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LETTERS OF BIOMEDICAL AND CLINICAL RESEARCH

Diagnostic performance of Sjögren's Syndrome Screening Questionnaire (SSSQ)

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Sjögren's disease (pSS) is a chronic, systemic autoimmune rheumatic disease characterized by lymphocytic infiltration of the lacrimal and salivary glands, causing sicca symptoms. Differentiating pSS from other causes of sicca symptoms is essential as pSS patients may develop a systemic disease, progress to malignant lymphoproliferation, and finally are commonly managed by immunomodulatory treatment. Standard questionnaires on sicca symptoms used traditionally in outpatient settings as well as an entry criterion (positive response to at least one of the five questions) in the current 2016 ACR/EULAR classification criteria [1] were actually designed for research purposes. However, a "Preliminary Questionnaire for Sjögren's Syndrome in the Rheumatology Setting" (SSSQ) [2] was proposed for use in daily clinical settings recently. The SSSQ encompasses 5 questions extracted from 88 baseline questions. They were scored and the final score ranges from 0 to 11. By setting a cutoff point at ≥ 7 points, the SSSQ enabled the discrimination of pSS from non-pSS patients with a 64% sensitivity and 58% specificity (area under receiver operating characteristic curve, 0.65) [2]. We therefore aimed to evaluate performance of the SSSQ in our sicca population.

In a 3-year period from 2016 to 2019, 535 consecutive subjects, all Caucasians, were referred to our outpatient clinic due to sicca symptoms. At the time of their first evaluation, all patients fulfilled the standard questionnaire [1] to assess both ocular and oral involvement. In all patients, Schirmer-I test, unstimulated salivary flow (USF) test, Rose Bengal scoring, and immunoserological tests were performed. Patients with at least one pathological test also underwent minor salivary gland biopsy. In September and October 2021, after the SSSQ was published, all 535 subjects were contacted by telephone to take the SSSQ. The interviews were all performed by the same two medical students. The performance of SSSQ in recognizing pSS was assessed using the 2016 ACR/EULAR classification criteria as the gold standard test for pSS diagnosis. Out of the 535 sicca subjects, 415 agreed to participate (384 (92.5%) females, mean (SD) age 57.6 (13.8) years); the others either declined (21), died (2), or could not be reached (97). Based on the 2016 ACR/EULAR criteria, pSS was diagnosed in 127/415 (30.6%) and not confirmed in 288/415 (69.4%) subjects. While 376/415 (90.6%) subjects responded positively to standard sicca questions (110 with and 266 without pSS), 129 (31.1%) subjects (54 with and 75 without pSS) responded positively when using the SSSQ (they obtained ≥ 7 points). The performance of SSSQ in recognizing pSS is presented in Table 1 and Fig. 1.

This is, as to our knowledge, the first attempt to evaluate the performance of SSSQ in the all Caucasian population of subjects with sicca symptoms. SSSQ showed a higher specificity and accuracy for pSS compared to the standard sicca questionnaire and therefore performed better as far as identifying patients without pSS is concerned. Both questionnaires were unbalanced as far as sensitivity and specificity are concerned and both had high NPV. Sicca symptoms are a common complaint, especially in population over 65 years [3–5] and pSS is only one of the potential causes. During the COVID-19 pandemic, performing the full functional diagnostic pSS workup (including non-stimulated and/or stimulated salivary flow tests and/or minor salivary gland biopsies (MSGB)) is challenging. Furthermore, standard questions on sicca symptoms used traditionally in the outpatient settings as well as an entry criterion in the current 2016 ACR/EULAR classification criteria were designed for research purposes. Having a more pSS-specific sicca questionnaire, designed primarily for use in daily clinical practice, could help in optimizing clinical practice and, in that respect, SSSQ performed well in our cohort. Our study faces some limitations because of its retrospective data retrieval; furthermore, both questionnaires were not applied simultaneously and SSSQ was performed on the phone. Other studies addressing those issues are therefore needed.

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Table 1 The diagnostic performance of SSSQ (Sjögren's Syndrome Screening Questionnaire) and the 2016 ACR/EULAR sicca questions in our sicca cohort

Statistics	SSSQ	Standard sicca questions
	Value (95% CI)	Value (95%CI)
Sensitivity	42.5% (33.8–51.6%)	86.6% (79.4–92.0%)
Specificity	73.7% (68.5–78.9%)	7.6% (4.9–11.3%)
PLR	1.4 (1.2–2.2)	0.9 (0.9–1.0)
NLR	0.8 (0.7–0.9)	1.8 (1.0–3.2)
PPV	41.9% (35.2–48.8%)	29.3% (27.7–30.9%)
NPV	74.5% (71.2–77.5%)	56.4% (41.6–70.2%)
Accuracy	64.3% (59.5–69.0%)	31.8% (27.4–36.5%)
AUC	0.58 (0.52–0.64)	0.52 (0.46–0.57)

PLR, positive likelihood ratio; *NLR*, negative likelihood ratio; *PPV*, positive predictive value; *NPV*, negative predictive value; *AUC*, area under receiver operating characteristic curve

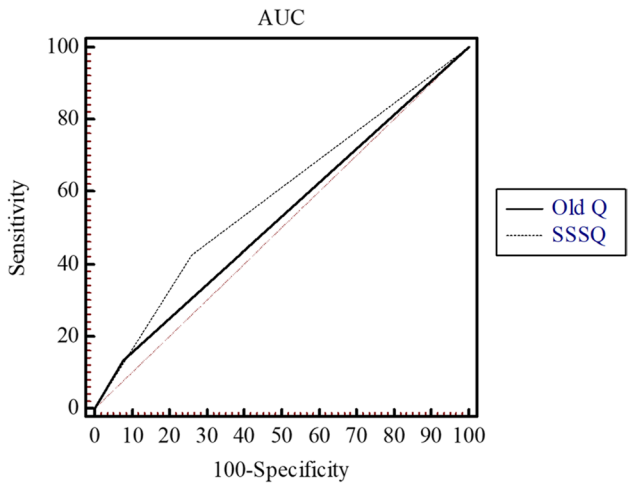

Fig. 1 Sensitivity and specificity of the old (Q) and new (SSSQ) questionnaires

To conclude, we tested the diagnostic performance of SSSQ in a cohort of Slovenian sicca subject. The SSSQ showed better specificity and accuracy for SS as the standard sicca questions and could indeed help in optimizing management of sicca subjects in daily clinical practice.

Author contribution AH, KPP, ZT, KF, and JK contributed to the acquisition, analysis, and interpretation of data. AH and KPP wrote the draft manuscript. ZT, KF, and JK helped revise the manuscript and gave suggestions. All 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 authors meet the authorship requirements and have read and approved the manuscript for submission.

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Declarations

Ethics approval The study was approved by the Slovenian National Medical Ethics Committee, Approval Number 0120–470/2019/4.

Disclosures None.

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