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BRIEF REPORT

Effect of an 8-Week Tailored Physiotherapy Program on Sexual Health in Women with Scleroderma and Myositis: A Controlled Pilot Study

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ABSTRACT

Introduction: Systemic sclerosis (SSc) and idiopathic inflammatory myopathies (IIM) are very rare rheumatic diseases burdened by a high prevalence of sexual dysfunctions. However, no specific treatment has been proposed to date. To our knowledge, this is the first (pilot) study aiming to investigate the effect of an 8-week tailored physiotherapy program on the sexual health of women with SSc and IIM.

Methods: In total, 12 women with SSc and 4 women with IIM were enrolled in the study. Based on the patients' capability to participate in the program, they were divided into an intervention group (IG) (mean ± SD age 46.8 ± 8.6 years) and a control group (CG) (mean ± SD age 46.3 ± 8.5 years). IG underwent the 8-week program (1 h of supervised physiotherapy twice weekly), whereas CG received no physiotherapy. At weeks 0 and 8, all patients filled in questionnaires assessing sexual function (Female Sexual Function Index [FSFI], Brief Index of Sexual Functioning for Women [BISF-W]), sexual quality of life (Sexual Quality of Life-Female [SQoL-F]), functional ability (Health Assessment Questionnaire [HAQ]), quality of life (Medical Outcomes Short Form-36 [SF-36]), and depression (Beck's Depression Inventory-II [BDI-II]). The changes were analyzed with two-way ANOVA and Friedmann's test.

Results: Compared to the statistically significant deterioration in CG over weeks 0–8, we found statistically significant improvements in the total scores of FSFI and BISF-W, and some of their domains, functional status, and the physical component of quality of life.

Conclusion: Our 8-week physiotherapy program not only prevented the natural course of progressive deterioration of functional ability but also led to a significant improvement in sexual function and quality of life in women with SSc and IIM. However, due to the lack of randomization and a relatively small sample size resulting from the strict inclusion criteria, further validation of our results is needed.**Trial**

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s40744-023-00559-9.

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Keywords: Female sexual dysfunction; Idiopathic inflammatory myopathies; Sexual dysfunction intervention; Pelvic floor exercise; Physiotherapy; Systemic sclerosis

Key Summary Points

To date, no study devoted to the therapy of sexual dysfunctions in rare rheumatic diseases such as SSc and IIM has been conducted, even though the prevalence of female sexual dysfunctions has been determined to be high in both diseases

This pilot project aimed to investigate the effect of an 8-week tailored physiotherapy program on sexual function in women with SSc and IIM

In this pilot study, a significant improvement was observed in the total scores of both questionnaires assessing sexual function (Female Sexual Function Index, Brief Index of Sexual Functioning for Women) and in the domains of arousal, frequency of sexual activity, initiation/receptivity of/to sexual activity, and a statistical trend of improvement in the domains of sexual pain and lubrication

Tailored, supervised 8-week physiotherapy may not only improve functional ability and the physical component of overall quality of life but also enhance the sexual function of women with SSc and IIM

Further validation of our pilot results could introduce a potential therapeutic modality for managing these conditions and provide patients the opportunity to actively participate in their treatment

INTRODUCTION

Systemic sclerosis (SSc) and idiopathic inflammatory myopathies (IIM) are rare rheumatic autoimmune diseases characterized by multiorgan involvement including the musculoskeletal system, which are chronic and progressive in nature [1, 2]. Severe clinical manifestations of SSc and IIM often lead to significant disability and reduced quality of life [3, 4], including sexual life [5, 6]. In our recent studies [5, 6], we demonstrated significantly lower sexual function in women with SSc and IIM compared to the healthy population as well as an association with greater functional impairment, poorer physical condition, and worse psychoemotional status. In addition, our studies provided new information regarding possible pelvic floor dysfunction, which was significantly worse in patients compared to healthy controls. Although it appears that sexual dysfunction may be highly prevalent in women with SSc [6] and IIM [5], no specific therapy has been proposed to date.

It is generally known that physical activity provides significant health benefits. According to the World Health Organization, regular physical activity can improve muscular and cardiorespiratory fitness, bone and functional health, and cognitive and mental health including alleviation of anxiety and depression symptoms [7]. Due to these multiple health benefits, physical exercise is also commonly used in the prevention and treatment of various chronic conditions [8]. The effect and safety of exercise therapy were also investigated in patients with SSc who reported improvements in hand and mouth function, aerobic capacity, and muscle strength [9, 10], as well as in myositis patients who demonstrated protection against progressive deterioration and improvement in muscle strength, endurance, stability, and disability [11, 12].

Several studies have also identified the positive effect of exercise on women's sexual health in the general population. The study of Lorenz et al. demonstrated that regular exercise improved sexual desire and global sexual function in women with antidepressant-induced

sexual dysfunction [13] or another study showed that 12-week Pilates interventions have a significant positive effect on sexual function in healthy women [14]. It is also well known that pelvic floor muscle (PFM) training can improve female sexual function [15]. For instance, the study by Nazarpour et al. showed that PFM exercises could improve the sexual function of postmenopausal women, especially in the domains of sexual arousal, orgasm, and satisfaction [16]. These findings led us to hypothesize that targeted physiotherapy including PFM exercises could lead to improvements in sexual function also in patients with SSc and IIM. Although physiotherapy could be suggested as a simple, non-invasive, and inexpensive way to treat sexual dysfunctions also in women with SSc and IIM, no relevant evidence has been published to date. Therefore, to our knowledge, this is the first pilot study aiming to investigate the effect of targeted physiotherapy on sexual function in women with SSc and IIM.

METHODS

Study Design

The present study is a pilot single-center, non-randomized controlled trial approved by the Ethics Committee of the Institute of Rheumatology in Prague (no. 12898/2019) and registered under the ISRCTN registry (no. ISRCTN91200867) in May 2021. Patients were non-selectively recruited from June 2021, and based on their capability and willingness to participate in the physiotherapy program, they were allocated to an intervention group (IG) and a control group (CG). The IG underwent the 8-week tailored physiotherapy program, including pelvic floor exercises and physiotherapy that target musculoskeletal problems subjectively limiting the patients' sexual function (twice weekly for 1 h per session), whereas the CG received no specialized therapy to treat sexual dysfunctions for the duration of this study. To ensure that the patients in the CG received no such therapy, at week 0, they were specifically instructed not to undergo any guided therapy (physiotherapy or other therapy targeted to improve sexual function). Furthermore, at week 8, in the CG we checked and confirmed that no prescriptions for physiotherapy at the Institute of Rheumatology were made by the attending rheumatologists, and no physiotherapy sessions were listed in our medical records. Patients in both groups were allowed to continue using stable doses of standard-of-care pharmacological therapy and to continue with any usual physical (leisure) activities that they routinely do.

Patients and Controls

The population of the study consists of female patients with SSc or IIM who participated in our previous projects evaluating the prevalence and severity of sexual and pelvic floor dysfunctions [5, 6]. For those with detected sexual dysfunction, we offered the intervention program. The inclusion criteria were as follows:

- The subject meets the 2013 EULAR (European League Against Rheumatism)/ACR (American College of Rheumatology) classification criteria for the diagnosis of SSc [17] or the 2017 EULAR/ACR classification criteria for dermatomyositis or polymyositis [18].
- The subject is regularly followed by the attending rheumatologist at the Institute of Rheumatology in Prague, treated with standard-of-care pharmacotherapy.
- The subject participated in the first part of our research evaluating the prevalence and severity of sexual and pelvic floor dysfunctions and filled out all the attached questionnaires and required medical data.
- The subject meets the criterion of sexual dysfunction in at least two of the three questionnaires assessing sexual function: Female Sexual Function Index (FSFI), Brief Index of Sexual Functioning for Women (BISF-W), and Sexual Function Questionnaire (SFQ-28). Sexual dysfunction was determined based on the diagnostic cut-off score for FSFI and SFQ-28 [19–21], and for BISF-W the threshold was set to the 15th percentile of the healthy population.

- The subject is currently sexually active (at least one sexual activity per week), and sexual activity includes sex with or without penetration or masturbation.
- The subject is a female over the age of 18 years.
- The subject has signed the informed consent prior to participating in the study approved by the independent ethics committee of the Institute of Rheumatology in Prague.

Exclusion criteria included:

- The subject suffers from any other severe chronic comorbid diseases or other systemic rheumatic disease as specified elsewhere [5].

Assessment Methods and Outcome Measures

At weeks 0 and 8, all patients filled in six well-established and validated questionnaires. Primary outcome measures included the Female Sexual Function Index (FSFI) [22] and Brief Index of Sexual Functioning for Women (BISF-W) assessing female sexual function [23] and the Sexual Quality of Life Questionnaire-Female (SQoL-F) [24] evaluating the impact of female sexual dysfunction on the quality of a patient's life. Secondary outcome measures were comprised of Health Assessment Questionnaire (HAQ) [25] assessing functional ability, Medical Outcomes Short Form-36 (SF-36) [26] evaluating the overall quality of life, and Beck's Depression Inventory-II (BDI-II) [27], which assesses depression. Further details, including appropriate references regarding the above-mentioned patient-reported outcomes and their Czech versions, have been provided elsewhere [5].

Prior to the intervention program, both groups underwent a clinical and laboratory examination performed by a board-certified rheumatologist blinded to the group allocation. A detailed medical, gynecological, and sexual history was taken from all patients, and the following data were collected:

1. Demographic characteristics: age at recruitment, education level (primary, secondary, tertiary), place of living, and current partnership status (single, in a relationship, married, divorced, widow).
2. Clinical characteristics: disease duration, disease activity determined by the European Scleroderma Study Group (ESSG) SSc activity score [28] or Myositis Intention to Treat Activity Index (MITAX) and Myositis Disease Activity Assessment Visual Analog Scale (MYOACT) [29], involvement of the skin evaluated by the modified Rodnan skin score (mRSS) [30], and current medical therapy. All disease-related features were also recorded. The functional limitations caused by SSc symptoms were assessed by Scleroderma Health Assessment Questionnaire (SHAQ) [31].
3. Laboratory data: serum concentrations of C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR) were evaluated [5].
4. Gynaecologic and sexual history: menstrual status, previous pelvic surgery, contraception use, hormone replacement therapy, number of childbirths, sexual activity, reasons for lack of sexual activity, and disease-related symptoms affecting sexual life. Patients were also asked about their subjective evaluation of sexual life importance, which was measured by a visual analog scale (VAS) graded from 0 (not important at all) to 10 (extremely important).

Only patients in the IG were examined at the baseline by a physiotherapist who performed a comprehensive kinesiological assessment including the pelvic floor function examination, which was carried out transvaginally using digital palpation following the standardized PERFECT scheme. The PERFECT scheme provides a simple standardized method to evaluate the main components of PFM contractility. It is an acronym for "Power, Endurance, Repetitions, Fast contractions, Elevation, Co-contraction and Timing" [32]. These qualities are assessed in three positions: supination, sitting, and standing position. In addition, the assessment of Manual Muscle Testing of eight muscle groups (MMT-8) [33] and Functional Index-2 (FI-2) [33] was evaluated in patients with IIM.

Intervention Program

Patients in IG underwent an 8-week intensive (1 h twice weekly) physiotherapy program consisting of individually focused therapy on musculoskeletal problems limiting the patient's sex life and, where needed, also of PFM exercise. They were initially asked to identify at least five main disease-associated symptoms that limited them the most in their sexual activities. The physiotherapy was subsequently targeted at the listed difficulties for each patient according to the scheme shown in Table 1. For each of the symptoms, the targeted physiotherapy, autotherapy, and regimen (if applicable) were proposed and individually performed depending on the capability and needs of each patient. In the case of objectively assessed findings on PFM, therapy was also devoted to the treatment of PFM according to the clinical findings. Patients with PFM weakness received a PFM-strengthening device (Educator®, Performance Health International Ltd, Huthwaite, UK) and instruction for home-based PFM exercise. The exact exercise unit was derived from the results of the PERFECT-scheme assessment; for example, a measurement of 4/6/5//9 means good contraction, held for 6 s, repeated five times, followed by nine fast contractions, and the home exercise would be based precisely on these values, as they determine the exact number of contractions required to stress the muscles for the exercise to be effective. This particular exercise is performed in all three positions (supination, sitting, and standing position), and it has to be checked to ensure no compensatory involvement of other muscle groups such as the gluteal, abdominal, or hip adductor muscles is present. In addition, PFM contraction during stress maneuvers (laughing, sneezing, coughing) is also practiced in all three positions. Visual biofeedback on the correctness of PFM contraction during exercise is provided by the Educator® device. To control whether the patients in the IG followed the protocol of the home exercise, patients in the IG kept a diary of the autotherapy they performed (how often they were able to perform the exercises, for how long, and how successful they felt), and at each visit, the ability to perform home exercises and the progress were checked.

Statistical Analysis

Basic descriptive statistics (mean, median, standard deviation [SD], inter-quartile range [IQR]) were computed for all variables, and the normality of the data distribution was analyzed using Shapiro-Wilk and Kolmogorov-Smirnov normality tests. To determine the baseline differences between IG and CG in selected parameters, the Mann-Whitney *U* test (for continuous variables) and chi-square test (for categorical variables) were used. Parameter changes over time between the IG and CG (inter-group analysis) were analyzed using two-way repeated measures ANOVA (interaction: group × time), and the intra-group changes between weeks 0–8 (in IG and CG separately) were determined by one-way repeated measures ANOVA (Friedmann's test). Inspired by the IMACS preliminary definitions of improvement using the core set measures [34], an absolute percentage change over weeks 0–8 was also calculated, according to which the patients were stratified into five categories: (1) improved by $> 20\%$, which is considered clinically significant; (2) improved by $\leq 20\%$ and $> 0\%$; (3) unchanged; (4) deteriorated by $\leq 20\%$ and $> 0\%$; (5) deteriorated by $> 20\%$. The differences in the distribution of IG and CG in these five categories were tested using the chi-square test. *P*-values < 0.05 were considered statistically significant. Data are presented as median (IQR) unless stated otherwise. All analyses and graphs were conducted using GraphPad Prism (version 6; GraphPad Software, La Jolla, CA, USA).

RESULTS

Of 36 women with SSc and 12 women with IIM who met the inclusion criteria and none of the exclusion criteria, 14 women with SSc and 4 women with IIM were willing to participate in the study. These patients were allocated to the IG (mostly patients living in Prague and its vicinity, $n = 7$ SSc/2 IIM) and the CG (mostly patients living further away, $n = 7$ SSc/2 IIM)

Table 1 Scheme of the therapeutic plan based on subjective symptoms and pelvic floor examination

Joint pain namely:	PT	Joint centering, traction/approximation, stabilization, optimization of muscle tension around the joint and associated functional changes, optimization of movement stereotypes
	AT	Home exercises following the instructions of the physiotherapist
	R	Planning sexual activity with minimal joint pain (if any), choosing appropriate sexual positions/activities
Muscle pain namely:	PT	Optimization of muscle tension of the functional unit, stabilization, and centering of functionally related segments, optimization of movement stereotypes
	AT	Home exercises following the instructions of the physiotherapist
	R	Planning sexual activity with minimal muscle pain (if any), choosing appropriate sexual positions/activities
Muscle weakness	PT	Analytical and functional strengthening of weak muscle groups, exteroceptive and proprioceptive stimulation, optimization of movement stereotypes
	AT	Home exercises following the instructions of the physiotherapist
	R	Choosing alternative sexual positions/activities that do not place high demands on posture biomechanics and maintaining the position
Limited range of motion namely:	PT	Joint mobilization, passive movements, active functional exercises, soft tissue tension optimization, breathing techniques, stretching of shortened muscles
	AT	Home exercises following the instructions of the physiotherapist
	R	Choosing alternative sexual positions/activities that do not place high demands on joint biomechanics and range of motion
Finger flexion contractures	PT	Mobilization of hand and wrist joints, myofascial techniques, muscle tension relaxation and stretching of wrist and finger flexors, functional hand exercises
	AT	Selected exercises with therapeutic rubber which the patient receives for home exercises
	R	Use of alternative touches of the partner (e.g., back part of the hand)
Raynaud's phenomenon	PT	Positive thermotherapy (paraffin), phototherapy (bio lamp), soft tissue techniques
	AT	Application of heat in the home environment
	R	Keep the affected part warm, warm showers/baths, use enough blankets
Microstomia	PT	Mobilization of the temporomandibular joint, muscle tension relaxation of masticatory muscles, soft tissue techniques in the orofacial area
	AT	Auto-relaxation of masticatory muscles, practicing exaggerated grimacing
	R	–
Psychoemotional status	PT	Relaxation techniques: elements of progressive relaxation, autogenic training, breathing exercises, yoga, Feldenkrais method, mindfulness meditation
	AT	Home exercise of relaxation techniques
	R	Mental hygiene, avoiding stress

Table 1 continued

Body image dissatisfaction	PT	Body awareness exercises, exteroceptive/proprioceptive stimulation, Feldenkrais method
	AT	Home exercises following the instructions of the physiotherapist
	R	–
Fatigue	PT	Relaxation techniques, breathing exercises, active exercises
	AT	Home exercises following the instructions of the physiotherapist
	R	Planning sexual activity with minimal fatigue
Sexual pain	PT	Optimization of PFM tension and associated functional changes, mobilization/stabilization of functionally related joints, optimization of breathing stereotype, the functional connection of the pelvis with functionally associated segments
	AT	Home exercises following the instructions of the physiotherapist
	R	Use of a suitable lubricating gel, choosing alternative sexual positions/activities that do not cause pain
Decreased lubrication	PT	–
	AT	–
	R	Use of a suitable lubricating gel
Vaginal tightness	PT	Optimization of PFM tension and associated functional changes
	AT	Vaginal auto-massage
	R	Use of a suitable lubricating gel, choosing alternative sexual positions/activities that do not cause pain
Objectively assessed PFM dysfunction	PT	Optimization of PFM tension and associated functional changes, mobilization/stabilization of functionally related joints, optimization of breathing stereotype, the functional connection of the pelvis with functionally associated segments
	AT	For PFM weakness: strengthening with the Educator® device following the instructions
	R	–

PT physiotherapy; *AT* autotherapy; *R* regimen; *PFM* pelvic floor muscles

based on their capability and motivation to undergo the 8-week intervention. One woman with SSc did not complete the study because of the time requirements of the project. Thus, in the final analysis, one patient with SSc from CG, who matched in age to the dropped-out SSc from IG, was excluded, and the final number of patients analyzed in the study was six women with SSc and two with IIM in each group (Fig. 1).

The clinical characteristics of both groups are shown in Table 2. The average age of women in IG and CG was almost identical. We observed a numerical difference between the groups in the ratio of patients with limited cutaneous (lc) SSc to diffuse cutaneous (dc) SSc, where IG had equal proportions of both subtypes, whereas in CG, most patients had dcSSc. This could explain the numerically higher disease activity score (ESSG) in CG, as well as skin score (mRSS), CRP, and significantly higher ESR, the functional limitations caused by SSc symptoms (SHAQ), and the physical component of quality of life (SF-36 PCS). In addition, there was a statistically

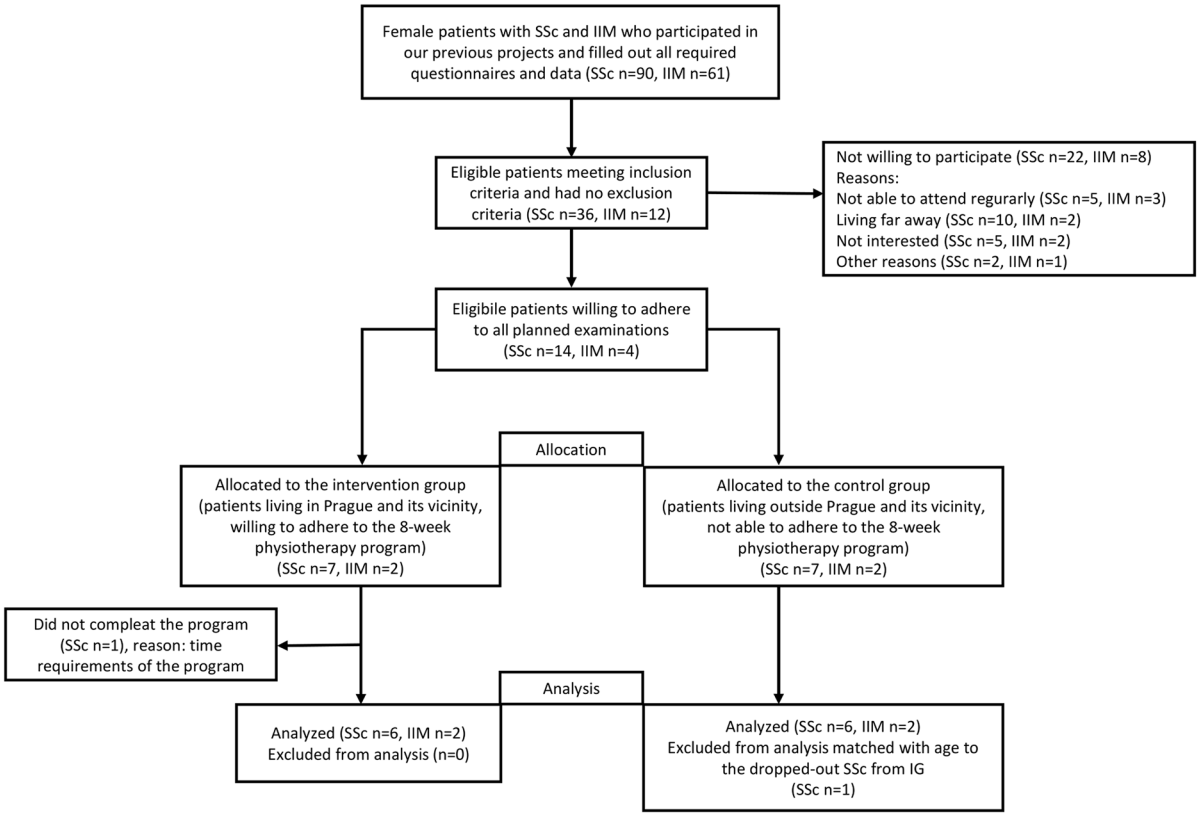

Fig. 1 Recruitment of patients with systemic sclerosis (SSc) and idiopathic inflammatory myopathies (IIM) into the 8-week physiotherapy program

significant difference in the subjective assessment of the sex life importance (VAS) when patients in IG considered sexual activity to be quite an important part of their lives, whereas in CG it was not considered that important.

The levels of overall sexual function (assessed by FSFI and BISF-W) were comparable in the two groups at the baseline. Over weeks 0–8, we observed a trend towards an improvement in IG in the FSFI total score compared to the numerical deterioration in CG. The inter-group difference for the FSFI total score was statistically significant. Clinically meaningful improvement (by $\geq$ 20%) in the FSFI total score was observed in 25% of IG vs. 12.5% of CG, whereas clinically meaningful deterioration (by $\geq$ 20%) was observed in 0% of IG vs. 37.5% of CG (Supplementary table S2). Similarly, significant changes were also found in the arousal domain of FSFI at the levels of both inter-group differences and intra-group improvement in IG. Moreover, we detected a trend towards differences between IG and CG at domains of lubrication and sexual pain of the FSFI, although the changes over weeks 0–8 within IG and CG were non-significant. We did not observe any significant changes in the domains of sexual desire, orgasm, and sexual satisfaction (Fig. 2 and Supplementary table 1).

Consistent with the above-described results, statistically significant inter-group changes were also shown in overall sexual function represented by the BISF-W total score. In CG we observed a noticeable trend towards a progressive deterioration of sexual function measured by BISF-W, whereas in IG, the numerical improvement did not reach statistical significance. Clinically meaningful improvement in the BISF-W total score was observed in 62.5% of IG vs. 12.5% of CG, whereas clinically meaningful deterioration was observed in 12.5% of IG vs. 50% of CG (Supplementary table S2).

Table 2 Sociodemographic, clinical and laboratory characteristics of patients in the intervention and control groups

Parameters	IG (n = 8)	CG (n = 8)	p-value
Sociodemographic variables			
Age, years	46.5 (39.0–54.3)	46.5 (28.0–55.0)	0.982
Having a partner, n (%)	7 (89)	8 (100)	> 0.999
Education, n (%) (primary/secondary/tertiary)	0 (0)/4 (50)/4 (50)	0 (0)/6 (75)/2 (25)	0.608
Sexual health features			
Menopause, n (%)	5 (62)	5 (62)	> 0.999
Pelvic surgery, n (%)	2 (25)	0 (0)	0.608
VAS: sexual life importance	8.0 (7.3–8.0)	4.5 (2.3–5.0)	**0.010**
Clinical features			
Disease duration, years	4.7 (2.7–6.5)	6.2 (5.0–8.7)	0.167
SSc subtype: lcSSc/dcSSc, n (%)	3 (50)/3 (50)	1 (16)/5 (83)	0.546
IIM subtype: PM/DM, n (%)	1 (50)/1 (50)	1 (50)/1 (50)	> 0.999
ESSG activity index (n = 6)	0.5 (0.5–1.0)	1.3 (0.4–3.3)	0.843
mRSS (n = 6)	2.0 (0.0–14.5)	10.0 (5.5–16.5)	0.251
MITAX (n = 2)	0.11 (0.11–0.11)	0.06 (0.02–0.10)	0.333
MYOACT (n = 2)	0.05 (0.03–0.06)	0.04 (0.0–0.08)	> 0.999
SSc/IIM-related symptoms: n (%)			
ILD/PAH, n (%)	3 (38)/1 (12)	4 (50)/2 (25)	> 0.999/ > 0.999
OD/P, n (%)	2 (25)/1 (12)	0 (0)/1 (12)	0.467/1.000
RI/RP, n (%)	0 (0)/6 (75)	2 (25)/6 (75)	0.467/1.000
DU/A, n (%)	1 (12)/0 (0)	1 (12)/1 (12)	1.000/ > 0.999
MW/SR, n (%)	4 (50)/1 (12)	2 (25)/1(12)	0.608/1.000
PROs (score range worst–best)			
BDI-II (63–0)	10.0 (0.3–22.3)	11.0 (5.7–20.5)	0.752
HAQ (3–0)	0.26 (0.00–1.22)	0.88 (0.44–1.74)	0.219
SHAQ (3–0), (n = 6)	0.69 (0.26–1.10)	1.54 (0.94–2.19)	**0.026**
Global SHAQ (3–0), (n = 6)	0.30 (0.11–1.15)	1.12 (0.84–1.87)	0.064
SF-36 PCS (16.6–57.9)	36.5 (27.4–51.8)	25.8 (21.5–27.2)	**0.049**
SF-36 MCS (5.5–63.6)	37.1 (27.4–51.8)	52.6 (39.6–57.9)	0.161
Laboratory features			
CRP, mg/l	1.7 (0.6–2.8)	3.1 (1.3–3.9)	0.128
ESR, mm/h	5.5 (4.3–13.0)	18.5 (11.8–34.0)	**0.033**

Table 2 continued

Parameters	IG (n = 8)	CG (n = 8)	p-value
Current treatment			
Prednisone equivalent dose, mg/day	1.5 ± 1.3	2.5 ± 1.3	0.843
GC/MTX, n (%)	3 (38)/1 (12)	1 (12)/1 (12)	0.608/1.000
CPA/LEF, n (%)	2 (25)/0 (0)	3 (38)/1 (12)	> 0.999/ > 0.999
RTX/ CCB, n (%)	1 (12)/1 (12)	1 (12)/ 3 (38)	1.000/0.569
AntiHT/bosentan, n (%)	0 (0)/0 (0)	3 (38)/1 (12)	1.000/ > 0.999
Epoprostenol, n (%)	0 (0)	1 (12)	> 0.999

Data are presented as median (IQR) or mean ± SD, unless stated otherwise. Statistically significant differences ($p < 0.05$) are highlighted in bold. If a selected parameter was evaluated only in a limited number of patients, n is specified in parentheses. *antiHT* antihypertensives; *A* arthritis; *BDI-II* Beck's Depression Inventory-II; *CCB* calcium channel blockers; *CG* control group; *CPA* cyclophosphamide; *CRP* C-reactive protein; *dcSSc* diffuse cutaneous SSc; *DM* dermatomyositis; *DU* digital ulceration; *ESR* erythrocyte sedimentation rate; *ESSG* European Scleroderma Study Group; *GC* glucocorticoids; *Global SHAQ* global SHAQ score; *HAQ* Health Assessment Questionnaire; *IG* intervention group; *IIM* idiopathic inflammatory myopathies; *ILD* interstitial lung disease; *lcSSc* limited cutaneous SSc; *MITAX* Myositis Intention to Treat Activity Index; *MYOACT* Myositis Disease Activity Assessment Visual Analog Scale; *mRSS* modified Rodnan Skin Score; *MTX* methotrexate; *MW* muscle weakness; *OD* esophageal dysmotility; *P* palpitation; *PAH* pulmonary arterial hypertension; *PM* polymyositis; *RI* renal involvement; *RP* Raynaud's phenomenon; RTX, rituximab; *SF-36 PCS* medical outcomes study Short Form 36–physical component summary; *SF-36 MCS* medical outcomes study Short Form 36–mental component summary; *SHAQ* Scleroderma Health Assessment Questionnaire–total score; *SR* skin rash; *SSc* systemic sclerosis; *VAS* visual analog scale

Similarly, in the domain of sexual arousal, we could recognize a statistical trend towards differences between IG and CG, where patients in CG showed a tendency towards gradual deterioration while in IG, the average score numerically improved despite the lack of statistical significance. In addition, we observed statistically significant inter-group changes in the frequency of sexual activity and initiation or receptivity of/to sexual activity. Women in IG showed a statistical trend to be more sexually active during our program, whereas patients in CG remained unchanged or slightly less involved in sexual activities during this period of time. No statistically significant changes were detected in the domains of sexual thoughts/desire, pleasure/orgasm, relationship satisfaction, and problems affecting sexual function (Fig. 3 and Supplementary table 1).

In addition to noticeable changes in sexual function between the groups, we also found statistically significant inter-group changes in the functional ability measured by HAQ. However, the intra-group differences did not reach statistical significance. In line with the improvement of physical abilities of patients in IG during our program, we also observed a statistically significant improvement in the physical component of the quality of life (SF-36 PCS) in IG, while in CG the values remained unchanged. Inter-group changes in SF-36 PCS reached statistical significance (Fig. 4 and Supplementary table 1). Clinically meaningful improvement in HAQ and SF-36 PCS was observed in 25% and 50% of IG vs. 0% and 37.5% of CG, whereas clinically meaningful deterioration was observed in 12.5% and 0% of IG vs. 50% and 25% of CG, respectively (Supplementary table S2).

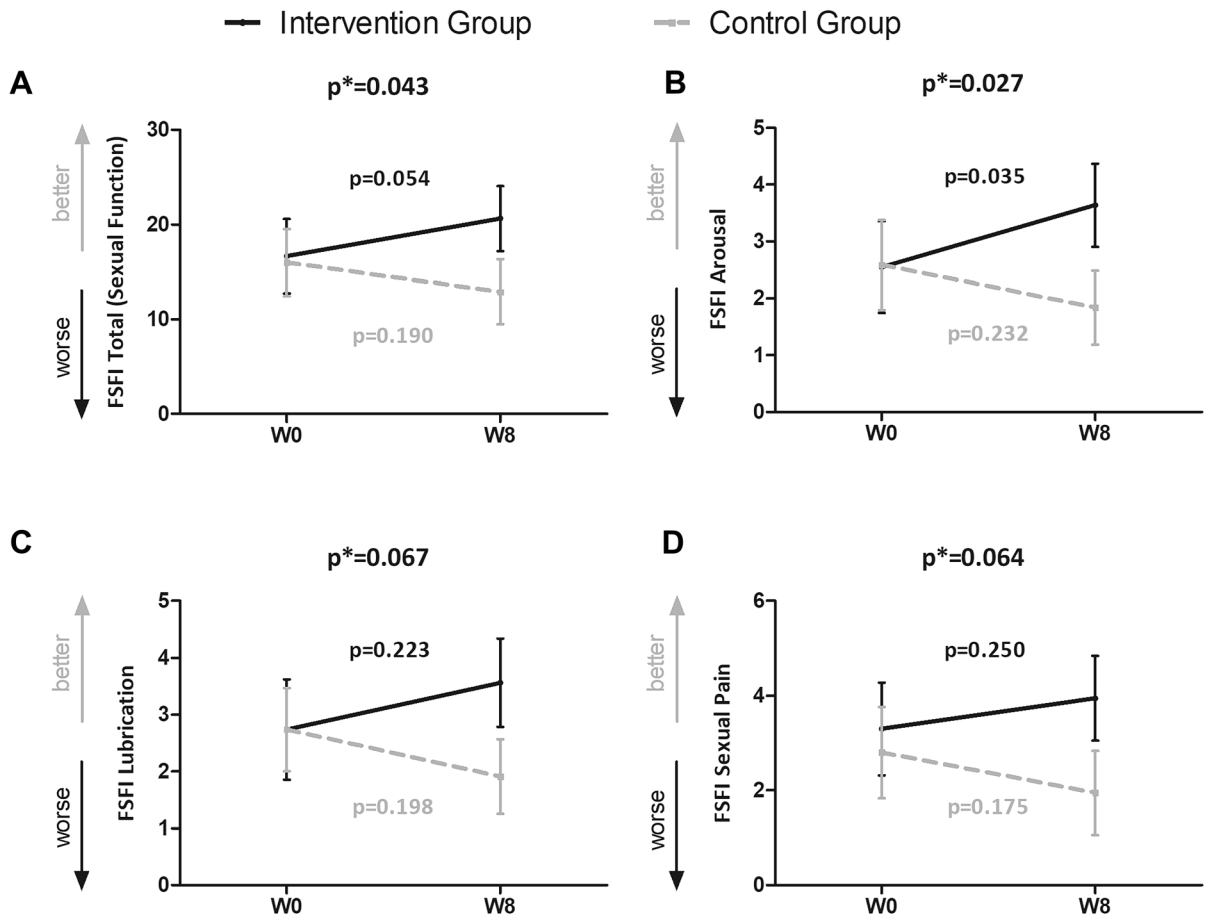

Fig. 2 Results of inter-group and intra-group analyses on sexual function assessed by the Female Sexual Function Index in the intervention and control group. **A** Significant inter-group difference in the change of overall sexual function assessed by the Female Sexual Function Index (FSFI) total score and a trend towards improvement in the intervention group over the weeks 0–8. **B** Significant improvement in sexual arousal observed during the 8-week physiotherapy program in the intervention group compared to the numerical deterioration in the control group and significant differences between both groups. A trend towards differences in inter-group changes in vaginal lubrication (**C**) and sexual pain (**D**). *p**, inter-group analysis by two-way repeated measures ANOVA (interaction: group × time); *p*, intra-group analysis by one-way repeated measures ANOVA; *w* week

DISCUSSION

The findings of our pilot intervention project suggest that physiotherapy could not only improve patients' functional abilities and their physical component of quality of life but also improve their sexual health. We are aware that these conclusions may be premature, as this was only a pilot project with a small sample size and two heterogeneous diagnoses, and further validation of our results is undoubtedly required. On the other hand, we would like to emphasize that the inclusion criteria were so narrowly defined that it was not achievable in our conditions to include a larger number of patients with such rare diagnoses. However, despite such a modest sample, some of the primary outcomes reached statistical significance; even more remarkable are the significant changes in total scores of both questionnaires assessing sexual functions (FSFI, BISF-W) and some of their domains, which definitely deserve attention and should spark further discussions. Perhaps less surprising are the secondary outcomes

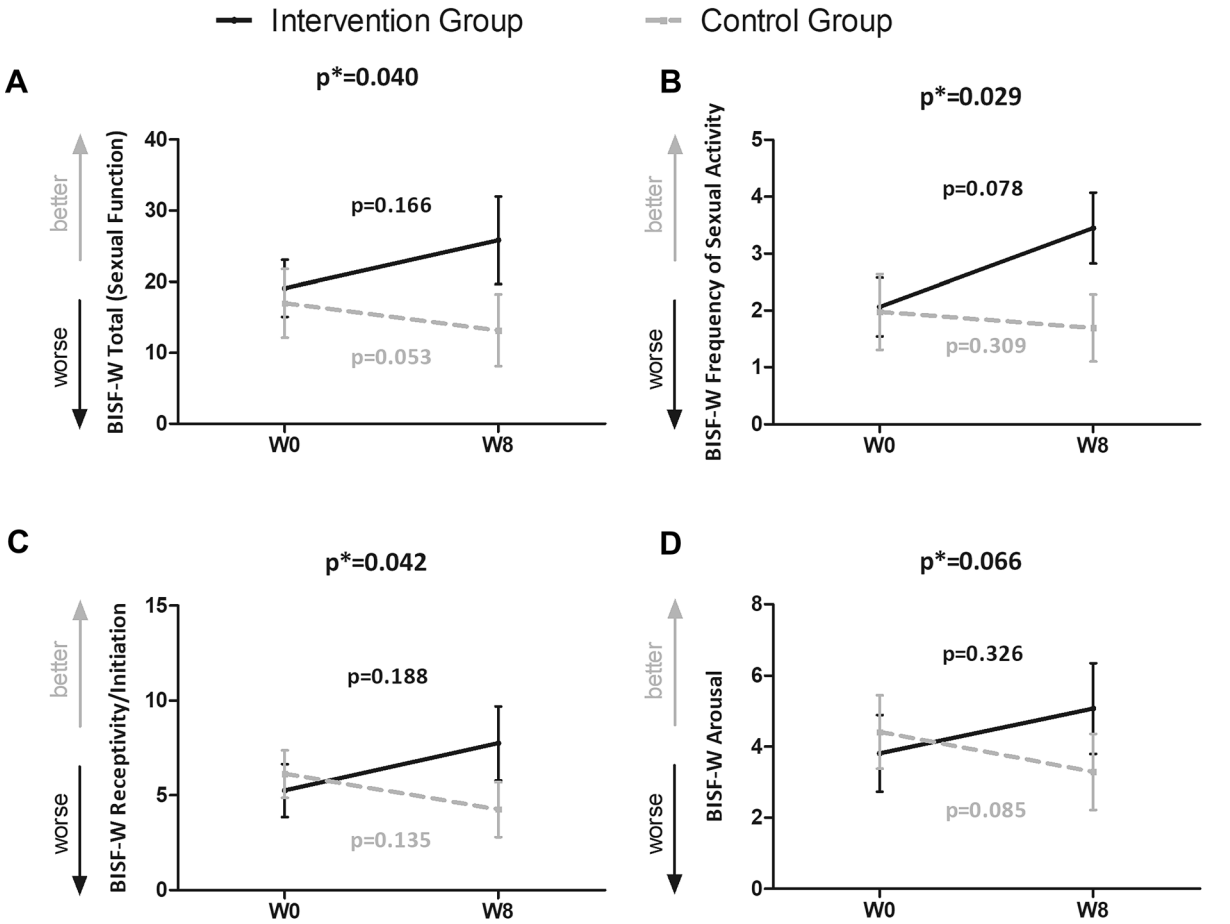

Fig. 3 Results of inter-group and intra-group analyses on sexual function assessed by the Brief Index of Sexual Functioning for Women in the intervention and control group. **A** Significant inter-group difference in changes in overall sexual function assessed by the Brief Index of Sexual Functioning for Women (BISF-W) total score and a trend towards deterioration in the control group over the weeks 0–8. **B** Significant difference between the intervention and control group in changes in frequency of sexual activity during the 8-week physiotherapy program and a trend towards improvement in the intervention group. **C** Significant inter-group difference in changes in receptivity or initiation of/to sexual activity. **D** A trend towards inter-group differences in changes of sexual arousal and a trend towards deterioration in sexual arousal over 8 weeks in the control group. *p**, inter-group analysis by two-way repeated measures ANOVA (interaction: group × time); *p*, intra-group analysis by one-way repeated measures ANOVA; *w* week

addressing improvements in functional ability and quality of life that are in agreement with findings concerning the effect of exercise/physiotherapy on overall health in patients with chronic diseases [8] as well as in patients with SSc [9, 10] and IIM [11, 12]. Moreover, we found statistically significant differences between IG and CG in the distribution of patients with clinically meaningful improvement and deterioration (by ≥ 20%) in FSFI total, BISF-W total, HAQ, SF-36 PCS, and MCS; however, these data need to be interpreted with caution because of the low numbers of patients in both cohorts. If we would like to compare our results of the effect of physiotherapy on sexual function in SSc and IIM, our options are narrowly limited, since this is the first study that addresses this topic to our knowledge. Our results are in agreement with the findings of relatively recent studies assuming the positive

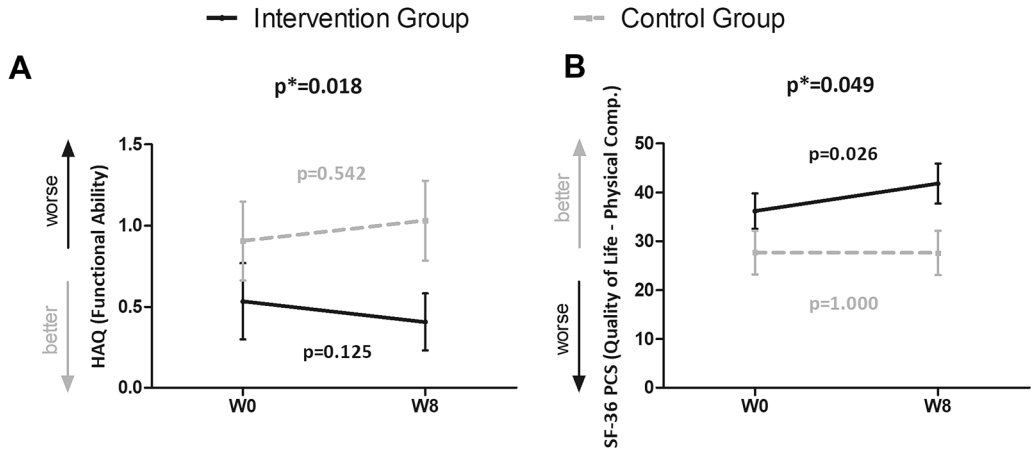

Fig. 4 Results of inter-group and intra-group analyses on functional ability and the physical component of overall quality of life in the intervention and control group. **A** Significant inter-group differences in changes in functional abilities assessed by the Health Assessment Questionnaire (HAQ). **B** Significant difference between the intervention and control group in the changes of the physical component of quality of life determined by the Medical outcomes study Short Form 36-Physical Component Summary (SF-36 PCS) and significant improvement during the 8-week physiotherapy program in the intervention group. *p**, inter-group analysis by two-way repeated measures ANOVA (interaction: group × time); *p*, intra-group analysis by one-way repeated measures ANOVA; *w* week

effect of physical exercise/activity on female sexual function in the general population [35, 36]. Contrarily, notably the recent Iranian randomized controlled trial (RCT) [37] evaluating the effect of PFM exercises and mindfulness on the sexual health of women with multiple sclerosis did not prove any significant impact on sexual dysfunction in these patients. However, if we take a closer look at the design of that study, its conclusions may be misleading because participants were assigned to three groups (a PFM exercise group, mindfulness group, and group combining the PFM exercise and mindfulness), and only multiple comparisons were performed to show no differences between the above-mentioned groups. Nevertheless, an improvement in the overall mean score of sexual function measured by FSFI and its subscales in each group was detected after 8 weeks of any intervention compared to baseline [37]. Another RCT studied the effect of a 16-week intervention including intermittent and continuous aerobic physical training on the sexual function of women with polycystic ovary syndrome and concluded that both protocols improved sexual function (measured by FSFI) and reduced anxiety and depression of these patients. Specifically, significant improvements were observed in the total score of FSFI for both exercise types in the domains of sexual pain and satisfaction for the continuous aerobic training group and in the domains of sexual desire, arousal, lubrication, orgasm, and satisfaction for intermittent aerobic training [38]. In our study, significant changes were observed in the total score of FSFI and its domain of arousal as well as a tendency towards improvements in domains of sexual pain and lubrication, which is consistent with the results of the study of Lopes et al. [38].

To explain the significant improvement in sexual health parameters in our study, we assume a combination of several factors. We can presume that, in addition to the generally positive effects of exercise on sexual function, the relaxation of the skin and soft tissues especially around the hands and mouth can make kissing, foreplay, or masturbation more pleasant for women with SSc. Furthermore, increased joint range of motion could improve the ability to assume certain sexual positions, and improved proprioception and body awareness might lead

to greater self-acceptance, as the appearance changes induced by SSc can be a frequent reason for body image dissatisfaction [39]. Moreover, pelvic floor exercises can increase proprioception in the pelvic area and increase blood flow in the vaginal mucosa, thus reducing discomfort during sexual intercourse, which is commonly experienced by women with SSc because of the tightening around the vaginal introitus, vaginal mucosal changes, and reduced lubrication [40]. Patients with IIM are likely to have weak pelvic floor muscles since the proximal muscle weakness is a typical manifestation [41], particularly in the muscles of the thighs and abdomen, which are anatomically and functionally connected to the pelvic floor. Also, psychological factors may play a key role in the improvement of sexual health. We can imagine that the opportunity itself to talk openly about this taboo topic with a health professional, and the specific therapy in which patients can actively participate, can improve patients' overall psychoemotional conditions, reduce feelings of frustration, and subsequently positively affect patients' sex lives. It is also possible that women who are willing to participate in the program and are more motivated to improve their sexual health may also be more likely to open up a discussion about their sexual issues with their partners and generally pay more attention to their sex lives. This alone can lead to positive changes. On the other hand, the greater willingness of patients in IG to engage in discussion about their sexual problems could have been a crucial reason why patients in IG had such remarkably improved sexual functioning in such a short time, while women in CG showed a gradual deterioration. However, the design of our study does not allow us to determine which of all possible factors might have contributed most significantly to the observed changes.

As mentioned above, the greatest limitation of this pilot study is the small sample size and the inclusion of women with two different diagnoses. Furthermore, the duration of the intervention was relatively short, and no follow-up was monitored. Another weakness of this project is that we could not afford to randomize the allocation into groups, first because it would have been unethical; second, we had to consider the feasibility of patients regularly commuting to the intervention program. Admittedly, the geographic disparity (patients allocated in IG were mostly from Prague and its vicinity whereas patients in CG lived far away from Prague) may have biased our results. Moreover, the effect of exercise could not be assessed by comparison to a healthy control population. This was because the therapeutic interventions tailored to SSc-/IIM-related subjective symptoms could not be applied to healthy controls who lack the disease-related manifestations as the dominant cause of sexual dysfunction. Another limitation of this study is that it did not directly engage gynecology or sexology specialists and that the routine gynecological examination was not included. Nevertheless, a cooperating sexologist, gynecologist, and specialists from the fields of psychosomatic medicine, and urogynecological physiotherapy were consulted about the study design. Moreover, some patients refused to be examined by another gynecologist than their attending one; thus, only the examination of the pelvic floor function (i.e., not a gynecological assessment) was performed (by the physiotherapist), which was fully within the competence of the physiotherapist.

Although our results may be limited because of these constraints, there is no doubt that they could serve as a basis for further more extensive analysis, ideally with multicenter collaborations, which would evaluate the effect of physiotherapy on sexual health in a larger sample of patients, for each diagnosis separately, optimally in a randomized manner, with a longer duration of the intervention and follow-up monitoring.

CONCLUSION

Our 8-week physiotherapy program not only prevented the natural course of progressive deterioration of functional ability but also led to a significant improvement in sexual function and quality of life in women with SSc and IIM. Since sexual dysfunction is a relatively common but at the same time an often neglected

problem in women with these conditions, future validation of our results could bring a potential therapeutic modality for managing sexual dysfunctions in women with SSc and IIM and provide patients the opportunity to actively participate in their treatment.

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Author's Contributions. MT, BH, and MŠ designed the study. SO, HŠ, BH, MT, LŠ, KP, HM, RB, and JV collected patients' data. BH carried out the physiotherapy intervention. SO, RB, HM, and JV performed the examination of the patients; HŠ processed the results of the laboratory tests. MT and BH performed the statistical analysis. BH and MT prepared the original draft of the manuscript. All authors critically interpreted the results, reviewed the drafts, and approved the final manuscript.

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Compliance with Ethics Guidelines. All relevant study documentation and amendments were approved by the independent Ethics Committee of the Institute of Rheumatology Prague with reference number 12898/2019, and the study was prospectively registered in the ISRCTN registry, no. ISRCTN91200867, in May 2021. It was conducted following the principles outlined in the Declaration of Helsinki, the Guidelines of the International Council for Harmonisation (ICH) on Good Clinical Practise (GCP) Guideline E6 (R2) (EMA/CPMP/ICH/135/95) European Union (EU) Directive 95/46/EC, and other applicable regulatory requirements. Patients provided informed written consent before enrollment in the study and consent for publication of the anonymized data.

Disclosures. Barbora Heřmánková, Maja Špiritović, Sabína Oreská, Hana Štorkánová, Heřman Mann, Karel Pavelka, Ladislav Šenolt, Jiří Vencovský, Radim Bečvář, Michal Tomčík have nothing to declare.

Data Availability. The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

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