

Gynaecological symptoms and sexual disability in women with primary Sjögren's syndrome and sicca syndrome

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Abstract

Objectives

This paper aims to investigate women with primary Sjögren's syndrome (pSS) and sicca syndrome (SS), focusing on the prevalence of disease-related symptoms and their impact on sexual ability, relationship, communication about sexuality with partner and health professionals (HP).

Methods

Sixty-two women with pSS and 33 with SS were assessed for sexual activity, relationship with partner, communication about sex; for physical disability and body esteem, fatigue, disability, quality of life (QoL), anxiety and depression.

Results

Around 55% patients had a relationship; >79% and around 70% at least 1 gynaecological (especially dryness), and 1 muscle-skeletal symptom, respectively; around 60% sex disability for disease-related symptoms, mainly dryness ($p=NS$ for all comparisons between pSS and SS). In both groups, disease changed sexual activity (around 50%), causing limitation (around 50%) and reduced frequency (>80%) in sexual intercourses; sex pleasure and satisfaction were around 30% and 25% ($p=NS$ for pSS vs. SS). Around 55% patients discussed with partner disease-effects on relationship; despite in around 70% partner understood difficulties, in around 34% disease altered relationship ($p=NS$ for pSS vs. SS). Around 16% patients were asked by HP if disease affected sexuality, around 30% never approached anyone to discuss about sex ($p=NS$ for pSS vs. SS). Disability, QOL, mood, fatigue, similar in pSS versus SS ($p=NS$), were not affected by xerostomia and xerophthalmia, but by sex concerns and sex disability.

Conclusion

Patients with pSS and SS present, often and at the same extent, gynaecological symptoms, leading to impaired sexual intercourse, affecting pleasure, satisfaction, sexual ability.

Key words

Sjögren's syndrome, sicca syndrome, sexual ability, gynaecological symptoms, quality of life

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Received on December 15, 2012; accepted

in revised form on February 13, 2013.

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EXPERIMENTAL RHEUMATOLOGY 2013.

Introduction

Sjögren's syndrome is a systemic autoimmune disorder in which inflammation causes atrophy of epithelial tissues and exocrine glands, reducing glandular secretion, especially at salivary and lachrymal glands, leading to xerostomia and xerophthalmia. It commonly involves also other epithelia, including skin, urogenital, respiratory, and gastrointestinal tracts and primarily affects women, with a female-male ratio of 9:1, and may occur in patients of all ages but typically has its onset in the fourth to sixth decades of life (1).

According to the American and European Consensus Group (AECG) classification criteria, primary Sjögren's syndrome (pSS) is defined in the presence of any 4 of 6 listed criteria as long as either histopathology or serology (antibodies to Ro[SSA] and/or to La[SSB]) is positive (2). Patients not fulfilling criteria but presenting ocular and salivary symptoms are considered as affected by not-Sjögren's sicca syndrome (SS).

Sexual health is defined as "a state of physical, emotional, mental and social health in relation to sexuality" (3). Sexuality is an integral part of an individual's self identity, body image, quality of life (QoL) (4), regarded as an important aspect of life for the majority of people in all stages of health and illness (5, 6).

Living with illness impacts on sexuality of patients with systemic rheumatic diseases such as rheumatoid arthritis (RA) (7, 8) ankylosing spondylitis (9), systemic lupus erythematosus (10), and systemic sclerosis (SSc) (11). Genital symptoms such as vulvar and vaginal dryness (12, 13), dyspareunia, pruritus, genital pain, increased susceptibility to infection and dysuria are frequent since pSS onset.

However, till now, only few data have been published about these symptoms, and, especially, about their potential impact on sexual ability, relationship with partners (14), global disability and QoL. Moreover, to the best of our knowledge, no data have been published about genital symptoms and sex ability in SS.

Our aim was to investigate sexual disability in women with pSS and SS,

evaluating by a specific questionnaire the prevalence of gynaecological and muscle-skeletal symptoms related to pSS and SS and their impact on sexual ability, the relationship and communication about sexuality with the partner and health professionals (HP).

Patients and methods

One hundred and thirty women attending the outpatient clinic of the Division of Rheumatology of the University of Florence were invited to participate in an observational transversal study, after signing a written informed consent according to the Declaration of Helsinki, after that a permission of ethical committee of AOUC (Azienda Ospedaliera Universitaria Careggi) was obtained.

The only inclusion criterion was the presence of xerophthalmia and xerostomia. Diagnosis of pSS (or SS) was made according to AECG criteria (2).

Exclusion criteria were the presence of current or previous rheumatic (secondary SS) and gynaecological diseases, and assumption of diuretic and anticholinergic drugs.

At enrolment, patients were investigated for: age, disease duration, oral and ocular symptoms, lachrymal and salivary glands function (by Schirmer, lissamine green, break-up time of lacrimal film tests, salivary scintigraphy), minor salivary gland biopsy, antibodies to SS-A(Ro) and SS-B(La), concurrent autoimmune diseases.

They were investigated for gynaecological anamnesis and assessed by some questionnaires to evaluate sexual ability, body esteem, fatigue, disability, QoL and psychological distress.

A control group including 50 healthy women comparable for age with patients with pSS and SS were investigated for gynaecological anamnesis and administered with questionnaires assessing fatigue, QoL and psychological distress.

Modified Hill Questionnaire

Created for RA, the Hill Questionnaire evaluates the impact of RA on sexual activity, relationship with the partner, and communication about sex issues by 12 questions (7). For this study, the tool was modified and adapted to pSS and SS. The revised questionnaire consists

Competing interests: none declared.

of 16 questions grouped according to the following areas: sexual life and disease-related symptoms impacting on it, relationship and communication with the partner, communication with HP. The 4 added questions investigate the presence of an actual sexual relationship and how the disease influenced frequency of intercourse, pleasure and satisfaction during sexual activity. The symptoms potentially impacting on sexual life and relationship included both gynaecological (vulvar and vaginal dryness, genital pain, dysuria, reduced sexual drive) and muscle-skeletal symptoms (arthralgia, myalgia, fatigue). Unlike the original version, evaluating the answers qualitatively, we chose to evaluate the answers quantitatively.

As in the original study (7), the modified questionnaire was proposed to 10 randomly selected patients with pSS and SS attending our Department.

As already in the Hill study (7), the questionnaire was not administered to healthy controls because it is meant to investigate the impact of pSS and SS on sexual activity, relationship with the partner, and communication.

Physical Disability Sexual and Body Esteem (PDSBE) scale

It evaluates, by 10 questions, the impact of physical disability on self esteem and affectivity. Each item is scored from 1 to 5, with total score ranging from 10 to 50, and lower values reflecting a more negative evaluation of affectivity and body esteem due to the disease (15).

Clinimetric evaluations

Fatigue was assessed by FACIT-F (Functional Assessment of Chronic Illness Therapy-Fatigue) (16, 17), disability by Health Assessment Questionnaire (HAQ) (18), QOL by Summary Physical and Mental Indexes (SPI and SMI) of Short Form 36 (SF-36) (19), anxiety and depression by Hospital Anxiety and Depression Scale (HADS-a and HADS-d) (20, 21).

Statistical analysis

Data were presented as mean $\pm$ standard deviation and as numbers and percentages. To compare for the clinical and clinimetric characteristics of

Table I. Demographical and clinical features of patients with Sjögren's syndrome and sicca syndrome and healthy controls.

	Sjögren's syndrome (62 subjects)	Sicca syndrome (33 subjects)	Healthy controls (50 subjects)	p-value
Age (years) (mean $\pm$ SD)	62.82 $\pm$ 13.74	65.45 $\pm$ 8.95	61.66 $\pm$ 8.6	NS
Disease duration (years) (mean $\pm$ SD)	6.45 $\pm$ 2.66	5.43 $\pm$ 3.1	–	NS
Menopausal state	54/62 (87.1%)	31/33 (93.9%)	40 /50 (80%)	0.036 [#]
Age at menopause (years) (mean $\pm$ SD)	47.08 $\pm$ 4.65	49.56 $\pm$ 5.21	50.31 $\pm$ 5.0	NS
Early menopause ($<$ 40 years old)	3/54 (5.5%)	2/31 (6.4%)	2/40 (5%)	NS
Hormone replacement therapy	11/54 (20.4%)	9/31 (29.03%)	10/40 (25%)	NS
Sexual activity	34/62 (54.8%)	20/33 (60.6%)	41/50 (82%)	0.0003 [§] 0.04*
Vaginal dryness	35/62 (56.4%)	23/33 (69.7%)	14/50 (28%)	0.004 [§] 0.0003*
Vulvar dryness	27/62 (43.5%)	19/33 (57.6%)	10/50 (20%)	0.009 [§] 0.0008*
Vulvar or vaginal dryness	42/62 (67.7%)	27/33 (81.8%)	16/50 (32%)	0.0003 [§] $<$ 0.0001*
Concurrent autoimmune diseases	17/62 (27.4%) 13 with Autoimmune thyroiditis	10/33 (30.3%)	–	NS

SD: standard deviation; NS: non significant; autoimmune diseases = autoimmune thyroiditis, autoimmune atrophic gastritis, inflammatory bowel disease, vitiligo, coeliac disease.

[#]p-value: significant for primary Sjögren's syndrome *versus* sicca syndrome; [§]p-value: significant for primary Sjögren's syndrome *versus* healthy controls; *p-value: significant for sicca syndrome *versus* healthy controls.

groups, Fisher's exact or χ^2 tests (when appropriate) were used to test for binomial variables, and Student *t*-test for continuous variables. Data were analysed by SPSS 18 for Windows.

Results

The 10 pSS and SS patients preliminarily interviewed by the Modified Hill Questionnaire found it feasible, not embarrassing and easy to be read and completed.

Seven out of 130 patients invited to participate refused, 28/123 patients who accepted did not return filled questionnaires and dropped out. Out of the 95 patients who completed the survey, 62 were diagnosed with pSS and 33 with SS (1).

In order to reduce the biases caused by missed responses, for each question we considered the percentage of the true number of answers and not the percentage of total number of patients.

Demographical and clinical features of the patients and healthy controls are shown in Table I. The groups of pa-

tients were similar in all the characteristics except for the number of patients in menopause, higher in SS than in pSS group ($p=0.036$).

Sexual activity was significantly higher in healthy controls than in patients with pSS and SS ($p<0.05$ for both comparisons) and, on the contrary, the prevalence of vaginal and vulvar dryness were significantly higher in patients with pSS and SS than in healthy controls ($p<0.05$ for all comparisons).

Modified Hill questionnaire

The results of the questionnaire are presented in Table II. No significant differences between pSS and SS patients were found in the prevalence of genital and muscle-skeletal symptoms, in the prevalence of symptoms interfering on sexual ability, in the variables assessing sexual activity and ability and in the items evaluating communication on sexual ability with partners and HP.

Genital and muscle-skeletal symptoms

Fifty-four point eight per cent of pSS

and 60.6% of SS patients were involved in a sexual relationship; 95.2% and 100%, respectively, of them had at least 1 gynaecological or muscle-skeletal symptom: 79.7% (pSS) and 90.9% (SS) had, at least, 1 gynaecological and 71.2% (pSS) and 72.7% (SS) 1 muscle-skeletal symptom.

Dryness was the most frequent symptom: vulvar or vaginal dryness were present in 67.7% and in 81.8% of patients, respectively with pSS and SS, followed by reduced sexual drive in 66.7%, only in SS patients, and fatigue (54.8% in pSS and 60.6% in SS). Vaginal dryness was reported by 56.4% (pSS) and 69.7% (SS) and vulvar dryness by 43.5% (pSS) and 57.6% (SS) of the patients. Arthralgias were referred in 46.8% (pSS) and in 51.5% (SS) and myalgias in 45.2% and 39.4% of the patients, respectively with pSS and SS.

Symptoms interfering on sexual ability

Sixty-seven point eight per cent of pSS and 50% of SS patients referred alterations in sexual ability due to symptoms: vulvar or vaginal dryness was reported by 94.7% (pSS) and 93.3% (SS). Moreover, 80.5% of pSS and 75% of SS patients referred vaginal and vulvar dryness, while in 73.3% of pSS and 93.3% of SS patients vaginal and vulvar dryness affected sexual ability.

Overall, 55.3% of pSS and 60% of SS patients referred association of vulvar or vaginal dryness, dyspareunia and reduced sexual drive.

Muscle-skeletal symptoms had minor effects on sexual ability, with fatigue and joint pain influencing sexual ability in 21% and muscular pain in 18.4% of pSS patients, while arthralgias and myalgias influenced sexual ability in 20% and fatigue in 33.3% of SS patients.

Sexual activity and ability

Sexual activity was regarded as important or very important by 61.5% and by 72.4% of pSS and SS patients, respectively.

Since disease onset, 50.9% of pSS and 48.4% of SS patients referred changes in sexual relationship; 55.3% and 51.7% reported limitation in sexual intercourse. Accordingly, reduced frequency

Table II. Results of the Modified Hill questionnaire in patients with Sjögren's syndrome and sicca syndrome.

	Sjögren's syndrome (62 patients)	Sicca syndrome (33 patients)	p-value
Are you involved at now in a sexual relationship?			
No	28/62 (45.2%)	13/33 (39.4%)	NS
Yes	34/62 (54.8%)	20/33 (60.6%)	
GENITAL AND MUSCLE-SKELETAL SYMPTOMS			
Do you have any of these problems?			
Gynaecological symptoms			
Vulvar dryness	27/62 (43.5%)	19/33 (57.6%)	NS
Vaginal dryness	35/62 (56.4%)	23/33 (69.7%)	NS
Vulvar or vaginal dryness	42/62 (67.7%)	27/33 (81.8%)	NS
Spontaneous genital pain	3/62 (4.8%)	3/33 (9.1%)	NS
Dyspareunia	31/62 (50%)	18/33 (54.5%)	NS
Dysuria	7/62 (11.3%)	8/33 (24.2%)	NS
At least 1 gynaecological symptom	47/59 (79.7%)	30/33 (90.9%)	NS
Reduced sexual drive	32/62 (51.6%)	22/33 (66.7%)	NS
No symptoms	13/62 (20.96%)	7/33 (21.21%)	NS
Muscle-skeletal symptoms			
Joint pain	29/62 (46.8%)	17/33 (51.5%)	NS
Muscle pain	28/62 (45.2%)	13/33 (39.4%)	NS
Fatigue	34/62 (54.8%)	20/33 (60.6%)	NS
At least 1 muscle-skeletal symptom	42/59 (71.2%)	24/33 (72.7%)	NS
No symptoms	3/62 (4.8%)	0/33 (0%)	NS
At least 1 gynaecological or muscle-skeletal symptom	59/62 (95.2%)	33/33 (100%)	NS
SYMPTOMS INTERFERING ON SEXUAL ABILITY			
Do any of the above problems affect your sexual ability?			
Answer	56/59 (94.9%)	30/33 (90.9%)	NS
Yes	38/56 (67.9%)	15/30 (50%)	NS
Vulvar dryness	27/38 (71%)	11/15 (73.3%)	NS
Vaginal dryness	29/38 (76.3%)	14/15 (93.3%)	NS
Vulvar or vaginal dryness	36/38 (94.7%)	14/15 (93.3%)	NS
Spontaneous genital pain	3/38 (7.9%)	3/15 (20%)	NS
Dyspareunia	23/38 (60.5%)	12/15 (80%)	NS
Dysuria	1/38 (2.6%)	3/15 (20%)	NS
Reduced sexual drive	21/38 (55.3%)	10/15 (66.7%)	NS
At least 1 gynaecological symptom	21/38 (55.3%)	9/15 (60%)	NS
Pain in your joint	8/38 (21%)	3/15 (20%)	NS
Pain in your muscles	7/38 (18.4%)	3/15 (20%)	NS
Fatigue	8/38 (21%)	5/15 (33.3%)	NS
Fatigue + joint or muscle pain	7/38 (18.4%)	4/15 (26.7%)	NS
SEXUAL ACTIVITY AND ABILITY			
How important is your sexual activity to you?			
Answer	52/62 (83.9%)	29/33 (87.9%)	NS
Not important	5/52 (9.6%)	2/29 (6.9%)	NS
Of little importance	14/52 (26.9%)	6/29 (20.7%)	
Important	29/52 (55.8%)	21/29 (72.4%)	
Very important	4/52 (7.7%)	0/29 (0%)	
Important/very important	32/52 (61.5%)	21/29 (72.4%)	NS
Not important/of little importance	19/52 (36.5%)	8/29 (27.6%)	
Do you think your condition has altered your relationship at sexual level in any way?			
Answer	55/62 (88.7%)	31/33 (93.9%)	NS
Yes	28/55 (50.9%)	15/31 (48.4%)	
Do you think your condition limits your sexual intercourse in any way?			
Answer	56/62 (90.3%)	29/33 (87.9%)	NS
Yes	31/56 (55.3%)	15/29 (51.7%)	NS

	Sjögren's syndrome (62 patients)	Sicca syndrome (33 patients)	p-value
<i>Has the frequency of your sexual intercourse changed since disease onset ?</i>			
Answer	46/62 (74.2%)	27/33 (81.8%)	NS
Yes	38/46 (82.6%)	26/27 (96.3%)	NS
	<50%: 4/38 (10.5%)	<50%: 1/26 (3.8%)	
	> 50%: 34/38 (89.5%)	>50%: 25/26 (96.2%)	NS
<i>Can you feel pleasure during your sexual activity since disease onset?</i>			
Answer	40/62 (64.5%)	26/33 (78.8%)	NS
Yes	17/40 (42.5%)	10/26 (38.5%)	NS
Not every time	7/40 (17.5%)	10/26 (38.5%)	NS
<i>Are you satisfied of your sexual activity since disease onset ?</i>			
Answer	41/62 (66.1%)	26/33 (79%)	NS
Yes	10/41 (24.4%)	7/26 (26.9%)	NS
Not every time	8/41 (19.5%)	10/26 (38.5%)	NS
COMMUNICATION ON SEXUAL ABILITY WITH PARTNERS AND HEALTH PROFESSIONALS			
<i>Do you think your condition has put a strain on your relationship with your partner?</i>			
Answer	54/62 (87.1%)	29/33 (87.9%)	NS
Yes	18/54 (33.3%)	10/29 (34.5%)	NS
<i>Have you discussed with your partner the effects of your disease to your sexual relationship?</i>			
Answer	48/62 (79%)	27/33 (81.8%)	NS
Yes	26/48 (54.2%)	17/27 (63%)	NS
<i>Does your partner understand the difficulties that your condition causes on your sexual relationship?</i>			
Answer	48/62 (77.5%)	26/33 (78.8%)	NS
Yes	32/48 (66.7%)	21/26 (80.8%)	NS
<i>Do you think that any of the drugs you take for your condition affect your sexual ability?</i>			
Answer	49/62 (79%)	28/33 (84.9%)	NS
Yes	6/49 (12.2%)	4/28 (14.3%)	NS
<i>Has any health professional before us ever asked if your condition has affected your sexual life?</i>			
Answer	51/62 (82.3%)	31/33 (93.9%)	NS
Yes	9/51 (17.6%)	5/31 (16.1%)	NS
	Gynaecologist: 8/9 (88.9%)	Gynaecologist: 4/5 (80%)	
	General practitioner: 1/9 (11.1%)	General practitioner: 1/5 (20%)	NS
<i>Have you ever approached anyone to discuss any problems that you have experienced with your sexual life?</i>			
Answer	52/62 (83.9%)	30/33 (90.9%)	NS
Yes	14/52 (26.9%)	10/30 (33.3%)	NS
	Medical doctor: 11/14 (78.6%)	Medical doctor: 8/10 (80%)	
	Friend: 5/14 (35.7%)	Friend: 2/10 (20%)	NS
	Relative: 0/14 (0%)	Relative: 0/10 (0%)	
	Nurse: 0/14 (0%)	Nurse: 0/10 (0%)	
	Association of patients: 0/14 (0%)	Association of patients: 0/10 (0%)	
<i>If you have any problems would you consider talking to any of the above people for help?</i>			
Answer	52/62 (83.9%)	26/33 (78.8%)	NS
Yes	41/52 (78.8%)	18/26 (69.2%)	NS

of sexual intercourse was reported by 82.6% of pSS and 96.3% of SS patients, with 89.5% and 96.1%, respectively, of them referring a reduction $\geq 50\%$. Only 42.5% of pSS and 38.5% of SS patients reported to feel pleasure during sexual activity, with the percentage falling respectively to 24.4% and 26.9% if related to sexual satisfaction.

Communication on sexual ability with partners and health professionals
Fifty-four point two per cent of pSS and 63% of SS patients discussed with their partner disease effects on sexual relationship and 66.7% of pSS and 81.8% of SS patients referred their partner as understanding the difficulties caused by the disease; 33.3% (pSS) and 34.5%

(SS) of patients thought the condition put a strain on their relationship. Only 12.2% of pSS and 14.3% of SS patients thought that drugs could affect sexual ability. Only 17.6% of pSS and 16.1% of SS patients referred that any HP before this survey asked if their condition affected sexual life, while 26.9% (pSS) and 33.3% (SS) approached someone to discuss about the problems related to sexual life; 78.6% and 80% of them spoke with a medical doctor; 78.8% and 69.2% of patients considered to talk to someone about sexual problems.

Modified Hill Questionnaire according to menopausal state in pSS and SS

In pSS, the answers to the Modified Hill Questionnaire were not different between patients not in menopause and patients in menopause ($p=NS$ for all comparisons). It was not possible to perform the same statistics in patients with SS, due to the extremely low number (2 patients) of pre-menopause subjects (data not shown).

Physical Disability Sexual and Body Esteem Scale

The scores of this scale are 32.59 ± 7.283 in pSS and 34.76 ± 8.367 in SS ($p=NS$) (Table III).

Clinimetric evaluations

No significant differences were found between the two populations in PDS-BE, FACIT, HADS-a and HADS-d, HAQ, SF-36 scores.

In both pSS and SS patients, FACIT scores were significantly higher, and SF36 SPI and SMI significantly lower, with respect to the control group ($p<0.05$ for all comparisons). Differently, HADS-a and -d scores in pSS and SS were similar to those found in controls (Table III).

Clinimetric values compared according to the answers to Hill

Modified Questionnaires – SPI and SMI of SF 36.

Patients with pSS not having a sexual relationship and not feeling pleasure during sexual activity presented lower scores in SPI than those having sexual relationship and feeling pleasure in sexual activity. In SS, patients with

Table III. Assessment of clinimetric measures in patients with Sjögren syndrome and sicca syndrome.

Questionnaire	Sjögren's syndrome (62 pts)	Sicca syndrome (33 pts)	Healthy controls (50 pts)	<i>p</i> -value (Sjögren's syndrome vs. Sicca syndrome)	<i>p</i> -value (Sjögren's syndrome vs. controls)	<i>p</i> -value (sicca syndrome vs. controls)
PDSBE (mean±SD)	32.59 ± 7.28	34.76 ± 8.37	–	NS	NA	NA
HAQ (mean±SD)	0.31 ± 0.39	0.35 ± 0.44	–	NS	NA	NA
FACIT-F (mean±SD)	13.17 ± 10.39	16.6 ± 10.39	9.6 ± 4.87	NS	0.0291	0.0001
<i>SF-36 (mean±SD)</i>						
SPI	42.33 ± 8.75	40.97 ± 8.68	48.9 ± 6.82	NS	<i>p</i> <0.0001	<i>p</i> <0.0001
SMI	39.32 ± 9.82	37.13 ± 10.3	44.16 ± 11.21	NS	0.0190	0.0065
<i>HADS (mean±SD)</i>						
HADS-a	7.54 ± 4.55	7.52 ± 4.49	6.86 ± 4.29	NS	NS	NS
HADS-d	7.06 ± 3.75	7.03 ± 4.11	5.70 ± 3.57	NS	NS	NS

SD: standard deviation; NS: not significant; PDSBE: Physical Disability Sexual and Body Esteem Scale; FACIT-F: Functional Assessment of Chronic Illness Therapy-Fatigue; HAQ: Health Assessment Questionnaire; SF-36: Medical Outcomes Survey Short Form 36; SPI: summary physical index; SMI: summary mental index; HADS-a: Hospital Anxiety and Depression Scale for anxiety; HADS-d: Hospital Anxiety and Depression Scale for depression.

dysuria had lower SPI than those not presenting the symptom ($p<0.05$ for all comparisons).

In SS, reduced SMI was found in patients with dysuria and fatigue and in those not feeling pleasure during sex in respect to patients without these features ($p<0.05$ for all comparisons).

– PDSBE

In pSS, lower PDBSE scores were found in patients in which disease limited sexual intercourse, in those referring dysuria and in patients not feeling pleasure during sex with respect to patients without these features ($p<0.05$ for all comparisons).

SS patients referring sexual relationships and intercourses as altered by the disease, with muscle pain, with sexual ability affected by disease-related symptoms and patients not satisfied about sexual activity showed lower PDBSE than patients without these features ($p<0.05$ for all comparisons).

– HADS: depression subscale

In pSS, higher HADS-d scores were found in patients with vulvar dryness than in those without this symptom, while, in SS, higher HADS-d was shown in patients with fatigue and in those with a reduced frequency of intercourses in respect to patients without these features ($p<0.05$ for all comparisons).

– HADS: anxiety subscale.

In SS, higher HADS-a scores were found in patients with vulvar dryness, with dysuria and in those not feeling pleasure during sex and not satisfied

about sexual activity in respect to patients without these features ($p<0.05$ for all comparisons).

– FACIT

Higher scores of FACIT were found in patients with pSS and SS referring fatigue than in those without this symptom ($p<0.05$ for all comparisons).

– HAQ

No difference in HAQ according to the answers to the Hill questionnaire was found in pSS and in SS.

Correlation of ocular and oral signs and symptoms with clinimetric values

No significant correlation of ocular tests (Schirmer, lissamine green, break-up time of lacrimal film tests) and oral tests (salivary scintigraphy) to scores of PDSBE, SF36, HAQ, FACIT, HADS-a and HADS-d was found.

Discussion

Our survey produced a 77% response rate, similarly to the 80% shown by Hill in RA (7) and higher than the 54% obtained with Female Sexual Function Index (FSFI) in SSc (11). Such a large response confirms feasibility of the Hill instrument, also in the present modified version.

We found no differences in women affected by pSS and SS in prevalence of gynaecological symptoms and on their impact on sexual ability, relationship and communication about issues related to sexuality and disease. At the best of our knowledge, our survey is the first assessing the prevalence of gynaecological symptoms and sexual issues

in women with SS, which, interestingly, results as high as in pSS patients. According to some authors, due to high specificity and low sensitivity of AEGC criteria (2), patients fulfilling them and those presenting only with sicca symptoms and signs may be regarded as affected by the same condition, yielding a different disease expression (22). Recent studies have shown that, in pSS, sicca symptoms are the predominant features in the majority of patients and that subjects presenting with sicca symptoms and developing pSS after a long follow-up have a favourable disease course (23-25). For this reason, it was proposed to consider patients fulfilling AECG-criteria as affected by Sjögren disease and those with negative antibodies and biopsy as affected by Sjögren syndrome (22, 26).

In course of pSS, gynaecological concerns, especially vaginal dryness (27-29) and dyspareunia (12, 26, 27, 30-32) are frequent in female patients, also potentially preceding the onset of ocular or oral symptoms by many years (30-32). In our population, 79.7% of pSS and 90.9% of SS patients referred almost one gynaecological symptom, with vulvar and vaginal dryness as the most reported concerns.

Although in literature a high prevalence of gynaecological symptoms in pSS is reported, there are scanty data on their impact on sexual ability and activity. Skoupuoli *et al.* showed that, despite the higher presence of dyspareunia and

vaginal dryness in pSS than in controls, no difference in the frequency of intercourse and libido, investigated just by single questions, between the two groups was found (33). Differently, our work, using a specific instrument, demonstrates that, in women with pSS and SS, genital concerns more than muscle-skeletal symptoms, interfere with sexual activity and ability.

In rheumatology, sexual disability is scarcely evaluated in clinical studies and in daily practice, although women with rheumatic diseases are known to have impaired sexual ability and activity, caused both by physical and psychological factors, potentially reducing sexual desire, satisfaction and frequency of intercourses (34).

In our study, sexual activity is considered important by the majority of patients with pSS (61.5%) and SS (72.4%), in agreement with RA patients, where it is regarded as important by 58% (7).

However, in our subjects, all the aspects related to sexual behaviour, from relationship and intercourse to pleasure and satisfaction, are severely impaired by the disease. This is concordant with data published about sexual disability in other chronic rheumatic diseases.

Around a half of pSS and SS patients refer, since disease onset, changes in sexual relationship and limitation in intercourses, whose frequency is reduced $\geq 50\%$ in around 90% of them, thus in a higher percentage than in RA, in which a reduction is reported in 70% of patients (7, 35-40).

Sexual drive is reduced in more than a half of pSS and SS and sexual ability is altered from disease symptoms in the majority of them in a similar percent in respect to RA (7, 35-40) and more than in SSc, in which desire and intercourse frequency are reduced in 57% and 52% of patients (37).

Differently from RA (7, 35-41) and partially in agreement with SSc (11, 37), in our pSS and SS patients sexual ability is more affected by gynaecological than by muscle-skeletal concerns.

In patients with a chronic illness, the diminishing of sexual activity can also be caused by a decrease in pleasure and a reduced interest in sexual activ-

ity, due to the disease-related problems (42, 43). Around 35% of the patients feel pleasure during intercourse, with the percentage further reducing if referred to satisfaction. These results are concordant with RA, in which reduced desire and satisfaction are referred by 60-90% of patients (35, 37-39), and in SSc, in which satisfaction is impaired with respect to controls (11).

In pSS women, circulating levels of the androgen dehydroepiandrosterone sulphate were shown as positively related to the quality of sexual life in women with pSS (44), but not with fatigue, well-being and functioning (45). The lack of data on sex hormonal profile and their potential influence on sex function and on other clinimetric measures may be seen as a limitation of the study. However, we found no differences in gynaecological symptoms and in sex activity and ability in pre-menopausal and post-menopausal women with pSS. Thus, we can hypothesise that the disease itself more than sex hormones may influence gynaecological concerns and impair sex function, fatigue, and QoL in pSS.

Accordingly to results shown for sexual activity and ability, also scores of PDSBE are similarly reduced in pSS and SS, demonstrating a notable impact of disease-related disability on body esteem and sexual expression, as well as on sexual ability and relationships. Interestingly, PDSBE is reduced in pSS and SS patients referring disease as limiting intercourses and also in those reporting a reduced sex ability and confidence.

Neither in pSS and in SS, QoL impairment, as well as disability, anxious and depressive symptoms, PDSBE are related to ocular and oral signs and symptoms, but are influenced by sexual ability and more by gynaecological than muscle-skeletal concerns. In fact, the low scores of HAQ, assessing mainly disability due to muscle-skeletal symptoms, show that both in patients with pSS and SS physical ability is scarcely altered.

In our pSS and SS patients, physical and mental QoL are similarly impaired and reduced with respect to controls. Moreover, SPI is lower in pSS patients

not having sexual relationship and not feeling pleasure during sexual activity and in SS subjects with dysuria. Similarly, patients with SS with dysuria and fatigue and not feeling pleasure during sex have lower scores in SMI.

Concordantly with data previously published in pSS (17), fatigue is equally increased in pSS and SS and higher than in controls. It is referred as a symptom in more than 50% of pSS and SS and reported as impairing sex function in more than 20% of our patients.

Contrarily to SSc and SLE, in our cohort, both anxiety and depression are not higher than controls (10, 11) but are, however, influenced by gynaecological concerns and sex issues more than by muscle-skeletal problems.

For what concerns the impact of gynaecological issues of pSS and SS on couple's relationship, in our study, the majority of patients discusses about their disease with the partner, who, in most cases, is sensitive to difficulties caused by their condition. This allows, in the majority of cases, not to put a strain on the relationship.

The many unanswered questions in items assessing relationship, pleasure, satisfaction and frequency of sexual intercourse are probably due to the embarrassment in speaking about sexuality (7). Accordingly, our results also highlight the reciprocal lack of communication about disease-related gynaecological and sexual problems between HP and women with pSS and SS, referring that, before our survey, no HP ever asked if their condition affected sexual life and the that majority of them, as already shown for RA patients (7), have spoken with a medical doctor. These mutual problems in communication are probably due to a variety of issues: the cultural heritage; the relationship between patient and physician; the scarce clinic importance given in the context of pSS and SS to gynaecological and sexual issues, considered less important than other features.

Conclusion

Despite the difficulty in speaking about sexuality, in women with pSS and SS we found a high and similar prevalence of gynaecological symptoms,

leading to reduced frequency of sexual intercourse, impaired pleasure and satisfaction, and altered sexual ability, and the need to talk about these issues with partners and HP. Rheumatologists should make patients aware that gynaecological symptoms and altered sex ability may arise in the course of pSS and SS, take them into account and, if needed, direct patients to obstetrics or gynaecologists, for proper management of these problems.

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