

A case report on integrative medicine management of suspected Sjogren's syndrome

Megan E. Jolicoeur[^], Grant Chu, Ka-Kit Hui

Center for East-West Medicine, University of California, Los Angeles (UCLA), Los Angeles, CA, USA

Correspondence to: Megan E. Jolicoeur. Center for East-West Medicine, University of California, Los Angeles (UCLA), Los Angeles, CA 90404, USA.

Email: MJolicoeur@mednet.ucla.edu.

Abstract: This case report provides evidence that integrative medicine and traditional Chinese medicine (TCM) techniques can aid in the treatment of Sjogren's syndrome (SS). SS is a late-onset chronic systemic autoimmune disease that can result in dry mouth (xerostomia), dryness and atrophy of conjunctiva and cornea (keratoconjunctivitis sicca), and occasional glandular enlargement. The ramifications of SS can severely impact a patient's quality of life and multiple studies suggest that acupuncture and other integrative modalities can be an effective contribution to management. A 31-year-old female with arthralgias and muscle aches presented with fatigue, dry eyes and mouth, occasional oral sores and concerns for suspected SS. On clinical exam, she had several trigger points and her tongue was red and cracked. After a thorough evaluation by several specialists, she was suspected to have seronegative Sjogren's. From a TCM perspective, the patient was diagnosed with Liver and Kidney Yin Deficiency with (internal) Dryness, Qi Stagnation, and Blood stasis. This diagnosis was made based on her history, clinical exam, and tongue diagnosis. She was treated with acupuncture, cupping, trigger point injections, mind body medicine modalities, and dietary recommendations. After five treatments, she reported improvement in pain, energy, mood, and dry mouth. She was educated on how to continue to perform self-care at home. This case demonstrates how an integrative approach can be beneficial for patients with suspected SS.

Keywords: Integrative medicine; acupuncture; traditional Chinese medicine (TCM); Sjogren's syndrome (SS); case report

Received: 26 September 2021; Accepted: 11 November 2021; Published: 30 March 2022.

doi: 10.21037/lcm-21-61

View this article at: <https://dx.doi.org/10.21037/lcm-21-61>

Introduction

Sjogren's syndrome (SS) is a chronic, autoimmune disease characterized by lymphocytic infiltration of the exocrine glands. The hallmark of SS is ocular and mouth dryness secondary to autoimmune exocrinopathy which can eventually lead to keratoconjunctivitis sicca and xerostomia. It may occur in two forms—primary and secondary—and is associated with other autoimmune diseases such as rheumatoid arthritis. Current medical treatments are aimed at reducing symptoms and typically do not include integrative modalities (1). Acupuncture, nutrition, lifestyle

changes, and mind body medicine offer an alternative approach to treat the symptoms of SS but are not the typical intervention. Here we present a case of suspected SS that uniquely improved with acupuncture and other integrative medicine modalities.

We present the following case in accordance with the CARE reporting checklist (available at <https://lcm.amegroups.com/article/view/10.21037/lcm-21-61/rc>).

Case presentation

A 31-year-old female with history of attention deficit

[^] ORCID: 0000-0002-8886-6153.

Table 1 Timeline of patient events

Date	Event
12/09/2020	♦ Initial virtual visit
12/29/2020	♦ Performing qigong and self-care
	♦ Working on anti-inflammatory diet
	♦ Tendon sheath injections performed
1/4/2021	♦ She started using TENS machine
	♦ Stress improved
1/13/2021	♦ She underwent salivary biopsy that was negative
	♦ Sleeping well
1/19/2021	♦ Improvement in pain, energy, and sleep
	♦ Her dry mouth has improved
1/26/2021	♦ Significant improvement
	♦ Performing self-care

TENS, transcutaneous electrical nerve stimulation.

hyperactivity disorder, seasonal allergies, blepharitis, human papilloma virus, and anxiety was referred by her rheumatologist for diffuse generalized joint and muscle pain, feeling of dry eyes and dry mouth, occasional oral sores and concern for suspected SS. Additionally, she reported severe fatigue, hypermobile joints, and occasional skin rash. She noted symptomatic flares every other month and stated her symptoms were recently progressive in severity. Weather changes and stress traditionally exacerbated her symptoms, and she has been under a lot of stress at work recently. She reported taking acetaminophen for analgesia and over-the-counter eye drops for her dry eyes. She had a remarkable family history consisting of a maternal aunt with SS, maternal grandmother with rheumatoid arthritis, and her mother was diagnosed with systemic lupus erythematosus.

Review of the patient's lifestyle habits revealed a diet consisting predominantly of salty, processed foods. She reported minimal exercise. Her sleep was poor and not refreshing.

On physical exam, she was noted to have hypermobile joints as well as trigger points in the neck and gluteal area. Her tongue was dry, cracked and red.

The patient was previously evaluated by an allergy and immunology specialist at Cedars-Sinai Medical Center and workup revealed immunoglobulin A deficiency, recurrent sinusitis, multiple food allergies, possible specific antibody deficiency. She also had a workup by a

dermatologist and ophthalmologist at UCLA who both concluded her symptoms may be related to allergies. Her Schirmer test was reduced per ophthalmology. Her rheumatologist performed a thorough connective tissue panel, which was ultimately negative with the exception of an elevated sedimentation rate; however, given her clinical symptoms and family history, there was a high likelihood of seronegative SS. She was referred to physical therapy as well as otolaryngology for a salivary gland biopsy. The salivary gland biopsy did not meet the histopathologic criterion for SS although her rheumatologist still suspected seronegative Sjogren's (*Table 1*).

From a TCM perspective, patient was diagnosed with Liver and Kidney Yin Deficiency with (internal) Dryness, Qi Stagnation, and Blood Stasis. This diagnosis was made based on her history, clinical exam, and tongue diagnosis. She responded very well to an integrative approach. She was treated with acupuncture, cupping, mind body medicine modalities, and dietary recommendations. Acupuncture was performed at LI 4, LV 3, LI 11, SP 10, YINTANG, LU 7, K 6, ST 6, SI 3, K 3, ANMIAN, LI 10, UB 2. The acupuncture points were chosen to support her Yin deficiency (i.e., LU 7, K 6, LI 4, K 3), improve her blood stasis (i.e., SP 10), enhance her sleep and energy (i.e., LI 10, LI 11, ANMIAN), improve her mood (YINTANG), and more local points to reduce her symptoms of dry eyes and mouth (i.e., UB 2 and ST 6). DU 20 was added into the acupuncture prescription to help calm the spirit. Eventually auricular battlefield acupuncture was also added. She received trigger point injections to the neck, gluteal muscles, and piriformis muscles and cupping was performed on the neck and lower back. Her mind body practices included qi gong, breathing exercises, and meditation. We recommended starting an anti-inflammatory diet with the addition of goji berries, chrysanthemum tea, ginseng tea, and Asian pear for the dry mouth. Additionally, she was started on a vitamin D and magnesium supplement. Her symptoms significantly improved after five treatments. She specifically noted subjective improvement in musculoskeletal pain, mood, fatigue, sleep and dryness. She continued to perform self-care and dietary modifications at home.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). All interventions were performed in accordance with current ethical medical standards and without coercion. No investigational therapy

or intervention was performed. After her improvement was noted, the patient's informed consent was obtained. She was informed of the intent to publish medical history, intervention, and outcomes, to which she agreed. A copy of the written consent is available for review.

Discussion

SS is a late-onset chronic systemic autoimmune disease characterized by lymphocytic infiltration and destruction of the salivary and lacrimal glands. This can result in dry mouth (xerostomia), dryness and atrophy of conjunctiva and cornea (keratoconjunctivitis sicca), and occasional glandular enlargement. The disease can also affect other glands as well including the stomach, pancreas, and intestine. Some patients do present with systemic manifestations to include fatigue, rash, musculoskeletal symptoms or joint pain (1).

Primary Sjogren's will affect the parotid and lacrimal glands in contrast to secondary Sjogren's which is typically associated with an underlying connective tissue disease such as rheumatoid arthritis, systemic lupus erythematosus, and systemic sclerosis. Sicca complex includes primarily xerophthalmia and xerostomia and is not specific to underlying etiology (1,2).

Sjogren's was first described in the early 1900s by Swedish physician Henrik Sjogren. Between 400,000 and 3.1 million adults have SS. This condition will typically present between the ages of 45 and 55. Women are affected ten times more than men. About one third of patient present with systemic manifestations (3).

The pathogenesis is not completely understood; however, it is thought to be multifactorial with a combination of genetic and environmental factors. Studies indicate that the triggering event may be initiated by a viral or bacterial infections in a genetically predisposed individual. The infection appears to either stimulate or prime the immune system, which inadvertently draws lymphocytes to salivary glands and eyes (1). Once CD4⁺ T cells have infiltrated glands, they produce cytokines and further drive the inflammatory response. The inflammatory response may remain localized or can potentially drive systemic inflammatory processes that result in autoimmune exocrinopathy with extra-glandular manifestations. In approximately 6 to 10 percent of localized cases, the unabated lymphocytic activation may undergo transformation to lymphoma (4).

Numerous criteria were proposed for the diagnosis of SS. Most widely accepted are American and European

group developed international classification criteria for SS. These criteria include six different criteria including ocular symptoms, oral symptoms, ocular signs, positive histopathology, salivary gland involvement and sialography, or antibodies to anti-SSA, anti SS-B. The classification requires four of the six criteria, one of which must be positive biopsy of minor salivary or antibody test (5).

The goal of therapy from a standard medical approach is to ameliorate symptoms of dry eye and mouth, prevent complications of mucosal dryness, and detect any systemic manifestations. Patients would benefit from a multidisciplinary team to include a rheumatologist, eye care professional, and dentist. Patients with moderate to severe SS, including extra glandular involvement, may also benefit from systemic therapies, including the use of immunosuppressive or biologic agents (1). All patients benefit from nonpharmacologic and preventive interventions, including patient education regarding self-care measures, counseling regarding diet and medication use, and routine preventive care. However, some SS patients treated with conventional medicine still experience discomfort, such as pain and fatigue, and these individuals have sought complementary and alternative medicine (CAM) for relieving symptoms (1).

There is little scientific evidence on dietary interventions for the treatment of SS. Nonetheless, many of the symptoms that directly impact food intake and quality of life for Sjogren's patients can be managed with dietary modifications. Studies do support benefit from an anti-inflammatory diet. This diet favors fruits and vegetable, whole grains, lean protein, healthful fats and encouraged foods with omega-3 fatty acids. It recommends reducing or excluding the consumption of processed foods, red meats and alcohol (6). From a TCM perspective, dairy is also considered inflammatory and is recommended to reduce or exclude.

Chinese herbal medicine and acupuncture are common CAM therapies that are used. SS, from a TCM perspective, is characterized as "dry-Bi" or "dryness impediment". It manifests as Yin deficiency, fluid exhaustion and Blood dryness. The Yin deficiency manifests as "dry heat" and "consumption and deficiency of Qi and Body Fluid changes in salivary" (7). Patient with fibrotic changes in salivary glands are a result of "stasis". The general treatment principle is to nourish Yin, cool the Blood, and remove Blood stasis. Furthermore, replenishing Qi can help Yin grow (7).

Acupuncture combined with herbal medicine has

been applied in a variety of disorders, including clinical symptoms of dryness. In 2019, researchers compared the therapeutic effects of acupuncture versus herbs with hydroxychloroquine. The study included 60 patients: 30 in the TCM group and 30 in the conventional medicine group. The TCM group received acupuncture and the herbal formula Jie Du Tong Luo Shen Jin Tang (toxin resolving channel freeing liquid engendering decoction). The drug group received hydroxychloroquine in tablet form. The acupuncture plus herbs group achieved an 83.33 percent total treatment effective rate versus a 60 percent total effective rate for the drug group. The results demonstrated that acupuncture and herbal medicine treatments yield significant positive outcomes for patients with SS (8).

A nationwide population-based study was conducted between 2002 and 2011 which aimed to evaluate frequencies and patterns of Chinese herbal medicine used for SS. They analyzed 4,867 visits. Most patients were between the ages of 50 and 59 years old. The most common herbs included Qi Ju Di Huang Wan (Lycium Chrysanthemum Rehmannia Pill), Gan Lu Yin (Sweet Dew Drink), Xuan Shen (Radix Scrophulariae), Mai Men Dong (Ophiopogon japonicus), and Sheng Di Huang (Rehmannia glutinosa) (9).

The benefits of acupuncture for dry eyes have been widely reported. A study published in *Chinese Journal of Practical Ophthalmology* “found that patients receiving acupuncture plus lubricant had a 76.2% improvement in rate of dry eye symptoms versus those receiving just lubricants (58.6%)” (10). Furthermore, a double-blinded randomized controlled trial conducted in 2018, showed that acupuncture performed by the Niemtzow Protocol established that true acupuncture out-performed sham acupuncture based of a comprehensive patient questionnaire evaluating ocular surface staining, tear flow, and tear break-up time (11). A study published in *Ophthalmology Times*, found that acupuncture “significantly improved patients’ subjective assessment of dry eye symptoms”. Patients treated with acupuncture showed significant use reduction of artificial tears at six months and benefits lasting 9–12 months (12). It is theorized that acupuncture may regulate serotonin, a hormone that is fundamental to appropriate tear production. Additive benefits from acupuncture may also include decreased inflammation (or inflammatory response), increased circulation, and stimulated the nervous system. From these studies and others, the recommended points for dry eyes include: UB 1, UB 2, EX-HN 4, TB 23, GB 14, ST 2 (5).

These findings recapitulated a 2011 study from the

United States Navy who investigated how acupuncture can be used to treat the problem of dry mouth or xerostomia. The team found that needling a combination of three acupuncture points on bilateral ears and one point on each hand improved xerostomia for 80 percent of the patient cohort, who reported a marked increase in salivation. Notably, many of these patients’ dry mouths were the result of radiation therapy; however, some did report a diagnosis of SS (13).

Alternative and adjunct modalities have been tested. In 2015, there was a pilot randomized placebo-controlled study that evaluated the effects of laser acupuncture on salivary flow rates in patient with severe hyposalivation due to SS. This was a prospective cohort of 26 females that underwent laser acupuncture to the following points: LI 2, ST 5, ST 6, ST 7, SI 19, UB 13. The true laser acupuncture led to a significant higher amount of saliva production after 5 weeks that was sustained for 6 months (14). Results from studies like this indicate that alternative or adjunct approaches to Sjogren’s may have equitable results and that each regimen should be tailored to the patient’s needs.

Although scientific evidence has supported the efficacy acupuncture in relieving symptoms of xerostomia and xerophthalmia, there is a lack of well-controlled and standardized clinical studies (12). An ongoing randomized and controlled trial of 50 participants may determine the efficiency of acupuncture as a therapeutic option for SS patients’ symptoms relief. The SS patients will be randomly assigned to an experimental group that includes selected acupuncture points: R 6, E 6, E 2, Ig 4, VC 24, TA 23, UB 2. They will be compared to sham acupuncture control group for 8 weeks over 20 min sessions (15). Additional well-designed, high-quality studies are required to further determine the efficacy of acupuncture in the management of SS.

We presented a case of suspected seronegative SS with acupuncture and other integrative medicine modalities resulting in significant improvement. The ramifications of SS can severely impact a patient’s quality of life and multiple studies suggest acupuncture and herbal therapy can be incorporated into SS management with notable efficacy. Although further high-quality research is needed, it is important that clinicians are aware of the available integrative medicine treatment modalities.

Acknowledgments

Funding: This work was supported by the University of

California Los Angeles (UCLA) Center for East-West Medicine.

Footnote

Provenance and Peer Review: This article was commissioned by the editorial office, *Longhua Chinese Medicine* for the series “UCLA Case Studies”. The article has undergone external peer review.

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at <https://lcm.amegroups.com/article/view/10.21037/lcm-21-61/rc>

Peer Review File: Available at <https://lcm.amegroups.com/article/view/10.21037/lcm-21-61/prf>

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <https://lcm.amegroups.com/article/view/10.21037/lcm-21-61/coif>). The series “UCLA Case Studies” was commissioned by the editorial office without any funding or sponsorship. KKH served as the unpaid Guest Editor of the series and serves as an unpaid editorial board member of *Longhua Chinese Medicine* from May 2020 to April 2022. GC served as the unpaid Guest Editor of the series. KKH serves as the unpaid co-chair for the Integrative and Mental Health Expert Panel. The authors have no other conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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doi: 10.21037/lcm-21-61

Cite this article as: Jolicoeur ME, Chu G, Hui KK. A case report on integrative medicine management of suspected Sjögren's syndrome. Longhua Chin Med 2022;5:10.