

Do Juvenile Idiopathic Arthritis Patients Benefit From an Exercise Program? A Pilot Study

TIM TAKKEN, JANJAAP VAN DER NET, AND PAUL J. M. HELDERS

Introduction

It is known from studies of adults with rheumatoid arthritis that they benefit from exercise (1). There is some evidence that children with juvenile idiopathic arthritis (JIA) can benefit from exercise as well (2,3); at least there is evidence that exercise does not exacerbate the arthritis (4,5).

Children with a rheumatic disease, and other chronic diseases, often live an inactive life. This leads to deconditioning and functional deterioration, which again promote an inactive lifestyle (6). A training program could prevent the deconditioning due to hypoactivity and break the vicious circle, thus preventing this major risk factor for comorbidity (7).

Until recently, the main focus of exercise interventions for patients with a rheumatic disease was to study adverse effects on inflammation and joint disease and the effects on physical fitness. Previous research found that children with arthritis benefit from an aquatic exercise program (2,3,8). Recently, Klepper (4) found that land-based exercise could improve physical fitness in JIA patients. But whether exercise could influence patient-centered outcome measures (i.e., functional ability and health-related quality of life) in JIA patients is relatively unknown. Therefore, the aim of the current study was to investigate whether an aerobic aquatic training program could improve not only the endurance of JIA patients but also the functional ability and health-related quality of life if added to the usual medical and physical therapeutic care.

Tim Takken, MSc, Janjaap van der Net, PhD, and Paul J. M. Helders, MSc, PhD, Department of Pediatric Physical Therapy, University Hospital for Children and Youth, "Het Wilhelmina Kinderziekenhuis," University Medical Center Utrecht, Utrecht, The Netherlands.

Address correspondence to Paul J. M. Helders, MSc, PhD, Professor, Department of Pediatric Physical Therapy, University Hospital for Children and Youth, "Het Wilhelmina Kinderziekenhuis," University Medical Center Utrecht, Room KB.02.056, P.O. Box 85090, 3508 AB Utrecht, The Netherlands.

Submitted for publication May 31, 2000; accepted in revised form October 28, 2000.

Methods

The study started in January 1999. Twenty-five patients (age range 5–12 years) who lived within 25 km of our hospital and were diagnosed with JIA (International League of Associations of Rheumatology criteria [9]) in the pediatric immunology/rheumatology outpatient clinic were asked to participate in a pilot study. Ten patients with JIA, all females, and their parents agreed to participate in the study. The parents signed their informed consent during the first visit to our department. The patient characteristics are shown in Table 1.

Joint status was assessed by the number of tender and swollen joints. 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 Pediatric Escola Paulista de Medicina Range of Motion Scale (pEPMROM [10]). The pEPMROM measures mobility in children with JIA based on the evaluation of joint range of motion. Ten joint movements (cervical spine lateral 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 10. Both joint status and joint mobility were scored by the same senior pediatric physical therapist with 20 years of experience in pediatric rheumatology (JvdN).

The Dutch translation of the Childhood Health Assessment Questionnaire (CHAQ), Pediatric Rheumatology International Trials Organization (PRINTO) version, was used as a self-administered pencil and paper questionnaire for the parents (proxy), as an index of functional ability. The CHAQ (11) 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 to 19 years. The CHAQ was recently cross-culturally adapted and validated for the Dutch language by the PRINTO group (12). 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; time frame was last week) determined

Table 1. Subject characteristics

Subject	Sex*	Age	DS†	YA‡	NSTJ§	pEPMROM¶	Medication#	PT**
1	F	10	SJIA	7.4	0	0.0	—	+
2	F	10	SJIA	2.3	2	0.1	PRE, MTX, NSAID	-
3	F	10	OJIA	5.5	5	0.1	DMARD, NSAID	-
4	F	7	OJIA	5.5	2	0.1	NSAID	-
5	F	9	PJIA	2.7	4	0.6	DMARD, NSAID	+
6	F	8	PJIA	6.8	7	0.65	PRE, MTX, NSAID	+
7	F	12	PJIA	6.5	11	0.1	DMARD, NSAID	+
8	F	10	OJIA	7.6	3	0.8	NSAID	+
9	F	12	SJIA	7.8	2	0.4	MTX, NSAID, CIC	+
10	F	5	OJIA	4	1	0.0	NSAID	-

* All subjects were female.

† DS = disease subclass; SJIA = systemic juvenile idiopathic arthritis (JIA); OJIA = oligoarticular JIA; PJIA = polyarticular JIA.

‡ YA = total years of arthritis.

§ NSTJ = number of swollen and tender joints.

¶ pEPMROM = score on the Pediatric Escola Paulista de Medicina Range of Motion Scale.

PRE = prednisone; MTX = methotrexate; NSAID = nonsteroidal anti-inflammatory drugs; DMARD = disease-modifying antiheumatic drugs; CIC = cyclosporin.

** PT = physical therapeutic care; + = yes; - = no.

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 on the 8 domains provided the CHAQ disability scale (range 0 to 3).

Endurance was measured with the 6-minute walking test, as recommended for patients with arthritis (13). The 6-minute walking test was performed on an 8-meter track in a straight corridor. The patients were instructed to walk at their own chosen walking speed from one side of the corridor to the other, turn, and walk back. The total distance covered in 6 minutes was calculated as the counted "repetitions" and multiplied by 8 meters. Time was measured with a stopwatch. During the test standardized verbal encouragements from the test leader were used to encourage the subjects.

Health-related quality of life was assessed with a Dutch translation of the Juvenile Arthritis Quality of Life Questionnaire (JAQQ). The JAQQ is a recently developed disease-specific health-related quality of life questionnaire for children with arthritis (14). The JAQQ was administered to the patients. The JAQQ consists of 74 items divided into 5 subclasses (gross motor function, fine motor function, psychosocial function, general symptoms, and unclassified). The children scored each item on a 7-point Likert scale according to how often they 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). Pain was measured on a 10-cm double anchored visual analog scale (VAS). In the original article the JAQQ was only scored on 5 particular items of interest per domain. This approach results in a different questionnaire on each occasion for each patient and makes group statistics difficult to interpret (15). Therefore, the score on the JAQQ was calculated as (a) the sum of the scores on all 74 items of the questionnaire, (b) the score on its subscales, and (c) the VAS for pain. A higher score indicates a worse health-related quality of life.

The children in this study received a 15-week aerobic aquatic training program. The exercise groups (2–4 pa-

tients) were supervised by a pediatric physical therapist (JvdN) and an exercise physiologist (TT). The training took place in a heated swimming pool (temperature of water \pm 32°C). The training activities were predominantly such exercises as swimming, aerobics, and ball games. Lessons were based on the same framework. The training started with a warm-up, followed by a conditioning part. After this conditioning part a small rest period was included. This period was followed by a second conditioning part. The training ended with a cooling down. The warm-up, rest, and cooling down period consisted of low-intensity

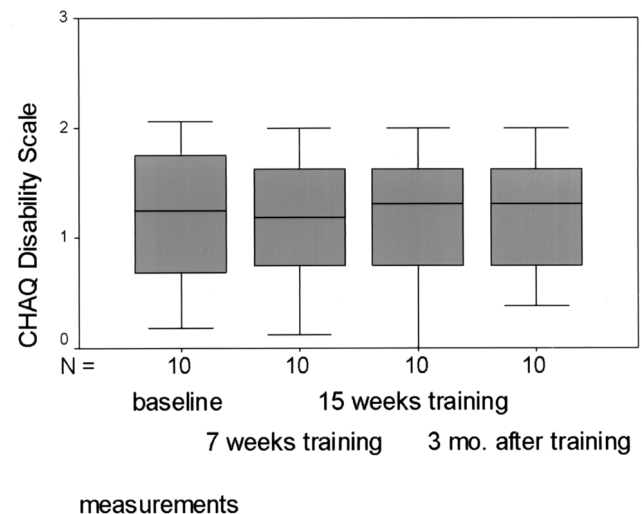


Figure 1. Box plot of the scores in functional ability (disability scale of the Childhood Health Assessment Questionnaire [CHAQ]) during the 4 assessments. Differences between the measurements are not statistically significant. Measurements: baseline, after 7 weeks of training, immediately after 15 weeks, and 3 months after the end of the training program. The whiskers represent the highest and lowest values within 1.5 times the interquartile range (box length, IQC). The lower horizontal line of the box indicates the 25th percentile. The upper horizontal line indicates the 75th percentile. The middle horizontal bar in the box indicates the median.

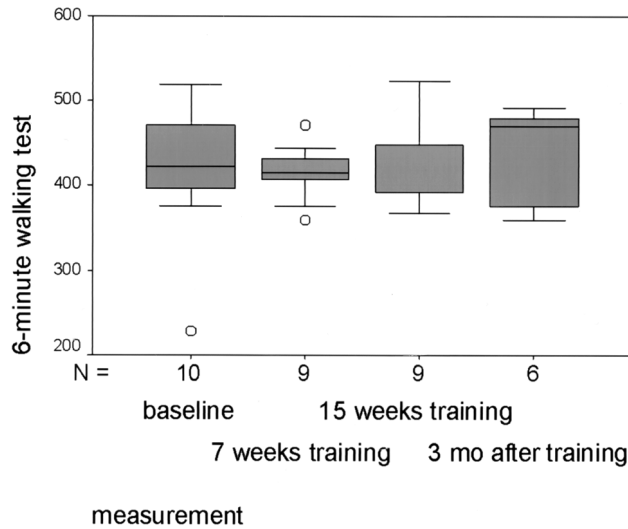


Figure 2. Box plot of the scores on the 6-minute walking test during the 4 assessments. Differences between the measurements are not statistically significant. Measurements: baseline, after 7 weeks of training, immediately after 15 weeks, and 3 months after the end of the training program. The O symbol indicates an outlier between 1.5 and 3 times the interquartile range (IQR) from the box. The whiskers represent the highest and lowest values within 1.5 times the IQR (box length, IQR). The lower horizontal line of the box indicates the 25th percentile. The upper horizontal line indicates the 75th percentile. The middle horizontal bar in the box indicates the median.

swimming, aquarobics, play, flexibility exercises, or ball games. The conditioning parts consisted mainly of high-intensity swimming, diving, walking through the water, aquajogging, or splashing with the legs. The duration and intensity of both conditioning parts increased stepwise during the program. During the training sessions the heart rates of the children were measured using a portable heart rate monitor (Polar Accurex Plus, Polar Oy, Kempele, Finland) to get an impression of the training intensity and prevent overreaching. The children received one training session per week, each lesson lasting 60 minutes.

There were 4 measurements: baseline (mean of 2 assessments), after 7 weeks of training, immediately after 15 weeks, and 3 months after the end of the training program.

Statistical analysis was performed with SPSS 8.0 for Windows (SPSS, Chicago, IL). The non-parametric Friedman test (16) was used for analyzing the 4 repeated measurements on all the measures (CHAQ, 6-minute walking test, and JAQQ). When the differences between the groups appeared to be significant ($P < 0.05$), the Wilcoxon signed-rank test was used to detect the significant differences. The study was approved by the human ethical committee of the University Medical Center Utrecht, Wilhelmina Children’s Hospital.

Results

The attendance during the training sessions was 85%. The major reasons for absence were disease, no available transportation, and other social peer activities. At the fourth measurement, because of nonadherence of one patient, the respective parents returned the CHAQ by mail. The mean

CHAQ disability scale showed a trend to improve from 1.21 (SD \pm 0.6, baseline) to 1.16 (SD \pm 0.6) after the training program. Three months after finishing, the CHAQ disability scale decreased to baseline level (1.21; SD \pm 0.6) as shown in Figure 1. However, these changes were not statistically significant.

The covered distances during the 6-minute walking test—baseline, during and immediately after the end of the training program, and 3 months after the end of the training program—were 440 meters (SD \pm 54.0), 415.6 meters (SD \pm 33.7), 436 meters (SD \pm 50.4), and 436 meters (SD \pm 50.4). The changes are shown in Figure 2. The values did not differ significantly from baseline and from each other. One patient was not able to accomplish the 6-minute walking test in the second, third, and fourth assessments because of arthritis-related problems in the ankle. Two other patients did not accomplish the 6-minute walk on the fourth assessment because of pain and fatigue. One patient did not comply with the fourth assessment.

The mean JAQQ subscale scores improved by 16% (NS—not significant), 19% (NS), 16% (NS), 12% ($P < 0.05$), and 17% (NS), respectively, for physical function, psychosocial function, general symptoms, and the VAS for pain after the end of the training program compared with baseline. The overall score on the JAQQ is shown in Figure 3. The improvements in the JAQQ during the training program (assessments 1, 2, and 3) were not statistically significant. However, the relapse after the training program (the differences between assessments 3 and 4) reached statistical significance. Two patients did not accomplish the JAQQ on the fourth assessment. One patient did not

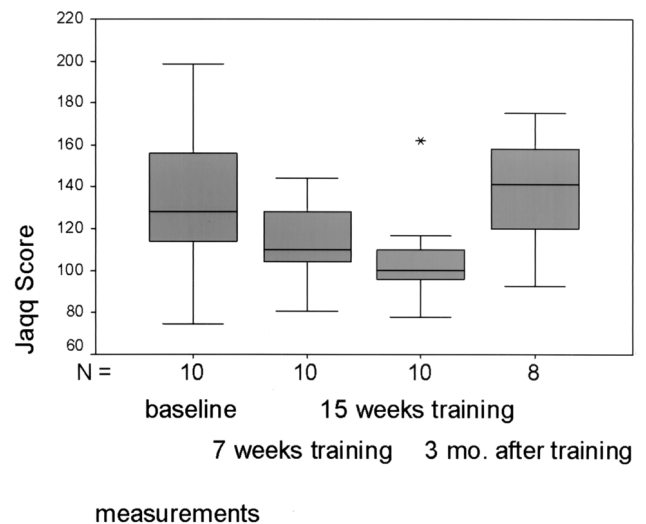


Figure 3. Box plot of the score changes in health-related quality of life (mean score on the total Juvenile Arthritis Quality of Life Questionnaire [Jaqq]) during the 4 assessments. The differences between the third and fourth assessments are significant. Measurements: baseline, after 7 weeks of training, immediately after 15 weeks, and 3 months after the end of the training program. The * symbol indicates an extreme lying more than 3 times the interquartile range (IQR) from the box. The whiskers represent the highest and lowest values within 1.5 times the IQR (box length, IQR). The lower horizontal line of the box indicates the 25th percentile. The upper horizontal line indicates the 75th percentile. The middle horizontal bar in the box indicates the median.

comply with the fourth assessment, and one patient failed to complete the entire questionnaire.

Conclusions

Our main interest was to investigate whether a 15-week aerobic aquatic training program could improve the functional ability, endurance, and health-related quality of life of JIA patients if added to the common medical and physical therapeutic care.

The high attendance rate of the current training program reflected good adherence of the parents and patients to our training intervention. It also reflects the high interests of parents and patients with JIA in adapted physical activity programs, which may be a more effective and economic method of providing therapeutic and recreational exercises to this population.

The lack of change of the CHAQ disability scale could be explained by the fact that the CHAQ was originally developed as a discriminative instrument for disability and not as an outcome measure for effect studies (8). Flato and colleagues (17) concluded that the Norwegian version of the CHAQ was sensitive to clinical change. However, the way in which the CHAQ disability scale is calculated (the highest score within a domain determining the score of that domain, the requirement of aids or assistance raising the score on that domain to a minimum of 2) makes it difficult to detect changes in functional ability in the "home situation."

Recently, the 6-minute walking test was proposed as a potential outcome measure for hydrotherapy (18). However, our subjects did not improve as to the distance they covered. It is difficult to explain the results for the 6-minute walking test, because these sorts of tests not only depend on physical fitness but are also highly influenced by pain, fatigue, motivation, muscle strength, and encouragement (19). Further experience will reveal its validity.

The JAQQ subscale scores on the gross motor function and fine motor function confirmed the results from the CHAQ disability score, indicating that there were no significant changes in functional ability. Our program was also not able to significantly improve the psychosocial function domain and pain. On the other hand, the general symptoms domain improved, and the change was statistically significant; thus, the patients encountered fewer disease signs and symptoms during the exercise program. This has been confirmed in a land-based aerobic intervention as well (4). However, it has been questioned whether a separate analysis on the different domains within one quality of life instrument is appropriate (20). The results on the JAQQ might have been different if this instrument had been used in the original manner. In that approach patients can pick or add items of interest in each domain of the JAQQ.

The trend toward an improvement of the overall health-related quality of life score during the training program is in accordance with the existing literature on adults with a chronic disease (20). Surprisingly, this improvement did not reach statistical significance. This could be explained by the large differences in response between subjects due to the heterogeneous patient population and the small

sample size that we used for this study. The decreased health-related quality of life in the 3 months after the end of the training program suggests that JIA patients can benefit from an exercise program or a more active lifestyle.

The changes in the outcome of our measurements can be explained by the exercise program, the seasonal influences, the repeated-measurements design of our study (21), the limited number of patients, or a combination of these factors. A randomized design, a control group, and a larger number of patients are necessary to take these factors into account.

In conclusion, an aquatic training program positively influenced the health-related quality of life of JIA patients, but it had no significant effects on endurance and functional ability. Further study should investigate these effects in a randomized design and with a larger number of patients.

REFERENCES

1. Van den Ende CH, Vliet Vlieland TP, Munneke M, Hazes JM. Dynamic exercise therapy in rheumatoid arthritis: a systematic review. *Br J Rheumatol* 1998;37:677-87.
2. Bacon MC, Nicholson C, Binder H, White PH. Juvenile rheumatoid arthritis: aquatic exercise and lower-extremity function. *Arthritis Care Res* 1991;4:102-5.
3. Baldwin J. Pool therapy compared with individual home exercise therapy for juvenile rheumatoid arthritic patients. *Physiotherapy* 1972;58:230-1.
4. Klepper SE. Effects of an eight-week physical conditioning program on disease signs and symptoms in children with chronic arthritis. *Arthritis Care Res* 1999;12:52-60.
5. Kirchheimer JC, Wanivenhaus A, Engel A. Does sport negatively influence joint scores in patients with juvenile rheumatoid arthritis: an 8-year prospective study. *Rheumatol Int* 1993;12:239-42.
6. Bar-Or O. Pediatric sports medicine for the practitioner. In: Katz M, Steihm ER, editors. *Comprehensive manuals in pediatrics*. New York: Springer-Verlag; 1983. p. 66-85.
7. Bell RD, Macek M, Rutenfranz J, Saris WHM. Health indicators and risk factors of cardiovascular diseases during childhood and adolescence. In: Rutenfranz J, Mocellin R, Klimt F, editors. *Children and exercise XII*. Champaign (IL): Human Kinetics; 1986. p. 19-27.
8. Oberg T, Karsznia A, Gare BA, Lagerstrand A. Physical training of children with juvenile chronic arthritis: effects on force, endurance and EMG response to localized muscle fatigue. *Scand J Rheumatol* 1994;23:92-5.
9. Petty RE, Southwood TR, Baum J, Bhattay E, Glass DN, Manners P, et al. Revision of the proposed classification criteria for juvenile idiopathic arthritis: Durban, 1997. *J Rheumatol* 1998; 25:1991-4.
10. Len C, Ferraz MB, Goldenberg J, Oliveria LM, Araujo PP, Quresma MR, et al. Pediatric Escola Paulista de Medicina Range of Motion Scale: a reduced joint count scale for general use in juvenile rheumatoid arthritis. *J Rheumatol* 1999;26: 909-13.
11. Singh G, Athreya BH, Fries JF, Goldsmith DP. Measurement of health status in children with juvenile rheumatoid arthritis. *Arthritis Rheum* 1994;37:1761-9.
12. Ruperto N, for the Pediatric Rheumatology International Trials Organisation (PRINTO). An international survey in 29 countries on health related quality of life (HRQOL) in juvenile idiopathic arthritis (JIA). I. A disease specific instrument: the Childhood Health Assessment Questionnaire (CHAQ) [abstract]. *Ann Rheum Dis* 2000;59:726.
13. Minor MA, Kay DR. Arthritis. In: Durstine JL, editor. *ACSM's exercise management for persons with chronic diseases and*

- disabilities. Champaign (IL): Human Kinetics; 1997. p. 149–54.
14. Duffy CM, Arsenault L, Duffy KN, Paquin JD, Strawczynski H. The Juvenile Arthritis Quality of Life Questionnaire—development of a new responsive index for juvenile rheumatoid arthritis and juvenile spondyloarthritis. *J Rheumatol* 1997; 24:738–46.
 15. Wright JG. Evaluating the outcome of treatment: shouldn't we be asking patients if they are better? *J Clin Epidemiol* 2000; 53:549–53.
 16. Friedman M. The use of ranks to avoid the assumption of normality implicit in the analysis of variance. *J Am Stat Assoc* 1937;32:675–701.
 17. Flato B, Sorskaar D, Vinje O, Lien G, Aasland A, Moum T, et al. Measuring disability in early juvenile rheumatoid arthritis: evaluation of a Norwegian version of the Childhood Health Assessment Questionnaire. *J Rheumatol* 1998;25:1851–8.
 18. Gowans SE, deHueck A, Voss S. Six-minute walk test: a potential outcome measure for hydrotherapy. *Arthritis Care Res* 1999;12:208–11.
 19. Rowland TW. Oxygen uptake and endurance fitness in children: a developmental perspective. *Pediatr Exerc Sci* 1989;1:313–28.
 20. Rejeski WJ, Brawley LR, Shumaker SA. Physical activity and health-related quality of life. *Exerc Sport Sci Rev* 1996;24:71–108.
 21. Beunen G, Simons J, Ostyn M, Renson R, van Gerven D. Learning effects in repeated measurement designs. In: Berg K, Eriksson BO, editors. *Children and exercise IX*. Baltimore: University Park Press; 1980. p. 139–45.