What is the Best Initial Screening for Suspected Sjögren's Disease?

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Introduction

During the COVID-19 pandemic the routine diagnostic procedure in Sjögren's disease (pSS) was highly hampered.
The aim of our study was to determine in a cohort of well-defined sicca patients the best screening tool for pSS, obviating, when possible, the initial exposure to potentially infectious body fluids (eg. saliva, tears) and/or invasive procedures (lip biopsy) We also evaluated the performance of the newly proposed 5-item Sjögren's Syndrome Screening Questionnaire (SSSQ) derived from SICCA study data, that was shown to better differentiate pSS from non-pSS sicca patients compared to conventional sicca questionnaire[1].

Methods

Our cohort consisted of 282 subjects (109 pSS and 173 non-pSS sicca patients) who completed a standardized diagnostic procedure for pSS based on the ACR/EULAR 2016 classification criteria[2] (standard sicca questions, Schirmer's test, Rose Bengal test, unstimulated salivary flow test (USF), immunoserological test (anti-SSA) and lip biopsy) between Jan/2016 to Dec/2018. In addition, major salivary gland ultrasound (SGUS) was performed in each subject at the time of diagnostic procedure[3]. Afterwards all subjects were contacted by phone to fulfil the new SSSQ. Descriptive statistics and ROC curve analysis, calculating area under the curve (AUC) were used to analyse data.

Results

The results of standard sicca questions, SSSQ, Schirmer's test, Rose Bengal test, USF, SGUS, anti-SSA and lip biopsy were positive in 90.4%, 34.0%, 50.4%, 30.1%, 63.8%, 19.1%, 27.7% and 33.3%, respectively. The Table 1 shows the distribution of positive tests according to final diagnosis (pSS vs. non-pSS). The new SSSQ had a sensitivity of 41.3% and a specificity of 70.5% for pSS (in comparison, the traditional sicca questions were 85.3% sensitive but only 6.4% specific for pSS). Table 2 and Figure 1 show the performance of combinations of various tests that were easily available in the daily clinical practice for supporting pSS diagnosis.

Table 1. Distribution of positive tests based on final diagnosis

	Standard questionnaire	SSSQ	Schirmer's test	Rose Bengal test	USF	SGUS	SSA	Lip biopsy
pSS	85.3%	41.3%	60.6%	45.0%	65.1%	39.4%	65.1%	84.4%
Non-pSS	93.6%	29.5%	43.9%	20.8%	63.0%	6.4%	4.0%	1.1%

Figure 1. ROC cur
analysis of differen
combinations for
diagnosing pSS

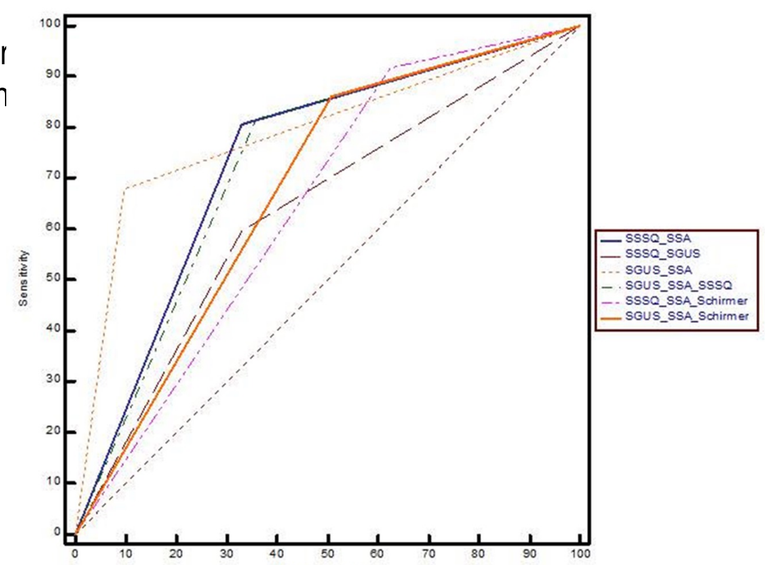

Table 2. The performance of different combinations tests in pSS screening

Positive tests	SGUS or a-SSA		SGUS or SSSQ		a-SSA or SSSQ		SGUS or a-SSA or SSSQ		SGUS or a-SSA or Schirmer		SSSQ or a-SSA or Schirmer		SGUS or a-SSA or Rose Bengal test		SSSQ or SSA or Rose Bengal test	
Statistic	Value	95% CI	Value	95% CI	Value	95% CI	Value	95% CI	Value	95% CI	Value	95% CI	Value	95% CI	Value	95% CI
Sensitivity	67.9%	58.3% 76.5%	59.6%	49.8% 68.9%	80.7%	72.1% 87.7%	81.7%	73.1% 88.4%	86.2%	78.3% 92.1%	91.7%	84.9% 96.2%	80.7%	72.1% 87.7%	86.3%	78.0% 92.3%
Specificity	90.2%	84.7% 94.2%	67.1%	59.5% 74.0%	67.1%	59.5% 74.0%	64.2%	56.5% 71.3%	49.1%	41.5% 56.8%	37.6%	30.3% 45.2%	71.1%	63.7% 77.7%	56.1%	48.3% 63.6%
PLR	6.9	4.3; 11.1	1.8	1.4; 2.4	2.5	1.9; 3.1	2.3	1.8; 2.8	1.7	1.4 2.0	1.5	1.3 1.7	2.8	2.2; 3.6	2.0	1.6; 2.4
NLR	0.4	0.3; 0.5	0.60	0.5; 0.8	0.3	0.2; 0.4	0.3	0.2; 0.4	0.3	0.2 0.5	0.2	0.1 0.4	0.3	0.2; 0.4	0.24	0.2; 0.4
PPV	81.3%	73.1% 87.4%	53.3%	46.7% 59.7%	60.7%	55.1% 66.1%	58.9%	53.6% 64.1%	51.7%	47.5% 55.7%	48.1%	44.9% 51.3%	38.7%	32.9% 44.6%	37.1%	31.4% 43.1%
NPV	81.7%	77.2% 85.5%	72.5%	67.2% 77.2%	84.7%	78.8% 89.2%	84.7%	78.6% 89.3%	85.0%	77.6% 90.3%	87.8%	78.9% 93.3%	63.8%	57.8% 69.4%	53.7%	49.0% 58.2%
Accuracy (*)	81.6%	76.5% 85.9%	64.2%	58.3% 69.8%	72.4%	66.7% 77.5%	70.9%	65.2% 76.2%	63.5%	57.6% 69.1%	58.5%	52.5% 64.3%	85.4%	79.8% 89.7%	87.4%	80.7% 92.0%
AUC	0.79	0.74 0.84	0.63	0.57 0.69	0.74	0.68 0.79	0.73	0.67 0.78	0.68	0.62 0.73	0.65	0.59 0.70	0.77	0.72 0.82	0.73	0.67 0.78

Conclusion

Our data suggest, that screening with a combination of anti-SSA and SGUS provides the highest AUC and specificity for pSS, while a combination of SSSQ, anti-SSA and Schirmer's test yielded the best sensitivity among the compared combinations.

Literature::

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