morton B.G., Stone E.F., Corbin C.B., Epstein L.H., Faucette N., … Taylor W.C (1992). *Determinants of physical activity and interventions in youth*. Med Sci Sports Exerc, 24, pp. S248–S257

Sanders P. & Korf J. (2008) *Neuroaetiology of chronic fatigue syndrome: an overview*. The world journal of biological psychiatry; 9(3): 165-171.

Schwarzer R. & Fuchs R. (1995) *Self-Efficacy and Health behaviour*. In Conner, M., & Norman, P. (1995). Predicting Health Behaviour: Research and Practice with Social Cognition Models. Buckingham: Open University Press

Schweitzer R., Kelly B., Foran A., Terry D, Whiting J. (1995). *Quality of life in chronic fatigue syndrome*. Soc Sci Med. Nov;41 (10):1367-72

Shapiro S.S., & Wilk, M.B (1965). An analysis of Variance tes for Normality (complete Samples). Biometrika, 52(3/4), 591-611.

Smith A.L & Biddle S. (Edt) (2008). *Youth Physical Activity and Sedentary Behavior: Challenges and Solutions*. Human Kinetics.

Smith M.S., Mitchell J., Corey L., Gold D., McCauley E.A., Glover D. & Tenover F.C (1991). *Chronic fatigue in adolescents*. Pediatrics 1991 Aug;88(2): 195-202

Snell C. R., Stevens S. R., Davenport T. E., & Van Ness J. M. (2013). *Discriminative validity of metabolic and workload measurements for identifying people with chronic fatigue syndrome*. Physical Therapy 93(11):1484-1492

Stajkovic, A. D. & Luthans, F. (1998). Self-efficacy and work-related performance: A meta-analysis. Psychological Bulletin, 124, 240-261.

Straus S.E., Tosato G., Armstrong G., Lawley T., Preble O.T., Henle W., … Blaese M (1985) *Persisting illness and fatigue in adults with evidence of EpsteinBarr virus infection*. Ann Intern Med 102:716.

Stulemeijer M., de Jong L.W.A.M., Fiselier T.J.W., Hoogveld S.W.B & Bleijenberg G (2005). *Cognitive behaviour therapy for adolescents with chronic fatigue syndrome: randomised controlled trial*. BMJ. Available from: http://www.bmj.com/cgi/reprint/330/7481/14

Sulheim D., Fagermoen E., Winger A.R.N., Andersen A.M., Godang K.G., Müller F … Whyller V.B. (2014). *Disease Mechanisms and Clonidine Treatment in Adolescent Chronic Fatigue SyndromeA Combined Cross-sectional and Randomized Clinical Trial*. JAMA Pediatr. 2014;168(4):351-360. doi:10.1001/jamapediatrics.2013.4647.

Taylor R.R., O'Brien J., Kielhofner G., Lee S.W., Katz B. &Mears C. (2010). *The Occupational and quality of life consequences of chronic fatigue syndrome/myalgic encephalomyelitis in young people*. Br J Occup Ther. 2010 Nov 1;73(11):524-530.

The ME association (2010). *Managing my M.E. What people with ME/CFS and their carers want from the UK's health and social services*. The ME association May 2010: Available from: http://www.meassociation.org.uk/wp-content/uploads/MEA-Management-Survey-2010.pdf

Thomas J.R., Nelson J.K and Silverman S.J (2005). *Research methods in physical activity*. (5th ed), Champaign, IL,Human Kinetics.

Torres-Harding S.R., Jordan K., Jason L.A., Arias R (2006). *Psychosocial and physical impact of chronic fatigue in a community-based sample of children and adolecents*.Vol. 13 No. 2-3, p 55-74.

Tremblay M.S., Colley R.S., Saunders T.J., Healy G.N. & Owen N. (2010). *Physiological and health implication of a sedentary lifestyle*. Appl Physiol Nutr Metab. 2010 Dec; 35(6):725-40.

Trost S.G, McIver K.L, Pate R.R (2005). *Conducting accelerometer-based activity assessments in field-based research*. Med Sci Sports Exer;37(suppl 11):S531-S543.

VanElzakker M.B. (2013) *Chronic fatigue syndrome from vagus nerve infection: a psychoneuroimmuniological hypothesis*. Med Hypothesis. Sep; 81(3):414-23.

Virtanen P (2014). *Polar Loop Activity tracker*. Polar R&D Physiological Research, White Paper. Polar Electro Oy. Domicile:Kempele

Welk G.J., Schaben J.A.. & Morrow J.R. (2004) *Reliability of accelerometry based activity monitors: a generalizability study*. Med Sci Sports Exerc;36(9):1637–45.

White P.D, Goldsmith K.A, Johnson A.L, Potts L, Walwyn R., DeCesare J.C., … Sharpe M. (2011). *Comparison of adaptive pacing therapy, cognitive behaviour therapy, graded exercise therapy, and specialist medical care for chronic fatigue syndrome (PACE): a randomised trial*. The Lancet Vol 377, Issue 9768, 5–11, 823–836. Available from: http://www.sciencedirect.com/science/article/pii/S0140673611600962

White P.D, Sharpe M.C., Chalder T., DeCesare J.C., Walwyn R., & the PACE trial group. (2007). *Protocol for the PACE trial: A randomised controlled trial of adaptive pacing, cognitive behaviour therapy, and graded exercise as supplements to standardised specialist medical care versus standardised specialist medical care alone for patients with the chronic fatigue syndrome/myalgic encephalomyelitis or encephalopathy*. BMC Neurology Vol 7.

Whiting P., Bagnall A., Sowden A., Cornell J., Mulrow C. & Ramirez G. (2001*). Interventions for the treatment and management of chronic fatigue syndrome: a systematic review*. JAMA, 286, pp. 1360–1368.

Wickstrom, G., Bendix, T. (2000) *The "Hawthorne effect" – what did the original Hawthorne studies actually show?* Scandinavian Journal of Work, Environment and Health, 26: pp 363-67

Wilbur J.E., Vassalo A., Chandler P., McDevitt J., & Miller A. M. (2005). *Midlife women's adherence to home-based walking during maintainance*. Nursing Research, 54 (1), 33-40.

World Health Organization [WHO] (2010), *Global Recommendation on Physical Activity for Health*. Geneva

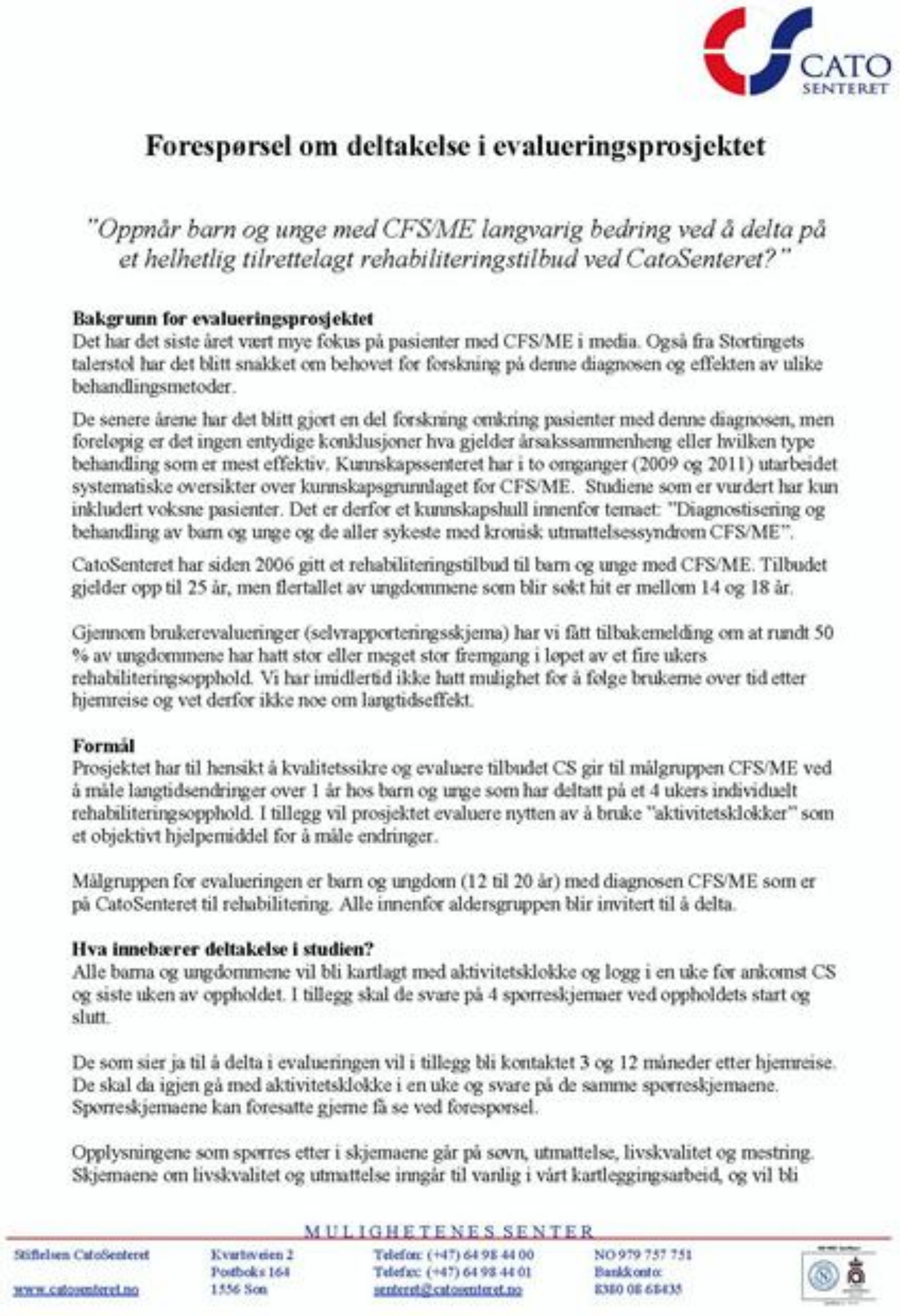
World medical association [WMA] (2013) *World medical association Declaration of Helsinki, Ethical Principles for Medical research involving human subjects*. Clinical Review & Education. Vol 310 nr.20

Wyller V.B (2008). *Kronisk utmattelsessyndrom hos barn og ungdommer Myalgisk encefalopati (CFS/ME).[Chronic fatigue syndrom in children and adolecents. Myalgisk Encephalomyelitis (CFS/ME)]*. Barneklinikken, Rikshospitales HF

Wyller V.B. & Helland I.B. (2013). *Relationship between autonomic cardiovascular control, case definition, clinical symptoms, and functional disability in adolescent chronic fatigue syndrome: an exploratory study*. Biopsychosoc Med. 2013 Feb 7;7(1):5

Zachariae R. (2009). *Psychoneuroimmunology: Abio-psycho-social approach to health and disease*.Scandinavian Journal of Psychology, 2009, 50, 645-651.

Appendix 1: Informed consent



CATO
SENTERET

Forespørsel om deltakelse i evalueringsprosjektet

"Oppnår barn og unge med CFS/ME langvarig bedring ved å delta på et helhetlig tilrettelagt rehabiliteringstilbud ved CatoSenteret?"

Bakgrunn for evalueringsprosjektet
Det har det siste året vært mye fokus på pasienter med CFS/ME i media. Også fra Stortingets talerstol har det blitt snakket om behovet for forskning på denne diagnosen og effekten av ulike behandlingsmetoder.

De senere årene har det blitt gjort en del forskning omkring pasienter med denne diagnosen, men foreløpig er det ingen entydige konklusjoner hva gjelder årsakssammenheng eller hvilken type behandling som er mest effektiv. Kunnskapssenteret har i to omganger (2009 og 2011) utarbeidet systematiske oversikter over kunnskapsgrunnlaget for CFS/ME. Studiene som er vurdert har kun inkludert voksne pasienter. Det er derfor et kunnskapshull innenfor temaet: "Diagnostisering og behandling av barn og unge og de aller sykeste med kronisk utmattelsessyndrom CFS/ME".

CatoSenteret har siden 2006 gitt et rehabiliteringstilbud til barn og unge med CFS/ME. Tilbudet gjelder opp til 25 år, men flertallet av ungdommene som blir søkt hit er mellom 14 og 18 år.

Gjennom brukerevalueringer (selvrapporteringsskjema) har vi fått tilbakemelding om at rundt 50 % av ungdommene har hatt stor eller meget stor fremgang i løpet av et fire ukers rehabiliteringsopphold. Vi har imidlertid ikke hatt mulighet for å følge brukerne over tid etter hjemreise og vet derfor ikke noe om langtidseffekt.

Formål
Prosjektet har til hensikt å kvalitetssikre og evaluere tilbudet CS gir til målgruppen CFS/ME ved å måle langtidsendringer over 1 år hos barn og unge som har deltatt på et 4 ukers individuelt rehabiliteringsopphold. I tillegg vil prosjektet evaluere nytten av å bruke "aktivitetsklokker" som et objektivt hjelpemiddel for å måle endringer.

Målgruppen for evalueringen er barn og ungdom (12 til 20 år) med diagnosen CFS/ME som er på CatoSenteret til rehabilitering. Alle innenfor aldersgruppen blir invitert til å delta.

Hva innebærer deltakelse i studien?
Alle barna og ungdommene vil bli kartlagt med aktivitetsklokke og logg i en uke før ankomst CS og siste uken av oppholdet. I tillegg skal de svare på 4 spørreskjemaer ved oppholdets start og slutt.

De som sier ja til å delta i evalueringen vil i tillegg bli kontaktet 3 og 12 måneder etter hjemreise. De skal da igjen gå med aktivitetsklokke i en uke og svare på de samme spørreskjemaene. Spørreskjemaene kan foresatte gjerne få se ved forespørsel.

Opplysningene som spørres etter i skjemaene går på søvn, utmattelse, livskvalitet og mestring. Skjemaene om livskvalitet og utmattelse inngår til vanlig i vårt kartleggingsarbeid, og vil bli

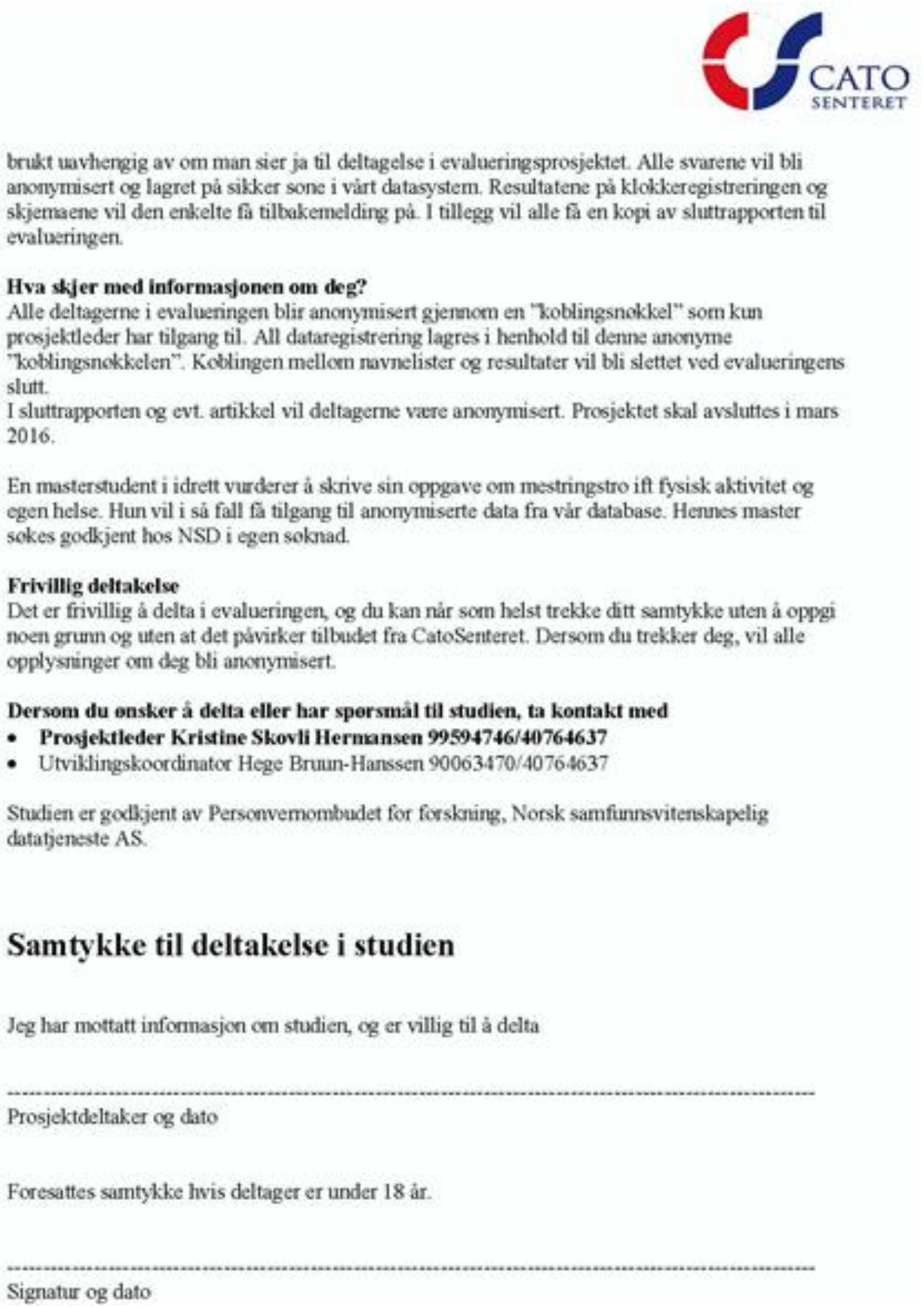
MULIGHETENES SENTER

Stiftelsen CatoSenteret
www.catosenteret.no

Kvartsveien 2
Postboks 164
1556 Son

Telefon: (+47) 64 98 44 00
Telefax: (+47) 64 98 44 01
senteret@catosenteret.no

NO 979 757 751
Bankkonto:
8380 08 68435



brukt uavhengig av om man sier ja til deltagelse i evalueringsprosjektet. Alle svarene vil bli anonymisert og lagret på sikker sone i vårt datasystem. Resultatene på klokkeregistreringen og skjemaene vil den enkelte få tilbakemelding på. I tillegg vil alle få en kopi av sluttrapporten til evalueringen.

Hva skjer med informasjonen om deg?

Alle deltagerne i evalueringen blir anonymisert gjennom en "koblingsnøkkel" som kun prosjektleder har tilgang til. All dataregistrering lagres i henhold til denne anonyme "koblingsnøkkelen". Koblingen mellom navnelister og resultater vil bli slettet ved evalueringens slutt.
I sluttrapporten og evt. artikkel vil deltagerne være anonymisert. Prosjektet skal avsluttes i mars 2016.

En masterstudent i idrett vurderer å skrive sin oppgave om mestringstro ift fysisk aktivitet og egen helse. Hun vil i så fall få tilgang til anonymiserte data fra vår database. Hennes master søkes godkjent hos NSD i egen søknad.

Frivillig deltakelse

Det er frivillig å delta i evalueringen, og du kan når som helst trekke ditt samtykke uten å oppgi noen grunn og uten at det påvirker tilbudet fra CatoSenteret. Dersom du trekker deg, vil alle opplysninger om deg bli anonymisert.

Dersom du ønsker å delta eller har spørsmål til studien, ta kontakt med

- **Prosjektleder Kristine Skovli Hermansen 99594746/40764637**
- Utviklingskoordinator Hege Bruun-Hanssen 90063470/40764637

Studien er godkjent av Personvernombudet for forskning, Norsk samfunnsvitenskapelig datatjeneste AS.

Samtykke til deltakelse i studien

Jeg har mottatt informasjon om studien, og er villig til å delta

----

Prosjektdeltaker og dato

Foresattes samtykke hvis deltager er under 18 år.

Signatur og dato

Appendix 2: Approval letter from Norwegian Social Science Data Services (NSD), Pilot Study

Norsk samfunnsvitenskapelig datatjeneste AS
NORWEGIAN SOCIAL SCIENCE DATA SERVICES

Kristine Skovli Hermansen
CatoSenteret
Kvartsveien 2
1555 SON

Vår dato: 06.10.2014 Vår ref: 39722 / 3 / KH Deres dato Deres ref

TILBAKEMELDING PÅ MELDING OM BEHANDLING AV PERSONOPPLYSNINGER

Vi viser til melding om behandling av personopplysninger, mottatt 08.09.2014. Meldingen gjelder prosjektet:

39722	*Oppnår barn og unge med CFS langvarig bedring ved å delta på et helhetlig tilrettelagt rehabiliteringstilbud ved CatoSenteret?*
Behandlingsansvarlig	*Stiftelsen CatoSenteret, ved institusjonens øverste leder*
Daglig ansvarlig	*Kristine Skovli Hermansen*

Personvernombudet har vurdert prosjektet, og finner at behandlingen av personopplysninger vil være regulert av § 7-27 i personopplysningsforskriften. Personvernombudet tilrår at prosjektet gjennomføres.

Personvernombudets tilråding forutsetter at prosjektet gjennomføres i tråd med opplysningene gitt i meldeskjemaet, korrespondanse med ombudet, ombudets kommentarer samt personopplysningsloven og helseregisterloven med forskrifter. Behandlingen av personopplysninger kan settes i gang.

Det gjøres oppmerksom på at det skal gis ny melding dersom behandlingen endres i forhold til de opplysninger som ligger til grunn for personvernombudets vurdering. Endringsmeldinger gis via et eget skjema, http://www.nsd.uib.no/personvern/meldeplikt/skjema.html. Det skal også gis melding etter tre år dersom prosjektet fortsatt pågår. Meldinger skal skje skriftlig til ombudet.

Personvernombudet har lagt ut opplysninger om prosjektet i en offentlig database, http://pvo.nsd.no/prosjekt.

Personvernombudet vil ved prosjektets avslutning, 31.03.2016, rette en henvendelse angående status for behandlingen av personopplysninger.

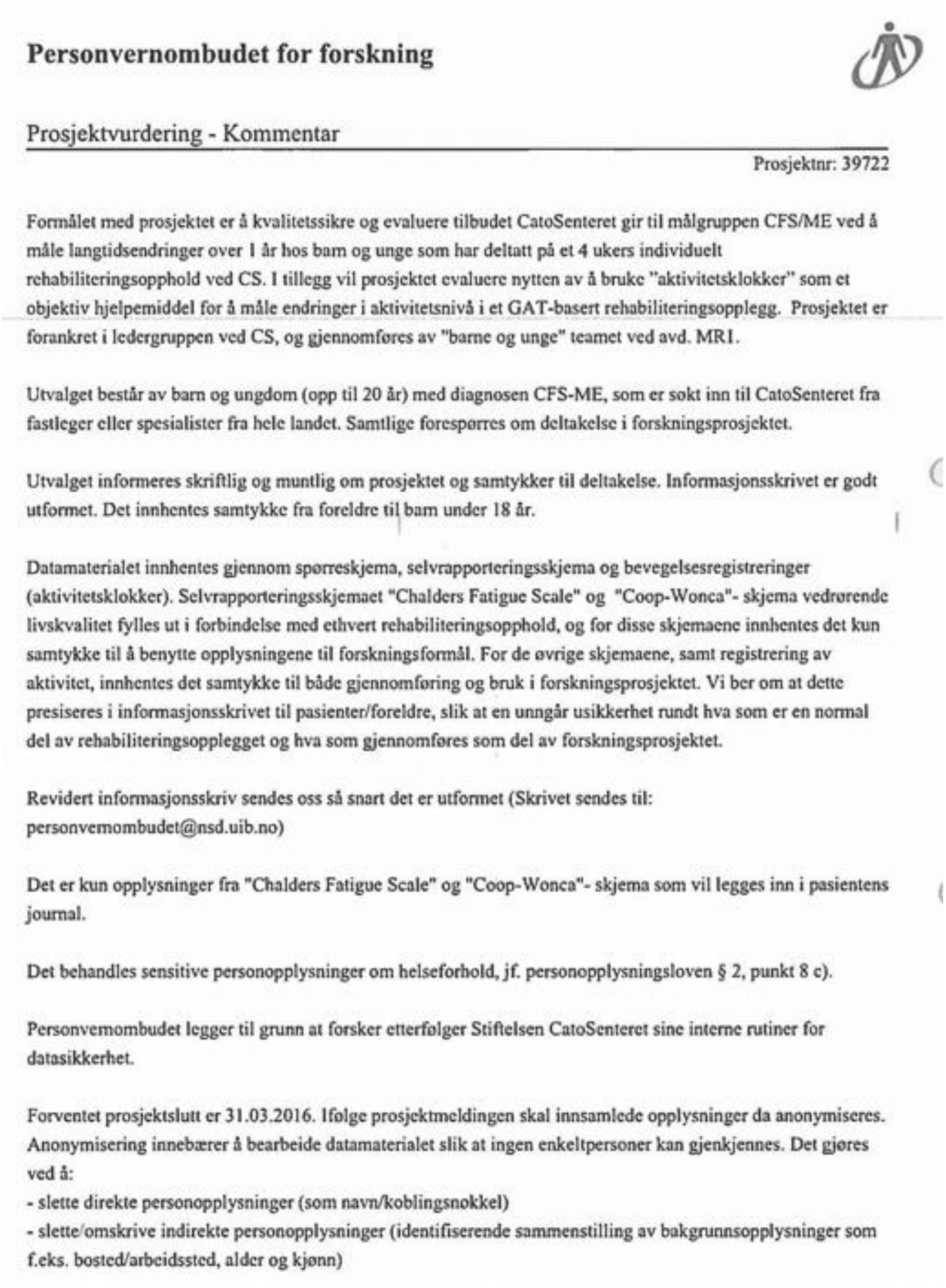
Vennlig hilsen

Katrine Utaaker Segadal

Kjersti Haugstvedt

Kontaktperson: Marie Strand Schildmann tlf: 55 58 31 52
Vedlegg: Prosjektvurdering

Dokumentet er elektronisk produsert og godkjent ved NSDs rutiner for elektronisk godkjenning.



Personvernombudet for forskning

Prosjektvurdering - Kommentar

Prosjektnr: 39722

Formålet med prosjektet er å kvalitetssikre og evaluere tilbudet CatoSenteret gir til målgruppen CFS/ME ved å måle langtidsendringer over 1 år hos barn og unge som har deltatt på et 4 ukers individuelt rehabiliteringsopphold ved CS. I tillegg vil prosjektet evaluere nytten av å bruke "aktivitetsklokker" som et objektiv hjelpemiddel for å måle endringer i aktivitetsnivå i et GAT-basert rehabiliteringsopplegg. Prosjektet er forankret i ledergruppen ved CS, og gjennomføres av "barne og unge" teamet ved avd. MRI.

Utvalget består av barn og ungdom (opp til 20 år) med diagnosen CFS-ME, som er søkt inn til CatoSenteret fra fastleger eller spesialister fra hele landet. Samtlige forespørres om deltakelse i forskningsprosjektet.

Utvalget informeres skriftlig og muntlig om prosjektet og samtykker til deltakelse. Informasjonsskrivet er godt utformet. Det innhentes samtykke fra foreldre til barn under 18 år.

Datamaterialet innhentes gjennom spørreskjema, selvrapporteringsskjema og bevegelsesregistreringer (aktivitetsklokker). Selvrapporteringsskjemaet "Chalders Fatigue Scale" og "Coop-Wonca"- skjema vedrørende livskvalitet fylles ut i forbindelse med ethvert rehabiliteringsopphold, og for disse skjemaene innhentes det kun samtykke til å benytte opplysningene til forskningsformål. For de øvrige skjemaene, samt registrering av aktivitet, innhentes det samtykke til både gjennomføring og bruk i forskningsprosjektet. Vi ber om at dette presiseres i informasjonsskrivet til pasienter/foreldre, slik at en unngår usikkerhet rundt hva som er en normal del av rehabiliteringsopplegget og hva som gjennomføres som del av forskningsprosjektet.

Revidert informasjonsskriv sendes oss så snart det er utformet (Skrivet sendes til: personvernombudet@nsd.uib.no)

Det er kun opplysninger fra "Chalders Fatigue Scale" og "Coop-Wonca"- skjema som vil legges inn i pasientens journal.

Det behandles sensitive personopplysninger om helseforhold, jf. personopplysningsloven § 2, punkt 8 c).

Personvernombudet legger til grunn at forsker etterfølger Stiftelsen CatoSenteret sine interne rutiner for datasikkerhet.

Forventet prosjektslutt er 31.03.2016. Ifølge prosjektmeldingen skal innsamlede opplysninger da anonymiseres. Anonymisering innebærer å bearbeide datamaterialet slik at ingen enkeltpersoner kan gjenkjennes. Det gjøres ved å:
- slette direkte personopplysninger (som navn/koblingsnøkkel)
- slette/omskrive indirekte personopplysninger (identifiserende sammenstilling av bakgrunnsopplysninger som f.eks. bosted/arbeidssted, alder og kjønn)

Appendix 3: Approval letter from Norwegian Social Science Data Services

Norsk samfunnsvitenskapelig datatjeneste AS
NORWEGIAN SOCIAL SCIENCE DATA SERVICES

Harald Hårfagres gate 29
N-5007 Bergen
Norway
Tel: +47-55 58 21 17
Fax: +47-55 58 96 50
nsd@nsd.uib.no
www.nsd.uib.no
Org.nr. 985 321 884

Anders Farholm
Seksjon for coaching og psykologi Norges idrettshøgskole
Postboks 4014 Ullevål Stadion
0806 OSLO

Vår dato: 15.01.2015 Vår ref: 41195 / 3 / AMS Deres dato: Deres ref:

TILBAKEMELDING PÅ MELDING OM BEHANDLING AV PERSONOPPLYSNINGER

Vi viser til melding om behandling av personopplysninger, mottatt 13.12.2014. Meldingen gjelder prosjektet:

41195	*Endringer i mestringstro og fysisk aktivitetsnivå hos hos barn og unge med CFS etter ett 4-ukers rehabilieringsopphold ved CatoSenteret*
Behandlingsansvarlig	*Norges idrettshøgskole, ved institusjonens øverste leder*
Daglig ansvarlig	*Anders Farholm*
Student	*Anita Monsen*

Personvernombudet har vurdert prosjektet, og finner at behandlingen av personopplysninger vil være regulert av § 7-27 i personopplysningsforskriften. Personvernombudet tilrår at prosjektet gjennomføres.

Personvernombudets tilråding forutsetter at prosjektet gjennomføres i tråd med opplysningene gitt i meldeskjemaet, korrespondanse med ombudet, ombudets kommentarer samt personopplysningsloven og helseregisterloven med forskrifter. Behandlingen av personopplysninger kan settes i gang.

Det gjøres oppmerksom på at det skal gis ny melding dersom behandlingen endres i forhold til de opplysninger som ligger til grunn for personvernombudets vurdering. Endringsmeldinger gis via et eget skjema, http://www.nsd.uib.no/personvern/meldeplikt/skjema.html. Det skal også gis melding etter tre år dersom prosjektet fortsatt pågår. Meldinger skal skje skriftlig til ombudet.

Personvernombudet har lagt ut opplysninger om prosjektet i en offentlig database, http://pvo.nsd.no/prosjekt.

Personvernombudet vil ved prosjektets avslutning, 01.07.2015, rette en henvendelse angående status for behandlingen av personopplysninger.

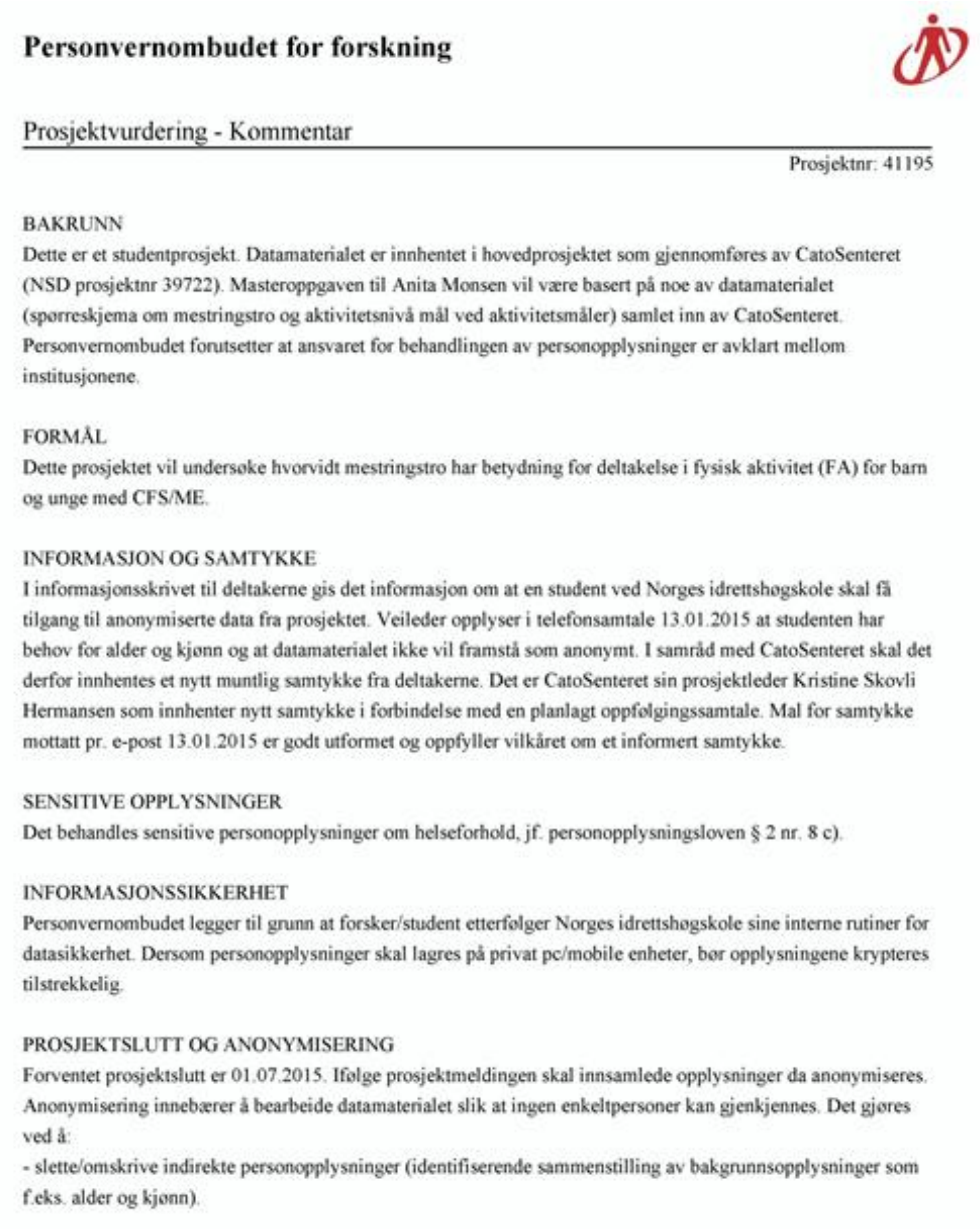
Vennlig hilsen

Vigdis Namtvedt Kvalheim

Anne-Mette Somby

Kontaktperson: Anne-Mette Somby tlf: 55 58 24 10

Dokumentet er elektronisk produsert og godkjent ved NSDs rutiner for elektronisk godkjenning.



Personvernombudet for forskning

Prosjektvurdering - Kommentar

Prosjektnr: 41195

BAKRUNN

Dette er et studentprosjekt. Datamaterialet er innhentet i hovedprosjektet som gjennomføres av CatoSenteret (NSD prosjektnr 39722). Masteroppgaven til Anita Monsen vil være basert på noe av datamaterialet (spørreskjema om mestringstro og aktivitetsnivå mål ved aktivitetsmåler) samlet inn av CatoSenteret. Personvernombudet forutsetter at ansvaret for behandlingen av personopplysninger er avklart mellom institusjonene.

FORMÅL

Dette prosjektet vil undersøke hvorvidt mestringstro har betydning for deltakelse i fysisk aktivitet (FA) for barn og unge med CFS/ME.

INFORMASJON OG SAMTYKKE

I informasjonsskrivet til deltakerne gis det informasjon om at en student ved Norges idrettshøgskole skal få tilgang til anonymiserte data fra prosjektet. Veileder opplyser i telefonsamtale 13.01.2015 at studenten har behov for alder og kjønn og at datamaterialet ikke vil framstå som anonymt. I samråd med CatoSenteret skal det derfor innhentes et nytt muntlig samtykke fra deltakerne. Det er CatoSenteret sin prosjektleder Kristine Skovli Hermansen som innhenter nytt samtykke i forbindelse med en planlagt oppfølgingssamtale. Mal for samtykke mottatt pr. e-post 13.01.2015 er godt utformet og oppfyller vilkåret om et informert samtykke.

SENSITIVE OPPLYSNINGER

Det behandles sensitive personopplysninger om helseforhold, jf. personopplysningsloven § 2 nr. 8 c).

INFORMASJONSSIKKERHET

Personvernombudet legger til grunn at forsker/student etterfølger Norges idrettshøgskole sine interne rutiner for datasikkerhet. Dersom personopplysninger skal lagres på privat pc/mobile enheter, bør opplysningene krypteres tilstrekkelig.

PROSJEKTSLUTT OG ANONYMISERING

Forventet prosjektslutt er 01.07.2015. Ifølge prosjektmeldingen skal innsamlede opplysninger da anonymiseres. Anonymisering innebærer å bearbeide datamaterialet slik at ingen enkeltpersoner kan gjenkjennes. Det gjøres ved å:

- slette/omskrive indirekte personopplysninger (identifiserende sammenstilling av bakgrunnsopplysninger som f.eks. alder og kjønn).

Appendix 4: Self-efficacy scale (SES)

Hvor sikker er du på at du klarer å være i fysisk aktivitet

Fysisk aktivitet i dette tilfellet vil si **å være i bevegelse**. For eksempel når du går til skolen, sykler, rydder, trener eller leker er du i fysisk aktivitet. Under følger noen spørsmål om dine tanker og holdninger til det å være i fysisk aktivitet. Vær vennlig og sett ring rundt det tallet på skalaen hvor utsagnene under stemmer med dine tanker om å være fysisk aktiv.

	Jeg er sikker på at...	Stemmer ikke									Stemmer svært godt
1.	...jeg kan være fysisk aktiv selv om jeg er trett og sliten	1	2	3	4	5	6	7	8	9	10
2.	...jeg kan være fysisk aktiv selv om jeg føler meg trist eller deprimert	1	2	3	4	5	6	7	8	9	10
3.	...jeg kan være fysisk aktiv selv om jeg ikke får støtte fra familie eller venner	1	2	3	4	5	6	7	8	9	10
4.	...jeg kan være fysisk aktiv uten hjelp av en fysioterapeut, instruktør eller trener	1	2	3	4	5	6	7	8	9	10
5.	...jeg kan motivere meg til å starte med fysisk aktivitet igjen etter å ha sluttet en periode	1	2	3	4	5	6	7	8	9	10
6.	...jeg kan være fysisk aktiv selv om jeg ikke har tilgang til treningsstudio eller andre trenings- eller rehabiliteringsfasiliteter	1	2	3	4	5	6	7	8	9	10
7.	...jeg kan finne måter å være fysisk aktiv på	1	2	3	4	5	6	7	8	9	10
8.	...jeg kan gjennomføre treningsmål jeg setter meg	1	2	3	4	5	6	7	8	9	10
9.	...jeg kan være fysisk aktiv selv om jeg har CFS/ME	1	2	3	4	5	6	7	8	9	10
10.	...jeg greier å være fysisk aktiv de fleste dager	1	2	3	4	5	6	7	8	9	10
11.	...jeg greier å være fysisk aktiv de fleste dager selv når jeg har mulighet til å se TV, bruke PC/Ipad eller lignende i stedet	1	2	3	4	5	6	7	8	9	10
12.	...jeg greier å være fysisk aktiv de fleste dager selv om det er dårlig vær ute	1	2	3	4	5	6	7	8	9	10
13.	...jeg greier å få med meg vennene mine på fysisk aktivitet de fleste dager	1	2	3	4	5	6	7	8	9	10
14.	...jeg klarer å være mer fysisk aktiv	1	2	3	4	5	6	7	8	9	10

Appendix 5: Polar Loop information for participants

CATO SENTERET

Informasjonsskriv Polar Loop Aktivitetsmåler

En aktivitetsmåler er et måleinstrument, som registrerer bevegelse i tre plan. Man får dermed data for grad av bevegelse og søvn. Brukeren går med den som ei klokke. Apparatet kan også måle skritt og puls når det er kombinert med en pulsmåler. Aktivitetsmåler brukes mye i utredning av søvnforstyrrelser og til forskning.

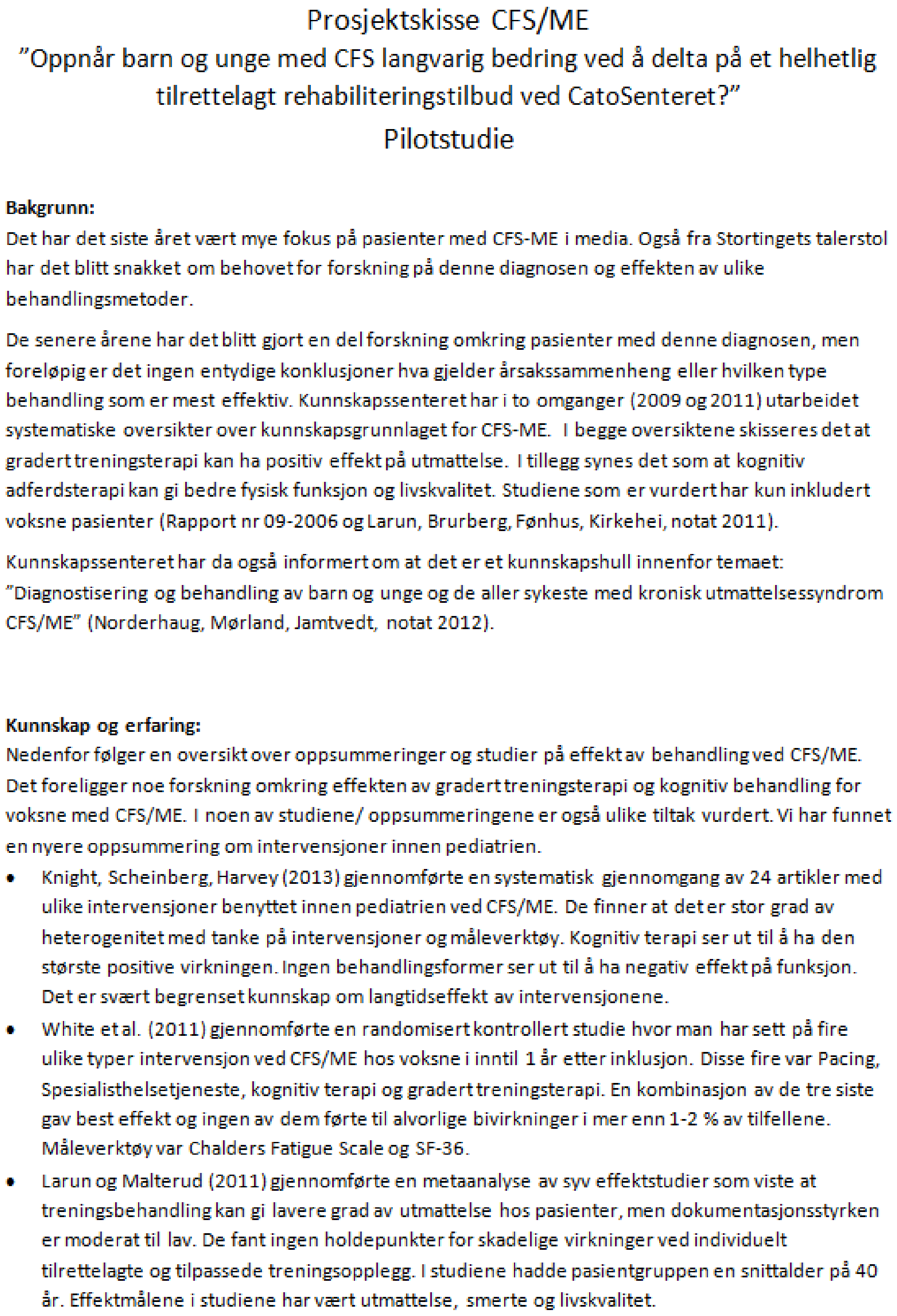
CatoSenteret ønsker å prøve ut verdien av aktivitetsmåler for å registrere aktivitetsnivå før oppholdet og mot slutten av oppholdet. Det er vanskelig å finne gode måter å måle aktivitet objektivt og vi håper at en slik aktivitetsmåler kan være et godt hjelpemiddel.

Viktig å vite om aktivitetsmåleren:

- Skal sitte på hele døgnet.
- Bør lades hver 3. dag i ca 1 time – lader følger med.
- Kan bades/dusjes med.
- Den må ikke kobles opp mot egen datamaskin, da klokken er ferdig registrert ved CatoSenteret.
- Klokken skal sitte på høyre arm.
- Vi har tilpasset klokkene i 3 standard størrelser. Vær så snill å ikke endre disse tilpasningene.
- Ignorer meldinger, opplysninger og oppfordringer som kommer på displayet. Vi vil at du skal leve helt som vanlig.
- Målingen skal foretas i en uke før ankomst CatoSenteret og ved slutten av rehabiliteringsoppholdet. For de som vil være med i prosjektet vil det i tillegg bli målt 3 mnd. og 12 mnd. etter hjemreise.

Lykke til!

Appendix 6: Project plan for overall pilot-study



Prosjektskisse CFS/ME
”Oppnår barn og unge med CFS langvarig bedring ved å delta på et helhetlig tilrettelagt rehabiliteringstilbud ved CatoSenteret?”
Pilotstudie

Bakgrunn:
Det har det siste året vært mye fokus på pasienter med CFS-ME i media. Også fra Stortingets talerstol har det blitt snakket om behovet for forskning på denne diagnosen og effekten av ulike behandlingsmetoder.

De senere årene har det blitt gjort en del forskning omkring pasienter med denne diagnosen, men foreløpig er det ingen entydige konklusjoner hva gjelder årsakssammenheng eller hvilken type behandling som er mest effektiv. Kunnskapssenteret har i to omganger (2009 og 2011) utarbeidet systematiske oversikter over kunnskapsgrunnlaget for CFS-ME. I begge oversiktene skisseres det at gradert treningsterapi kan ha positiv effekt på utmattelse. I tillegg synes det som at kognitiv adferdsterapi kan gi bedre fysisk funksjon og livskvalitet. Studiene som er vurdert har kun inkludert voksne pasienter (Rapport nr 09-2006 og Larun, Brurberg, Fønhus, Kirkehei, notat 2011).

Kunnskapssenteret har da også informert om at det er et kunnskapshull innenfor temaet: ”Diagnostisering og behandling av barn og unge og de aller sykeste med kronisk utmattelsessyndrom CFS/ME” (Norderhaug, Mørland, Jamtvedt, notat 2012).

Kunnskap og erfaring:
Nedenfor følger en oversikt over oppsummeringer og studier på effekt av behandling ved CFS/ME. Det foreligger noe forskning omkring effekten av gradert treningsterapi og kognitiv behandling for voksne med CFS/ME. I noen av studiene/ oppsummeringene er også ulike tiltak vurdert. Vi har funnet en nyere oppsummering om intervensjoner innen pediatrien.

- Knight, Scheinberg, Harvey (2013) gjennomførte en systematisk gjennomgang av 24 artikler med ulike intervensjoner benyttet innen pediatrien ved CFS/ME. De finner at det er stor grad av heterogenitet med tanke på intervensjoner og måleverktøy. Kognitiv terapi ser ut til å ha den største positive virkningen. Ingen behandlingsformer ser ut til å ha negativ effekt på funksjon. Det er svært begrenset kunnskap om langtidseffekt av intervensjonene.
- White et al. (2011) gjennomførte en randomisert kontrollert studie hvor man har sett på fire ulike typer intervensjon ved CFS/ME hos voksne i inntil 1 år etter inklusjon. Disse fire var Pacing, Spesialisthelsetjeneste, kognitiv terapi og gradert treningsterapi. En kombinasjon av de tre siste gav best effekt og ingen av dem førte til alvorlige bivirkninger i mer enn 1-2 % av tilfellene. Måleverktøy var Chalders Fatigue Scale og SF-36.
- Larun og Malterud (2011) gjennomførte en metaanalyse av syv effektstudier som viste at treningsbehandling kan gi lavere grad av utmattelse hos pasienter, men dokumentasjonsstyrken er moderat til lav. De fant ingen holdepunkter for skadelige virkninger ved individuelt tilrettelagte og tilpassede treningsopplegg. I studiene hadde pasientgruppen en snittalder på 40 år. Effektmålene i studiene har vært utmattelse, smerte og livskvalitet.

- Pinxsterhuis (2011) gjennomførte en systematisk litteraturgjennomgang for å vurdere effekten av fysisk aktivitet på utmattelse og aktivitetsnivå hos voksne over 16 år med ME. Totalt 14 studier ble inkludert. Hun anbefaler en kombinasjon av spørreskjema og actigraf for å oppnå en mer kontrollert beskrivelse av aktivitetsnivå og utbytte av behandlingsopplegg. Det ser ut til at kombinasjonsprogrammer med ulike typer aktiviteter som også inkluderer bruk av kognitiv terapi har best effekt. Effektmålene i studiene var utmattelse og fysisk aktivitetsnivå, alt målt med egenrapporterte spørreskjemaer.
- Berge og Dehli (2009) refererer til ulike studier som viser effekt av kognitiv terapi ved CFS/ME. Effekten vedvarte over en periode på 5 år. Kognitiv terapi brukt i behandling innebærer en gradvis økning av aktivitetsnivå med bruk av aktivitetsplan. Aktivitetsplanen gir balanse i aktivitetene gjennom dagen og kan motvirke symptomfokusering og store svingninger i aktivitet. De formidler en forståelse av årsaken til CFS/ME som tre-delt; sårbarhetsfaktorer, utløsende faktorer og opprettholdende faktorer. Kognitiv terapi retter seg mot de siste faktorene, de onde sirklene som bidrar til at utmattelsen blir kronisk, og målet med terapien er mestring og økt livskvalitet.
- Njølstad, Sveen og Bruun Wyller (2012) har undersøkt bruken av aktivitetsplan som del av gradert aktivitetstilpasning til ungdom 12-18 år. Aktivitetsplan blir relativt lite brukt, til tross for at tiltaket er anbefalt og en sentral del av anbefalt rehabiliteringsopplegg. Blant pasienter og behandlere som hadde benyttet den, ble planen oppfattet som nyttig for å bevisstgjøre energi og prioritering av aktiviteter.
- Knoop et al (2008) fant at behandlingsprognosen til ungdom med CFS/ME var påvirket av morens grad av utmattelse og av familiens forstillinger om hva utmattelse er. Dette bør derfor være tema i samtaler.
- I en Cochrane oppsummering viser Edmonds et al (2004) at fysisk aktivitet med lav intensitet sammen med et opplæringsprogram (kognitiv terapi) har bedre effekt på utmattelse, livskvalitet og depresjon enn fysisk aktivitet alene. Intervensjonene hadde en varighet på 12 uker. De etterspør målinger og oppfølging over lengre tid. Studiene benyttet utmattelse, fysisk funksjon og depresjon som effektmål.
- Anette Winger (2012) skriver i sin masteroppgave at ungdommene ønsker å bli tatt på alvor, de etterspør kunnskap og forklaring på sine symptomer. De opplever det som vanskelig å gå glipp av skole og sosialt samvær med venner. De ønsker seg bedre tilrettelegging i forhold til skole.

Det har vist seg svært vanskelig å finne oppsummeringer eller studier som viser effekt av rehabilitering hos barn og unge med CFS/ME. Kontakt med Nasjonal kompetansetjeneste for CFS/ME ved UiO, Rikshospitalet bekrefter dette. Dette blir støttet av Knight og kollegaer (2013) som understreker at forskning på barn og unge med CFS/ME er helt i startfasen.

CatoSenteret har siden 2006 gitt et rehabiliteringstilbud til barn og unge med CFS-ME. Tilbudet gjelder opp til 25 år, men flertallet av ungdommene som blir søkt hit er mellom 14 og 18 år. Forekomsten av diagnosen i Norge er ikke endelig fastslått, men tall fra utenlandske studier tyder på mellom 9 000-18 000 mennesker. Andelen barn og unge (under 18 år) er uavklart, men 600 pasienter i denne aldersgruppen er et forsiktig anslag (Bruun Wyller, 2011). Forholdet mellom jenter og gutter er ca. 3:1. Tilstanden er sjelden hos barn under 10 år. Funksjonstapet hos disse ungdommene fører til frafall/ sykemelding fra skole hos 2 av 3 pasienter (Crawley, Sterne, 2009). Ungdommene som søkes inn til CatoSenteret har et moderat funksjonstap – dvs. at all aktivitet er sterkt redusert og hvileperioder er nødvendige. Allmennsymptomer, smerte og dårlig søvnkvalitet gir redusert livskvalitet (Helse Bergen, 2011).

Gjennom brukerevalueringer (selvrapporteringsskjema) har vi fått tilbakemelding om at rundt 50 % av ungdommene har hatt stor eller meget stor fremgang i løpet av et fire ukers rehabiliteringsopphold. Men vi har ikke hatt mulighet for å følge brukerne over tid etter hjemreise og vet derfor ikke noe om langtidseffekt. Vi har ikke gjennomført lignende effektevalueringer med mer objektive metoder.

Prosjektets målsetting:

Prosjektet har som mål å bidra til økt kunnskap om effekt av et rehabiliteringstilbud hvor gradert aktivitetstilpasning (GAT) er sentralt. Dette er den type tilbud som blir anbefalt fra Nasjonal Kompetansetjeneste for CFS/ME (Njølstad, Sveen, Bruun Wyller, 2012).

1. Prosjektet har til hensikt å måle langtidsendringer over 1 år hos barn og unge med diagnosen CFS/ME som deltar på et 4 ukers individuelt rehabiliteringsopphold ved CatoSenteret.
2. Prosjektet vil evaluere nytten av å bruke "aktivitetsklokker" som et objektiv hjelpemiddel for å måle aktivitetsnivå i et GAT basert rehabiliteringsopplegg.
3. Prosjektet vil fungere som en pilotstudie for å vurdere metode og måleverktøy for senere å kunne gjennomføre en større og kontrollert studie. Til dette studiet søkes det samarbeid med HF, avdelinger som bistår i diagnostisering av CFS/ME. Det vil i denne studien være aktuelt å se på mulighetene for å randomisere deltagerne.

Kriterier for måloppnåelse:
Prosjektets målsettinger sammenfaller med tre kriterier for måloppnåelse:

- Gjennom intern evaluering av effekt og utprøving av et nytt måleverktøy har prosjektet som mål å bidra til økt kunnskap om effekt av et rehabiliteringstilbud hvor "gradert aktivitetstilpasning" (GAT) er sentralt. Det er et kunnskapshull om diagnostisering og behandling til prosjektets målgruppe (Norderhaug, Mørland, Jamtvedt, notat 2012).
- Behandlingslinjen for rehabilitering av CFS/ME ved CatoSenteret beskriver samarbeidet mellom skole og helse internt på senteret, og videre samhandling med de samme etater i 1. linjetjenesten. En oppfølging over 12 mnd vil avdekke om denne samhandlingen har fungert i samsvar med brukers behov.
- Resultatene som kommer frem i prosjektet vil bli en del av den årlige evalueringen av behandlingslinjen og dermed implementert i daglig drift.

Metoder:
Prosjektets målgruppe:
Målgruppen er barn og ungdom (opp til 20 år) med diagnosen CFS-ME. De er søkt inn til CatoSenteret fra fastleger eller spesialister fra hele landet. Hovedtyngden av brukerne kommer fra Helse Sør-Øst regionen.

- Pinxsterhuis (2011) gjennomførte en systematisk litteraturgjennomgang for å vurdere effekten av fysisk aktivitet på utmattelse og aktivitetsnivå hos voksne over 16 år med ME. Totalt 14 studier ble inkludert. Hun anbefaler en kombinasjon av spørreskjema og actigraf for å oppnå en mer kontrollert beskrivelse av aktivitetsnivå og utbytte av behandlingsopplegg. Det ser ut til at kombinasjonsprogrammer med ulike typer aktiviteter som også inkluderer bruk av kognitiv terapi har best effekt. Effektmålene i studiene var utmattelse og fysisk aktivitetsnivå, alt målt med egenrapporterte spørreskjemaer.
- Berge og Dehli (2009) refererer til ulike studier som viser effekt av kognitiv terapi ved CFS/ME. Effekten vedvarte over en periode på 5 år. Kognitiv terapi brukt i behandling innebærer en gradvis økning av aktivitetsnivå med bruk av aktivitetsplan. Aktivitetsplanen gir balanse i aktivitetene gjennom dagen og kan motvirke symptomfokusering og store svingninger i aktivitet. De formidler en forståelse av årsaken til CFS/ME som tre-delt; sårbarhetsfaktorer, utløsende faktorer og opprettholdende faktorer. Kognitiv terapi retter seg mot de siste faktorene, de onde sirklene som bidrar til at utmattelsen blir kronisk, og målet med terapien er mestring og økt livskvalitet.
- Njølstad, Sveen og Bruun Wyller (2012) har undersøkt bruken av aktivitetsplan som del av gradert aktivitetstilpasning til ungdom 12-18 år. Aktivitetsplan blir relativt lite brukt, til tross for at tiltaket er anbefalt og en sentral del av anbefalt rehabiliteringsopplegg. Blant pasienter og behandlere som hadde benyttet den, ble planen oppfattet som nyttig for å bevisstgjøre energi og prioritering av aktiviteter.
- Knoop et al (2008) fant at behandlingsprognosen til ungdom med CFS/ME var påvirket av morens grad av utmattelse og av familiens forstillinger om hva utmattelse er. Dette bør derfor være tema i samtaler.
- I en Cochrane oppsummering viser Edmonds et al (2004) at fysisk aktivitet med lav intensitet sammen med et opplæringsprogram (kognitiv terapi) har bedre effekt på utmattelse, livskvalitet og depresjon enn fysisk aktivitet alene. Intervensjonene hadde en varighet på 12 uker. De etterspør målinger og oppfølging over lengre tid. Studiene benyttet utmattelse, fysisk funksjon og depresjon som effektmål.
- Anette Winger (2012) skriver i sin masteroppgave at ungdommene ønsker å bli tatt på alvor, de etterspør kunnskap og forklaring på sine symptomer. De opplever det som vanskelig å gå glipp av skole og sosialt samvær med venner. De ønsker seg bedre tilrettelegging i forhold til skole.

Det har vist seg svært vanskelig å finne oppsummeringer eller studier som viser effekt av rehabilitering hos barn og unge med CFS/ME. Kontakt med Nasjonal kompetansetjeneste for CFS/ME ved UiO, Rikshospitalet bekrefter dette. Dette blir støttet av Knight og kollegaer (2013) som understreker at forskning på barn og unge med CFS/ME er helt i startfasen.

CatoSenteret har siden 2006 gitt et rehabiliteringstilbud til barn og unge med CFS-ME. Tilbudet gjelder opp til 25 år, men flertallet av ungdommene som blir søkt hit er mellom 14 og 18 år. Forekomsten av diagnosen i Norge er ikke endelig fastslått, men tall fra utenlandske studier tyder på mellom 9 000-18 000 mennesker. Andelen barn og unge (under 18 år) er uavklart, men 600 pasienter i denne aldersgruppen er et forsiktig anslag (Bruun Wyller, 2011). Forholdet mellom jenter og gutter er ca. 3:1. Tilstanden er sjelden hos barn under 10 år. Funksjonstapet hos disse ungdommene fører til frafall/ sykemelding fra skole hos 2 av 3 pasienter (Crawley, Sterne, 2009). Ungdommene som søkes inn til CatoSenteret har et moderat funksjonstap – dvs. at all aktivitet er sterkt redusert og hvileperioder er nødvendige. Allmennsymptomer, smerte og dårlig søvnkvalitet gir redusert livskvalitet (Helse Bergen, 2011).

Gjennom brukerevalueringer (selvrapporteringsskjema) har vi fått tilbakemelding om at rundt 50 % av ungdommene har hatt stor eller meget stor fremgang i løpet av et fire ukers rehabiliteringsopphold. Men vi har ikke hatt mulighet for å følge brukerne over tid etter hjemreise og vet derfor ikke noe om langtidseffekt. Vi har ikke gjennomført lignende effektevalueringer med mer objektive metoder.

Prosjektets målsetting:

Prosjektet har som mål å bidra til økt kunnskap om effekt av et rehabiliteringstilbud hvor gradert aktivitetstilpasning (GAT) er sentralt. Dette er den type tilbud som blir anbefalt fra Nasjonal Kompetansetjeneste for CFS/ME (Njølstad, Sveen, Bruun Wyller, 2012).

1. Prosjektet har til hensikt å måle langtidsendringer over 1 år hos barn og unge med diagnosen CFS/ME som deltar på et 4 ukers individuelt rehabiliteringsopphold ved CatoSenteret.
2. Prosjektet vil evaluere nytten av å bruke "aktivitetsklokker" som et objektiv hjelpemiddel for å måle aktivitetsnivå i et GAT basert rehabiliteringsopplegg.
3. Prosjektet vil fungere som en pilotstudie for å vurdere metode og måleverktøy for senere å kunne gjennomføre en større og kontrollert studie. Til dette studiet søkes det samarbeid med HF, avdelinger som bistår i diagnostisering av CFS/ME. Det vil i denne studien være aktuelt å se på mulighetene for å randomisere deltagerne.

Kriterier for måloppnåelse:
Prosjektets målsettinger sammenfaller med tre kriterier for måloppnåelse:

- Gjennom intern evaluering av effekt og utprøving av et nytt måleverktøy har prosjektet som mål å bidra til økt kunnskap om effekt av et rehabiliteringstilbud hvor "gradert aktivitetstilpasning" (GAT) er sentralt. Det er et kunnskapshull om diagnostisering og behandling til prosjektets målgruppe (Norderhaug, Mørland, Jamtvedt, notat 2012).
- Behandlingslinjen for rehabilitering av CFS/ME ved CatoSenteret beskriver samarbeidet mellom skole og helse internt på senteret, og videre samhandling med de samme etater i 1. linjetjenesten. En oppfølging over 12 mnd vil avdekke om denne samhandlingen har fungert i samsvar med brukers behov.
- Resultatene som kommer frem i prosjektet vil bli en del av den årlige evalueringen av behandlingslinjen og dermed implementert i daglig drift.

Metoder:
Prosjektets målgruppe:
Målgruppen er barn og ungdom (opp til 20 år) med diagnosen CFS-ME. De er søkt inn til CatoSenteret fra fastleger eller spesialister fra hele landet. Hovedtyngden av brukerne kommer fra Helse Sør-Øst regionen.

Inklusjonskriterier:

- Alder mellom 12 og 20 år
- Utredet av sykehusavdeling eller privatpraktiserende spesialist i barnesykdommer
- Har fått diagnosen Chronic Fatigue Syndrome (CFS) og/eller Myalgisk Encephalopathi (ME)
- Er rettighetsvurdert og vurdert av vårt inntaksteam

Eksklusjonskriterier:

- Pasienten ønsker ikke selv et opphold på CatoSenteret
- Ikke ferdig utredet. Pågående utredning eller "alternativ behandling" må være avsluttet/ gjennomført før inntak.

Beskrivelse av gjennomføring:

Denne pilotstudien vil inkludere 3 grupper á 8 ungdommer med CFS/ME og ha fire ulike målepunkter: **T1)** Formøte /Uke 1: rehabilitering start, **T2)** Uke 4: rehabilitering slutt, **T3)** 3 måneder etter rehabilitering slutt, og **T4)** 12 måneder etter rehabilitering slutt. Det vil bli gitt informasjon til deltagerne før, underveis og i etterkant. Dette inkluderer samtykkeskjema i henhold til retningslinjer fra NSD (for deltagere under 18 år, vil også pårørende bli bedt om å skrive under), innsyn i sine egne resultater i sluttrapporten og oppsummering av resultater i etterkant av studien.

Det finnes ingen universelle målverktøy eller test som kan si noe om alvorlighetsgraden av CFS/ME. Når vi skal undersøke endringer etter rehabiliteringsoppholdet har vi derfor tatt utgangspunkt i Nijhof et al (2012) fire komponenter som undersøker sykdomsbildet. Disse fire er: 1) utmattelse, 2) fysisk funksjon, 3) skoledeltagelse og 4) selvrapportert fremgang. I tillegg har vi på bakgrunn av egen erfaring med brukergruppen valgt å legge til kartlegging av søvn og ernæring. Dette fordi vi har erfart at døgnrytmen hos disse ungdommene ofte er veldig forstyrret. Da er ofte måltidsrytmen forskjøvet i tillegg. Mange ungdommer strever også med kvalme og manglende matlyst – noe som kan gi et ensidig og endret kosthold. Det vil derfor bli målt på følgende parametere: Søvn/ døgnrytme, aktivitetsnivå (fysisk aktivitet, sosial aktivitet og skole aktivitet) målt ved logg og aktivitetsmåler, ernæring, livskvalitet og utmattelse.

Intervensjonen (rehabiliteringstilbudet) er i tråd med det tilbudet som er beskrevet i våre avtaler med Helse Sør-Øst: (Avtalen varer frem til 31.12.14 og er søkt forlenget fra CS)

Kort beskrivelse av tilbudet ved CS:

Siden 2009 har senteret valgt å ta imot 6-8 brukere samtidig, slik at man kan bruke samtalegrupper og få en "likemannseffekt" selv om dette tilbudet er individuelt tilrettelagt. I 2013 var 56 ungdommer inne på tilbudet. Det blir gitt et tilbud om samtalegruppe for de pårørende.

Så langt det lar seg gjøre starter alle opphold med et "formøte". Dette holdes på senteret, eller som videokonferanse. Hensikten med møtet er å gi informasjon om tilbudet, møte behandlerteamet, avklare forventninger (begge veier) og møte noen av de andre ungdommene som skal delta samtidig.

Døgntilbudet har fokus på normalisering av døgnrytme, søvnhygiene, ernæring, skole og lekpreget fysisk aktivitet (gradert treningsterapi) med veiledning i gruppe og individuelt. Som del av den graderte aktivitetstilpasningen, må ungdommene utarbeide en aktivitetsplan som de skal bruke når de kommer hjem. Det er to "obligatoriske" samtalegrupper i uken. Første uke er "Bli kjent" tema for samtalene – etablere og bli trygg i gruppen. Uke 2 er det å bli bedre kjent med seg selv, individuelt og i gruppe som er tema. Det jobbes med å finne balansen mellom aktivitet og hvile. I uke 3 er "nyorientering" tema – utforske nye aktiviteter, egne tanker og grenser, hvordan sette gode grenser. Den siste uken brukes mye av tiden på å utarbeide en god aktivitetsplan som kan brukes videre hjemme, sette mål for den første tiden og overføre kunnskap til hjemmemiljøet. Det gjennomføres "overføringsmøter" med ungdom, familie og kontakter fra hjemmetjeneste/skole så langt det er praktisk mulig (evt. som videomøter). Rehabiliteringstilbudet er godt beskrevet i en behandlingslinje. Tilbudet er populært og vi har (dessverre) omtrent ett års venteliste.

Datainnsamling:

T1:

Ved formøtet 3-4 uker før rehabiliteringsstart vil det bli gitt både skriftlig og muntlig informasjon om studien. De som ønsker å delta leverer samtykkeerklæring og blir utstyrt med en aktivitetsmåler og beskjed om å skrive aktivitetslogg slik at en kan etablere en baseline. Denne målingen av aktivitet vil vare en uke. Ved ankomst vil det bli målt på livskvalitet, utmattelse og søvn. For å minske belastningen på deltagerne vil det videre ikke bli målt på søvn hvis dette ikke oppleves som problematisk.

T2:

Den siste uken av rehabiliteringsoppholdet vil deltagerne igjen gå med aktivitetsmåleren samt skrive aktivitetslogg. Før avreise vil det bli målt på livskvalitet, utmattelse, brukertilfredshet og evt. søvn ved behov.

T3:

Ved 3-måneders oppfølging vil deltagerne få tilsendt aktivitetsmåler og logg i posten en uke før et telefonintervju finner sted. Intervjuet vil være basert på loggen og aktivitetsmålingen, samt måle livskvalitet, utmattelse og evt. søvn. Samtalen gir mulighet for å undersøke om deltageren har opplevd fremgang/tilbakegang og eventuelle årsaker til dette.

T4:

12-måneders oppfølging vil være identisk med 3-måneders oppfølging.

Måleverktøy

Følgende målemetoder vil bli benyttet:

1) Adolescent Sleep-Wake Scale (12-18 år), et skjema med 5 dimensjoner som måler ulike typer søvnvansker hos ungdom. Til norsk ved Ståle Pallesen. (LeBourgeois et al. (2005).
2) Fatigue Scale, et selvrapporteringsskjema med 13 punkter som kartlegger grad av utmattelse og som også kan skille mellom fysisk og mental utmattelse. Mye brukt i ulike prosjekter. (T. Chalder, G. Berelowitz, T. Pawlikowska, L. Watts, S. Wessely, D. Wright, and E. P. Wallace, 1993)
3) Aktivitetsnivå - målt med aktivitetsmåler. En aktivitetsmåler er et måleinstrument, som måler bevegelse i tre plan. Man får dermed data for grad av bevegelse eller søvn. Brukeren går med den som ei klokke. Apparatet kan også måle skritt og puls når det er kombinert med en pulsmåler. Aktivitetsmåler brukes mye i utredning av søvnforstyrrelser og til forskning.
4) Aktivitetsnivå – mål med selvrapportering i logg. Herunder skoledeltagelse, måltider, sosial aktivitet og døgnrytme. Bruken av logg sammen med aktivitetsmåler gir oss mulighet for å validere ungdommenes egen rapportering i loggen.
5) Livskvalitet, modifisert Coop-Wonca.

6) Brukerevalueringsskjema for barn og unge fra CatoSenteret. Måler brukertilfredshet. Pårørende til ungdom under 16 år blir også spurt om å fylle ut at pårørendeskjema.

Spørreskjemaene ligger som vedlegg.
Metodene 2,4,5 og 6 er allerede i bruk ved tilbudet på CS.

Prosjektets betydning:
CFS/ME er en langvarig, til dels invalidiserende tilstand som representerer et av de viktigste helseproblemene blant ungdom i Norge i dag. De underliggende årsakene er for en stor del uavklarte, og det er også diskusjon om hva som er den beste formen for behandling og rehabilitering (Njølstad, Sveen, Bruun Wyller, 2012).

CatoSenteret, sammen med Valnesfjord Helsesportsenter, er de eneste rehabiliteringssentrene i Norge som tilbyr døgnopphold for denne målgruppen pr i dag. Det estimeres et antall på 600 barn og ungdommer som har diagnosen i Norge, CatoSenteret tar imot 50 av dem hvert år (altså drøyt 8 %). Dette burde bety at senteret er et velegnet sted å gjennomføre denne evalueringen. Det er fra flere hold understreket at det er et kunnskapshull knyttet til behandling av barn og unge med CFS/ME. Resultatene fra denne pilotstudien vil derfor i seg selv kunne være av potensielt stor betydning. En slik langtidsoppfølging vil videre gi en unik mulighet til å undersøke endringer over tid, samt å kunne justere behandlingsopplegget. I tillegg vil en slik pilotstudie være et første steg mot en større randomisert kontrollert studie som med større sikkerhet vil kunne si noe om effekten av et slikt opplegg. Pilotstudien vil her kunne bidra med erfaringer knyttet til bruk av ulike måleverktøy, inkludert en aktivitetsmåler som vil kunne gi en objektiv tilbakemelding på aktivitet, logistikk rundt innsamling av data, hvordan en kan implementere et forskningsprosjekt i en klinisk hverdag på et rehabiliteringssenter samt være grunnlag for en styrkeanalyse som vil kunne si noe om hvor mange deltagere som bør inkluderes i studien.

Resultatene etter denne pilotstudien vil også kunne være interessante fordi det gir en mulighet til å sammenligne et rehabiliteringsopplegg basert på GAT med andre tilbud barn og unge med CFS/ME mottar.

Resultatet av prosjektet vil bli spredd i relevante nasjonale fagmiljøer gjennom presentasjoner på konferanser, artikler i fagtidsskrift og gjennom nasjonale nettverk.

Framdriftsplan/tiltaksplan:

2014:

- Oppstart av målinger på formøte 18. august
- 3 grupper à 8 ungdommer vil bli igangsatt i løpet av høsten 2014

2015:

- Innhenting av 3 mnd data (T3) fra 1. januar til 1. mars
- Bearbeiding av data og sammenligning med data fra T1 og T2
- Innhenting av 12 mnd data (T4) fra 6. oktober til 1. desember
- Fortløpende bearbeiding av data T1 – T4

2016:

- Skriving av sluttrapport/oppgave/artikkel medio januar og februar

Risikofaktorer:

- Ungdommene ønsker ikke å delta i pilotstudien
- Aktivitetsklokkene fungerer ikke til å måle aktivitet
  – klokkene har allerede vært i salg en periode og tilbakemeldingene så langt er gode. I tillegg vil tre ansatte ved senteret prøve ut klokkene før oppstart.
- Bruk av aktivitetsklokker som måleverktøy kan føre til "stress". Det kan medføre ikke-reelle målinger
  - ved å ha klokken på en uke av gangen, mener vi at evt. stress vil avta mot slutten av måleperioden.
- Tidsaspektet – det er belastende for ungdom å følges opp over lang tid. Noen kan bli friske i prosjektperioden og ikke ønske spørsmål om tidligere sykdom.
- Prosjektleder får langvarig fravær. Organiseringen med prosjektkoordinator som skal kjenne prosjektet og arbeidet med dette godt, vil gi mulighet for å sette en evt. vikar inn i arbeidet.

Kontrolltiltak:

- CS har utarbeidet en egen prosjektstyringsmodell etter PM (Project Management) metoden. Denne ligger på vårt kvalitetssystem og er en beskrivelse av prosjektforløpet med tilhørende skjema for mandat, prosjektlederplan, tidsplan osv. Alle prosjekt blir forankret i ledergruppen, med eget mandat og styringsgruppe med deltagelse også fra ledelsen. Rapportering foregår til ansvarlig leder månedlig.
- Senterets utviklingsavdeling er organisert med en koordinator som har som oppgave å veilede prosjektleder underveis, samt koordinere i forhold til andre prosjekter.
- Det utnevnes controller fra administrasjonen som bistår med økonomisk oversikt og hjelper med regnskapet.
- En Ph.D stipendiat er ansatt som veileder og vil kunne gi råd om databearbeiding med mer.

Budsjett:

Innkjøp av "Polar Loop" aktivitetsmåler (20 stk) 700,-	14 000
Lønn med sosiale utgifter – prosjektleder i 100 % stilling – 01.08.14 til 01.04.15 (8 mnd) – 01.10.15 til 31.12.15 (2 mnd) – 01.01.16 til 31.03.16 (3 mnd)	 450 000 112 500 168 750
Controller	10 000
Prosjektkoordinator/ veiledningstjenester	50 000
Trykking, publikasjoner, utsending/distribusjon	10 000
Reiseutgifter, arrangement, møter, konferanser	50 000
Forbruksmateriell, kontortjenester, driftsutgifter	15 000
Sum utgifter	**880 250**
Omsøkt tilskudd fra H-Dir	697 500
Tilskudd fra CatoSenteret*	182 750
Sum inntekter	**880 250**

* Det er viktig å ha en lang oppfølging i innsamling av data. CS vil derfor bruke av sin egeninnsats for å gjøre bearbeiding av data og sluttrapport ferdig til 31.03.2016. I tillegg vil senteret selv kjøpe inn aktivitetsmålere.

Litteraturliste:

"Diagnostisering og behandling av kronisk utmattelsessyndrom/ myalgisk encefalopati (CFS/ME)". Rapport fra Kunnskapssenteret nr 09 – 2006. V.B.Wyller med flere.

"Kronisk utmattelsessyndrom. Kunnskap, aktivitet og utfordringer". Avhandling Universitetet i Bergen 2011. Lillebeth Larun.

"Behandling av kronisk utmattelsessyndrom CFS/ME". Kunnskapssenteret notat 2011. Larun, Brurberg, Fønhus, Kirkehei.

"Informasjon om kunnskapshull fra Kunnskapssenterets systematiske oversikter og HTA-rapporter publisert i 2011". Kunnskapssenteret notat 2012. Norderhaug, Mørland, Jamtvedt.

"Kronisk utmattelsessyndrom". Utposten nr 3, 2011. Vegard Bruun Wyller.

"Assiciation between school absence and physical function in pediatric chronic fatigue syndrome/ myalgic encephalopathi". Arch Dis Child 2009; 94: 752-756. E.Crawley, JAC.Sterne.

"Intervensions in Pediatric Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis: A Systematic Review." Journal of adolescent health, 53, 154-165, 2013. Knight, Scheinberg, Harvey.

Comparison of adaptive pacing therapy, cognitive behaviout therapy, graded exercise therapy, and specialist medical care for chronic fatigue syndrome (PACE): a randomised trial. The Lancet, vol 377, March 5, 2011. White, Goldsmith, et.al.

"Treningsbehandling ved kronisk utmattelsessyndrom". Tidsskriftet nr. 3, 2011; 131:231-6. Lillebeth Larun, Kirsti Malterud.

"Fysisk aktivitet som behandling ved Myalgisk Encefalopati". Ergoterapeuten 01.11. Irma Pinxsterhuis.

"Kognitiv terapi ved kronisk utmattelsessyndrom/ME". Tidsskrift for Norsk Psykologforening, Vol 46, nummer 9, 2009, side 828-836. Torkil Berge og Lars Dehli.

"Gradert aktivitetstilpasning hos ungdom med kronisk utmattelsessyndrom". Ergoterapeuten 03.12 (2012). Njølstad, Sveen, Bruun Wyller.

"Efficacy of cognitive behavioral therapy for adolescents with chronic fatigue syndrome: Long-term follow-up of a randomized, controlled trial". Pediatrics, 121, 619-625, 2008. Knoop,H., Stulemeijer,M., de Jong,L., Fiselier,T., Bliejenberg,G.

"Exercise therapy for chronic fatigue syndrome". Cochrane Library 2004, Issue 3. Edmonds,M., McGuire,H., Price,J.

"Ungdommers opplevelse av å leve med CFS/ME". Mastergrad i Helsefag 2012. Anette Winger, foredrag på Nasjonal konferanse om Kronisk utmattelsessyndrom, april 2013.

"Adolescent Sleep-Wake Scale". Til norsk ved Ståle Pallesen. LeBourgeois et al. (2005). Pediatrics, 115, 257-265. Fra Nasjonalt kompetansesenter for søvn, Haukeland sykehus HF.

"Adolescent Sleep Hygiene Scale". Til norsk ved Ståle Pallesen. LeBourgeois, et al. (2005). Pediatrics, 115, 257-265. Fra Nasjonalt kompetansesenter for søvn, Haukeland sykehus HF.

" Development of a fatigue scale ". T. Chalder, G. Berelowitz, T. Pawlikowska, L. Watts, S. Wessely, D. Wright, and E. P. Wallace.. J.Psychosom.Res. 37 (2):147-153, 1993.

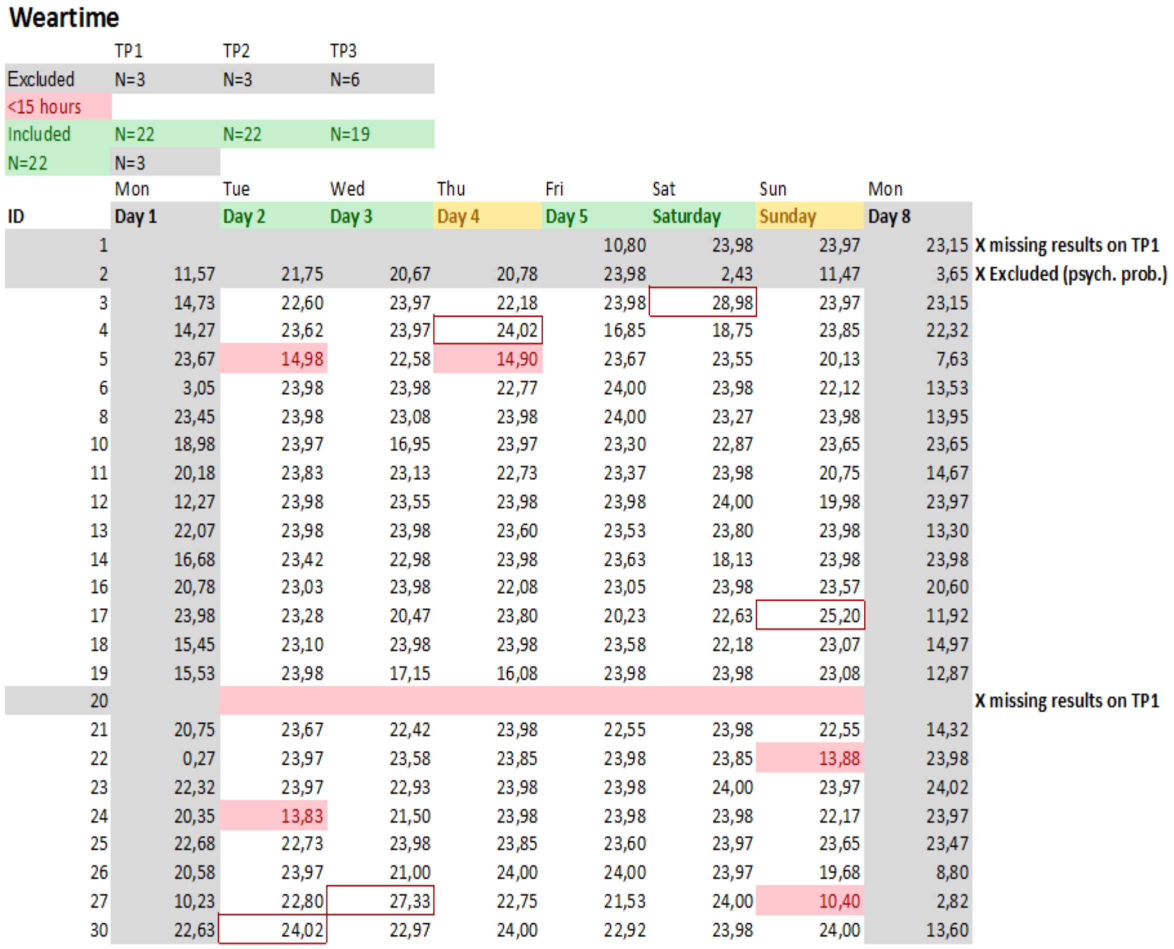
"Kronisk utmattelsessyndrom (CFS/ME) og arbeidsrettet rehabilitering". Helse Bergen, Haukeland sykehus, 2011.

"Effectiveness of internet-based cognitive behavioural treatment for adolescents with chronic fatigue syndrome (FITNET): a randomised controlled trial". The Lancet, Vol 379, April 14, 2012. Nijhof, Bleijenberg, Uiterwaal, Kimpen, van de Putte

Son 12.05.14

Bjørn Nordby	Hege Bruun-Hanssen
Styreleder	Kvalitets- og utviklingskoordinator
Stiftelsen CatoSenteret	Stiftelsen CatoSenteret

Appendix 7: Polar Loop wear time for all participants



Weartime

	TP1	TP2	TP3						
Excluded	N=3	N=3	N=6						
<15 hours									
Included	N=22	N=22	N=19						
N=22	N=3								
	Mon	Tue	Wed	Thu	Fri	Sat	Sun	Mon	
ID	**Day 1**	**Day 2**	**Day 3**	**Day 4**	**Day 5**	**Saturday**	**Sunday**	**Day 8**	
1					10,80	23,98	23,97	23,15	**X missing results on TP1**
2	11,57	21,75	20,67	20,78	23,98	2,43	11,47	3,65	**X Excluded (psych. prob.)**
3	14,73	22,60	23,97	22,18	23,98	28,98	23,97	23,15	
4	14,27	23,62	23,97	24,02	16,85	18,75	23,85	22,32	
5	23,67	14,98	22,58	14,90	23,67	23,55	20,13	7,63	
6	3,05	23,98	23,98	22,77	24,00	23,98	22,12	13,53	
8	23,45	23,98	23,08	23,98	24,00	23,27	23,98	13,95	
10	18,98	23,97	16,95	23,97	23,30	22,87	23,65	23,65	
11	20,18	23,83	23,13	22,73	23,37	23,98	20,75	14,67	
12	12,27	23,98	23,55	23,98	23,98	24,00	19,98	23,97	
13	22,07	23,98	23,98	23,60	23,53	23,80	23,98	13,30	
14	16,68	23,42	22,98	23,98	23,63	18,13	23,98	23,98	
16	20,78	23,03	23,98	22,08	23,05	23,98	23,57	20,60	
17	23,98	23,28	20,47	23,80	20,23	22,63	25,20	11,92	
18	15,45	23,10	23,98	23,98	23,58	22,18	23,07	14,97	
19	15,53	23,98	17,15	16,08	23,98	23,98	23,08	12,87	
20									**X missing results on TP1**
21	20,75	23,67	22,42	23,98	22,55	23,98	22,55	14,32	
22	0,27	23,97	23,58	23,85	23,98	23,85	13,88	23,98	
23	22,32	23,97	22,93	23,98	23,98	24,00	23,97	24,02	
24	20,35	13,83	21,50	23,98	23,98	23,98	22,17	23,97	
25	22,68	22,73	23,98	23,85	23,60	23,97	23,65	23,47	
26	20,58	23,97	21,00	24,00	24,00	23,97	19,68	8,80	
27	10,23	22,80	27,33	22,75	21,53	24,00	10,40	2,82	
30	22,63	24,02	22,97	24,00	22,92	23,98	24,00	13,60	

N=22	N=3								
	Thu	Fri	Sat	Sun	Mon	Tue	Wed	Thu	
ID	Day 1	Day 2	Saturday	Sunday	Day 5	Day 6	Day 7	Day 8	
1	23,57	23,98	23,65	22,83	12,97	23,67	23,98	9,63	**X missing results on TP1**
2	23,72	21,97	22,23	5,60	10,88	23,80	17,28	22,78	**X Excluded (psych. prob.)**
3	23,98	23,98	23,98	20,53	12,77	23,98	22,93	5,97	
4	23,63	23,92	23,98	22,32	23,70	15,52	30,65	23,98	
5	23,98	23,98	20,38	23,98	23,80	30,48	22,02	0,00	
6	22,30	23,98	23,98	21,35	23,98	23,98	23,98	14,63	
8	24,30	23,98	23,55	28,98	22,92	23,98	23,98	14,73	
10	23,63	23,97	23,57	19,97	18,62	21,90	23,98	11,85	
11	11,38	22,50	23,77	22,77	23,98	22,33	24,03	12,73	
12	10,50	23,98	23,98	17,60	15,95	23,98	23,65	13,58	
13	23,98	23,67	23,97	23,97	23,65	23,98	13,65	14,63	
14	10,90	23,73	17,42	6,70	0,00	12,03	17,82	12,23	
16	12,85	6,73	15,55	23,28	24,85	17,48	21,38	5,85	
17	0,00	9,23	23,97	15,00	23,80	24,00	21,15	23,98	
18	11,38	23,60	23,80	23,47	23,98	19,02	23,45	13,60	
19	8,50	22,67	9,55	23,98	23,98	20,68	23,98	13,52	
20	11,37	23,98	23,88	23,85	23,85	23,48	21,98	13,33	**X missing results on TP1**
21	0,00	9,85	23,50	23,73	23,10	23,98	22,65	13,27	
22	10,80	23,97	17,30	16,38	8,30	23,50	22,70	1,70	
23	10,18	23,97	23,75	24,00	17,47	23,98	23,97	9,70	
24	10,80	23,98	23,98	23,98	23,98	23,98	23,87	9,67	
25	10,80	23,97	24,00	22,32	10,80	23,98	23,97	9,73	
26	10,80	23,35	13,35	23,98	20,67	23,97	21,93	0,57	
27	10,78	23,98	22,95	22,35	22,93	22,08	23,63	9,62	
30	10,82	24,32	23,98	23,98	24,45	23,97	23,97	9,78	

N=18	N=7								
	Mon	Tue	Wed	Thu	Fri	Sat	Sun	Mon	
ID	Day 1	Day 2	Day 3	Day 4	Day 5	Saturday	Sunday	Day 8	
1	23,98	23,73	16,72	9,53	23,98	21,90	10,22	9,10	X missing results on TP1
2									X Excluded (psych. prob.)
3	23,97	23,98	23,98	0,58	24,00	23,98	24,00	23,98	
4	15,22	23,37	9,30	23,42	23,53	23,98	23,13	8,65	
5	18,90	23,82	23,98	22,70	24,00	22,78	23,98	22,52	
6	0,00	0,00	23,12	23,98	23,98	23,98	20,07	12,70	X missing results
8	23,03	23,98	23,98	22,07	23,98	23,98	23,98	14,83	
10	24,32	14,98	22,97	23,98	23,98	23,98	23,98	16,62	
11	11,52	24,15	23,97	23,47	22,50	21,95	23,43	12,67	
12	18,35	23,98	23,55	23,48	11,98	23,98	23,98	23,15	
13	23,98	23,85	23,73	9,85	23,98	21,85	14,72	0,07	
14	23,13	23,32	23,98	19,97	22,77	23,98	23,98	17,75	
16	0,00	0,00	0,00	0,00	0,00	0,00	0,00	0,00	X (bedbound)
17	14,18	15,82	20,28	19,98	22,97	23,97	23,43	23,10	
18	9,17	23,98	22,32	23,05	23,98	23,97	21,48	12,55	
19	15,37	12,70	4,13	23,97	23,98	23,98	23,98	13,60	
20	21,37	20,97	23,05	23,27	16,37	19,08	23,05	13,12	X missing results on TP1
21	23,22	23,55	23,28	23,33	23,37	23,65	23,98	17,07	
22	7,92	23,98	24,32	23,82	10,33	13,75	24,32	16,22	
23	3,15	23,98	23,97	24,00	21,67	0,00	0,00	0,00	X?
24	21,93	23,97	21,53	23,98	23,58	20,72	0,00	19,25	
25	23,63	24,30	23,98	23,98	22,17	23,77	23,25	11,50	
26	21,85	23,42	21,60	23,63	0,00	0,00	0,00	0,00	X?
29	13,40	23,98	24,00	19,22	23,97	16,50	21,10	12,60	
30	10,60	22,97	23,98	23,98	22,13	24,30	23,98	19,52	