Opleiding Revalidatiewetenschappen en Kinesitherapie

The effect of hydrotraining on adults with Inclusion Body Myositis: Quality of Life and muscle endurance

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DM	Dermatomyositis
FI-2Test	Functional Index-2 Test
h-IBM	Hereditary Inclusion Body Myositis
IBM	Inclusion Body Myositis
IIm	Auto-immune skeletal Myopathy
IMACS	International Myositis Outcome Assessment Collaborative Study Group
IS	Immunosupressive treatment
PM	Polymyositis
reps	Repetitions
s-IBM	Sporadic Inclusion Body Myositis
SF-36	36-item Short Form Survey
UZA	University Hospital of Antwerp/ Universitair Ziekenhuis Antwerpen

1 Positioning statement

This research continues on master's thesis part 1: "Suitable therapies for patients who suffer from Dermatomyositis or Polymyositis: a systematic review". Initially, this master's thesis was started to find a physiotherapy for people with Dermatomyositis or Polymyositis (continuing with Master's thesis part 1- a systematic review). Masterproef part 2 was carried out with IBM patients, because the effect on the wounds/skin rashes of the hot water during hydrotherapy is not yet proven in DM, besides people with these two types of muscle diseases can almost completely restore through medication, which is not so for IBM patients [1][2][3]. The presentation of this disease is progressively declining. As current medical treatment has proven ineffective for IBM patients, there is a strong need to identify alternative treatment methods for these patients [3]. This is the reason why master thesis part 2 is continued with: The effect of hydrotherapy in adults with Inclusion Body Myositis: Quality of Life and muscle endurance.

A monthly update is sent from database PubMed with all new published articles that end up under the search strategy, which is used to screen articles for Master's Thesis Part 1. A final screening took place on 19 May 2019. The new literature was reviewed every month and added if it was relevant. One article was published in 2018 that is supplementary to the study of Master's thesis part 1. "Community exercise is feasible for neuromuscular diseases and can improve aerobic capacity", written by Wallace A. et al. This randomized single-blinded crossover trial design containing a 12-week aerobic training program with a control period. They used exercise bicycles. The training was carried out by patients with Charcot-Marie-Tooth disease (CMT) type 1 and IBM patients. They concluded that twelve weeks of aerobic training in community gyms was feasible, safe, and improved aerobic capacity in people with CMT and IBM. The screening strategy is depicted in Figure 7.3 [4].

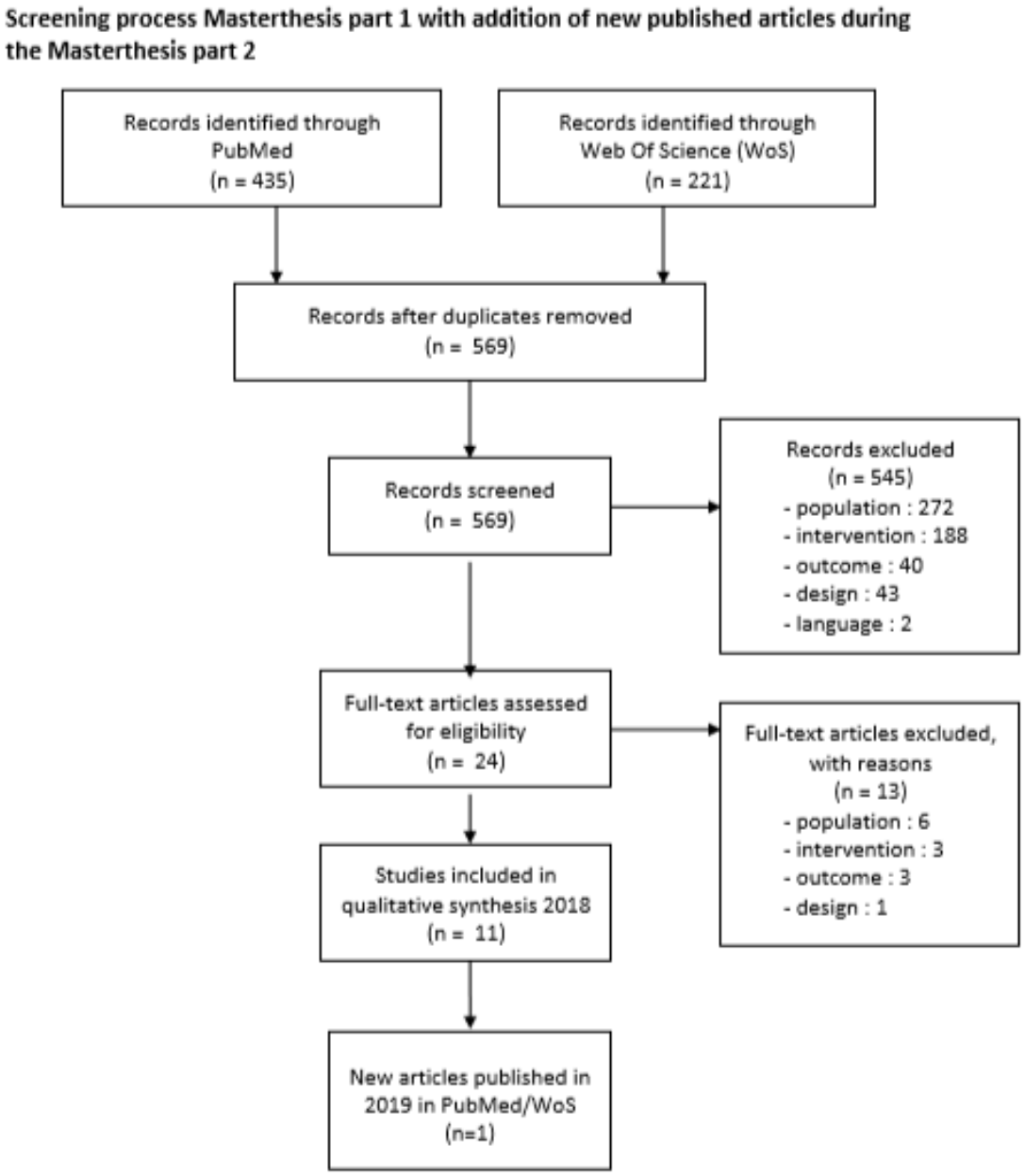

Figure 1.1: Schematic overview of the used screening process of Masterthesis part 1, continued with part 2

Very little is known about the best physiotherapy treatment for patients suffering from IBM. This is partly due to the high variability of symptoms in this disease. The aim of this study is to be able to help people with this disease in the safest and most effective way to

keep their Quality of Life as high as possible.

During my internship as a physiotherapist in Finland, I came into contact with a patient suffering from IBM. She suffered from this disease for years and was getting weaker. This process was slowed down when a Finnish physiotherapist started hydrotherapy with her. She became stronger, gained confidence in her abilities and her Quality of Life improved significantly. Before hydrotherapy she often had pneumonia. This means her mucus got stuck in her lungs and trachea. Together with the physiotherapist, the patient practised variable exercises in the water. These exercises in combination with her hard effort improved the functioning of her lungs significantly. This particular case made me want to study this physiotherapy on a scientific basis.

Research has already been carried out to find the most appropriate therapy for IBM patients; it has been proven that exercise therapy (both endurance/ strength/ as a combination) improves the general health of the patient[5][6] [7] [8]. Hydrotherapy was however never investigated for these patients. This master's thesis will dive into this subject and verify whether hydrotherapy has a beneficial effect on the clinical presentation of the disease.

2 Abstract in het Nederlands

Doelstelling: Het doel van deze experimentele studie was de haalbaarheid van oefeningen in het water te objectiveren bij volwassenen met Inclusion Body Myositis (IBM). Dit aan de hand van de veranderingen in de levenskwaliteit en spieruithouding.

Methode: Een klinische proef werd voor 4,5 weken uitgevoerd bij personen met IBM (n=2). Trainingsessies werden 2 keer per week uitgevoerd in het Revalidatie Centrum Nottebohm. De patiënten voerden oefeningen uit onder begeleiding van een studente kinesitherapie en een begeleider van de UAntwerpen. Er werd een individuele vergelijking gemaakt voor en na de interventie. Primaire outcomes waren enerzijds de levenskwaliteit (beoordeeld met de Short Form-36 vragenlijst) en de spieruithouding anderzijds (geobjectiveerd met de Functional Index-2 test). Als secundaire outcome werden bevindingen van de patiënten zelf kort bevraagd.

Resultaten: Er werd een verbetering gevonden in de volgende domeinen van de SF-36 voor beide patiënten: "physical functioning and role limitations due to physical functioning". Eén patiënt bleef gelijk op alle andere subdomeinen van de SF-36. De tweede patiënt gaf verbetering aan op alle subdomeinen en bleef gelijk op het subdomein "pain". Bij alle IBM patiënten werd een verbetering gezien in alle spiergroepen met de FI-2 test. De patiënten zelf rapporteerden een verbetering in conditie en minder bewegingsangst.

Conclusie: Oefeningen in het water bij patiënten met IBM kunnen voor deze patiënten beschouwd worden als een waardevolle, toepasbare behandeling.

Keywords: Inclusion Body Myositis, inflammatory myopathies, exercise therapy, physical treatment modalities, aerobic exercise, resistive exercise, hydrotherapy, Quality of Life, muscle strength, treatment outcome, outcome measure.

3 Abstract in English

Objective: The aim of this experimental physiotherapeutic training was to ascertain the feasibility and effect of hydrotraining on Quality of Life and muscle endurance for people with Inclusion Body Myositis (IBM).

Methods: A clinical trial design was used to compare a 4,5-week aerobic training program in water for IBM patients (n=2). The training occurred 2 times a week in a Revalidation Center. Supervision was carried out by a physiotherapy student and her promotor. The patients were analyzed individually before and after the experiment. The primary outcome measures were Quality of Life, observed by the Short Form 36-questionnaire, and muscle endurance, objectified by the Functional Index-2 test. Secondary outcome was patient-reported findings.

Results: An improvement in the following subdomains of the SF-36 was seen in both participants: "physical functioning and role limitations due to physical functioning". One patient remained equal on all other subdomains. The second patient with IBM improved on every subdomain, except "pain", which remained the same. All IBM patients improved in the FI-2 test on all exercises. Both patients reported an improvement in physical condition and less fear of movement.

Conclusion: Four and a half weeks of aerobic training in water (hydrotherapy) resulted as safe, feasible, improved endurance capacity of all muscle groups and progressed Quality of Life in people with IBM.

Keywords: Inclusion Body Myositis, inflammatory myopathies, exercise therapy, physical treatment modalities, aerobic exercise, resistive exercise, hydrotherapy, Quality of Life, muscle strength, treatment outcome, outcome measure.

4 Introduction

Inclusion Body Myositis (IBM), Polymyositis (PM) and Dermatomyositis (DM) are rare, chronic, idiopathic, inflammatory and auto-immune skeletal myopathies (IIM). The idiopathic inflammatory myopathies are a heterogeneous group of rare diseases that primarily affect skeletal muscles. [5] [9]. PM, DM and IBM lead to inflammatory cells between the muscle fibers, but some clinical differences exist. DM can be distinguished from the others because it affects not only the muscles but the skin as well (leads to rashes). An important difference between PM and DM lies in the fact that IBM is progressive [6].

There are 2 types of IBM, sporadic Inclusion Body Myositis (s-IBM) and familial or hereditary Inclusion Body Myositis (h-IBM). Inflammation is almost invariably seen in s-IBM, in contrast to h-IBM, with rarely encountered inflammation [10]. Yet, it is suggested that in IBM disease, there is a primary inflammatory mechanism that can partly respond to immunosuppressive treatment (IS). In very weak muscles there may be other factors of interest, such as degenerative mechanisms, which are not affected by treatment with IS [11].

IBM is the most common inflammatory skeletal muscle disease in patients above the age of 50. The late-onset myopathy is a slowly but steadily progressive disorder in which disability increases over the course of many years. The precise cause is not defined [12]. The progression in muscle weakness in IBM patients approximately corresponds to an annual loss in skeletal muscle strength of 5–16%, while in healthy elderly an age-related muscle loss is seen of 0.5-1.5% per year [3]. The slow advancing muscle weakness and atrophy experienced by patients with IBM is mainly found in finger flexors, M. Quadriceps and distal muscle groups of the limbs [13].

As a result, IBM patients experience degradation: a higher incidence of falls and they may increasingly rely on help from caregivers to perform activities in their daily life. Ultimately, IBM patients demonstrate lower Quality of Life compared to the general elderly population, especially in terms of physical functioning [3][14] [5].

In order to diagnose IBM, doctors use laboratory tests and an anamnesis concerning the family history [10]. This is elaborated in detail in Appendix A: "Proposed diagnostic Criteria for Inclusion Body Myositis" (Griggs RC, 1995). A prevalence 1-9 / 100 000 is estimated, but the true prevalence of IBM is unknown [12] [15].

IBM patients often feel fatigue. This is because their muscles are weakened by the disease and to compensate, other muscles have to work harder. As a result, patients want to avoid movement to prevent pain and fatigue. This causes even more weakness and atrophy. When muscles atrophy, they cannot be built back up again [16]. Exercise training is increasingly utilized as a non-pharmacological intervention in the clinical management of patients with IBM, DM and PM. Recent studies confirm the safety and efficacy of both resistance training and aerobic exercise in adults suffering from IBM. Exercises can improve muscle strength, even in very affected muscle groups. It can also improve secondary problems, such as the active range of motion, balance, fatigue, Quality of Live, muscle function,... [17][13]

IBM patients appear to have asymmetric weakness of both proximal and distal upper and lower limb muscles. The slowness of muscle deterioration appears to be asymmetric as well [18]. The loss of average muscle strength appears not to be influenced by factors such as gender, age and serum CK level at diagnosis. The region where the first clinical symptoms of muscle weakness are experienced and the initial muscle strength at the start of the therapy do not influence the loss of average muscle strength. Furthermore, the prognosis for older patients with IBM is not worse than the average, with regard to the speed of muscle decrease or wheelchair use [11].

It has already been proven that exercises are safe and beneficial for them, but there is still nothing published about exercises in the water [19]. The idea of training in the water is that people can move without pain and fright. Hydrotherapy is performed in a heated

swimming pool (32 degrees). This heat often has a positive effect on joint stiffness and excessive muscle tension. The water serves as assistance and resistance during movement (relieving), it is a medium to minimize gravity. The patient feels less pressure, so that he can perform physical training in which he/she experiences limitations on the ground floor. Thanks to the water, patients feel the opportunity and freedom to overcome their limitations and fears they experience on the ground [20][21].

Various goals can be achieved depending on the intensity of the exercises. The patients can walk, jump, cycle in the water and these activities help them in improving lung function, blood circulation, balance and coordination, strengthening of the not affected muscles, ... They have the opportunity to push their limits, mentally and physically [16][21].

Therefore the objective of this study is to investigate the effect of aquatic exercises on muscle endurance and the function of daily activities without overuse of involved muscles in a 4,5 week therapeutic experiment. The patients in this physiotherapy scientific experiment are people who suffer from IBM. Following questions will be answered in the further course of this work to check whether the given training has had the desired effect:

- Are exercises in the water provided as a safe and beneficial treatment alternative for adults with IBM?
- Does muscle endurance increase?
- Has this training led to an experienced Quality of Life improvement?

5 Method

5.1 Subjects

The patients were selected by prof. dr. em. De Jonghe P. and Prof. dr. Baets J. from the neurology department at the UZA Hospital. In order to participate in the study, the following inclusion and exclusion criteria were drawn up for the patients:

- The population consisted of adults, equal or older than eighteen year.
- Patients were diagnosed with the Inclusion Body Myositis disease according to Grigg's criteria (Grigg's IBM and myopathies, see Appendix A).
- Patients have the possibility of coming into contact with water and are being able to perform exercises in the water.
- Patients have a "stable" display of the disease/ well-being so that training could take place on a regular basis (twice a week).
- Patients need to be able to walk independently.
- Patients experienced difficulties in their daily occupations, which was assessed by a motivational interview.
- Patients need to sign the Informed Consent.

The exclusion criteria for persons, relevant for this study were as follows:

- Persons under the age of eighteen.
- Too weak or not mobile enough to stand up independently in the water.
- Severe cognitive impairment.
- Co-morbidities preventing safe exercise training (e.g. uncontrolled hypertension (systolic blood pressure > 140 mmHg, diastolic blood pressure > 90 mmHg).[22]

5.2 Aim

The purpose of this work is to examine whether aerobic training in aquatic environment facilities can be used as a safe, feasible and acceptable new physiotherapy method for patients with IBM. The variables are Quality of Life (social, physical, mental) and muscle endurance (muscles of the upper- and lower extremity, and neck), after a period of 4,5 weeks.

5.3 Design

This research is a case report with 2 patients. The progress of the patients with IBM was evaluated by checking their capabilities and struggles in daily life before and after the hydrotherapy sessions by the Short Form-36 questionnaire (SF-36) [23] and Functional Index-2 test (FI-2 test) [24]. The scores of the SF-36 questionnaire were calculated as a score to the physical component, the mental component and the social component. It contains subcategories: Physical functioning, Role Limitations due to Physical Problems, Role Limitations due to Emotional Problems, Vitality, Mental health, Social functioning, Pain, General Health Experience and Health Change. The higher the score on the questionnaire, the better the health status [23].

5.4 Recruitment procedures

The recruitment of patients started on the 26th of July 2018. The method of contacting was done by telephone, emails or personal visit. The following instances/persons were contacted in search of adequate patients for the study:

- Rehabilitation Centers such as the Nottebohm in Brecht and Mick in Brasschaat
- Hospitals in which patients suffering from IBM rehabilitate, more specific UZA and Klina
- The chairman of REVAKI, who arranges internships for physiotherapy students
- The Polymyositis Liga, a group for patients suffering from the disease.
- Personal contact with patients by means of personal message via a closed Facebook group
- A brochure was shared several times on social media

Despite of all the above attempts, only few positive responses were received. Moreover, due to practical limitations, some of the patients that responded positively could not participate because they live in e.g. the UK or Texas. However, close contact is kept with these

patients to inform them of the results of the study. Finally, two patients adequate for the study could be recruited via the UZA. The patient flowchart is depicted in Figure 5.1.

5.5 Ethical approval

The Ethics Committee of the UZA/UAntwerpen, Belgium approved the study with admission number B300201940113. The test patients were finally recruited via the UZA. Before the start of the study, a Dutch written and verbal informed consent was obtained from the patients, including the permission to take photographs during the sessions and to publish them.

5.6 Course of the experiment / exercise program

Both the questionnaire and the clinical endurance test were conducted in a therapy room at the University of Antwerp. Training took place at the swimming pool of the Rehabilitation Center Nottebohm, with a controllable bottom that can be set deeper or shallower. This way the patient could safely enter the swimming pool and the height could be adjusted for each exercise. The protocol consisted of 4,5 weeks of training, twice a week. The details of these exercises can be found in Appendix B.

The training sessions were supervised by the physiotherapist F. Sluiter, with the assistance of one additional supervisor (Prof. dr. H. Bortier/F. Ego). Training was designed for every patient individually, because the physical needs of the patients were very different. The goal was to strengthen the muscles that are not affected by the disease process, and preventing more weakness of the muscles that are affected [16].

The patients wore a heart rate monitor during all sessions (Garmin Forerunner 235). The severity of the effort was determined by the combination of the following 3 factors: intensity, duration and frequency:

- **Frequency**: The elderly (after the age of 50) need more recovery time after a workout.

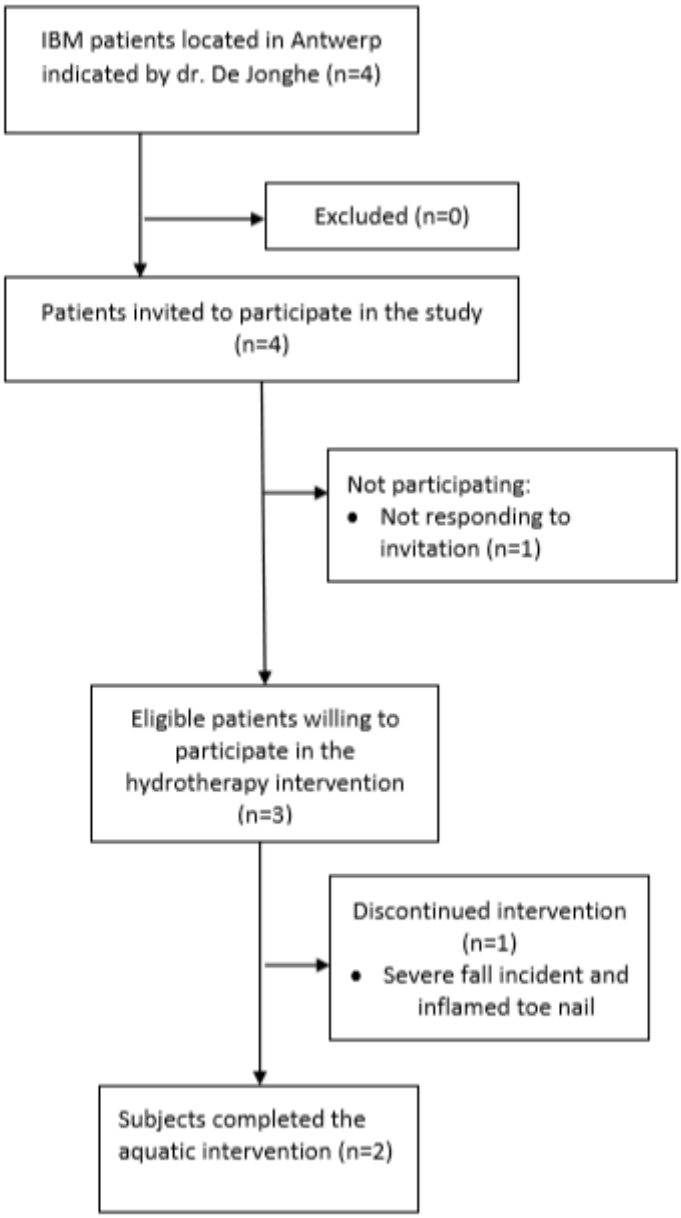

Figure 5.1: Patient flowchart

Training, 2 times a week ensures sufficient intensity it builds muscle, but in many cases prevents muscle strain and injuries [25].

- **Intensity**: The intensity was determined based on Karvonen's formula. Training was done at an intensity of 70% of the maximum heart rate, determined via the Karvonen formula [26]. Attention was paid to keeping the frequency around this heartbeat. An important rule was that the patient should be able to keep talking while swimming, so that the exercise was aerobic. The intensity, speed of movements and endurance was built up throughout the sessions.

- **Duration**: The first session took 30 minutes. The patient soon indicated that he could practice for an hour, so the training lasted 1 hour from the second session onwards.

Before starting the training, the patients walked 4 lengths with large steps and high knees to warm up. Sufficient rest was taken between exercises. Training was alternated between leg exercises and arm exercises, heavy and light exercises. When the patients had reached a fatigue or suffered from cramps, they switched to proprioception exercises, coordination or another muscle group was trained. Training ended with passively stretching Hamstrings, M. Gastrocnemius and M. Quadriceps. The sessions themselves consisted of exercises prepared individually for each patient. The sessions consisted of coordination exercises with a cognitive stimulus, balance exercises and resistance exercises. Intensity was reduced when a patient indicated that he felt very tired the day after a training or when he noticed stiffness longer than 24h [27].

	Patient 1	Patient 2
Age (Yr)	77	65
Sex (M/F)	M	M
Disease activity	Chronic	Chronic
Year of diagnosis	2016	2009
Walking aids	Cane	Cane
Medication	• asaflow • burinex • coveram • d-cure amp • ezetrol • glucophage • uni diamicron • jardiance	• enalparil • depakine chrono 500 • allopurinol
Types of exercises in physio	• cycling • taking stairs • balance exercises • sit to stand exercises	• cycling • strength exercises for the core, legs, forearms and shoulders • stability exercises
Physio (days/week)	2	2
Sports	Daily (fitness 1h)	/
Personal goals	• Endurance • Strength of M. Quadriceps bilateral	Fine and gross motor skills

Table 5.1: Medical record patients

5.7 Outcome assessments

For the outcome of the assessment a distinction is made between the primary outcome and the secondary. The primary outcomes were collected at baseline and at 4,5 weeks' follow-up. Secondary outcomes were only collected after 4,5 weeks of training in the water. FI-2 test was carried out at the same hour so that the fatigue was as representative as possible. The remainder of this section will discuss these separately, starting with the primary outcome.

5.7.1 SF-36 Questionnaire

The change from baseline to follow-up concerning self-reported Quality of Life, was evaluated with the reliable and valid SF-36 Questionnaire [28][29]. This survey is proposed as the QoL assessment tool by the International Myositis Outcome Assessment Collaborative Study Group (IMACS), used in IIm patients [3]. The SF-36 questionnaire consists of thirty-six items that surveys three domains: physical, mental and social health. These main domains are then subdivided in: Physical functioning, Role functioning (physical), Role functioning (emotional), Energy/fatigue, Emotional well-being, Social functioning, Pain, General Health and Health change [23]. The results of this questionnaire can be found in Section 6. The number of questions per domain are listed in table 5.2, this is important in assessing the results.

Domain	Number of questions
Physical functioning	10
Role functioning/physical	4
Role functioning/emotional	3
Energy/fatigue	4
Emotional well-being	5
Social functioning	2
Pain	5
General Health	5

Table 5.2: Scoring of patient 1

The questionnaire is filled in by giving scores to the extent by which the patient agrees to a certain statement. These scores (mostly ranging from 1-5) are then awarded a score reflecting the overall health. A higher score always reflects a better state of health. The scores can be inversely related to the response choice of the patient. When for example the question would be: "How often do you eat at a fast food restaurant?", a response choice of 5 (indicating very often) will be awarded a score 0 on the test whilst a response choice of 1 (indicating never) will be awarded a score of 100 as this is more favorable for the overall health. Scores are always graded from 0-100 (100 most favorable for health and 0 least favorable for health). Intermediate response choices are linearly graded in between these values. This means for example that when the patient has 5 response choices (1,2,3,4,5) and 5 is the most favorable health state, the value 2 corresponds to a score of 25 (0,25,50,75,100). However, when the patient has only 3 possible response choices (1,2,3) where 3 is the most favorable health state, the number 2 corresponds to a score of 50 (0,50,100). [30].

5.7.2 FI2-test

Objective measures of muscle endurance (FI-2 Test) were completed by patients [24]. This test is conducted specifically for persons with Dermatomyositis and Polymyositis (as well

as IIM). The content of the FI-2 test represents measurement of muscle impairment of the upper and lower limbs and neck. The test includes the separate performance of shoulder flexion (with 1 kg weight cuff), shoulder abduction, head lift, hip flexion, step test, heel lift and toe lift. The type of exercises are in accordance with the Myositis disease phenotype [24].

The patient had to perform as many correct repetitions (reps) as possible of each task. A maximal score of all tasks was given when 60 correct reps are reached. An exception counts for the heel and toe lift: here was the maximal score 120 reps. A metronome was used to standardize the movement pace of each task. This pace of the metronome was 40 beats/ minute for all exercises, except for the heel lift and toe lift where it was 80 beats/ minute. The test was ended in 2 conditions, by patient or by observer.
The test was ended by the patient:

- when reaching maximal numbers of repetitions.
- due to muscle fatigue, pain or general fatigue.

The test was ended by the observer:

- when the patient could not keep up the given pace and was unable to correct this within three repetitions.
- when the patient started to compensate and was not able to correct this within three repetitions.

After each task a Borg CR-10 scale was used to rate muscle exertion per exercise [24]. The results of the FI-2 test for both patients can be found in tables 6.4 and 6.5 respectively. The table shows the number of repetitions the patient was able to perform before and after the training sessions [24].

5.7.3 Secondary outcomes

The secondary/supplementary outcomes were evaluated at the end of the training sessions. Input of patients was asked about the general experience of the new type of experi-

mental physiotherapy training. However, this is not been clinically validated. The specific questions that were asked can be found in the result section.

5.8 Statistical relevance

It is never feasible to study the whole population at once. A very important aspect in carrying out any clinical study is therefore the sample size calculation. Only a part of the population is studied but this part needs to be representative for the whole population in order to draw any conclusions. An excessive sample size leads to a waste of for example researching resources (as they have to study the whole population). The sample size of any study depends on following parameters. They will be explained separately below.

- Acceptable level of significance
- Power of the study
- Expected effect size
- Underlying event rate in the population
- Standard deviation in the population.

5.8.1 Acceptable level of significance

The level of significance is the p-value that is widely used to determine whether to accept or reject a hypothesis. Probably the most used p-value in all literature is 0.05. This means that we accept all results with a p value lower than 0.05. The lower the p-value, the more sure we are of our decision. Using a p-value of 0.05 means that we accept that there is a 5% chance that a significant improvement is reported, although there is no improvement in reality.

5.8.2 Power of the study

The inverse of the p-value is the power of a study. This means that we fail to detect for example an improvement although there is an improvement in reality. The power of a study

is represented by 1-. A widely used value for the power is 80%.

5.8.3 Effect size

The effect size in statistics is the difference between the studied variable in the test group. This effect size can be absolute or given as a percentage. Mostly the effect size, used to determine the sample size, is based on previous studies concerning the same subject. A smaller sample size is needed when the effect size is larger and vice versa. When no preliminary studies are made and hence no data is available, a small effective size should be assumed in calculating the sample size.

5.8.4 Standard deviation

Standard deviation is a measure for variability in data. When data is very homogeneous and thereby standard deviations are small, a smaller sample size is needed. When however data is very dispersed, a larger sample size is needed to draw conclusions.

5.8.5 Sample size calculation

For the final calculation of the sample size, following formula is used where Z_α is the Z-value corresponding to the p-value, Z_{1-}-β the Z-value corresponding to $1-\beta$, σ the standard deviation and Δ the effect size.

$$n = \frac{(Z_\alpha + Z_{1-\beta})^2 * \sigma^2}{\Delta^2} \tag{5.1}$$

When following conventions for many clinical studies, a p value=0.05 (thus α=0.05 and $Z_\alpha = 1.96$) and a power of 80% (thus $1-\beta$=0.2 and Z1-=0.8416) can be chosen. An effect size of 20% was estimated. The standard deviation in previous studies in not representative due to its limited size. Therefore a fairly high standard estimation should be taken for this study. It was decided to move forward assuming a standard deviation of 1. The calculated sample size is then:

$$n = \frac{2(1.96 + 0.8416)^2 * 1^2}{0.2^2} = 392 \tag{5.2}$$

Comparing this required sample size to the prevalence of the IBM (around 1-9/100000), it can be concluded that it is not feasible to acquire the needed amount of participants to carry out a statistical representative studies. Even with very optimistic parameters, for example a lower standard variation of 0.5 and an effect size of 40%, the required number of participants would still be:

$$n = \frac{2(1.96 + 0.8416)^2 * 0.5^2}{0.24^2} = 25 \tag{5.3}$$

This number seems already more feasible than in earlier studies, the standard deviation always proved high, this does not represent an adequate sample size.

5.8.6 Conclusion

After having calculated the sample size and comparing this sample size to the prevalence of the disease, it can be concluded that this study will not be able to acquire the needed number of participants to be statistically significant. This however does not mean it makes no sense to carry out the study. One should only be careful when drawing conclusions for the whole studied populations (meaning all patients suffering from IBM). This study can lay the groundwork for further research by providing already some first insights that can be tested later on in a more extensive study. This kind of study would also require sufficient funding, needed for own resources and finding an adequate number of participants as this is not straightforward given the low prevalence.

6 Results

In this section the results of the two methodologies used to assess the training, the SF-36 questionnaire and the FI-2 test, will be discussed. As described in the previous section it will not be possible to extrapolate these results to the whole population due to the limited amount of participants. However, qualitative conclusions can be drawn that will prove useful in further studies done on a larger scale.

6.1 SF-36 Questionnaire

The first assessment methodology that was used is the SF-36 questionnaire. In this section the results of the SF-36 questionnaire will be displayed. The used methodology can be found in Section 5.7.1. The results will be interpreted in the discussion section of this work.

The results will be displayed in several steps. For each patient the different domains of the SF-36 questionnaire are graded before the start of the training and after it. As such it can be seen in which domains the patients improved. Afterwards, the combined results of both patients will be given by taking the average scores for each of the domains. The results for patient 1 are displayed in Table 6.1. The results for patient 2 are displayed in Table 6.2.

Domain	Score before training	Score after training
Physical functioning	15	35
Role functioning/physical	75	100
Role functioning/emotional	100	100
Energy/fatigue	70	70
Emotional well-being	92	92
Social functioning	75	75
Pain	80	80
General Health	85	85

Table 6.1: Scoring of patient 1

Domain	Score before training	Score after training
Physical functioning	20	25
Role functioning/physical	0	75
Role functioning/emotional	0	100
Energy/fatigue	70	80
Emotional well-being	80	96
Social functioning	62.5	100
Pain	100	100
General Health	75	95

Table 6.2: Scoring of patient 2

The final table of this subsection gives an overview of the entire population that was investigated. Due to the low prevalence of the disease, the results have little statistical relevance and should be interpreted carefully. The mean value and the standard deviation ($SD = \sqrt{\frac{1}{N-1}\sum_{i=1}^{N}(x_i - \overline{x})^2}$) for the different domains is calculated before and after the training. The results are shown in Table 6.3.

Domain	Mean (before)	Mean (after)	SD (before)	SD (after)
Physical functioning	17.5	30	3.53	7.07
Role functioning/physical	37.5	87.5	53.03	17.68
Role functioning/emotional	50	100	70.71	0
Energy/fatigue	70	75	0	7.07
Emotional well-being	86	94	8.49	2.83
Social functioning	81.25	87.5	26.52	17.68
Pain	90	90	14.14	14.14
General Health	80	90	7.07	7.07

Table 6.3: Overall training statistics for the SF-36

6.2 FI-2 test

In this section the results of the FI2-test, as described in section 6.5 will be discussed. The results of this test will be further interpreted in section 7.2.

Muscle Group	Before training		After training	
	% of max repetitions	Muscle exertion Borg CR-10	% of max repetitions	Muscle exertion Borg CR-10
Shoulder flexion right	58.33	2-3	83.33	5
Shoulder flexion left	100	2	100	4
Shoulder abduction right	55	4-5	61.67	5
Shoulder abduction left	55	4-5	61.67	5
Head lift	30	3	73.33	2
Hip flexion right	18	4	50	3-4
Hip flexion left	43.33	4	50	2
Step test right	0	/	0	/
Step test left	0	/	0	/
Heel lift	50	5	72.5	5
Toe lift	50	5	66.67	5

Table 6.4: FI-2 Patient 1

Muscle Group	Before training		After training	
	% of max repetitions	Muscle exertion Borg CR-10	% of max repetitions	Muscle exertion Borg CR-10
Shoulder flexion right	100	4	100	5
Shoulder flexion left	100	2	100	5
Shoulder abduction right	73.33	8	83.33	5-6
Shoulder abduction left	73.33	9	83.33	8-9
Head lift	75	7	83.33	7
Hip flexion right	51.67	9-10	83.33	8
Hip flexion left	100	4	100	6
Step test right	0	/	0	/
Step test left	0	/	0	/
Heel lift	41	8	55	7
Toe lift	0	/	0	/

Table 6.5: FI-2 Patient 2

6.3 Secondary outcome: patient-reported findings

After the 4,5 weeks of training, the underneath questions were answered by the patients via Google Form. The general trends will be deduced from the answers and discussed in Section 7.3.

1. Which benefits did you notice through the exercises in the water?
2. Did you feel soreness of the muscles after the training?
3. Which exercises had little effect for you?
4. Did you feel a difference when conservative physiotherapy was the day before/ after the hydrotraining, e.g. more tired/ less soreness because of the water?
5. What is the big difference that you experienced between the first and the last hydrotraining?

6. Did you feel more tired after hydrotraining than after conservative physiotherapy on the ground?
7. Would you recommend this training in the water as a therapy on it's own for patients with IBM?
8. Do you feel an improvement in Quality of Life compared to 4,5 weeks ago?
9. What are the observed disadvantages of exercising in the water?
10. Which exercises were the most effective for you?

7 Discussion

7.1 SF-36 Questionnaire

For patient 1, the main problem at the start of the training was the physical functioning as this domain only had a score of 15. On the other domains the patient already scored quite high before starting the training. After a training period of only 4,5 weeks, we can see that the patient improved significantly on this aspect. The score increased from 15 to 35 which is remarkable. Moreover the role limitations due to physical health problems the patient experienced, were resolved during the course of the training. In the table with results, we also see that the social functioning decreased over the measured period. However, as indicated in Table 5.2, this result is only influenced by two questions. When looking at the questionnaire itself, it showed that this was due to 1 of the questions which was responded slightly more negatively. Taking the content of the training program into account, which was not expected to influence this aspect to a great extent, it is assumed that variation is due to random fluctuations.

For patient 2 a similar pattern in improvement can be noted. This patient improved less on the physical functioning itself but overcame all limitations he faced, those who were due to his physical disabilities and those due to emotional issues. This means that the training not only helped him to physically cope with his daily role, but also improved his mindset. He felt better and became more confident in his daily routine.

Although one should be careful in extrapolating these results for all patients suffering from IBM, some general conclusions can be drawn:

- The patients have improved or remained constant on all domains.
- The training had the most beneficial impact on the physical functioning of the patients and the limitations they experienced due to limited physical functioning.
- The training effectively addressed the problematic domains. The domains that ini-

tially had a low score improved the most.

7.2 FI2-test

In what follows the results of the FI-2 test will be discussed qualitatively. As the number of patients included was very limited, no statistical analysis is performed on the outcome. However, some interesting remarks can be made:

- Both patients drastically improved on all exercises. There is not a single exercise where they could do less repetitions after the training.
- The muscle exertion did not improve over the course of the training sessions. In general it even worsened. This is probably due to the fact that as they got further into some of the exercises, they were more rapidly exhausted in the other ones as well. Thereby, the test was observed the day after hydro-exercises. Which means there could be a relation for exhaustion.
- The results of the FI-2 test are in line with the ones of the SF-36 questionnaire. Patients improved significantly in terms of their physical capabilities but their energy level (indicated by muscle exertion for FI-2 and energy for SF-36) did not improve.

7.3 Secondary outcomes

In this section the most important takeaways that came forward from the survey will be discussed. The benefits of training in the water were the feeling of better physical condition, less fear of moving and more confidence. A possible explanation for this could be that moving is easier in the water and this results in a good blood flow circulation. Both patients indicated that the sessions had good progression in intensity, as well as lots of variation and challenging exercises. For this reason, both patients felt an improvement in Quality of Life. Also the exhausted feeling after training decreased after a few sessions, whilst exercises became more difficult. They both recommended the hydrotraining as a therapy for patients with IBM and could not name a disadvantage of hydrotraining. One person would rather

switch it for conventional physiotherapy. This was because he was capable to jump, run and take a step on a box without support (cardiovascular exercises), which is less/not possible on the ground. For this reason, cardiovascular exercises were liked the most.

7.4 Conclusion

This thesis proposed training in the water as an alternative solution for people suffering from IBM. Due to the low prevalence of the disease, only a limited group of patients could participate in the study. Moreover, the patients needed to be able to come biweekly to the training and they needed to be strong enough to carry out exercises. This is why it was decided not to include a control group as to help a maximal number of patients as possible.

The effects of the training were investigated by three means, namely the SF-36 questionnaire, the FI-2 test and a small survey on the patients experience. From all three investigation methods it could be concluded that the training has had a beneficial effect on the patients capabilities. They significantly improved on a physical level and overcame limitations they had during their daily routine. In spite of them following already physiotherapy on a regular basis, it is remarkably how much they improved over the course of only 4,5 weeks.

Although the study group was too small to draw statistical conclusions, hydrotraining certainly holds very promising results. These results could unfortunately not be compared to other trainings due to the low statistical value and the short training period in comparison to other studies. [7]. However, I am convinced that when this experiment can be extended to a larger group of patients, and similar results are obtained, hydrotraining can soon become a valued therapy and help IBM patients all over the world. Further investigation should be carried out with e.g. a larger training period, a follow-up of cardiorespiratory parameters, a double-blinded study, comparison with a conservative physiotherapy or a multicenter.

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Appendix A: Proposed Diagnostic Criteria for Inclusion Body Myositis [10]

Here are the characteristic features explained to diagnose IBM.

"A. Clinical features

1. Duration of illness > 6 months
2. Age of onset > 30 years old
3. Muscle weakness must affect proximal and distal muscles of arms and legs and patient must exhibit at least one
    - Finger flexor weakness
    - Wrist flexor > wrist extensor weakness
    - M. Quadriceps muscle weakness (= or < grade 4 MRC)

B. Laboratory features

1. Serum creatine kinase > 12 times normal
2. Muscle biopsy
    - Inflammatory myopathy characterized by mononuclear cell invasion of non-necrotic muscle fibers
    - Vacuolated muscle fibers
    - Either Intracellular amyloid deposits (must use fluorescent method of identification before excluding the presence of amyloid) or 15-18-nm tubulofilaments by electron microscopy
3. Electromyography must be consistent with features of an inflammatory myopathy (however, long-duration potentials are commonly observed and do not exclude diagnosis of sporadic inclusion body myositis)" (Griggs RC, 1995).

"C. Family history

1. Rarely, inclusion body myositis may be observed in families. This condition is different from hereditary inclusion body myopathy without inflammation.

2. The diagnosis of familial inclusion body myositis requires specific documentation of the inflammatory component by muscle biopsy in addition to vacuolated muscle fibers, intracellular (within muscle fibers) amyloid, and 15- 18-nm tubulofilaments.

D. Comorbidities

Inclusion body myositis occurs with a variety of other, especially immuno-mediated conditions. An associated condition does not preclude a diagnosis of inclusion body myositis if diagnostic criteria (below) are fulfilled.

E. Diagnostic criteria for inclusion body myositis

1. **Definite inclusion body myositis**: Patients must exhibit muscle biopsy features including invasion of non necrotic fibers by mononuclear cells, vacuolated muscle fibers, and intracellular (within muscle fibers) amyloid deposits or 15- 18-nm tubulofilaments. None of the other clinical or laboratory features are mandatory if muscle biopsy features are diagnostic.

2. **Possible inclusion body myositis**: If the muscle shows only inflammation (invasion of non necrotic muscle fibers by mononuclear cells) without other pathological features of inclusion body myositis, then a diagnosis of possible inclusion body myositis can be given if the patient exhibits the characteristic clinical (A 1,2,3) and laboratory (B1,3) features" (Griggs RC, 1995).

Appendix B: various hydrotraining exercises for IBM patients

Picture	Description + reps and sets	Progression
	• Sideways walk with noodle • 4 lengths each leg	• Without support at the bar
	• Abduction and adduction of arms in prone position • 15 reps, 3 sets	• With weight cuffs (1kg)
	• Swimming • 4 lengths	• With weightcuffs around wrists
	• Walking with a noodle • 2 lengths each leg	• Walking with a noodle under both feet

Table 7.1: Overview exercises part 1

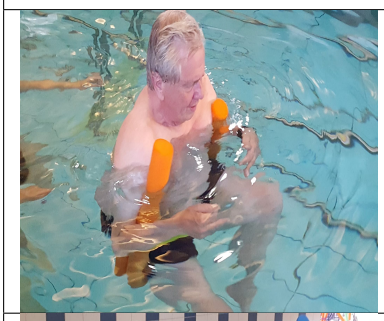
Picture P1	Description + reps and sets	Progression
	• Sit to stand position • 10 reps, 3 sets	• Without support of the bar
	• Hamstring curl with noodle • 10 reps each leg	• Without support of the bar
	• Cycling • 30”	• Cycling with noodles not between the legs but under the butt
	• Figure 8 movement	• Noodle under one foot and making a figure 8 in the water, do not let the noodle slip away
	• Walking without looking left and right • 4 lengths	• Walking backwards

Table 7.2: Overview exercises part 2

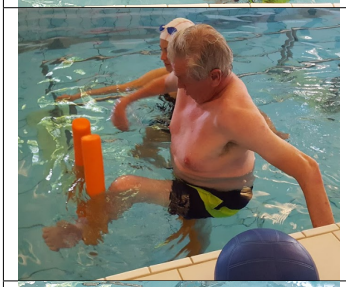
Picture P2	Description + reps and sets	Progression
	• Backstroke kicks with legs • 2 lengths	• 4 lengths
	• Jumping squat • 20 reps	• 30 reps and lower water level
	• Running at the same place • 3 sets of 30”	• Running back and forth
	• Walking sideways with one noodle	• Walking with noodle under each foot
	• Box step up • 15 reps with support of the bar	• No support
	• Pushing the shelf away and pulling back with 2 hands • 10 reps, 3 sets	• Fast rotations with the shelf

Table 7.3: Overview exercises part 3

Appendix C: Plagiarism check

Plagiarism introduction

Figure 7.1: Plagiarism check introduction

Plagiarism Discussion and conclusion

Figure 7.2: Plagiarism check discussion

Appendix D: Informed consent

Oproep voor deelname aan Wetenschappelijk onderzoek

Effect van watertherapie bij volwassenen met Inclusie Body Myositis: een pilootstudie

10 april 2019

Beste,

Ik wil jullie met deze brief graag uitnodigen om deel te nemen aan een wetenschappelijk onderzoek. Dit onderzoek is een samenwerking tussen de Universiteit Antwerpen en het Universitair ziekenhuis (UZA). Eén student Revalidatie en Kinesitherapie zal met dit onderzoek haar masterproef maken.

Het onderzoek betreft het effect van revalidatie in het water te onderzoeken. De therapie zal plaatsvinden in het revalidatie centrum Nottebohm te Brecht. Dit (indien mogelijk) 2x/week, voor 6 weken. De intensiteit zal aangepast worden naargelang uw fysieke fitheid, mogelijkheden en persoonlijke doelen. Het doel is ervoor zorgen dat u gemakkelijker al uw dagdagelijkse activiteiten kan uitvoeren en fysiek en mentaal verbetering merkt.

Vooraleer de interventie start, wordt kennis gemaakt d.m.v. een kort interview. Dit zal +-een half uur in beslag nemen en kan georganiseerd worden via een telefoongesprek.

Om het effect op spieruithouding te bestuderen, wil ik u graag een test (Functional Index 2) laten uitvoeren vóór en na een oefenprogramma. Zo kan nagaan worden of uw spieruithouding veranderd is door de therapie in het water. Deze test vindt plaats op de begane grond in een therapiezaal van de Universiteit van Antwerpen te Kleine Doornstraat, Wilrijk. U wordt alsook verzocht een vragenlijst (Short Form 36) in te vullen om een idee te krijgen over uw levenskwaliteit en verandering na 4,5 weken.

Het onderzoek zal er als volgt uitzien:
Van zodra u aangeeft geïnteresseerd te zijn om deel te nemen, wordt het interview (telefoongesprek) gehouden waarin alle vragen en bemerkingen worden uitgeklaard.
Indien u akkoord bent om deel te nemen, wordt een afspraak gepland in de Kleine Doornstraat te Wilrijk om allereerst een beeld te krijgen van de achtergrond van de patiënt. Het Word-document "informatie deelnemers" wordt ingevuld samen met de patiënt. Het invullen van dit document zal 30' in beslag nemen. Hierna mag de deelnemer een vragenlijst (Short Form 36) invullen en wordt verzocht een uithoudingstest (Functional Index 2-test) uit te voeren. De benodigdheden voor de FI2-test zijn:

- een gewicht van 0,5 kg,
- een stoel,
- een behandeltafel om op de rug te liggen
- een metronoom (gratis applicatie op gsm) die de snelheid aangeeft van de bewegingen.

Figure 7.3: Informed Consent part 1

Vervolgens wordt voor 4,5 weken 2x/week een oefenprogramma gepland te revalidatiecentrum Nottebohm Brecht. Dit is persoonlijk op uw mogelijkheden afgesteld. U krijgt begeleiding van een studente kinesitherapie van de Universiteit Antwerpen.
Na deze 4,5 weken wordt dezelfde meting uitgevoerd (FI2 test) en u wordt gevraagd om dezelfde vragenlijst in te vullen, beide in het therapiezaal van de UAntwerpen in de Kleine Doornstraat te Wilrijk. Deze dienen om te vergelijken of uw spieruithouding en/of levenskwaliteit veranderd is door de watertherapie.

Het is belangrijk om te weten dat er geen extra risico's zijn verbonden aan het trainen op zich, naast normale risico's zoals spierstijfheid, vermoeidheid,…
Ook zullen uw identiteit en alle gegevens die u ons verstrekt in het kader van de Wet ter bescherming van de persoonlijke levenssfeer ten opzichte van de verwerking van persoonsgegevens strikt vertrouwelijk behandeld worden. Uw deelname aan het onderzoek is volledig vrijwillig en u hebt op elk moment het recht, om welke reden dan ook, u uit het onderzoek terug te trekken. U heeft ook op elk moment het recht op inzage in de resultaten van het onderzoek. De gegevens worden 25 jaar lang bewaard op een beveiligde server van de Universiteit.

Dit onderzoek werd voorgelegd aan het Ethisch Comité van het UZA.

Indien u nog vragen heeft, aarzel dan niet om contact op te nemen. Dit kan u doen door een e-mail te sturen naar volgend e-mailadres: febe.sluiter@student.uantwerpen.be

Lijkt dit iets voor u en heeft u Inclusion Body Myositis? Bent u ouder dan 18 jaar? Schrijf u dan zeker in via een e-mail en stuur onderstaande pagina naar me terug.

Met vriendelijke groeten,

Het onderzoeksteam

Promotoren:
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Studente:
Febe Sluiter (studente master 2 Revalidatie en Kinesitherapie); de studente is betrokken bij alle fasen van het onderzoek.
Begeleiders:
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Figure 7.4: Informed Consent part 2

Wetenschappelijk onderzoek: Effect van watertherapie bij volwassenen met Inclusie Body Myositis: een pilootstudie

Toestemmingsformulier

Hierbij verklaar ik de bovenstaande tekst te hebben gelezen en begrepen. Ik heb de mogelijkheid gehad om vragen te stellen en deze werden naar mijn voldoening beantwoord. Ik begrijp het doel van het onderzoek. Ik ga akkoord met het verzamelen, de verwerking en het gebruik van de gegevens zoals hierboven beschreven.

Naam en voornaam:

Adres:

Telefoonnummer waarop we u kunnen bereiken:
E-mail adres:

Geboortedatum:

☐ verleent toestemming om deel te nemen aan het onderzoek
☐ verleent geen toestemming

Datum:

Handtekening: Handtekening van de onderzoeker:

Figure 7.5: Informed Consent part 3

Appendix F: Disclaimer