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Bartosz Radosław Wnuk[1], Wojciech Kroczek[1], Edyta Kroczek[1], Agnieszka Fajkierz[2], Andrzej Setlak[2], Klaudia Palka[3], Barbara Buc-Piorun[3]

[1]Department of Rehabilitation, Faculty of Health Sciences in Katowice, Medical University of Silesia, Katowice, Poland
[2]Medical Rehabilitation Department, Upper Silesian Medical Center, Katowice, Poland
[3]Department of Internal Diseases and Rheumatology, Upper Silesian Medical Center, Katowice, Poland

Everything you always wanted to know about systemic sclerosis but were afraid to ask: Part 7. Evaluation of the effectiveness of kinesitherapy conducted in a group of patients with systemic sclerosis based on a literature review

ABSTRACT

Background: Systemic sclerosis (SSc) is a chronic autoimmune disease of the connective tissue. Physical training as a planned activity can delay the process of disease changes. The aim of the study is to assess the effectiveness of kinesitherapy conducted in a group of patients with SSc based on a literature review.
Material and methods: The PubMed and Pedro databases were analyzed using the following keywords: "systemic sclerosis and physiotherapy or physical training". The university multi-search engine was also used, including the database of periodicals of the Medical University of Silesia in Katowice.
Results: Nine works out of 23 articles presenting the impact of various forms of training and physiotherapy methods in the treatment of people with SSc were qualified for the analysis.
Conclusions: The conducted analysis of the literature on physical training in people with SSc confirms the advisability of its use with the use of interval training and resistance training. These forms of training have a positive effect on the functions of the respiratory, circulatory and muscular systems. Physical training can be a valuable supplement to pharmacotherapy to support the treatment of this group of patients.

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KEY WORDS: systemic sclerosis; high-intensity interval training; resistance training; physical training; physiotherapy

INTRODUCTION

In systemic sclerosis (SSc; scleroderma), one of the early clinical signs, although not prominent, is Raynaud's phenomenon presenting as a vasomotor disorder. It causes blanching and, at a later stage, lividity and reddening of the fingers and toes. The most prominent change is the hardening of the skin resulting from collagen accumulation [1]. Microvascular dysfunction is a key element in the pathogenesis of SSc, while the contribution of large and medium vessel abnormalities has not yet been established. Parameters of macroangiopathy are strongly correlated with microvascular pathology in patients with SSc [2]. In this condition, the lesions with the worst prognostic of survival are pulmonary (16–22% of

Address for correspondence:
Bartosz Radosław Wnuk, MD, PhD, Department of Rehabilitation, Faculty of Health Sciences in Katowice, Medical University of Silesia, Ziołowa 45/47, 40–635 Katowice, Poland;
e-mail: bwnuk@sum.edu.pl

death rate) and renal (i.e., scleroderma renal crisis, 10% of death rate). Many patients develop muscle atrophy, mainly skeletal, accompanied by weakness of muscle strength. There are two forms of SSc. Limited SSc affects the skin of the hands, feet, forearms, shins and face. This type has a prolonged, chronic, progressive course, and up to several years pass from the onset of the disease expressed by Raynaud's phenomenon. Diffuse SSc affects the skin of the face, trunk and limbs proximal to the elbows and knees. This form has a rapid course, and it does not even take a year from the first episode of Raynaud's phenomenon to noticeable hardening of the skin. The treatment of patients with SSc requires the involvement of multiple specialists and should be comprehensive and include education, pharmacotherapy, physiotherapy and sometimes surgical treatment [3–5].

Due to organ complications, musculoskeletal and skin involvement, SSc in patients in the advanced stage can result in severe disability. Comprehensive and individually tailored therapy is essential. The overriding goal of treatment is to slow down the progression of organ lesions. Despite the many medications available, there is still a problem of progressive impairment in patients. Increasingly, attention is being paid to the need for rehabilitation treatment through programmes tailored to disease severity. Appropriate, individually tailored therapy helps counteract and reduce dysfunctions in the joints, soft tissues and skin. It also has a beneficial effect on improving the efficiency of internal organs. Despite the many drugs available, there is still the problem of progressive impairment in patients with SSc, which cannot be solved by drug therapy alone [6].

HIGH-INTENSITY INTERVAL TRAINING

This type of training is a unique form of exercise that improves overall health. It consists of repeated, short periods of high-intensity training (i.e., ≥ 85% of maximum heart rate) interspersed with periods of low-intensity training or rest [7]. Compared to typical training (e.g., aerobics, resistance training), high-intensity interval training (HIIT) appears to be a more effective strategy for obtaining a range of physiological health benefits. These include improved cardiorespiratory fitness [8, 9], improved insulin sensitivity [10] and reduced body fat and blood pressure [11]. In addition to these physiological changes, an effect on increasing the cardiac ejection fraction can be observed [12]. Exercise time should be no longer than 20–30 minutes for this type of training. This is important because of the strain on the nervous and cardiovascular systems.

RESISTANCE TRAINING

Impaired skeletal muscle function is one of the causes of poor exercise tolerance. Resistance training is recommended for prevention and cardiovascular disease. The physiological benefits of this training include an increase in muscle strength, a reduction in blood pressure and, as in HIIT, an improvement in insulin sensitivity [13, 14]. Flow-mediated dilation has been recognised as a major marker of healthy vessels. Flow-mediated dilation during muscle work is related to the release of nitric oxide by endothelial cells. Resistance training in people with cardiovascular and metabolic diseases impacts the increase of this marker [15].

Incorporating resistance training into physiotherapy is expected to prevent atrophy and weakening of skeletal muscles. Well-chosen resistance training can slow down the cachexia associated with the progression of various diseases. It complements endurance training. As a rule of thumb in resistance training, the training cycle starts with shorter sessions, during which you aim to reach at least the lower limit of your training load. Later, you gradually increase the training session duration by increasing the load. Patients should not exceed the appropriate exercise intensity determined by the Borg RPE scale. Traditional resistance training uses a specific external weight. In this case, the load is constant throughout the range of movement. In resistance training with elastic bands, there is a variable resistance that peaks at the extremes of movement. A systematic review of studies comparing the two types of training in healthy individuals does not differentiate between the final exercise effects achieved [16, 17].

OBJECTIVE

Physical training as a planned activity can delay disease progression. This study aims to evaluate the effectiveness of kinesitherapy in a group of patients with SSc. This paper presents a review of the literature on the use of different forms of physical training in the physiotherapy of individuals with SSc. It considers

the results of studies published over the last 14 years. The articles were reviewed using data from the PubMed database and a publication multi-search engine covering the periodicals database of the Medical University of Silesia in Katowice. The Physiotherapy Evidence Database (PEDro) was also analysed separately. Systemic sclerosis and physiotherapy or physical training were used as keywords. The effectiveness assessment included articles on adults. It was planned to exclude publications using manual therapy, massage and special techniques. The use of combined techniques (paraffin baths with exercise, i.e. combining elements of kinesitherapy with physical therapy) was also excluded. The terms physical training and physiotherapy were deliberately chosen. Endurance training, resistance training or any other type of active or passive exercise or special methods are often described in very general terms by various medical professionals without being precisely defined. These are most commonly referred to as physical training or physical activity (PA). For this reason, such keywords were used in the search. It was only during the literature review and selection that a revision was applied, which considered only stand-alone endurance and resistance training, rejecting all other techniques or combined techniques regarding the use of kinesitherapy with physical therapy.

RESULTS

The search yielded 86 articles in total. After reviewing the paper titles, abstracts and full texts, 12 articles met the first selection criterion. After a review of these articles, nine studies qualified for final analysis. Three studies out of 12 were rejected due to combining physical training methods with active and passive manual therapy techniques and occupational therapy [18–20] (Fig. 1).

The authors of the first article evaluated an 8-week programme of moderate-intensity aerobic exercise for 30 minutes on a treadmill. Oliveira's team of researchers reported a significant improvement in peak oxygen consumption ($p = 0.006$) and a reduction in peak blood lactate levels ($p = 0.01$). Peak oxygen saturation during exercise improved significantly compared with baseline values ($p = 0.048$), while oxygen saturation at rest was the same after the workout [21]. In

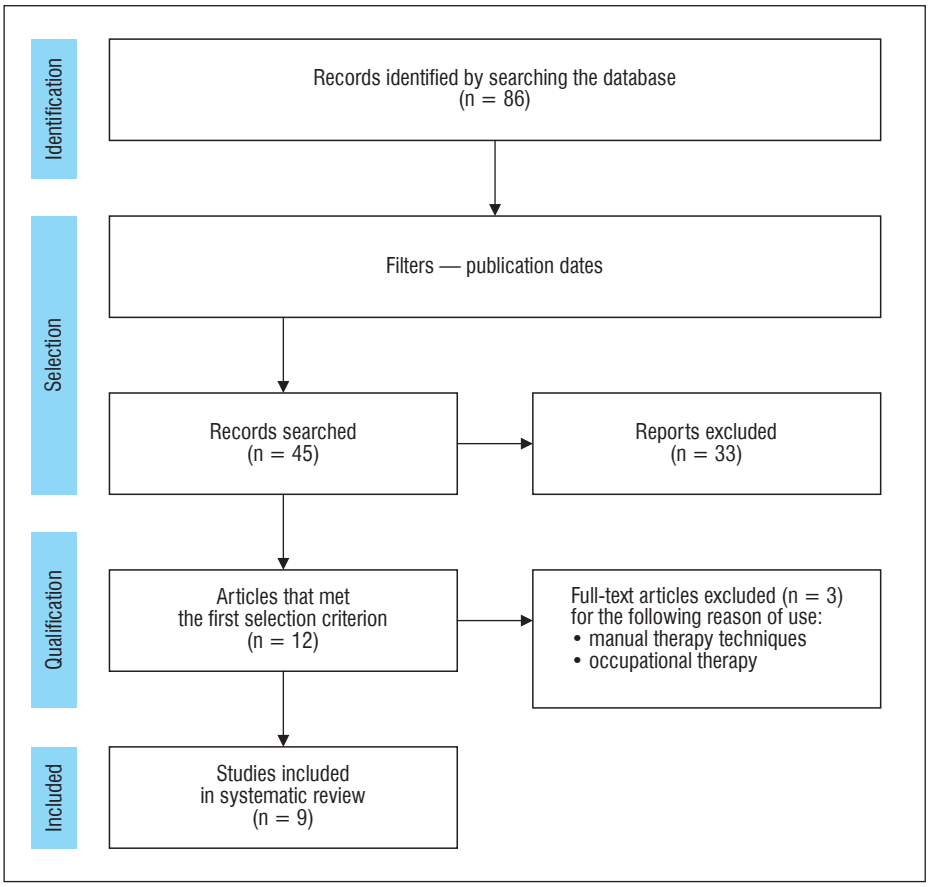

Figure 1. Scheme of literature search on physical training in a group of people with systemic sclerosis

the Pinto study, eleven patients with SSc participated in 12 weeks of combined resistance and aerobic training. The training routine was carried out twice a week. Significant improvements in muscle function and performance were recorded. A reduction in resting heart rate was observed, and better exercise tolerance was obtained ($p > 0.05$) [22]. Another study was conducted in a group of four patients with pulmonary dysfunction due to pulmonary fibrosis. In Alexanderson's 8-week research project, a significant improvement in muscular endurance was registered in 3 participants, and two participants had higher peak oxygen consumption ($p > 0.05$). The exercise tolerance of those exercising on the Borg RPE scale increased from 13 to 15. Other variables related to activity limitation — the Health Assessment Questionnaire (HAQ) and the Short Form (36) Health Survey (SF–36) did not change [23]. Another study conducted by Mitropoulos in 2018 looked at the effects of HIIT on the respiratory and vascular systems. Peak oxygen uptake increased in the group trained on the cycle ergometer and the group trained on the arm ergometer. Transcutaneous partial oxygen pressure was higher but not significantly in the arm ergometer group. The endothelium-dependent improvement in vasodilation was also significantly greater in the arm ergometer group ($p < 0.05$). An equally significant increase in life satisfaction and reduction in Raynaud's phenomenon discomfort was observed in both groups ($p < 0.05$) [24]. A second randomised study by the same author assessed the effect of combining HIIT with resistance training on microvascular function in a group of patients with SSc. The combination of these two forms of physical training resulted in a significant improvement in microcirculation as assessed by measuring endothelium-dependent reactivity and transcutaneous LDF of partial oxygen pressure $\Delta TcpO_2$ ($p < 0.05$) [25]. Another paper by this author evaluated the feasibility of performing this type of training consisting of HIIT with resistance training in a group of patients with SSc. The feasibility of this type of training consisted of measuring the quality of life and the level of perceived enjoyment for this type of physical training and examining the difficulty of performing individual exercises and the number of complications. The results confirm the feasibility and safety of this training protocol [26]. In his study, Defi used the same HIIT protocol as Mitropoulos to assess the effects of this activity in the measurement of inspiratory muscle strength (sniff nasal inspiratory pressure), walking speed, handgrip strength and quality of life according to the SHAQ-DI. After 12 weeks of training, beneficial changes were observed in inspiratory muscle strength, handgrip strength, walking speed, MET and the SSc-Health Assessment Questionnaire ($p < 0.05$). This confirms the effectiveness of this type of training. Training twice a week as part of a 12-week HIIT programme is considered safe for this group of patients [27]. A study by the next author, Yakut, compared the effects of supervised exercise and a home exercise programme in patients with SSc. Breathing, aerobic and resistance exercises were used in these two programmes over a 12-week period. Home physiotherapy has only provided instruction for these exercises. After a 12-week exercise period, a significant improvement in physical performance measured by the 6-minute test was observed in the supervised and home exercise groups ($p < 0.05$). An increase in quality of life and a decrease in fatigue levels were observed in the supervised exercise group. This demonstrates the greater benefits of supervised exercises [28]. A recent study is a case report of a 40-year-old SSc patient following interstitial pneumonia. In that study, the author of the paper — Alhowikan — used supervised strengthening and resistance training of the upper limbs, lower limbs and thoracic muscles. Spirometry test parameters, 6-minute test parameters, handgrip strength parameters and lower limb strength parameters were assessed. The 6-minute test measured distance, blood pressure, saturation, breathlessness level according to the Borg RPE scale. Eight weeks of supervised exercise improved spirometric parameters from 48.7% to 54%, 6-minute test distance by 15.4%. An increase in right handgrip strength of 79.4%, left handgrip strength of 25% and lower limb strength of 30.9% was recorded. The exercise programme proves its effectiveness under the supervision of trained personnel [29]. The individual physical training protocols of the eligible publications for review are shown in Tables 1 and 2.

DISCUSSION

Regular PA is increasingly promoted among people with rheumatic and musculoskeletal diseases, as well as in the general population. A systematic review of the research carried out on physiotherapy for people with

Table 1. Analysis of applied physical training in 2009–2018

First author, year, country, references	Participants in the study Type of study	Physical training programme	Training control Training period	Methods of performance evaluation
Oliveira, 2009, Brazil [21]	7 SSc patients without pulmonary dysfunction, 7 controls Observational study	Aerobic exercise (30 minutes walking on a treadmill at moderate intensity)	Supervised training 8 weeks (2 days a week)	VO_2max, saturation
Pinto, 2011, Brazil [22]	11 SSc patients without pulmonary dysfunction Observational study	Aerobic training (20 minutes of treadmill exercise to about 70% of peak VO_2), resistance training (30 minutes, four sets of 8–12 maximum repetitions for major muscle groups)	Supervised training 12 weeks (2 days a week)	VO_2max
Alexanderson, 2014, Sweden [23]	Four SSc patients with pulmonary fibrosis Experimental project	Aerobic exercise (cycling on a cycle ergometer for a maximum of 30 minutes. intensity was measured with the Borg RPE scale, muscular endurance training of the shoulder and hip joints, 30–50 minutes	Supervised training 8 weeks (3 days a week)	VO_2max 6-minute test Quality of life questionnaire HAQ, SF-36
Mitropoulos, 2018, England [24]	Randomisation of a group of 34 people with SSc into a training group on a cycle ergometer, a training group on a hand ergometer, a control group Randomised trial	30-minute interval training — 2 groups, control group without exercise	Supervised training 12 weeks (2 days a week)	VO_2max Transcutaneous partial oxygen pressure ΔTcpO2 EQ-5D-5-L questionnaire

SSc — systemic sclerosis; VO_2max — maximal oxygen uptake;TcPO2 — transcutaneous oxygen pressure; HAQ — Health Assessment Questionaire; SF-36 — Short Form Health Survey; EQ-5D-5-L — EuroQoL 5-Dimension 5-Level

SSc shows the health benefits and safety of the exercises, treatments carried out using this method. Physical activity through moderate-intensity and medium-load resistance exercises is recommended. Healthcare professionals should inform SSc patients about the importance of PA and avoiding a sedentary lifestyle [30, 31]. According to the recommendations of the American College of Sports Medicine, all healthy adults aged 18 to 65 years should participate in moderate-intensity aerobic PA exercise for at least 30 minutes 5 days a week. High-intensity aerobic exercise should be performed for 20 minutes 3 days a week [32]. In the studies analysed in the 6 completed research projects, the period of physical training was 12 weeks with a frequency of 2 exercise sessions per week. In the other completed research projects, the training period was smaller at 8 weeks with an exercise session frequency of 2 to 3. Exercise time oscillated between 20 and 30 minutes and was less than American College of Sports Medicine recommendations.

In the papers reviewed, physical training has a beneficial effect in stabilising SSc symptoms associated with digit ulceration and vasomotor disorders, including Raynaud's phenomenon. This may be related to improved aerobic capacity and the training implemented by Oliveira itself confirms its feasibility [21]. The optimal training model for SSc patients is unknown. Pinto introduced a combined programme of resistance and aerobic training as parallel training over a 12-week period, which significantly improved strength, function and muscle performance in SSc patients. After a period of 12 weeks, the Rodnan skin score remained unchanged, and muscle enzymes were at normal levels. Also, in that study, there were no changes in digit ulceration and Raynaud's phenomenon, indicating the safety of the training [22]. Parallel training consisting of intensive aerobic exercise and endurance training can also be used in a group of patients with reduced spirometry test values associated with pulmonary fibrosis. Improved exercise tolerance

Table 2. Analysis of applied physical training in 2019–2022

First author, year, country, references	Participants in the study Type of study	Physical training programme	Training control Training period	Methods of performance evaluation
Mitropoulos, 2019, England [25]	Randomisation of a group of 32 people with SSc into an experimental group and a control group Randomised trials	High-intensity interval — 30 minutes (30 sec effort, 30 sec break) resistance training (5 upper body exercises in a circuit system for three circles at 2–3 min intervals) Control group: no exercise	Supervised training 12 weeks (2 days a week)	VO_2max Microcirculation activity Transcutaneous oxygen pressure ΔTcpO2
Mitropoulos, 2020, England [26]	Randomisation of a group of 32 people with SSc into an experimental group and a control group Randomised trials	High-intensity interval — 30 minutes (30 sec effort, 30 sec break) Upper limb resistance training, intensity measured with the Borg RPE scale Control group: no exercise	Supervised training 12 weeks (2 days a week)	EQ-5D-5-L questionnaire Body composition analysis
Defi, 2021, Indonesia [27]	11 people with SSc were eligible for the study Observational study	High-intensity interval — 30 minutes (30 sec effort, 30 sec break)	Supervised training 12 weeks (2 days a week)	Sniff nasal inspiratory pressure, inspiratory muscle assessment Walking speed — 6-minute test Handgrip strength SHAQ-DI questionnaire
Yakut, 2021, Turkey [28]	37 SSc patients Supervised exercise group, home exercise group Randomised trial	Breathing exercises, aerobic Resistance exercises	Supervised training 12 weeks (2 days a week)	6-minute test HRQoL questionnaire
Alhowikan, 2022, Saudi Arabia [29]	A 40-year-old female patient with SSc after interstitial pneumonia — a case report	Strengthening and resistance training of upper limbs, lower limbs and chest	Supervised training 8 weeks (2 days a week)	Spirometry test 6-minute test assessment of muscle strength

SSc — systemic sclerosis; VO_2max — maximal oxygen uptake;TcPO2 — transcutaneous oxygen pressure; RPE — Rating of Perceived Exertion; EQ-5D-5-L — EuroQoL 5-Dimension 5-Level; SHAQ-DI — Scleroderma Health Assessment Questionnare — Disability Index; HRQoL — Health-Related Quality of Life

is associated with a tendency for reduced fatigue symptoms [23]. In a series of studies, Mitropoulos initially uses high-frequency interval training (HIT). This form of training improves vascular function, particularly in clinical conditions that have a strong macroangiopathic component. This form of training protocol performed on a hand ergometer has the potential to improve microvascular endothelial function in SSc patients. It is also worth noting that the recommended training dose (e.g., a 12-week HIIT programme twice a week) is sufficient and well tolerated for this patient population. In another study by this author, the implementation of HIT with resistance training supports vascular and microvascular function. This protocol was effective in improving endothelium-dependent reactivity in people with SSc. Engaging multiple large muscle parts with different types of muscle contraction in training can effectively support standard pharmacotherapy. A recent publication by this author emphasises the feasibility and safety without contingencies for this type of training [24–26].

The usefulness of HIT in this group of patients is supported by the work of Defi. In that clinical observation, there were improvements in the sniff nasal inspiratory pressure indices of inspiratory muscle function, gait speed and handgrip strength, which can also be considered as important biomarkers of health status [27]. The positive effects of training are influenced not only by well-targeted training, but also by proper supervision by qualified medical staff. Periodic measurement of quality of life is

also important to determine the patient's level of satisfaction with the forms of physical training presented [23, 24, 26–29].

Home physiotherapy is a promising alternative to outpatient care programmes or hospital-based physiotherapy because of the supportive family atmosphere. There is a lack of literature describing the impact of home rehabilitation programmes. Yakut's work compared a supervised exercise programme with an exercise programme completed at home. The supervised exercise programme proved superior to home exercise in observing breathlessness severity levels. However, both programmes had a positive effect on reducing fatigue after specific exercises [28, 33].

The review and analysis of available articles meeting the criterion as physical training have their limitations. These include a limited number of publications that do not exceed nine studies. Some of these are observational studies or case reports. In the papers presented, there is a small number of eligible people with SSc who have limitations in exercise tolerance associated with respiratory and vascular conditions. Some explanation is provided by the inclusion and exclusion criteria of the studies, which also aim to ensure the safety of the training. What is missing from the papers analysed is an assessment of the level of medications taken during a given training cycle. This information may be useful in assessing the effectiveness of physical training programmes in this group of patients.

CONCLUSIONS

An analysis of the literature on physical training in people with SSc confirms the advisability of its use with interval training and resistance training. These forms of training have a beneficial effect on respiratory, cardiovascular and muscular functions. These forms are safe and feasible for the SSc patient, provided that they are controlled by qualified medical personnel. Physical training can be a valuable adjunct to ongoing pharmacotherapy to support the treatment of this patient group.

CONFLICT OF INTEREST

None declared.

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None declared.

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