



Case Report

Yoga therapy as an adjunct to conventional management of systemic sclerosis: A case series

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ARTICLE INFO

Article history:

Received 28 January 2021

Received in revised form

16 June 2021

Accepted 16 June 2021

Available online 12 November 2021

Keywords:

Lifestyle

Scleroderma

Inflammation

Autoimmunity

Mind-body medicine

ABSTRACT

Systemic sclerosis (SSc) is an autoimmune disorder leading to significant disability and loss of Quality of Life (QoL). Yoga has become popular in recent times for its potential therapeutic benefits. Since there are no scientific reports on the use of Yoga for SSc, we present two female cases (aged 49 and 29 respectively) of limited SSc (duration of illness 4 and 3 years respectively) who underwent Yoga therapy as an adjunct to conventional management in a residential setting for a period of five and four weeks, respectively. During their stay, they underwent a specifically designed Yoga module. After their discharge, they were followed-up for four weeks, during which they were asked to continue practicing Yoga for 1 h every day. Both of them reported a reduction in pain, stiffness, symptom scores, and improved QoL on discharge and at the follow-up compared to the values on admission. Erythrocyte Sedimentation Rate (ESR) and C-Reactive Protein (CRP), as biomarkers of inflammation, reduced on the discharge when compared to the baseline. No adverse events were noted during the stay and the follow-up. Thus, the present case series indicate a possible beneficial role of Yoga as an adjunct therapy to conventional management of SSc. Further studies in the area are warranted to ascertain the efficacy of Yoga for SSc.

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1. Introduction

Systemic sclerosis (SSc) is a chronic disease of unknown etiology characterized by abnormal accumulation of fibrous tissue in the skin. It is also a multisystem disorder involving the gastrointestinal tract, kidneys, heart, muscles, and lungs. SSc is an autoimmune condition that leads to a cascading inflammatory response in the affected tissue [1]. The peak age of onset is in the fourth and fifth decades of life, and the overall prevalence is 10–20 per 100,000, with a 4:1 female preponderance [2]. Noticeable changes due to SSc occur in the skin, alimentary tract, musculoskeletal system, and kidneys. Besides, lesions are often present in the blood vessels, heart, lungs, and peripheral nerves [3]. Skin symptoms include skin thickening, particularly on the fingers' joints, tight or shiny skin on the face, arms and legs [4]. Arthralgia, morning stiffness, and flexor tenosynovitis are also common symptoms of SSc [5]. The standard treatments for SSc include administration of immunosuppressants,

proton pump inhibitors (PPI) and non-steroidal anti-inflammatory drugs (NSAIDs) [6]. Being a progressive condition with multisystem involvement, SSc is often associated with lowered Quality of Life (QoL) and psychological problems [7]. Use of Complementary and Alternative Medicine (CAM) is common among patients with SSc [8].

Yoga is an ancient mind-body practice from India that has become popular globally for its therapeutic and health-promoting effects. The practices recommended in Yoga include the practice of physical postures (*asana*), breathing practices (*pranayama*), cleansing procedures (*shatkarma*), meditation (*dharana* and *dhyana*). The ancient texts of Yoga also recommend dietary modifications making it a lifestyle intervention. Yoga is known to influence people's musculoskeletal system, immune system, nervous system, and autonomic balance. Specifically, it is found to help increase flexibility, muscle strength, balance, and alleviate pain [9]. Yoga practice is also associated with positive emotions, self-regulatory skills in dealing with pain, emotional regulation and re-appraisal of life situations. Studies indicate that elements of Yoga may lead to mental relaxation and influence aspects of the parasympathetic nervous

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Peer review under responsibility of Transdisciplinary University, Bangalore.

system [10]. Yoga is one of the popular CAM modalities, but there are no studies evaluating the effect of Yoga in patients with SSc. Thus, the current case series explored the effect of Yoga on the symptoms, pain, stiffness, QoL, and inflammatory markers in patients with SSc.

2. Patient information

2.1. Patient A

A 49-year-old female participant presenting with complaints of tightness of the skin, swelling, pain, and skin discoloration from 4 years was admitted to a residential Yoga therapy center. She also had intermittent episodes of breathlessness since 4 years, which was attributed to the weakness of respiratory muscle weakness due to SSc. She was diagnosed with limited SSc by a rheumatologist using clinical evaluation. She was taking Mycophenolate mofetil 500 mg – TD, Rabeprezol 20 mg – BD, and Calcium (500 mg) and Vitamin D (200 IU) supplements OD since the diagnosis was made.

2.2. Patient B

Another 29-year-old female was admitted with complaints of multiple joint stiffness, discoloration, and tightness of the skin for three years. She was also diagnosed by a rheumatologist to have limited SSc three years back and was on Mycophenolate mofetil 500 mg – TD, Nifedipine 10 mg – BD, and Omeprazol 120 mg – BD.

3. Clinical findings

On examination, both participants were having generalized pain and tightness of the skin, multiple joint pain, along with a restricted range of motion. Skin discoloration was noted for both the participants. Both participants were normotensive and had no other health issues. While interacting with the attending physician, both expressed that they lacked interest, energy, and enthusiasm in their routine activities.

4. Timeline

A brief timeline of the occurrence of first symptoms, diagnosis, and commencement of medications is indicated in Table 1.

5. Diagnostic assessment

Since SSc is an inflammatory disease, Erythrocyte Sedimentation Rate (ESR) and C-Reactive Protein (CRP) were taken as the markers of inflammation on the date of admission and discharge. Participants were asked to rate their overall pain and stiffness on a visual analogue scale. Also, a specific symptom score checklist was prepared, which included skin-related

symptoms such as skin tightness and pigmentation. The participants had to mark their symptom severity as 0 – no symptom, 1 – mild symptom, 2 – moderate symptom, and 3 – severe symptom. A composite score of sum of all symptom scores was assessed on a daily basis. The weekly change in the composite symptom score is presented in Fig. 1. QoL was assessed using SF-36 questionnaire. SF-36 is a validated and reliable questionnaire to assess QoL in multiple health situations. It has nine subscales and the values are reported in percentage. A higher percentage indicates better QoL [11]. The findings on the day of admission, discharge, and follow-up after four weeks are presented in Table 2.

6. Therapeutic intervention

Following informed consent, clinical history, thorough systemic examination, the patients were administered a Yoga module specifically designed for their physical and psychological state due to SSc (Table 3). The intervention included training in gentle Yoga-based stretching, *asana*, *pranayama* and meditation/relaxation techniques. The practices were divided and administered through multiple sessions over the day during the inpatient stay. They were also subjected to yogic counselling by the in-house counsellor. During their stay, a bland vegetarian diet, as recommended in the texts of Yoga, was served to them. An attending physician monitored their condition daily. Both patients reported improvements in their pain and symptoms, along with improvement in the QoL. At the time of discharge, both patients demonstrated a marked reduction in the immunological markers (ESR and CRP). Both reported no adverse events during the course of treatment.

7. Follow-up and outcomes

Following their discharge, the patients were followed-up for subsequent four weeks on the telephone, during which they were asked to continue the Yoga practices for at least 1 h/day. Following four weeks, symptom scores, VAS for pain and stiffness, and SF36 were assessed using telephonic communication. Although the patients were asked to repeat the ESR and CRP, both did not do it and thus, we were unable to ascertain the changes in immunological markers on follow-up. Both patients reported a decline in symptoms, pain and stiffness, and a better QoL (Table 2). Both of them self-reported good compliance with Yoga.

8. Discussion

The present case series is the first attempt to evaluate the effect of Yoga therapy for patients with SSc. We found a reduction in the symptoms, pain, stiffness along with the improved QoL. Also, the markers of inflammation, ESR and CRP were found to decline during the stay of the participants in the residential Yoga therapy

Table 1
Timeline of events for the participants.

Health Event	Patient A	Patient B
Occurrence of first symptom	June 2014 (skin discoloration and hyperpigmentation)	November 2015 (skin discoloration & tightness)
Diagnosis of SSc	September 2014 (Clinical diagnosis by the rheumatologist)	December 2015 (Clinical and serological findings by the rheumatologist)
Use of medications	September 2014	December 2015
Admission to Residential Yoga Therapy Centre	09 February 2018	27 April 2018
Discharge from Yoga Therapy Centre	15 March 2018	17 May 2018
Follow up after 4 weeks	14 April 2018	15 June 2018

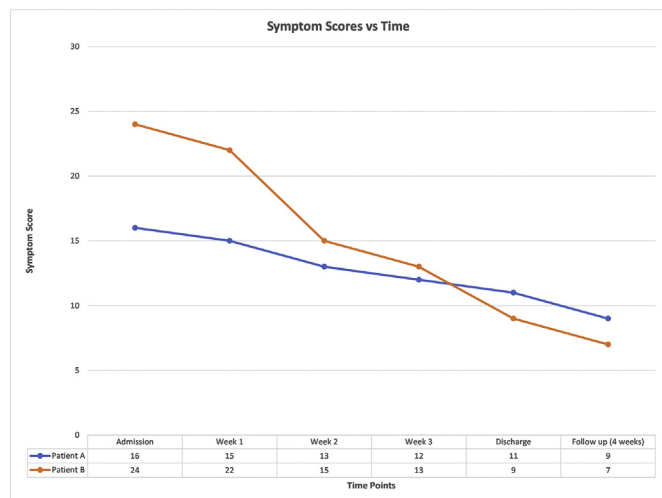


Fig. 1. The change of symptom scores for both patients over time. Note: Discharge of patient A was at week 5 and for patient B was at week 4.

program. Both the participants were advised to continue the practices during the four-week follow-up, during which the decline in symptoms, pain, and stiffness continued along with the better QoL. Since there were no adverse events during the entire period of intervention and follow-up, Yoga therapy may be considered a safe adjunct to conventional management of SSc.

According to the yogic understanding, autoimmune disorders are considered as *Adhija Vyadhi* (stress-borne diseases) [12]. In both the cases presented in this study, the psychological components affected were assessed using the emotional domains of SF-36 questionnaire. Both the participants had their scores of emotional subdomains lower at the baseline, which were enhanced following the practice of Yoga. This also indicated that Yoga practices not only work for symptom management, but also could be effective in working at the mental level of the disorder, i.e., *adhi* (stress). According to the *yoga sutras of Patanjali*, the root cause of mental stress is *avidya* (lack of knowledge) [13]. The patient education during their stay at our hospital worked at the causal state of removing *avidya*.

The findings of the current study are similar to other studies using Yoga as an intervention to manage chronic pain-related disorders. Yoga has been found useful in autoimmune conditions such as rheumatoid arthritis. Studies indicate the usefulness of Yoga in

reducing the serological markers of inflammation along with symptoms, both physical and psychological, when Yoga is administered along with medications [14–16]. A Cochrane review indicated positive outcomes of Yoga in patients with chronic pain [17]. Further, Yoga has been found useful in managing various autoimmune conditions such as multiple sclerosis [18,19], rheumatoid arthritis [15,20], Systemic Lupus Erythematosus [10], and pemphigus vulgaris [21].

The plausible mechanisms of action include the methodology of the practice of Yoga. The practices include gentle stretching with a focus on relaxation, mindful breathing, and the calming effect of meditation on the body–mind complex. Earlier studies indicate a beneficial role of Yoga in managing stress and thus, altering the neuroendocrine pathways [22]. Yoga brings down the levels of stress hormones that promote inflammation, lowers the pro-inflammatory cytokines such as IL-6, TNF- α , and CRP [23]. A recent review indicates Yoga as a feasible intervention in reducing inflammatory processes in chronic disorders [24].

Another critical aspect of reducing the QoL among patients with SSc is dissatisfaction with body image perception [25,26]. Studies indicate a positive role of Yoga in improving body image perception in different cohorts [27,28]. Therefore, one of the possible mechanisms to improve the QoL in these patients could be improved body image perception. However, since we did not record the body image perception as an assessment, it is difficult to state the effect.

Yoga is also known to help in symptom management, primarily pain, and improve QoL in conditions such as rheumatoid arthritis [16] and cancer [29]. This could be due to enhanced pain tolerance with Yoga practices. The mechanism of better pain tolerance appears to be the changes in the cerebral cortex, especially the insula [30]. Studies also indicate the role of Yoga in bringing autonomic balance [31], improved sensory-motor rhythm, and promote self-regulation [32,33]. However, it is not possible to ascertain a specific pathway through which the pain-reducing effects of Yoga were observed in the current cases.

There are several limitations of the current study. The sample size of two is too small to state the effects of Yoga clearly. We did not specifically assess the skin-related outcomes and body-image perception, which were the significant limitations of the study. Since SSc is a chronic degenerative autoimmune disorder, lack of long-term follow is yet another limitation of the study. Non-availability of the serological markers of inflammation during the follow-up also limits the understanding of the long-term effects of Yoga.

This preliminary case series indicates the feasibility of incorporating the Yoga module as an adjunct to conventional therapy to SSc. Further, large-scale clinical trials with robust design can be

Table 2

Changes in the variables from admission, discharge to follow-up period.

Variables	Patient A			Patients B			
	DOA	DOD (5 weeks)	Follow-up	DOA	DOD (4 weeks)	Follow-up	
VAS for pain	8	4	2	7	5	4	
VAS for stiffness	8	3	2	7	3	2	
Symptoms score	16	11	9	24	9	7	
Inflammatory Markers	ESR (mm/hour)	36	19	45	32	–	
	CRP (mg/L)	8	6	–	16	10	
SF-36 Questionnaire	Physical functioning	20	50	70	15	35	60
	Role limitations due to physical health	25	50	75	25	50	75
	Role limitations due to emotional problems	33.3	66.7	66.7	33.3	66.7	66.7
	Energy/fatigue	20	45	60	10	60	80
	Emotional well-being	48	52	60	32	60	64
	Social functioning	12.5	37.5	50	12.5	50	75
	Pain	10	55	77.5	10	45	77.5
	General health	15	55	65	10	60	75
	Health change	0	50	75	25	50	75

Note: VAS- Visual Analogue Scale; ESR- Erythrocyte Sedimentation Rate, CRP- C-Reactive Protein.

Table 3

Yoga practices administered to the participants (all practices were done on all days during the inpatient stay. The number of repetitions of the practice for follow up are mentioned in the table along with the practices).

Sl. No	Practices	Practice Name (Sanskrit and English)	Duration of Practice
1.	Breathing practices (any one or two/day)	Hands in and out breathing Hands stretch breathing Ankle stretch breathing	5 rounds (2 min) 5 rounds (2 min) 5 rounds (2 min)
2.	Loosening practices in standing (all practices to be repeated every day)	Twisting Side bending Waist rotation (clockwise and anticlockwise direction)	5 rounds (2 min) 5 rounds (2 min) 10 rounds each (2 min)
3.	Loosening practices in sitting (repeat section 3 and 4 on alternate days)	Knee cap tightening Passive patella movement (up and down, in and out, rotation) Knee bending Half butterfly Butterfly	5 rounds (2 min) each, both legs 10 rounds (4 min) both legs 5 rounds (2 min) each, both legs 5 rounds (2 min) each, both legs 3 min
4.	Loosening practices in supine (repeat section 3 and 4 on alternate days)	Folded leg lumbar stretch (left, right, both) Cycling Straight leg raising (left, right, and both)	5 rounds (2 min) 5 rounds (2 min) both legs 5 rounds (2 min)
5.	Shakti vikasaka (all practices to be performed every day)	Anguli shakti vikasaka (fingers) Kara tala shakti vikasaka (wrist) Kara prista shakti vikasaka (palm) Mani bandha shakti vikasaka (wrist) Kaphoni shakti vikasaka (elbow) Bhujabandha shakti vikasaka (upper arm) Griva shakti vikasaka (neck)	5 rounds (2 min) 5 rounds (2 min) 5 rounds (2 min) 5 rounds (2 min) 5 rounds (2 min) 5 rounds (2 min) 5 rounds (2 min)
6.	Yoga postures in standing (every day)	Ardha chakrasana (half wheel posture)	2 rounds (2 min)
7.	Yoga postures in sitting (every day)	Paschimottanasana (seated forward bend pose) Bhujanmanasana (earth salutation pose)	1 round (30 Sec) 1 round (30 Sec)
8.	Yoga postures in prone (every day)	Bhujangasana (cobra pose) Salabhasana (locust pose)	1 round (30 Sec) 1 round (30 Sec)
9.	Yoga posture in supine (every day)	Setubandhasana (bridge pose) Vipareetakarani (leg and hip raising pose)	1 round (30 Sec) 1 round (30 Sec)
10.	Relaxation techniques (to be performed in Shavasana: every day)	Instant relaxation technique Quick relaxation technique Deep relaxation technique	2 min 5 min 10 min
11.	Kriyas (cleansing techniques)	Jalaneti (nasal cleansing with water) (once a week) Vamanadhouti (internal cleansing by water) (once a week) Trataka (candle light gazing) (once a week) Kapalabhati (frontal brain cleansing) (every day)	10 min 10 min 10 min 1 min
12.	Pranayama (Yoga breathing)	Vibhagya pranayama (sectional breathing) Nadishuddhi (alternate breathing) Brahmari (humming bee breathing) Shitali pranayama (tongue breathing)	3 rounds (3 min) 9 rounds (3 min) 9 rounds (3 min) 9 rounds (3 min)
13.	Meditation (if possible, every day at least one practice should be done)	Nadaanusandhana (A, U, M and A-U-M kara chanting) Om meditation Cyclic meditation Mind sound resonance technique	10 min 10 min 30 min 10 min

employed to ascertain the efficacy of Yoga as an adjunct therapy for the management of SSC.

9. Patient perspective

Both the patients found the Yoga module to be helpful in managing their condition. During the follow-up, both indicated good compliance with the Yoga module. According to the participant A, “Yoga was easy to practice and could help in reducing stiffness and pain. I will continue with the practices that are helping for my long-standing health problem.” Participant B mentioned, “my symptoms have reduced drastically. I feel more comfortable with my own self. I wish to continue Yoga therapy and I hope that it will help me to give away or minimize my medications on a long run.”

10. Conclusion

The results from the current case series indicate the possible beneficial role of Yoga therapy as an adjunct to conventional SSC management. The limited evidence from the current study indicates that Yoga may reduce inflammation, pain, stiffness, and

symptoms and improve QoL. The findings warrant further large-scale clinical trials for ascertaining the efficacy of Yoga therapy as an adjunct to the management of SSC.

Informed consent

Signed informed consent was obtained from both the participants before administering residential Yoga therapy, data collection, and the use of data for scientific reporting.

Source(s) of funding

None.

Conflict of interest

None.

Author contributions

Apar Avinash Saoji: conceptualization; Data curation; Formal analysis; Investigation; Methodology; Project administration;

Resources; Software; Supervision; Validation; Writing and editing the manuscript. Pranab Das: Data curation; Formal analysis; Investigation; Writing and editing the manuscript. Naorem Subhadra Devi: Data curation; Formal analysis; Investigation; Methodology; Project administration; Resources.

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