

Juvenile Arthritis and Exercise Therapy: Current Research and Future Considerations

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Abstract

Juvenile Idiopathic Arthritis (JIA) is a chronic condition affecting significant numbers of children and young adults. Symptoms such as pain and swelling can lead to secondary conditions such as altered movement patterns and decreases in physical activity, range of motion, aerobic capacity, and strength. Exercise therapy has been an increasingly utilized component of treatment which addresses both primary and secondary symptoms. The objective of this paper was to give an overview of the current research on different types of exercise therapies, their measurements, and outcomes, as well as to make recommendations for future considerations and research. After defining the objective, articles involving patients with JIA and exercise or physical activity-based interventions were identified through electronic databases and bibliographic hand search of the existing literature. In all, nineteen articles were identified for inclusion. Studies involved patients affected by multiple subtypes of arthritis, mostly of lower body joints. Interventions ranged from light systems of movement like Pilates to an intense individualized neuromuscular training program. None of the studies exhibited notable negative effects beyond an individual level, and most produced positive outcomes, although the significance varied. Exercise and physical activity do not worsen the symptoms of juvenile idiopathic arthritis; in fact, they can be extremely beneficial in helping address both the primary symptoms of pain and swelling, and the secondary issues related to range of motion, cardiovascular fitness, and strength. Incorporation of an individual-specific activity plan should be strongly considered as an integral part of a treatment plan.

Keywords: Juvenile Idiopathic Arthritis (JIA); Exercise therapy; Physical activity; Review

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Introduction

Juvenile Idiopathic Arthritis (JIA) is a term that encompasses forms of arthritis (chronic auto inflammation of at least one joint) that are of unknown origin, with onset prior to the age of 16 and symptoms persisting longer than 6 weeks. JIA is the most common form of arthritis in children: studies based in developed countries have reported prevalence rates of between 16 and 150 per 100,000 [1], with an annual incidence between 0.008 and 0.226 per 1000 [2]. Due to this chronic inflammation, individuals can experience pain, stiffness, and a resulting difficulty with everyday activities. The pain and swelling can cause the child to alter their movement and/or posture, leading to muscular imbalances and decreased ranges of motion [3]. Studies examining gait found

that individuals with JIA had decreased walking speed, step length, hip and knee extension, and push-off plantar flexion than healthy controls [4].

These musculoskeletal issues often create additional difficulties for kids with JIA. Symptoms often deter them from being active to such a degree that physical activity and sport participation levels are significantly lower than that of their healthy peers [5]. This trend towards inactivity leads to deconditioning and weight gain, which in turn exacerbates arthritis symptoms, resulting in a difficult to break cycle. As a natural consequence of this deconditioning, JIA patients as a group also exhibit higher submaximal energy expenditure and lower aerobic fitness than non-affected controls [6,7]. Arthritis persists through adulthood in up to 55% of cases, and even when the disease itself is no

longer present, there are consequential long-term effects such as restrictions in joint motion, contractures, and local growth disturbances, and resulting limb length discrepancies [8].

The main treatment for JIA is medication, typically in the form of anti-inflammatories. These can range from over-the-counter NSAIDs to more powerful Disease-Modifying Anti-Rheumatic Drugs (DMARDs) and corticosteroids. Less commonly surgery, splints, and orthotics are used to correct deformities or postures. In the past decade, exercise has become increasingly recognized as a beneficial and vital component of a holistic treatment plan. The most common exercise-centred therapy for JIA initially involved types of aquatic exercise, on the basis that it would be less likely to aggravate joint pain. Eventually, as it was demonstrated that these types of programs could often be implemented without significant increases in pain, methods incorporating weight bearing and strength exercises became increasingly common as well, as have fitness systems like Pilates and qigong.

A physical activity program is essential to break the cycle of inactivity and deconditioning described previously, improving cardiovascular and musculoskeletal strength to levels more comparable to that of their non-diagnosed peers. Engagement in exercise is also beneficial for the same reasons it is in healthy children and adolescents, primarily among them impact loading for bone health, which is of increased significance given that the joints are primarily affected [9,10]. Part of the government initiative 'Healthy people 2020' includes a focus on keeping individuals with chronic conditions as healthy and active as possible [11]. Given that youth patterns of physical activity tend to carry over into adulthood [12-14] and the fact that arthritis persists into adulthood more often than not, developing effective exercise habits in these patients has clear short and long term implications.

This aim of this paper is to survey the current research on exercise interventions in children and young adults with JIA and examine the following: (1) What are the demographics of the populations being studied? (2) What types of interventions are being studied, and what measurements are being employed to determine the efficacy of these interventions? (3) What are the outcomes and (4) What are the future applications and implications of these studies?

Paper Identification

Electronic databases PubMed, CINAHL, Science Direct, and Web of Science were searched between 1990 and July 2015. Keywords included combinations of 'juvenile idiopathic arthritis', 'chronic juvenile arthritis', 'juvenile rheumatoid arthritis', and 'exercise', 'endurance', 'strength training', 'aerobic training', 'physical therapy', 'physical activity', 'aerobic', 'hydrotherapy', and 'aquatic therapy'. A hand search of the bibliographic references of the extracted articles was conducted to capture any publications missed by electronic databases. To be included, articles had to: Incorporate a juvenile population that had an identified subset of juvenile idiopathic arthritis (polyarticular, pauci/oligoarticular, systemic, psoriatic, or enthesitis-related) and have at least one physical-activity or exercise-based intervention with clearly

defined outcome measures. Single case and case studies were included, as was a prospective study on a comparison between children who only underwent physical therapy versus children who participated in sports in addition to their therapy. **Table 1** provides a summary of the existing literature on exercise and sports research involving patients with Juvenile Idiopathic Arthritis.

Study Designs

There were nineteen publications in total, over a 14-year time period (1991 to 2015). The majority were randomized control (eleven) or pilot (five) studies, and included comparisons of an intervention—some type of exercise therapy—to either continuation of current treatment, another type of exercise, or to healthy matched controls. The most common therapies were aquatic (hydro) therapy and/or some form of basic strength and/or aerobic training. Others included an investigation of the effects of Pilates, qigong (a Chinese-based practice focused on posture and breathing), and a highly individualized neuromuscular training program. Session durations all fell between 40 and 60 min. Intervention periods lasted between five weeks and six months, with three and six months being the most used durations.

Demographics

Overall 463 female and 178 male subjects participated in the studies. While the gender breakdown of arthritis varies with characteristics such as location and subset, it is generally accepted that with the exception of enthesitis-related arthritis, JIA is more common in girls. The subtype breakdown was as follows:

- 339 poly articular.
- 212 oligo/pauciarticular.
- 61 systemic.
- 28 enthesitis or 'other'.
- 22 psoriatic.

At 51% and 32%, the prevalence of polyarticular is overrepresented at the expense of oligo-articular-diagnosed individuals, by at least 10% when compared to recent estimates [1]. All the subjects were affected in a lower body joint, and as such most interventions focused on the core and lower body.

Primary Assessment Measures

The most common survey-based measures were pain and quality of life/functional ability scores. The use of a Visual Analog Scale (VAS) was most common for the former, as were the Juvenile Arthritis Quality of Life Questionnaire (JAQQ), Pediatric Quality of Life Inventory (PedsQL™), and the Childhood Health Assessment Questionnaire (CHAQ), and for the latter. The most common physical measurement was Range of Motion (ROM). Also common were joint scores (the number of joints affected), 6 or 9 min run walk tests, tests of oxygen consumption— VO_2 max and VO_2 peak, and isometric strength tests. Notable study-specific measures included gait measurements, bone mineral content, vertical jump tests, and EMG data.

Table 1: Summary of exercise intervention studies for individuals with juvenile arthritis.

Year	Author	Number of Participants (Control/alternative treatment)	Age Gender	Subset	Design	Intervention	Duration	Assessment Measures	Outcomes
1991	Bacon et al. [13]	11	4-13 7F/4M	4 poly, 4 oligo, 3 sys	Pilot	Aquatic exercise 2/week 1.75 h stretching, ROM, strengthening exercises +0.25 h free play	6 weeks	Joint ROM, balance tests, timed tasks, gait analysis	Improved hip rotation
1992	Kircheimer et al. [11]	62	6-17 7F/3M	35 poly, 11 oligo, 7 sys	Prospective monitoring sports versus only PT	Sporting activity	8 years	ROM, mal-alignment, joint scores	Joint scores significantly correlated with stage of disease, not with participation in sports. Disease duration longer than 10 years associated with low participation in athletic activities.
1994	Oberg et al. [14]	20 10 control	7-15	3 poly, 5 oligo, 1 psori, 1 sys	RCT (healthy controls)	40 min/2 week strength and endurance training incorporating static & dynamic exercises	3 months	EMG	No significant difference in force or endurance b/w patients and controls, same strength trainability, normalization of fatigue after training
1999	Klepper [15]	25	8-17 23F/2M	25 poly	Pilot	3/week low-impact weight-bearing physical conditioning program 1/week 60 min video-guided home exercise	8 weeks	ASI (pain on motion, tenderness, swelling, limitation of motion), joint count, pain (VAS), 9 min run walk test	Significant improvement in ASI, run walk test, and joint count, Mixed VAS scores
2002	Takken et al. [6]	10	5-13 38F/16M	3 poly, 4 oligo, 3 sys	Pilot	1/week 1 h aerobic aquatic exercise	15 weeks	Joint status & mobility, CHAQ, 6 min walking test, JAQQ, pain (VAS)	Improved quality of life, no change in other measures
2003	Takken et al. [16]	54 27 control	5-13 38F/16M	29 poly, 23 oligo, 2 sys	RCT	20 1 h sessions of pool-based aerobic exercise	6 months	Functional ability (CHAQ, JAFAS), Juvenile arthritis quality of life (JAQQ), Joint status (tenderness, swelling, movement), physical fitness (maximal exercise test, 6 min walking test)	Small improvement in joint status, physical function, and quality of life
2005	Epps et al. [17]	78 39 land/39 combined	4-19 53F/35M,	33 poly, 22 oligo, 10 sys, 12 enthesitis, 1 psori	RCT	Land -16 h sessions of muscle strengthening & stretching exercises combined -8 land +8 hydrotherapy	2 weeks intensive 2 months outpatient	Disease status (CHAQ, ROM, active joints, parent/physician assessment), isometric muscle strength, physical fitness (cycle ergometer), pain (VAS)	Slight increase in CHQ and mean muscle strength in combined; aerobic fitness, and endurance improved in both

Year	Author	Number of Participants (Control/alternative treatment)	Age Gender	Subset	Design	Intervention	Duration	Assessment Measures	Outcomes
2005	Myer et al. [12]	1	10 F	Oligo/pauci	Case study	2/week neuromuscular training warmup, plyometric training, core strengthening	5 weeks	Pain (VAS), ROM, gait, landing technique, vertical jump test, knee ext/flex strength, postural stability	Increased hamstring strength and hams: quads, movement towards more normal gait, decrease in peak force on drop/jump, improved stability
2006	Singh-Grewal et al. [18]	9	8-11 5F/4M	3 poly, 4 oligo, 2 sys	Pilot	2/week sessions pool warmup, aerobic gym stations (fitball, cycle, treadmill, strength exercises)	12 weeks	CHAQ, JASI, energy cost of locomotion, vo2peak peak/mean muscle power	Slight decrease in energy cost of locomotion, increase in VO ₂ peak, anaerobic leg power
2007	Singh-Grewal et al. [19]	80 aerobic/39 Qi gong	8-16 64F/16M	34 poly, 18 oligo, 7 sys, 11 enth, 8 psori, 2 other	RCT	3/week 45 min high intensity aerobic program vs. qigong	12 weeks	Vo2submax/peak, peak power, physical function (C-HAQ), QOL, HRSQOL, VAS	improved physical function (C-HAQ) w/ no difference between groups, no significant change in any fitness parameters
2009	Fragala-Pinnkham et al. [20]	4 (1 with JIA)	2 y/o F	oligo	Case Series	1/week 60 min pool exercise 1/week 60 min PT at home	6 months	Occupational Performance measure (COPM), GMFIM-66, Pediatric Evaluation of Disability Inventory, mobility functional skills, and caregiver assistance, energy expenditure, observational gait scale, functional reach test, timed single leg stance, floor to stand, manual muscle testing, isometric muscle strength, passive ROM, numerical pain scale, JAQQ	Clinically significant improvement in QOL and ROM, extension, and weight bearing on involved limb
2010	Lelieveld et al. [21]	33 control	8-12 29F/4M	9 poly, 20 oligo, 4 sys	RCT	Internet-based program teaching about basics of JIA, PT and its benefits, goal setting, etc.	17 weeks	C-HAQ, physical activity (diary), aerobic exercise capacity (bruce treadmill test)	Increased physical activity, time spent on moderate to vigorous activity, and days one week+hours of moderate to vigorous PA, increased max endurance time on treadmill

Year	Author	Number of Participants (Control/alternative treatment)	Age Gender	Subset	Design	Intervention	Duration	Assessment Measures	Outcomes
2012	Tarakci et al. [22]	93 38 control	5-17 29F/4M	46 poly, 30 oligo, 3 sys, 1 psori	RCT	1/week 20-45 min individualized land-based home exercise (strengthening, postural exercises, functional activities) 4/week at hospital	3 months	6 min walk test, CHAQ, pain (VAS), PedsQL,	Increased physical function and quality of life in exercise group, decreased pain in control group
2012	Sandstedt et al.[9]	54 21 control	9-21 41F/13M	31 poly, 17 oligo, 6 enth/psori	RCT	3/week 20+min of strength exercise, including jump rope, core, arm, and shoulder strength exercises	12 weeks	Foot BMC and BMC Z score	Total body BMD increased in exercise group
2013	Mendonca et al. [23]	50 25 standard exercise/25 pilates	8-18 32F/18M	12 poly, 24 oligo, 14 sys	RCT	2/week 50 min	6 months	Quality of life (PedsQL), functional ability (CHAQ), pain (VAS), ROM	Improved pain and CHAQ scores, improved ROM/w greater improvements in pilates group
2013	Van Oort et al. [24]	7	10-17 4F/3M	3 poly, 3 oligo, 1 enth	Pilot	3/week 40 min home based resistance training (band and body weight)	6 weeks	Pain, inflammation, muscle thickness (vastus lateralis and biceps brachii), muscle strength, functional ability (CHAQ)	Increased vastus lateralis thickness
2013	Sandstedt et al.	54 21 control	9-21 41F/13M	29 poly, 15 oligo, 4 enth/psori	RCT	3/week exercise program including jump rope, muscle strength/core exercises & free weights	12 weeks	ROM, balance, muscle strength, grip strength, step test, quality of life (CHAQ, CHQ-C87)	Significant increase in hip and knee extensor strength, small increase in CHAQ
2014	Dogru Apti et al. [28]	50 20 healthy controls	8-16 29F/18M	24 polyarticular, 16 oligo, 2 sys, 1 enth, 4 psori	RCT (healthy controls)	4/week aerobic walking, daily ROM exercises 3/week 40 min home-based resistance training	8 weeks 6 weeks	VO ₂ peak, hrs, RER, O ₂ uptake, ROM	Significantly increased ROM in shoulder (abd/flex), wrist (flex/ext), elbow (flex), hip (flex), knee (flex/ext), and ankle (PF/DF). moderate increase in VO ₂ peak, RER, VO _{2at'} resting hrs, resting sys BP, VE _{peak'} Max h, exercise duration
2015	Baydogan et al. [26]	30 15 strength/15 balance	6-18 21F/9M	16 poly, 11 oligo, 3 psori	RCT	3/week 45 min resistant bike ergometer, stretching (both groups) group 1: knee strengthening/ROM exercises group 2: proprioceptive/balance exercises	12 weeks	pain, passive ROM, muscle strength, balance, functional ability, walking stair climbing	INTRAGROUP: Improved strength in all except hip/ankle, increased proprioception INTERGROUP: improvements in all aspects in group 2 except NRS, CHAQ, PROM, and hip ext/knee flex

Outcomes

All of studies included demonstrated positive outcomes on some level, though the degree of significance of these outcomes was quite varied. Many studies took care to note that overall pain did not increase, and no group measure regressed over the study period. However, there were isolated instances of increased pain or discomfort, associated with individuals identified as experiencing more severe cases. These cases were either noted in results reporting, or in reporting subject drop out or loss to follow up.

Physical function, quality of life, and range of motion of at least some joints improved in essentially all studies where they were measured, significantly in many of them. Groups that were engaged in land-based aerobic activities showed more improvement in the various aerobic outcome measures than those in hydro or movement centred therapies. Although strength measures tended to improve slightly in cases when measured, results tended more towards levels of significance when this was a focus of the program, and at the muscles that were the focus of the program. Gait measures trended towards normal levels, and in the study examining bone content, total body bone mineral density increased in the exercise group.

Case Studies

A prospective overview of arthritic youths participating in sports activity over an eight year period found pain to be more correlated with the disease stage than with their level of participation in sports¹¹. Additionally, increased severity and length of disease preceded a decline in activity, as opposed to a change in activity level resulting in a change in severity. A second case study involved a highly individualized training program for a ten-year old with a quiescent case of bilateral knee arthritis [12]. A training program was created to improve the subject's landing technique, core strength, and balance and coordination, in order to address biomechanical and neuromuscular imbalances and deficits that would prevent her from safely participating in a high-impact sport (basketball). Following training, the subject's heel strike and step width were brought to within normal limits, peak force and imbalance on a box jump landing decreased, and flexor/extensor strength ratios and balance measures converged to normal and equal levels.

Discussion

The overall quality of existing research investigating the efficacy of exercise programs in mediating the symptoms of children with juvenile arthritis is generally high. Most studies involved a large subject pool, included children of both sexes, and individuals with varied types of arthritis. Additionally, the studies were fairly consistent in choosing outcome measures (CHAQ, VAS, joint ROM), making comparison of results a more productive endeavour. This review shows that including an exercise program as a component of treatment for juvenile arthritis is a worthwhile endeavour. However, additional research would be beneficial in that it could provide more definitive results. In

addition, more information would allow for the development of patient-specific programs, pairing exercises and programs with particular types or severities of arthritis. For example, given the existing research, a water-based program would likely be more highly recommended for children with more severe and multi-joint lower body arthritis. Conversely, children with more mild arthritis, or fewer involved joints may be given more freedom in choosing a program. This might increase retention, as it has been shown that enjoyment and engagement with an exercise program increases participation rates.

Essentially all of the present studies examining the effects of exercise on children with idiopathic arthritis have varied populations that include individuals with polyarticular, oligo/pauciarticular, systemic, and enthesitis-related and psoriatic JIA in the same study. Aside from single incidence studies, there were no articles that applied an intervention to a specific subtype. Therefore, we don't know whether exercise efficacy varies based on arthritis classification, since it is difficult to determine in a significant manner when all of these patient populations are treated as one entity. There are multiple reasons why it would be beneficial to design a study to examine the effect of an intervention on a specific subtype. For example, oligoarticular arthritis tended to be somewhat diminished in its study representation compared to its prevalence. This is potentially due to its higher prevalence in age ranges not captured, as study age ranges typically started at at least six. Separating by subtype would also be particularly useful in the cases of systemic arthritis, where patients tend to be less tolerant of more strenuous and weight bearing exercise. In this case, research may choose to focus more on combinations of medication and activity, or on less impact-heavy systems like Pilates and yoga.

In addition, studies mostly focused on patients who had involved lower body joints. While this coincides with trends in the overall population, and affection of the lower body tends to have more impact on mobility and gait, there are still a significant number of individuals with upper body affected JIA, particularly in the hands. Potential future research could examine if the aerobic capacity of these individuals is different when compared to lower extremity affected individuals or healthy counterparts. Upper body individuals would also be more readily able to engage in weight bearing activity without pain, and may benefit from a more strength focused program.

Some of the interventions required only once or twice weekly sessions. While this is sufficient if the exercise were purely therapy, to see more significant physiological results, it is probable that the exercise would have to be undertaken more frequently. Given the depressed levels of physical activity in this population, it is unlikely the subjects were being adequately active outside of the sessions, and would therefore not likely meet recommended amounts of daily activity. This is of additional significance given that JIA is more prevalent in girls, who tend to experience declines in physical activity as they progress through puberty, even in healthy populations [14,15]. For future studies, intervention duration of at least three months would be optimal when possible.

The reviewed articles were well-advised in their selection and application of outcome measures. Most employed measures that are universally important to arthritis patients—pain, functional ability, and quality of life. From that set, researchers added measures that were specific to the outcomes deemed of importance, or the particular type of intervention. While the subjective measures are clearly useful, it is important to continue to stress the use of the objective measures. Both gait analysis and EMG measurement were two assessment methods that were rarely used, but can provide unique insights into the biomechanical and neuromuscular issues that occur with JIA [16-26]. For example, the Oberg article was able to determine that affected joint muscles were more fatigable than healthy controls, and that this gap was largely diminished upon training. This is certainly an area that has great potential in identifying neuromuscular deficits and quantifying progress due to therapies. Gait analysis is important, as it is a more quantitative measure of mobility. Additionally, for individuals who have less severe cases of arthritis, they may have goals of being able to participate in competitive sports with their peers, but they first have to develop similar gait and postural control before they can safely perform more complicated and energy intensive movements.

Conclusion

The optimal exercise therapy program will address the patient's posture and range of motion issues, in addition to their aerobic capacity and strength. The former are important for pain and functional ability, while the latter are important for weight management, functional ability, and bone health. It is also recommended to match the system of activity to the disease status and goals of the patient. For example in extremely severe cases the goal may simply be an increase in range of motion, or more normal gait, while a well-managed or quiescent case may aim to participate in competitive sports, with the Myer et al. [12] providing an incredibly informative blueprint on how this might be achieved.

Encouraging exercise is a good in and of itself, with clear and well known benefits in the general population, and youth in particular. In children with arthritis, it can improve quality of life and close the gap between them and their healthy peers when it comes to things like pain, aerobic capacity, bone health, strength, gait, and everyday functional capacity, providing them with the enthusiasm and confidence to become lifelong participants in sports and exercise. The fact that juvenile idiopathic arthritis persists into adulthood in up to half of patients, underscores the need to assist patients in finding ways to manage their arthritis and be active that are effective for them.

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