# Curing ME with music?

May 8, 2020 · by melivet – Nina E. Steinkopf

**In a Norwegian study, a group of adolescents with chronic fatigue were treated with music therapy. The result is an article full of contradictions and illogical conclusions, which essentially disproves the researchers' own hypotheses and conclusions – and it seems the researches didn’t even discover it.**

Recently, a Norwegian research team at Akershus University Hospital published an article on a study in which young people with chronic fatigue were treated with a mental training program with elements of cognitive therapy and music therapy. The purpose was to see if the therapy could improve their level of function.

Three years ago, the CDC; US health authorities, removed the advice on cognitive behavioral therapy and graded exercise training as a treatment for ME. ME sufferers have reported zero efficacy and of deterioration due to these methods for 20-30 years. In this article, it is documented, and the researchers themselves don’t seem to have discovered it. Here are some key points.

This is a CFS/ME study although only 14% of the participants in the intervention group met strict CFS / ME diagnostic criteria. Participants were adolescents between the age of 12 and 20 with chronic fatigue after an acute EBV infection - mononucleosis. The level of function at baseline was 8000 steps per day, close to twice the level of ME in mild degree.

The study is based on a hypothesis from 2007 that CFS / ME is a sustained arousal response and that personality, emotions and psychological trauma contributes to disease development. The researchers claim that cognitive behavioral therapy and graded exercise therapy have a positive effect on CFS / ME and that there is no risk of side effects. There is no basis in neither the research literature or in broad patient surveys for such a claim.

The purpose of the study was to see if mental training with elements from cognitive therapy and music therapy could improve the level of functioning. The training program has many similarities with the pseudoscientific method Lightning Process.

Participants and their parents were told that personality, thoughts and feelings influence the development of the disease. They were informed of the importance of – as well as requirements of - motivation and an increasing effort from the patients to change the "situation". This was substantiated by a testimony from one person who had recovered himself.

The "situation", which was a long-term fatigue after an infection, is described by the research team as negative illness behavior, disease attribution and "avoidance". The hypothesis is that this contributes to maintaining the fatigue. The parents were involved in the therapy because the researchers believed that parents' influence could strengthen negative disease behavior and attribution, which in turn could have a negative impact on the prognosis.

The therapy aimed to control such negative thoughts and emotions that hinder increased activity. Music was used in order to evoke good feelings. The therapy was individually tailored, and the therapists knew in advance the results of extensive psychological tests that had been taken six months earlier. During the program, the focus was aimed at the personality of the children, the parents and family dynamics. Both participants and parents were thus implicitly made responsible for the "situation" and the outcome of the therapy.

The study reports on only one objective outcome measure; number of steps per day. The intervention group experienced a significant decline of 11% after 3 months, or 23% if another (per protocol) method of calculation is used. The mental training program provides no increase in measurable activity, actually the opposite. The results show a significant reduction in objectively measured activity level, even in participants who have a significantly higher level of function than the average ME patient. This surprised the researchers, but did not seem to change their conclusion but instead they recommend further studies. The researchers claim the high dropout rate is due to fear of school absence, and speculate that the reduced level of function is due to the participants not being treated with graded exercise therapy in addition.

The article defines "recovered" as a certain drop in subjectively reported fatigue. The study concludes with 65% recovery in the intervention group after 15 months. The results show that 8 participants recovered - compared to 7 in the control group. At the same time, participants in the same group became more depressed, more fatigued, had more pain and a lower level of function. 8 of the 21 participants withdrew along the way. The results of the clinical examinations and biomarkers at follow-up, cf. the study protocol, is not included in the article.

The number of "recovered" is oversold and negative effects are understated. The only objective measurement point disproves the researchers' hypothesis and the article's conclusion.

Representatives from the organisation Recovery Norway were involved in the design of the study design. The organisation was formally established in January 2018. The study design is dated June 2015. The organization started by amongst others a group of Lightning Process instructors, and is marketing and lobbying for the method.

The study report is peer-reviewed. One assessment is made by a member of the British research community referred to in the study. The second assessment is done by an IT researcher who has only read the abstract. Both state that the conclusions are exaggerated.

Although the results are negative, the researchers interpret them as positive and believe the study can justify a full-scale clinical study.

The study's findings largely contradict their own hypotheses and conclusions about the benefits of cognitive and music therapy - without this being commented on in the article. The researchers seek to confirm their own hypothesis by selectively selecting results, drawing unfounded conclusions, and overselling them. This is cherry-picking. The study is another example of poor research that reflects the researchers' predetermined psychosomatic view, rather than an honest scientific study.

This study is likely to be referred to by the health authorities. It can be used in treatment recommendations, and can be used in decisions made by the Norwegian Labour and Welfare Administration/NAV. This can lead to patients being treated with inactive or harmful treatment methods. ME patients deserve better research.

## Review of the study

I would like to point out that I have been in contact with the senior investigator, Vegard B. B. Wyller. He was willing to answer questions about the study, and at the same time was offered the opportunity to make a public comment on this analysis/review. As of yet, he has not answered any questions.

1. The study

The article is called “Cognitive – behavioral therapy combined with music therapy for chronic fatigue following Epstein-Barr virus infection in adolescents: a feasibility study”, BMJ Paediatrics Open 2020; 4: e000620. doi: 10.1136 / bmjpo-2019-000620

Research team: Sadaf Malik, Tarjei Tørre Asprusten, Maria Pedersen, Julie Mangersnes, Gro Trondalen, Betty van Roy, Eva Skovlund, led by Vegard B. B. Wyller. The study is approved by Regional Committees for Medical and Health Research Ethics (REK) and funded by Akershus University Hospital / Health South-East.

## 2. Study Design

The participants were 43 children and adolescents between the age of 12 and 20 who had developed post-infectious chronic fatigue following an acute EBV infection (mononucleosis). Of these, 21 participants were treated with therapy over 3 months (the intervention group), and were compared with the control group of 22 who did not receive treatment. The purpose of the study was to see if a recently developed mental training program, with elements from cognitive behavioral therapy and music therapy, could have a positive effect. Effects on symptoms, function and underlying disease markers would be examined immediately after treatment termination and 1 year later.

The project was approved as a blinded, randomised controlled trial (RCT) with high estimate of effect. The article, on the other hand, describes a feasibility study with estimate of effect.

## 3. Participants

The project is presented as a CFS / ME study. The article uses several different illness definitions, contributing to the confusion that often arise in discussions on ME research. Post-infectious chronic fatigue has diagnostic code 8E49 in WHO's diagnostic code system IDC-11, and includes Benign myalgic encephalomyelitis (ME) and Chronic Fatigue Syndrome (CFS). Norwegian health authorities use the term CFS / ME and recommend the Canadian criteria for diagnostics. In the intervention group, only 14% met these criteria.

Myalgic Encephalopathy (ME) has been classified as a neurological disease by the WHO since 1969. Among the core symptoms are debilitating fatigue, pain, brain fog, hypersensitivity to light and sound, non-refreshing sleep and post exertional malaise (PEM). The triggering factor is often an infection. Hypersensitivity to sound is typical to ME. It is described in both the Ramsay Criteria 1986, the Canada Criteria 2003, the International Consensus Criteria 2011, the IOM Report 2015, the Directorate of Health’s guideline of CFS / ME 2015, and the US health authorities' information on the disease from 2019.

Patients with a mild degree of the disease may use and enjoy music. However, for severe and very severe ME patients, all sounds - including beautiful music - can lead to deterioration.

## 4. Hypothesis

The study is based on the hypothesis that CFS / ME is a suspended stress response, sustained arousal, and that personality, thoughts, feelings and life events contribute to persistent illness after an EBV infection. The theory is that CFS / ME can occur as a result of classical and operant conditioning; that ME is a form of learned state / behavior. There is no basis for claiming this about ME defined by strict diagnostic criteria.

Participants were recruited from the CEBA study, which had 200 participants. It found no difference in scores on perfectionism or psychological trauma for those with EBV infection and healthy controls. The researchers refer to the 2015 U.S. IOM Report, which states that no other studies have suggested a higher rate of childhood trauma in those with confirmed ME / CFS as opposed to non-specific chronic fatigue. Also, Vegard B. B. Wyller's own study from 2009 showed no significant psychiatric or psychosocial differences between fatigued adolescents with or without autonomic dysregulation.

## 5. Background material

The research protocol states that “Despite strong research efforts, no pharmaceutical has been proven beneficial in CFS/ME. In contrast, evidence suggests a beneficial effect of mental techniques, and CBT in particular».

It is not true. Little biomedical research has been done on ME. Read more about the size of research funding in the US to ME in comparison to other diseases here.

Neither is there any basis in either the research literature or in broad patient experiences for the claim that: "Cognitive behavioral therapy (CBT) is effective in chronic fatigue syndrome".

The authors refer to a number of scandalous studies that have major methodological errors:

In the PACE study, participants were described as cured even though they had poorer functional levels at the end of the study compared with the start of study. The hypothesis is that ME patients have activity phobia.

Read more in Aftenposten: Does cognitive therapy improve ME?

In the FITNET studies, there was no effect of the intervention at long-term follow-up, and the results showing that there was no objective improvement are not included in the report.

In the SMILE study, no objective endpoint was used. It has a 3000 word long correction due to. violation of basic ethical and methodological principles. Half of the participants were registered retrospectively, and the endpoints were changed midway through the study.

Lillebeth Larun's Cochrane review has been revised several times and is now under a new, complete review.

Reference is also made to Bjarte Stubhaug's controversial study which claims to have cured 8 out of 10 ME sufferers in 4 days, but where it is uncertain whether there was ME sickness among the study participants.

In these studies, no diagnostic criteria have been used that require tPost Exertional Malaise (PEM) or where PEM is a mandatory symptom. If the assumption of PEM as a cardinal symptom by ME is considered by the Health Directorate and US health authorities, there are no studies showing that cognitive behavioral therapy, graded exercise therapy or similar interventions are effective in treating ME.

Read more in the AHRQ report; Diagnosis and Treatment of Myalgic Encephalomyelitis / Chronic Fatigue Syndrome, which summarises these methods, and does not find effects in studies with strict diagnostic criteria.

## 6. Treatment

The treatment was based on the hypothesis that CFS / ME is a suspended stress response, sustained arousal and that the disease may occur as a result of classical and operant conditioning; that ME is a form of learned state / behavior. The researchers believe it is not useful to look for the cause of the illness, as patients often tend to misinterpret. Important elements of the treatment were to identify and control negative thoughts and feelings.

The intervention consisted of 10 weeks with a recently developed mental training program with elements from cognitive behavioral therapy and music therapy. The introductory session was conducted by a researcher, a music therapist and a cognitive therapist, with both the patient and parents / guardians present. The participants and their parents received psychoeducation. Psychoeducation is information about mental disorders and how to beter master them. Read more about psychoeducation as treatment at Oslo University Hospitals website. Personal experiences were shared by a young adult volunteer who had recovered from CFS himself.

At the start of the program, music therapy elements were used to evoke good feelings. Part of the treatment consisted of visualization. The researchers claim that visualization has been shown to have a positive effect on CFS / ME and related conditions, pointing to a study that does not address CFS / ME - but somatization disorders.

Somatization disorder is, according to psychiatrist Ulrik Fredrik Malt, a chronic and debilitating psychosomatic disorder: «Early traumatization (sexual abuse, abuse) can lead to somatization disorder in adulthood. About one in five with somatization disorder has limited intellectual resources at their disposal for the development of somatization disorder».

There is no basis in science to claim that CFS / ME is a mental disorder or a somatization disorder. Also, as mentioned in Chapter 4, there is no indication that CFS / ME patients have an over-frequency of childhood trauma. When diagnosed according to The Canada criteria, psychological causes for symptoms must be excluded before diagnosis can be given. Thousands of research articles have been published all over the world that consistently show abnormalities in the immune system and in energy production in ME patients.

Participants were told that they had the possibility, ability and responsibility to influence the "situation" through self-effort. The therapist should possess specialist authority, trust as a therapist, empathy, and establish a trusting relationship with the children.

The "situation", which was prolonged fatigue after EBV infection, is described as negative disease behavior and disease attribution. Reduced activity levels as a result of illness are referred to as "avoidance" - avoidance of activity - and it is said that this will, after a while, contribute to maintaining the fatigue. The children were thus also implicitly made responsible for the fatigue and the result of the therapy.

The parents became involved in the therapy because the researchers believe that the parents' influence can help to reinforce negative illness behavior and attribution, which in turn can have a negative impact on the prognosis. The parents were thus also implicitly made co-responsible for contributing to maintaining the children's illness, and co-responsible for the outcome of the therapy. This can lead to significant and systematic bias in self-reporting.

The participants had filled in many questionnaires upon inclusion to another study, the CEBA study, six months earlier. In addition to questions about chronic fatigue and pain, they answered questions about worry, sleep, anxiety and depression, perception of illness, perfectionism, quality of life, functional disability, psychological trauma and difficulty in expressing thoughts and feelings. They also conducted a full IQ test. The results were used in the therapy. In the 4th meeting, with children, parents and 2 therapists present, the patient and family were surveyed. The therapist's task is described as follows:

“Mapping out the patient and the family: Before the conversation much information regarding the patient is already known from the questionnaires (particularily anxiety, depression, sleeping problems, problems with executive functions (attention, working memory), perfectionism and worries). Use this and anamnestic information to make a fundament for individual adapted treatment in the rest of the treatment program.”

“Go over the rationale once again and try to establish a common understanding.”

“Focus on building alliances – create good frames for learning.”

Patients had to be motivated to work on identifying negative thought patterns, practicing techniques to stop such thoughts, and increasingly working on illness behavior.

Throughout the program there is a clear expectation that the patient will improve. Not surprisingly, some report it in some areas, although the results are complex and the objective test (step measurement) shows otherwise. This is a phenomenon known as "pleasing", that in surveys people give the answer that they think interviewers want. In such studies where one develops a good relationship with the respondent - who in this case are easy-to-influence children and adolescents, one will be very vulnerable to such systematic bias.

## 7. Alternative Treatment

The treatment manual shows that the therapy has many common features with the alternative method Lightning Process (LP). LP is largely about taking control of thoughts and feelings, and responsibility for one's own health and recovery. The first part of the LP course is psychoeducation: that thoughts, feelings and behaviors affect the body. The method is broadly based on making choices: When the participants get symptoms, they should become aware of thoughts and feelings: that they are heading down the ditch. Then they’re supposed to say Stop! - take control, and then make a choice: They must choose between the ditch or turn their minds around and thus get the life they love. The participants must be motivated, they must want to be healthy, and be willing to make an effort themselves. Important elements are motivation and belief that you can succeed. Participants should tell themselves and others that they have recovered. The course may also include parents and their influence.

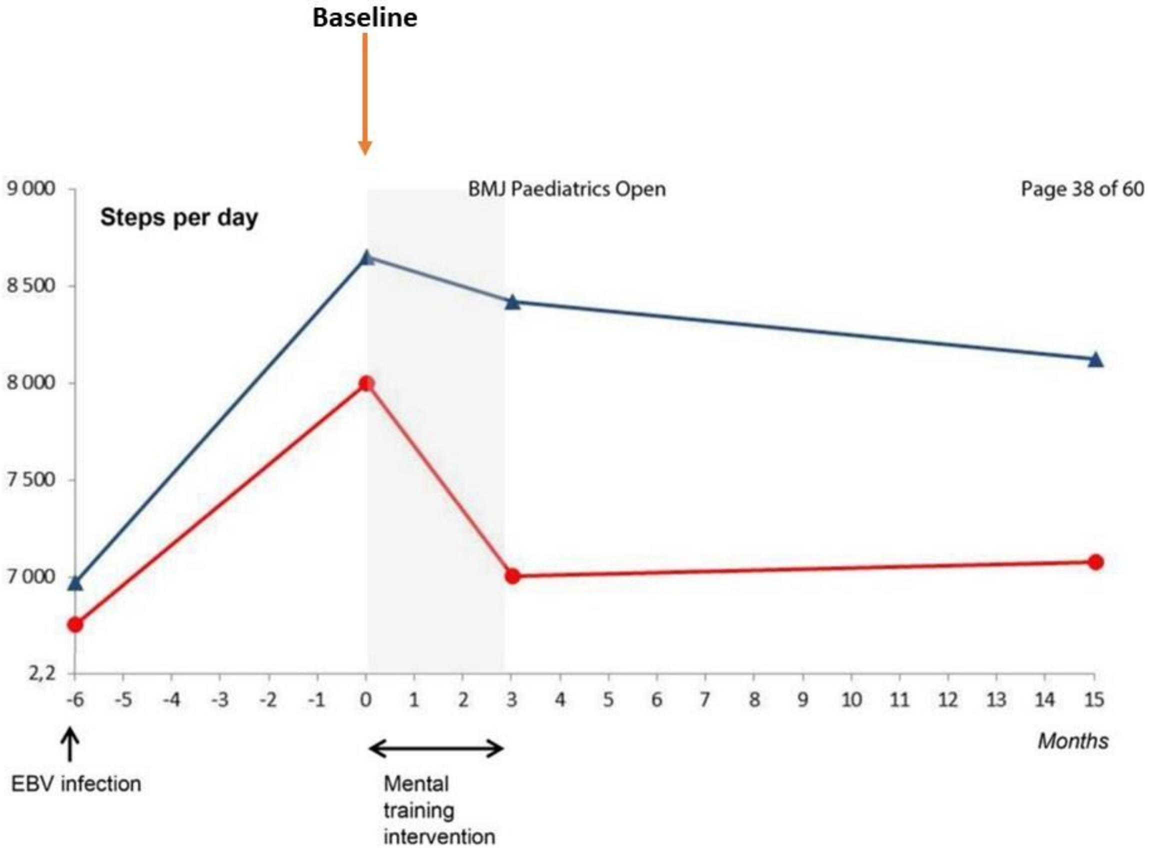
VG: LP instructor believes that: More children can get healthy if the parents manage to turn their attention from the children's symptoms to their own fears and despair.

Sadaf Malik, one of the therapists in the study, was an observer on an LP course in September 2014. She recommends LP: Aftenposten: Psychiatrist received ethics complaint from patient association, the Council for Medical Ethics praised the psychiatrist.

Project Manager Vegard B. B. Wyller, has long recommended children with CFS / ME to treated with LP: NRK: 13 year old attempted suicide after ME course.

## 8. Results

At baseline, participants had approx. 8000 steps per day. In comparison, participants in another CFS / ME study from the same research group had 4500 steps per day. The number of steps decreased to 6200 during the treatment period and the negative effect persisted.



Subjective measurement points are unreliable, especially when the purpose of the study is to change the participants' perception of their own illness. The results show that the changes at the subjective endpoints are marginal. In the intervention group, fatigue was reduced by 3% and PEM was reduced by 10% during the treatment period. The trend was similar to that of the control group at 15 months follow-up.

Pain increased in the intervention group after 15 months, but decreased in the control group. After three months, depression in the intervention group increased, but decreased in the control group.

9. Negative effect

According to Table 5 in the article, 27% of participants in the intervention group were more depressed at three months of follow-up than at baseline. In the control group this was only 5%. 23% of participants in the intervention group were more depressed at 15 months of follow-up than at baseline. In the control group this was 0%.

Despite the fact that 38% of participants withdrew from the study, despite the fact that the participants in the intervention group were more depressed, more exhausted, more in pain and had a lower level of functioning - it was concluded that no harmful effects were recorded.

The researchers were surprised that the level of activity was reduced, and wonder if it could be because the participants were not treated with additional graded exercise therapy.

"…… graded exercise therapy (GET) in itself is shown to have beneficial effects in CFS….. Our mental training programme did not contain this element, which may explain why physical activity to our surprise actually tended to decline in the intervention group during the treatment period.”

There are no grounds based basis on research studies or broad patient studies to claim that this type of therapy has a positive effect on CFS / ME, see also Chapter 5. On the contrary, it is well known that graded exercise therapy can be very damaging to ME. US health authorities have removed the recommendation of this type of treatment.

Instead of assessing whether the cause of the adverse development could be the therapy itself, the researchers consider whether the reason was that the participants were not treated with even more therapy.

10. Recovery

"Recovered" is considered a certain drop in scores on subjectively measured fatigue. The article states that "After 15 months of follow-up, there was a trend towards higher recovery rates in the intervention group (62% versus 37%)". This is correct, but the percentage is high because 38% of participants withdrew. Table 4 shows that:

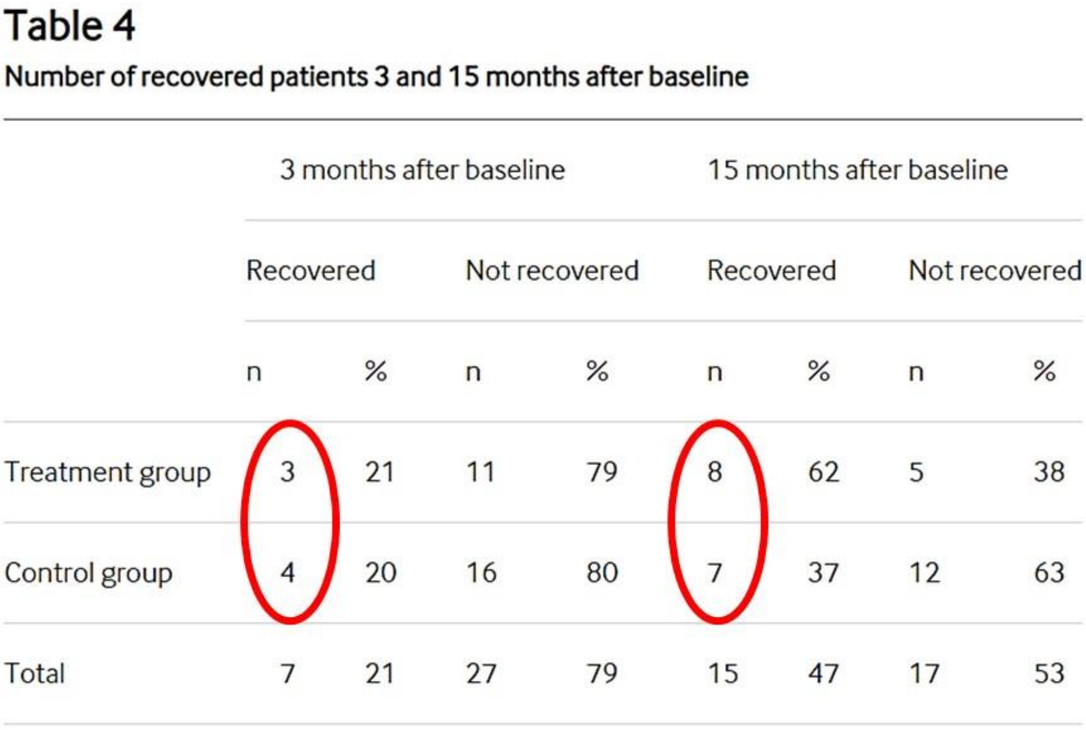


**Table 4**

**Number of recovered patients 3 and 15 months after baseline**

| | 3 months after baseline | | | | 15 months after baseline | | | |
|---|---|---|---|---|---|---|---|---|
| | Recovered | | Not recovered | | Recovered | | Not recovered | |
| | n | % | n | % | n | % | n | % |
| Treatment group | 3 | 21 | 11 | 79 | 8 | 62 | 5 | 38 |
| Control group | 4 | 20 | 16 | 80 | 7 | 37 | 12 | 63 |
| Total | 7 | 21 | 27 | 79 | 15 | 47 | 17 | 53 |

After three months, 3 in the intervention group had recovered, and 4 in the control group.

After 15 months, 8 in the intervention group had recovered, and 7 in the control group.

At baseline, there were 21 participants in the intervention group and 22 in the control group. The article provides no information on the 8 of the intervention group who withdrew along the way.

The differences are marginal and may as well have been random. Patients were trained in how to interpret the symptoms and how to interpret a deterioration. That is perhaps some of the explanation as to why some have responded that they were recovered - despite measurements showing that it might not be correct.

The figures must be seen in light of the fact that 38% of the intervention group withdrew from the study and that participants in the same group became more depressed, more exhausted, more in pain and had a lower level of functioning. The result is oversold by including only one of the subjective goals. The results would be more precise if the researchers also took into account the results of biomarkers and clinical studies at follow-up.

## 11. Secondary endpoint

According to the project description, a number of physiological and biological samples were supposed to be taken before and after treatment. In an application to REK to make changes to the study dated March 2018, it’s stated that: "We investigate effects on both symptoms (fatigue), function (number of steps) and underlying disease markers (immune function, nerve function, etc.) immediately after treatment termination and 1 year later." According to the protocol, patients should be thoroughly examined at week 12 in an assessment program identical to that performed at inclusion:

Biomarkers: plasma catecholamines, urinary-cortisol / creatinine ratio, cytokines, NK cells and gene expression profiles. Autonomic cardiovascular control: heart rates and standing blood pressure response. Cognitions / neurobiology: Working memory, cognitive impairment and brain imaging.

The results of these follow-up studies are not reproduced in the article. It’s crucial for the study's credibility to have objective tests both before and after, since the goal of the therapy is to change the course participants' thoughts, feelings and behaviors - and thus their level of function.

## 12. Dropouts

Eight of the 21 participants in the intervention group withdrew from the study. The researchers claim the high dropout rate is due to the fear of school absence. The article does not consider whether the youth - given the effect of the therapy and the expectations and responsibilities imposed upon them, may have blamed on school absence by politeness.

Neither has it been reported, as is often the case in scientific studies - whether the group that dropped out differs from the group that was still in the study.

## 13. Conclusions and recommendations

The results disprove the hypothesis. Nevertheless, the researchers conclude that an intervention study seems acceptable to participants and that "the tendencies of positive effect on patients' symptoms and recovery may justify a full-scale clinical trial".

## 14. User involvement

The protocol states that the Norwegian ME Association / MENIN should participate. The article states that representatives of the organization Recovery Norway (RN) were involved in the design of the study. RN was established in September 2017 and formally funded in January 2018. The study design is from *June 2015*.

RN is an interest organization for practitioners in alternative treatments, who, under the guise of being a "user organization", markets Lightning Process and lobbies with the help of satisfied customers. The organization was started by a number of LP instructors, claiming that the illness can be cured by changing thoughts, feelings, behaviors and relationships.

There are around 10,000 ME sufferers in Norway. The Norwegian ME Association has 5300 members. As of December 2019, RN had 181 members and is not representative of the patient population.

## 15. Peer review

Two peer reviews have been made. One is by Prof. Maria Loades. She is part of the British research community referred to in the article and the protocol, and has the same approach to the disease as the authors. It would be appropriate to have the study report evaluated by neutral peer reviewers.

The second review is by Jeremy Miles, an IT researcher at Google who has only read the abstract. He writes: "The conclusion in the abstract is overstated, in my opinion."

## 16 Reviews

The task of research is falsification; to try to disprove ones their own hypotheses. The article is a good illustration of the opposite; researchers who selectively pick results to support their own hypotheses. The result is a study full of contradictions and illogical conclusions, which essentially disproves own hypotheses and conclusions. The approach may be reminiscent of victim blaming, and is particularly serious since the positive effect of the treatment was not only absent - but because adolescents even experienced lasting deterioration.

How does it affect easily influenced sick children and adolescents being told that their illness is actually psychological - and that they themselves are responsible for recovering? How did parents feel about being co-responsible for recovery and contributing to "negative illness behavior"? How did the participants and parents feel when they thought the children would get treatments with music therapy - and discovered that the focus was on them personally, on the parents and on family dynamics?

Thousands of research articles have been published from all over the world that consistently show abnormalities in the immune system and in energy production in ME patients. It has been three years since US health authorities removed the advice on cognitive behavioral therapy and graded training as a treatment for ME. ME sufferers have reported zero effect and deterioration due to cognitive and graded exercise therapy for years. In this article, it is documented, without the researchers themselves seeming to have discovered it. ME patients deserve better research.

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