

Effects of a High-Intensity-Interval-Training (HIIT) in Charcot-Marie-Tooth – A Single Case Study

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Abstract:

Background: Up to now, Charcot-Marie-Tooth disease is not curable. Only symptoms are used to be treated with conventional therapy like physical exercise. Many studies have been conducted to investigate diverse training therapy concepts, but there is no study available that investigates the effect of a High Intensity Interval Training. This is the aim of this study.

Materials and Methods: A 40 year-old female Charcot-Marie-Tooth patient underwent a high intensity interval training intervention (HIIT) over 12 weeks, 1 x/week 30 minutes, each set lasting 45 seconds with a rest between sets of 15 seconds. Investigated were depression (BDI-II), fatigue (FAS), and motor fitness before and after the intervention.

Results: Improvements in raw values in ball grasping, figure-eight circles and walking backwards (coordination), stand & reach and shouldering out (flexibility), sit ups and push ups (strength). The rest of the variables shows no difference in the posttest compared to the pretest.

Conclusions: HIIT seems to be effective to strengthen trunk muscles, to improve flexibility and coordination abilities. The tasks standing longjump and 2 km walking were not improved, which could be associated with the foot lifter muscle weakness, so the strengthening of these muscles should be included into physical exercise training. For this purpose, Whole Body Vibration could be recommended.

Keywords: Charcot-Marie-Tooth, Hereditary Motor Sensory Neuropathy, Exercise therapy, Rehabilitation, High Intensity Interval Training

1. Introduction

Charcot-Marie-Tooth (CMT) is the most common neurogenetic disorder. Approximately 20 to 30 people per 100,000 population suffer from it. The disease is mostly inherited in an autosomal-dominant manner. Therefore, there are clusters in individual families. In most cases, the cause is a mutation on chromosome 17. In CMT, the peripheral nerves are affected. The insulating myelin layer of the axon of the nerve cell is damaged by a gene mutation. This layer acts like plastic insulation around an electrical wire. In CMT, the

transmission of nerve impulses in peripheral nerves is impaired. As a result, commands from the brain do not reach the muscles or do not reach them properly. Denervation leads to weakness and degeneration of the affected muscles. Typical early symptoms include weakness or paralysis of the foot and lower leg muscles, which can lead to difficulty lifting the foot and frequent stumbles or falls. Affected individuals also have problems with balance. Foot deformities are also common in CMT. As the disease progresses, weakness and atrophy in the hands can lead to difficulty with fine motor skills. Degeneration of sensory nerve axes can lead to a decreased ability to feel heat, cold, and tactile sensations. Proprioception is often diminished in people with CMT. The condition can also cause curvature of the spine (scoliosis) and hip displacement. Many people with CMT develop contractures that prevent joints from moving freely. Muscle spasms are common. Nerve pain can range from mild to severe. Symptoms progress gradually. The gene mutations in CMT are inherited in three different patterns: autosomal dominant, autosomal recessive, and X-linked, all of which are linked to a person's chromosomes [1,2]. The following figure 1 demonstrates the health problem CMT in the sense of the International Classification of Functioning, Disability and Health ICF by the World Health Organization WHO [3].

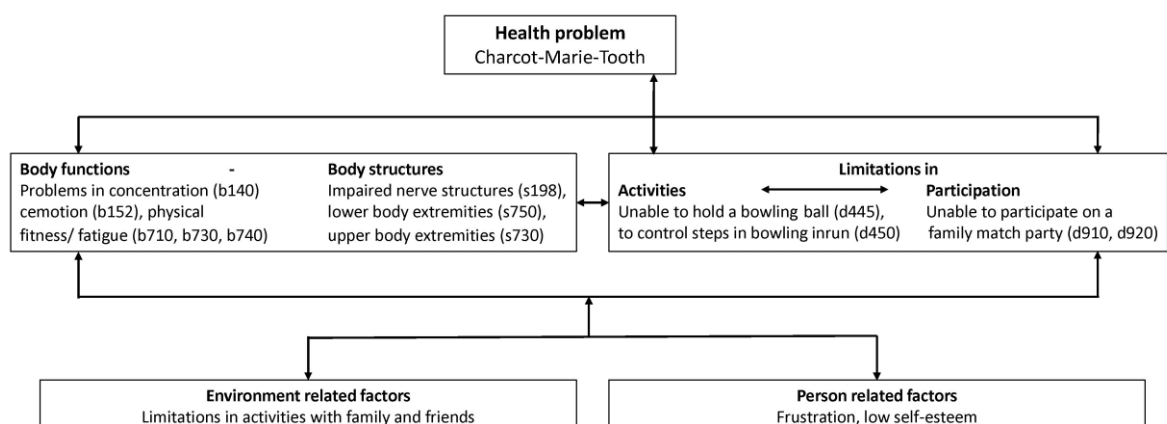


Figure 1. Meaning and impact of CMT in terms of ICF (adapted from [3])

A person suffering from CMT has damage to the nerve structures, lower (especially foot elevator muscle) and upper extremities (especially hand). As a result, problems in physical fitness and fatigue develop. Due to these problems, the person is unable, for example, to hold a bowling ball and coordinate the steps before the throw. Because of this, she is unable to participate in a bowling match at a family party. In the area of person-related factors, frustration and low self-esteem are evident, and in the area of environment-related factors, limitation in activities with the family.

CMT is not yet curable, one only tries to improve the symptoms or to keep the current state. This is done, for example, with orthoses or splints, but also with physiotherapy or sports therapy. In the meantime, so many studies have been conducted with

different training interventions that several reviews and meta-analyses have been made about the effectiveness of these trainings [4,5,6]. Here it was summarized that balance training has the strongest positive effect on gait parameters, balance and mobility [7]. However, it can be seen in these papers that no high intensity training has yet been tested for its effectiveness.

The positive effect of general physical exercise on the symptoms of various diseases has been demonstrated, such as depression [8,9] or fatigue [10,11].

A special form of physical exercise is high intensity interval training, or HIIT. This is characterized by the fact that the individual exercises are performed at high intensity (e.g., with heavy weights or as many repetitions as possible per time unit) in intervals with short breaks in between. Here, either exercises are performed with the own body weight or equipment such as medicine ball, dumbbells or theraband are added, so that an individual load is possible [12,13]. Some studies and reviews illustrate the effectiveness of this form of exercise on strength [14] and on depression [15] in different groups of individuals or patients. However, no study was found that explored this form of training in CMT patients, nor the effectiveness on flexibility and coordination. For this, the present single case study wants to investigate the effect of HIIT in CMT.

Hypothesis

A 12-week HIIT workout will improve physical fitness, depression, and fatigue in CMT.

2. Material and Methods

Sample of persons

One female participant, 40 years old, light physical activity for about four hours per week like working in house and garden and two hours per week moderate physical activity like walking and going by bicycle to work. No comorbidities, 1.75 m tall, 85 kg. CMT was diagnosed by a neurologist at the age of 20 because of foot lifter weakness and known disorder of the father.

Variable sample

BDI-II: Beck Depression Inventory, second edition. Here, a total of 21 questions are answered regarding pessimism, sadness, feelings of failure, loss of pleasure, self-worth, fatigue, energy, etc. Between 0 and 3 points are awarded per task. All points are added to a total score. A classification of the total score is made as follows: 0 to 13 points = no depression, 14 to 19 points = mild depression, 20 to 28 points = moderate depression, 29 and more points = severe depression [16]. The total score of the questionnaire is evaluated.

FAS: Fatigue Assessment Scale. The questionnaire consists of ten questions about physical and mental fatigue, tiredness, concentration, etc. The total score of the questionnaire is evaluated. Between 1 and 5 points are awarded for each task. All

points are added up to a total value. A classification of the total score is made as follows: 10 to 21 points = no fatigue, 22 to 34 points = moderate fatigue, 35 to 50 points = extreme fatigue. A minimally important difference is given as 4 points or 10% change [17]. The total score of the questionnaire is evaluated.

Motor fitness test: Testing of motor abilities (fitness) with the tasks hop run, ball grasping, ball throw with rotation, figure-eight circles in one-leg stand, walking backwards (coordination), sit-ups, push-ups, standing long jump (strength), shouldering out, stand & reach (flexibility) and 2 km walking (endurance). With the exception of walking, the subject has two attempts at each task. The items are scored according to age and gender [18]. The raw scores and age- and gender-normalized scores of the tasks as well as the total score are evaluated.

Treatment sample

High intensity interval training HIIT once a week for 30 minutes over a period of 12 weeks. Sets had a duration of 45 seconds with 15 seconds rest between the sets. In each session, there was one exercise of four sets for arms (e.g. dips or pushups, biceps curls), legs (e.g. squats or lunges), back (e.g. superman or hyperextensions, rowing) and abdominals e.g. crunches or leg raises).

Study flow

The study was approved by the ethics committee of Saarland University, application number 21-15 and registered at the Deutsches Register Klinischer Studien, application number DRKS00029150. All tests and interventions were performed at Saarland University.

3. Results

The following table 1 provides an overview of the results in pre- and posttest in BDI-II, FAS, and motor fitness test.

Table 1. Results in pre- and posttest of the Beck Depression Inventory BDI-II, Fatigue Assessment Scale FAS, and fitness test (RV = raw value, SV = standard value)

Variables	Pretest	Posttest	Change
BDI-II	5	6	+1
FAS	26	27	+1
Hop run RV (number of correct hops)	5	5	0
Ball grasping RV (number of correct grasps)	1	2	+1
Ball throw with rotation RV (number of correct rotations)	0	0	0
Figure-eight circles RV (number of correct eights)	3	5	+2
Walking backwards RV (m/s)	Not possible	0.33	+0.33
Stand & Reach RV (cm)	-17	-13	+4
Shouldering out RV (cm)	93	78	-15
Situp RV (number of correct situps)	0	8	+8
Push up RV (number of correct push ups)	6	8	+2
Standing long jump RV (cm)	65	63	-2
2 km Walking RV(min)	20:35	20:37	+0:02
Hop run SV	1	1	0
Ball grasping SV	0	0	0
Ball throw with rotation SV	0	0	0
Figure-eight circles SV	0	1	+1
Walking backwards SV	0	2	+2
Stand & Reach SV	0	0	0
Shouldering out SV	0	1	+1
Situp SV	0	0	0
Push up SV	0	1	+1
Standing long jump SV	0	0	0
2 km Walking SV	0	0	0
Fitness Total Value	0.50	2.85	+2.35

There are both decreases in values (shouldering out RV, standing long jump RV), and increases in values (BDI II, FAS, ball grasping RV, figure-eight circles RV, walking backwards RV, stand & reach RV, situp RV, push ups RV and SV, 2 km walking RV, figure-eight circles SV, walking backwards SV, shouldering out SV and Fitness total value). The rest of the variables shows no difference.

4. Discussion

This single case study wanted to investigate the effects of a HIT program on depression, fatigue, and motor fitness in CMT.

Improvements and consistent results are shown throughout.

The test person showed a stable result in the BDI-II. She already showed no depression in the pretest with a value of < 13 points [16], as well as in the posttest. Gu et al [15] describe a small effect of HIIT on depression, whereby the effect is stronger in comparison to usual care than in comparison to classic fitness training. However, since the test person showed no signs of depression in the pretest, it was not expected that the result would improve in the posttest.

In the FAS, a stable result can be observed. In the pre- and posttest, the result is to be classified as moderate fatigue [17]. Since the value is in the lower or moderate range, no improvement was expected at this point. Possibly a person with severe fatigue would show a positive change.

With an age of 40 years, the test person is in the range of middle adulthood, which is characterized by a decrease in motor performance if no training takes place [19]. There is a clear difference between exercisers and non-exercisers in motor abilities and skills [20].

In the area of physical fitness, there are improvements in the raw values in walking speed when walking backwards (in the pretest, this task could not be performed), in ball grasping and the figure-eight circles task. So, the standardized values in figure-eight circles (from zero to one point) and walking backwards (from zero to two points) are improved, too. No study has yet been found on the effect of HIIT on coordination ability. The positive change could be attributed to motor plasticity. Winter and Hartmann [19] describe that in middle adulthood, the coordinative performance decreases, but a constant or improved level can be achieved through sports activity.

The stand & reach as well as the shouldering out improved in raw values, even though in the standardized values, only an improvement in the shouldering out is visible. Similarly, no study was found on the effect of HIIT on flexibility. The positive change here could also be attributed to motor plasticity. Here it is described that exercisers of this age range can maintain or even improve their level [19].

An enormous increase can be found in the situps. Here, in the pretest, the task could not be performed. In the posttest, the test person performed eight; in the standardized value is no change, but now she is on the upper margin to reach the next class (one point). In the pushups, there were two more than in the pretest and this shows an improvement in the standardized value too by one point. The trunk strength has improved significantly. This was to be expected, since exactly these muscle groups were also trained. In late adulthood, strength continues to decline, although this decline can be slowed, or even prevented or reversed, through regular training [21]. The fact that there was no strong improvement in the push-ups can be attributed on the one hand to the fact that not enough strengthening exercises were performed for the

muscles involved here. Furthermore, the reduced nerve conduction velocity due to the damaged myelin structures could also be responsible for this [1].

Standing longjump remains in the lower range, there is no change visible, as well as in the 2 km walking time. The unchanged performance in the standing long jump and 2 km walk can be attributed to persistent foot lifter weakness [1].

The Fitness total score improved more than 500 % from 0.50 to 2.85, even though it remains in the lower range.

5. Conclusions

The present single case study was able to show that an improvement of physical fitness in CMT is possible, even if an exercise session of HIIT takes place only once a week. Therefore, a pilot study should follow to compare the effectiveness of one with two training sessions per week. In addition, the foot lifter weakness should be treated with physical exercise. Here, whole body vibration could be used. After that, an improvement in walking speed and standing longjump could be reached. Such a study is in the planning process.

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