

Paediatric Rheumatology/Series Editor: L. Wedderburn

## Aquatic fitness training for children with juvenile idiopathic arthritis

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**Objective.** To evaluate the effects of an aquatic training programme for JIA patients.

**Methods.** Fifty-four patients with JIA (age range 5 to 13 yr) participated in this study and were randomized into an experimental ( $n = 27$ ) and a control ( $n = 27$ ) group. The children in the experimental group received a training programme consisting of a 1 h per week supervised training programme in a local pool of approximately 20 sessions. Effects were analysed on the following domains: functional ability, health-related quality of life, joint status and physical fitness.

**Results.** Although all measures improved more in the experimental group than the control group, none of the differences was statistically significant.

**Conclusions.** The current research found no significant effect of an aquatic fitness training programme in children with JIA. Since there were no signs of worsening in health status, one can conclude that this was a safe exercise programme.

KEY WORDS: Randomized controlled trial, Exercise, Functional ability, Quality of life, Endurance.

Patients with juvenile idiopathic arthritis (JIA) show a decreased physical fitness compared with healthy children [1]. In addition, JIA patients perform less strenuous physical activities than their healthy peers [2], and are often restricted in their activities of daily living. They may experience a limitation in functioning in one or more joints, stiffness or fatigue due to the arthritis. This may have considerable impact on the patients' daily functioning. For decades, rest was thought to be, besides medication, the predominant treatment for children with JIA and adults with rheumatoid arthritis (RA) [3]. This attitude recently altered. In the past there were major concerns about detrimental effects of physical exercise; nowadays evidence is growing of the beneficial effects of regular exercise for arthritis patients [4, 5]. Since aquatic therapy has been used for centuries to treat musculoskeletal disorders [4], and has proven to be safe in adult RA patients [6], and a very enjoyable form of therapy for

children [7], we developed an aquatic aerobic physical training programme for JIA patients.

Previously published studies on the effects of physical training programmes for JIA patients were small pilot studies [8–11], were not controlled by randomization [7, 12, 13], and had small sample sizes. In a previous publication we described the pilot study for this larger randomized clinical trial [10]. In the previous conducted studies on this topic in JIA, the main focus was on improvements in physical fitness and on impairments like joint swelling and range of motion. None of the previous studies used contemporary, and recently validated, functional assessment tools (e.g. Childhood Health Assessment Questionnaire) as outcome measures for their study. As these instruments are increasingly accepted as outcome measures in clinical trials in paediatric rheumatology, it is important for comparison with other studies to include

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Submitted 25 October 2002; revised version accepted 4 March 2003.

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these instruments as an outcome measure in an intervention study. Therefore we studied the effects of an aquatic training programme on physical fitness, functional ability, joint status, and health-related quality of life in a randomized controlled design.

## Subjects and methods

### Subjects

Fifty-four patients (38 girls, 16 boys) diagnosed with JIA were recruited from the paediatric rheumatology out-patient clinics of the Wilhelmina Children's Hospital, University Medical Center Utrecht, the Netherlands and the University Hospital Groningen, the Netherlands. Inclusion criteria were: diagnosed with JIA by a medical specialist (EULAR criteria or ILAR criteria [14]); a phase of remission without medication of no longer than 6 months in the absence of joint pain, tenderness and/or morning stiffness, and an erythrocyte sedimentation rate within normal limits. All patients had received a local and/or systemic arthritis-related therapy consisting of non-steroidal anti-inflammatory drugs and/or disease-modifying anti-rheumatic drugs and/or immunosuppressive medication and/or steroids in the last 6 months prior to inclusion.

Exclusion criteria were: a systemic disease with fever, low haemoglobin level and a general feeling of malaise; exercise contraindication by a medical specialist; a recipient of a bone marrow transplant; and not feeling confident in water.

The characteristics of the patients are described in Table 1. The children and parents were informed of all aspects of the study, and written consent was obtained. The human ethical committees of the University Medical Center Utrecht/Wilhelmina Children's Hospital and the University Hospital Groningen approved the study.

### Design

Patients were stratified to disease subclass [two strata: an oligoarticular JIA group and a polyarticular (with or without systemic onset) JIA group] and individually randomly assigned to the assessment-only group (Con-group) or the training group (Exp-group) by an off-site data-manager. Three measurements were included in the study protocol: (T1) just before the start of the training programme, (T2) 3 months after the start, and (T3) immediately after the end of the training programme. The investigators and the subjects were blinded for previous measurements at each stage of the evaluation; the investigators and subjects were not blinded for the group allocation of each subject.

### Training programme

All patients received their usual care and medical treatment during the study. The patients in the Exp-group received an aquatic group (2–4 children/group) exercise programme, 1 h a week, supervised by an instructed community physical therapist. If there was only one patient at a certain training

location, the patient was allowed to bring a relative or friend to join the training programme. The training took place in heated (30–33°C) community-based pools at 20 different locations throughout The Netherlands. The training programme was conducted within a 6-month period. Owing to holidays, seasonal holidays and pool maintenance, the programme consisted of approximately 20 training sessions at each of the different locations.

The programme was available on paper and on instructional videotape, and consisted predominantly of aerobic exercises. Training sessions were based on the same framework. The training started with a warm-up, followed by an aerobic conditioning part, a short rest period, and then a second conditioning part. The training ended with a cooling-down. The warm-up, rest and cooling-down periods consisted of low intensity swimming, aquarobics, play, flexibility exercises or ball games. The conditioning parts consisted mainly of high intensity swimming, diving, walking through the water, aqua jogging or splashing with the legs. The duration and intensity of both conditioning parts increased stepwise throughout the programme. During the training sessions the heart rates of the patients were measured using a portable heart-rate monitor (Polar Accurex Plus, Polar Oy, Kempele, Finland) to assess training intensity.

### Functional ability

Functional ability was measured using the Childhood Health Assessment Questionnaire (CHAQ) [15] and the Juvenile Arthritis Functional Assessment Scale (JAFAS) [16]. The Dutch translation of the CHAQ [17] was used as a self-administered pencil and paper questionnaire for the parents (proxy). The CHAQ has been adapted from the Stanford Health Assessment Questionnaire so that at least one question in each domain is relevant to children aged 0.6–19 yr. The questionnaire consists of 30 items divided into eight domains. The question with the highest score within each domain (range 0 to 3; able to do with no difficulty = 0, able to do with some difficulty = 1, able to do with much difficulty = 2, unable to do = 3) determined the score for that domain, unless aids or assistance were required (raising the score for that domain to a minimum of 2). The mean of the scores in the eight domains provided the CHAQ disability scale (range 0 to 3) and was chosen as the primary outcome measure of this study.

The Juvenile Arthritis Functional Assessment Scale (JAFAS) [16] is a performance test assessing disability in children with JIA. This test includes 10 activities of daily living. Time to perform each individual item is recorded and compared with a reference time. Each item was scored on a scale from 0–2 (able to perform the task within reference time = 0, perform the task slower than reference time = 1, not able to perform the task = 2).

### Health-related quality of life

Health-related quality of life was assessed with a Dutch translation of the Juvenile Arthritis Quality of Life

TABLE 1. Characteristics of the study participants

Groups	Age (yr)	Weight (kg)	Height (m)	Skin folds (mm)	Onset type	Gender (m/f)
Exp-group	8.66 ± 2.29	31.86 ± 10.1	1.34 ± 0.12	86.0 ± 43.5	11 oJIA, 15 pJIA, 1 sJIA	11/16
Con-group	8.88 ± 1.86	30.3 ± 9.1	1.33 ± 0.11	80.7 ± 27.36	12 oJIA, 14 pJIA, 1 sJIA	3/24

Values are mean ± s.d. Skin folds = sum of seven different skin-fold sites; oJIA, oligoarticular JIA; pJIA, polyarticular JIA; sJIA,

Questionnaire (JAQQ) [18] and the Child Health Questionnaire (CHQ) [17].

The JAQQ is a recently developed disease-specific health-related quality of life questionnaire for children with arthritis [18]. The JAQQ was administered to the patients; it consists of 74 items divided into five subclasses (gross motor function, fine motor function, psychosocial function, general symptoms and a pain assessment section). The patients scored each item on a 7-point Likert scale according to how often they had encountered problems during the last 14 days (none of the time = 1, hardly any of the time = 2, some of the time = 3, half of the time = 4, most of the time = 5, almost all of the time = 6, all of the time = 7). The score on the JAQQ was calculated as the sum of the five highest scores in each domain of the JAQQ. A higher score indicates a worse health-related quality of life.

The Child Health Questionnaire Parent-Form 50 (CHQ) [17] is a parent proxy report for assessing general health. A Dutch translation of the CHQ was used in this study [17]. The questionnaire consists of 50 items in 14 dimensions. From these dimensions a summary can be calculated for physical (CHQ-PhS) and psychosocial health (CHQ-PsS). The CHQ was administered to the parents; a higher score reflects a better health status of the child.

### Joint status

Joint status was assessed by the number of tender and swollen joints and the range of motion. Tenderness and swelling were scored for the following joints: temporomandibular, sternoclavicular, shoulder, elbow, wrist, thumb, knee, ankle and toes. Joint mobility was scored on the Paediatric Escola Paulista de Medicina Range of Motion Scale (pEPMROM) [19]. The pEPMROM measures mobility in children with JIA based on the evaluation of joint range of motion. Ten joint movements [cervical spine rotation, shoulder abduction, wrist flexion and extension, thumb flexion (metacarpophalangeal), hip internal and external rotation, knee extension, and ankle dorsiflexion and plantar flexion] were examined using a goniometer and classified on a 4-point Likert scale (0 = no limitation, 3 = severe limitation). The final score was calculated as the sum of each movement score divided by 20.

### Physical fitness

The physical fitness of the patients was assessed using a maximal exercise test (MXT) and a submaximal 6-min walking test.

Subjects underwent a MXT on an electronically braked cycle ergometer (Lode Examiner, Lode BV, Groningen, The Netherlands). The patients who did not fit on this ergometer were tested on a mechanically braked ergometer (Tunturi, Finland). The seat height was adjusted to the patient's leg length. Three minutes of unloaded cycling preceded the application of resistance to the ergometer. Thereafter, the workload was increased in constant increments of 20 watts every 3 min. This protocol continued until the patient stopped owing to volitional exhaustion, despite strong verbal encouragement from the experimenters. During the test the subjects breathed through a facemask (Hans Rudolph Inc., Kansas City, MO, USA) connected to a calibrated metabolic cart (Oxycon Champion, Jaeger, Breda, The Netherlands). Expired gas was passed through a flow meter, an oxygen ( $O_2$ ) analyser and a carbon dioxide ( $CO_2$ ) analyser. The flow meter and gas analysers were connected to a computer, which calculated breath-by-breath minute ventilation ( $\dot{V}E$ ), oxygen consumption ( $\dot{V}O_2$ ), carbon dioxide production ( $\dot{V}CO_2$ ) and

respiratory exchange ratio (RER) from conventional equations (Jaeger Masterlab Software, Jaeger, Breda, The Netherlands). Heart rate (HR) was measured continuously during the maximal exercise test by a bipolar electrocardiogram. Absolute peak oxygen consumption ( $\dot{V}O_{2peak}$ ) was taken as the average value over the last 30 s during the maximal exercise test.

Submaximal endurance was measured using the 6-min walking test, as recommended for patients with arthritis [20]. The 6-min walking test was performed on an 8-m track in a straight corridor. The patients were instructed to walk at their own chosen walking speed from one end of the corridor to the other, turn and walk back. The total distance covered in 6 min was calculated as the counted 'repetitions' and multiplied by 8 m. Time was measured with a stopwatch. During the test, standardized verbal support from the test leader was used to encourage the subjects. Before the assessment of physical fitness, anthropometric measurements were taken: height, weight and skin-fold thickness, as previously described [21].

### Statistical analysis

All data were entered into SPSS data entry 3.0 and analysed using SPSS-base 10.0 for Windows (SPSS Inc, Chicago, IL, USA). The General Linear Model [repeated measures ANOVA group (2)  $\times$  time (3)] was used to analyse the effects of the aerobic aquatic training programme. Greenhouse-Geisser epsilon was used for adjusting the degrees of freedom when the sphericity assumption was not met. An alpha level  $< 0.05$  was considered as statistically significant.

## Results

Descriptive characteristics of the subjects at baseline are presented in Table 1. No significant differences were found at baseline between the Exp-group and Con-group on CHAQ, JAFAS, JAQQ, CHQ-PsS, CHQ-PhS, joint status or physical fitness levels.

The patients attended a mean number of  $19.6 \pm 3.9$  training sessions. There was one dropout during the training programme; one boy (13 yr old with an oligoarticular JIA onset type) stopped the training programme after 15 training sessions. Since he still met the 75% criteria of 20 sessions, his data were not excluded from the analysis.

### Functional ability

For the CHAQ and the JAFAS, mean scores and standard deviations of each group are presented in Table 2. A lower score on both instruments means better performance on functional ability and functional skills. Table 2 shows  $F$  values of the main effect of time, and the main effect of group  $\times$  time interaction. No significant effects were found; however, the Exp-group improved 27% in CHAQ score, while the Con-group improved only 5%.

The scores on the JAFAS were very low (a mean score of 0.15 at baseline for the EXP-group). The range of the JAFAS is from 0 to 2; a score of 0.15 is very close to the lowest possible score on this instrument and this phenomenon, a so-called floor effect, would make improvement on this instrument almost impossible.

TABLE 2. Mean scores and standard deviations for both groups and statistics of the analysis of variance for the functional ability instruments CHAQ and JAFAS

	Assessment (mean ± s.d.)			Effect			
				Time		Group × time	
	T1	T2	T3	F	P	F	P
CHAQ <sup>a</sup>							
Exp-group	0.65 ± 0.54	0.56 ± 0.39	0.47 ± 0.45	2.3	0.1	1.03	0.35
Con-group	0.875 ± 0.77	0.79 ± 0.74	0.83 ± 0.79				
JAFAS							
Exp-group	0.14 ± 0.12	0.14 ± 0.14	0.13 ± 0.12	2.2	0.11	0.6	0.55
Con-group	0.22 ± 0.36	0.24 ± 0.35	0.19 ± 0.34				

<sup>a</sup>Adjusted using Greenhouse–Geisser epsilon.

TABLE 3. Mean scores and standard deviations for both groups and statistics of the analysis of variance for the health-related quality of life instruments JAQQ and CHQ

	Assessment (mean ± s.d.)			Effect			
				Time		Group × time	
	T1	T2	T3	F	P	F	P
JAQQ <sup>a</sup>							
Exp-group	12.4 ± 5.6	11.5 ± 4.7	10.6 ± 5.2	1.57	0.22	1.71	0.19
Con-group	14.3 ± 5.0	14.6 ± 5.4	14.34 ± 5.9				
CHQ-PhS <sup>a</sup>							
Exp-group	48.79 ± 11.14	51.89 ± 7.25	52.87 ± 7.42	0.69	0.47	2.67	0.085
Con-group	44.27 ± 13.11	44.04 ± 14.95	42.42 ± 15.8				
CHQ-PsS							
Exp-group	50.24 ± 7.27	52.24 ± 7.48	53.58 ± 5.31	1.62	0.2	2.09	0.13
Con-group	49.32 ± 8.98	50.28 ± 7.46	48.91 ± 8.24				

<sup>a</sup>Adjusted using Greenhouse–Geisser epsilon.

*Health-related quality of life*

JAQQ and CHQ-50 (divided into a physical summary and a psychological summary), mean scores, standard deviations and the *F* values of the main effect of time, and the main effect of group × time interaction are presented in Table 3.

There was a trend for the Con-group to deteriorate in JAQQ score (−15%), as the Exp-group remained stable throughout the intervention period (0% change). However, differences between the two groups did not reach statistical significance.

The Exp-group showed a small improvement over time on the physical and psychological CHQ summaries (8.4 and 7%, respectively), while the Con-group decreased slightly (PhS −4%) or remained stable (0% PsS). These changes almost reached statistical significance.

*Joint status*

Mean scores, standard deviations and the *F* values of the main effect of time, and the main effect of group × time interaction of both swollen and tender joints, and pEPMROM are presented in Table 4. The number of swollen and tender joints decreased in the Exp-group (−55%), while it increased in the Con-group (+21%). These differences were almost statistically significant (*P* = 0.07).

For the range of motion (pEPMROM) there were no significant changes over time as both groups showed a very small decrease over time (18 and 30% for swollen and tender joints, and pEPMROM, respectively).

*Physical fitness*

Results of *VO*<sub>2peak</sub> and the distance covered on the 6-min walking test, mean scores, standard deviations, *F* values of the main effect of time, and the main effect of group × time interaction are presented in Table 5. *VO*<sub>2peak</sub> remained stable during the training period for both the Exp-group (0% change) and Con-group (−3% decrease). The 6-min walk improved slightly (+3%; non-significant) in the Exp-group, while it did not change in the Con-group.

**Discussion**

The aim of this study was to determine the effects of an aquatic training programme for JIA patients on functional ability, quality of life, joint status and physical fitness.

We found no statistically significant effects of a 20-week aquatic training programme for JIA patients on functional ability, quality of life, joint status or physical

TABLE 4. Mean scores and standard deviations for both groups and statistics of the analysis of variance for the joint status: number of swollen and tender joints and pEPMROM

	Assessment (mean $\pm$ s.d.)			Effect			
				Time		Group $\times$ time	
	T1	T2	T3	F	P	F	P
Swollen and tender joints							
Exp-group	2.5 $\pm$ 2.7	2.2 $\pm$ 2.5	1.11 $\pm$ 1.3	0.92	0.41	2.7	0.07
Con-group	2.9 $\pm$ 4.7	3.6 $\pm$ 5.1	3.6 $\pm$ 4.9				
pEPMROM							
Exp-group	0.11 $\pm$ 0.19	0.15 $\pm$ 0.26	0.13 $\pm$ 0.22	3.0	0.08	0.3	0.057
Con-group	0.23 $\pm$ 0.39	0.22 $\pm$ 0.42	0.3 $\pm$ 0.45				

TABLE 5. Mean scores and standard deviations for both groups and statistics of the analysis of variance for physical fitness measures  $\dot{V}O_{2peak}$  and the 6-min walking test

	Assessment (mean $\pm$ s.d.)			Effect			
				Time		Group $\times$ time	
	T1	T2	T3	F	P	F	P
$\dot{V}O_{2peak}^a$							
Exp-group	1.11 $\pm$ 0.32	1.21 $\pm$ 0.41	1.11 $\pm$ 0.41	7.746	0.001	0.736	0.46
Con-group	1.07 $\pm$ 0.35	1.12 $\pm$ 0.34	1.00 $\pm$ 0.34				
6-mwt <sup>a</sup>							
Exp-group	455.0 $\pm$ 71.8	471.1 $\pm$ 78.8	471.9 $\pm$ 67.58	1.281	0.28	0.395	0.63
Con-group	458.1 $\pm$ 76.9	469.4 $\pm$ 86.6	457.1 $\pm$ 105.4				

$\dot{V}O_{2peak}$ , maximum oxygen consumption (litre/min); 6-mwt, 6-min walking test.

<sup>a</sup>Adjusted using Greenhouse–Geisser epsilon.

fitness. However, there was a clear trend of an improved joint status during the training programme. The small, non-significant effects could be explained by the chosen outcome measures. We hypothesized the aquatic exercises would improve physical fitness. This increased physical fitness could result in a better functional ability and improvements in health-related quality of life. However, this transfer of the training effects seems very difficult. Numerous exercise studies in adult RA patients found improvements in physical fitness, joint pain, morning stiffness and fatigue, but no clear improvements in functional ability [22, 23]. In our study, the largest effects were found in the joint status domain.

The aquatic training did not improve the  $\dot{V}O_{2peak}$  of leg exercise on the bicycle ergometer; perhaps an arm-exercise protocol would have shown improvements since many exercises in the programme were performed using both legs and arms.

Moreover, the outcome instruments employed may have lacked the sensitivity to detect the training effects. The majority of the instruments (CHAQ, CHQ, JAFAS, pEPMROM, JAQQ) have been developed and validated as discriminative instruments for disability, and not yet as outcomes for effect studies. However, in clinical practice they are used frequently for evaluative purposes, and the Paediatric Rheumatology International Trials Organisation selected some of them for trial evaluation (CHAQ and CHQ). Van den Ende *et al.* [24] found a very limited sensitivity to change in the adult version of the

CHAQ, the HAQ, in an exercise trial with adult RA patients. The value of the CHAQ for detecting changes resulting from an exercise training programme might be limited. However, there was a positive relationship between the baseline CHAQ score and improvement after the intervention ( $r = 0.62$ ,  $P < 0.05$ ), indicating that patients with a higher level of disability benefited more from the aquatic training than the patients with a lower level of disability.

The lack of improvements in the current study could also be the result of a limited trainability of children with JIA. Even in healthy children, some investigators have found no or small effects on physical fitness of exercise training programmes [25]; our data more or less confirm this finding for JIA patients. A meta-analysis of all available exercise training programmes showed on average an 8% increase in physical fitness in healthy children [26]. Compared with adults, the degree of aerobic trainability is somewhat limited in children. Additionally, for detained, chronically ill patients it might not be possible to adhere to programmes based on adult standards for intensity and duration. They might be compromised in their level of trainability.

There are some guidelines available for children with arthritis, but they are based on preliminary evidence [27]. Moderate-intensity exercise, as in our programme, will facilitate most of the disease prevention and health promoting effects of exercise [28], but these are long-term effects of exercise training.

Our programme lasted approximately 6 months, but healthy peers participate for years in certain sport activities such as soccer, cycling, tennis or athletics. A long-term exercise training study (>2yr) should be performed with the current patient group.

However, our results clearly indicated that the programme, as measured by joint status, does not negatively influence the disease process. As parents and JIA patients were very keen to participate in adapted physical training programmes, this study shows that an aquatic training programme could be a valuable adjunct to the common medical care of children with JIA.

Limited information is available concerning changes in physical fitness and functional ability in JIA patients following an exercise training programme. Thus more research is needed in this area. From a practical point of view, it was not possible to increase the frequency of training (i.e. two training sessions a week) owing to busy family schedules and limited pool access. The inclusion of a home-based exercise component (land-based) should be considered in a future study. This might also support patients and parents in achieving a more active lifestyle. Moreover, studies on healthy adults have shown that a higher exercise frequency (more than 3 days/week) is needed for an improvement in physical fitness [29]. However, one must bear in mind that JIA patients do have a diminished load-bearing capacity because of their inflammatory disease and the immunosuppressive medication. Exercise training might improve immune function [30], but a training load which is too large might easily lead to overreaching or overtraining, as is often seen in athletes with a compromised immune system [31].

We tried to find a minimum level of exercise training that could improve functional ability and fitness, but did not impede the immune system.

The lack of significant improvements in the training group also shows the importance of an active lifestyle for children with JIA. The current research project shows that it is very difficult to improve physical fitness. The sound bite 'it is easier to maintain good health through proper exercise, diet and emotional balance than to regain it once it is lost' might also be true for JIA patients.

## Conclusion

With the advances in the medical treatment of JIA patients using new and successful treatments the interest in adapted physical training programmes is increasing. The effects of adapted training programmes have not been well investigated. The current research found small, non-significant effects of an aquatic fitness training programme in children with JIA. Since there were no signs of worsening in health status, one can conclude that this was a safe exercise programme. As there are many different exercise modalities, effort should be made to create exercise training programmes and investigate their safety and effectiveness in children with JIA.

## Acknowledgements

We would like to thank O. Lelieveld, L. Frolek and Dr W. van Luyck from the University Hospital Groningen, E. van Bostelen, A. Hamer, S. Holleman, E. Nijkamp, Dr C. Volman, and Dr N. van Veldhoven from the Faculty of Social Sciences, Utrecht University, and all participating patients, parents, trainers and physical therapists for their dedicated co-operation.

## Conflict of interest

The authors have declared no conflicts of interest.

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