

原文題目(出處)：	The role of a labial salivary gland biopsy in the diagnostic procedure for Sjögren's syndrome; a study of 94 cases
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報告日期：	103.09.09

內文：

Introduction

Sjögren's syndrome:

Sjögren's syndrome (SS) is a multiorgan, chronic autoimmune disease, primarily directed against exocrine glands(salivary and lacrimal glands)

1. Symptoms & signs:

Dry mouth(Xerostomia),dry eyes(xerophthalmia)

2. Complication:

Dry mouth → angular cheilitis, dental decay (cervical caries)

Salivary gland enlargement → retrograde bacterial sialadenitis

Fatigue, arthritis, kidney failure, non-Hodgkin lymphoma

3. Population prevalence: 0.5-1%

4. Male-to-Female rate : 1:9

5. Aged: Middle-aged adult

6. Primary & secondary Sjögren's syndrome

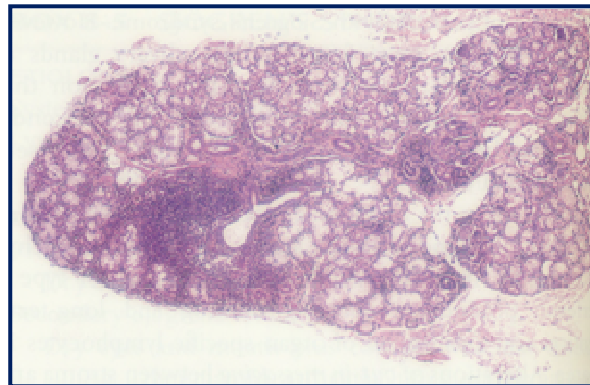
Classification	Explanation
Primary	no other autoimmune disorder
Secondary	other associated autoimmune disease, ex rheumatoid arthritis

7. Classification criteria: American-European Consensus Group (AECG), at least 4

Symptoms & signs	Explanation
Ocular symptoms	Dry eyes
Oral symptoms	Dry mouth
Ocular signs	Schirmer I test, Rose Bengal score
<u>Histopathology</u>	<u>Minor salivary glands, lymphocytes foci</u>
Salivary gland involvement	
Serologic findings	Serum, Anti- Ro(SS-A), Anti-La(SS-B)

8. Labial salivary gland biopsy (LSGB):

- a. Site: usually lower lip, normal labial mucosa
- b. Dimension: 1.5~2.0 cm incision, ≥ 5 accessory glands
- c. Histopathology:
 - i. Focal chronic inflammatory aggregates
 - ii. ≥ 50 lymphocytes and plasma cells within 4-mm² area
 - iii. Lymphocytes focus adjacent to normal-appearing acini
 - iv. Consistent in most of glands



Not 100% reliable

Purpose of present study:

Examine the role of the outcome of the LSGB in the diagnostic procedure

Material and Methods

1. In the files of the department of pathology of the VU university medical center, Amsterdam, the Netherlands, 139 labial salivary gland biopsies (LSGBs) could be retrieved in the period between 2000 and 2010.
2. Out of these 139 cases, 45 patients have been excluded because of incomplete data.
→ 94 patients have been included in the study
3. 74 females and 20 males
4. Mean age at the time of biopsy was 50 years (range 21 to 79 years old).
5. The histopathological assessment of the presence of lymphocytic foci in the labial salivary gland biopsies has been performed in a quantitative way
6. Statistical analyses: SPSS 21.0 for Windows, Pierson’s chisquared test

Result

Table 1. Results of the various diagnostic tests in 94 patients in whom a labial salivary gland biopsy has been performed.

		Positive SS (n=26)	Negative SS (n=68)	Total (n=94)
AECG Criteria (4)				
I. Ocular complaints	Positive	22	41	63
	Negative	4	27	31
II. Oral complaints	Positive	25	52	77
	Negative	1	16	17
III. Schirmer I test	Positive	18	23	41
	Negative	1	11	12
	Not performed	7	34	41
IV. Labial biopsy	Positive	24	6	30
	Negative	2	62	64
V. Salivary flow	Positive	2	6	8
	Negative	2	1	3
	Not performed	22	61	83
VI. Auto - antibodies	Positive	20	13	33
	Negative	5	35	40
	Not performed/ unclear	1	20	21

1. Sensitivity of the labial biopsy : 0.92
2. Specificity of the labial biopsy : 0.91
3. Positive predictive value : 0.80
4. Negative predictive value : 0.97

Table 2. The results of the various diagnostic tests in 20 patients with primary SS (#1-20) and two patients with secondary SS (#21 and 22).

Patient number	I. Ocular complaints	II. Oral complaints	III. Schirmer I test	IV. Labial biopsy	V. Salivary flow	VI. Auto-antibodies	M/F
1	positive	positive	positive	positive	negative	negative	F
2	positive	positive	-	positive	-	positive	F
3	positive	positive	positive	positive	-	positive	F
4	positive	positive	-	positive	-	positive	F
5	positive	positive	-	positive	-	positive	F
6	negative	positive	positive	positive	-	positive	F
7	positive	negative	positive	positive	-	positive	F
8	positive	positive	-	positive	-	positive	F
9	positive	positive	positive	positive	-	negative	F
10	positive	positive	positive	negative	-	positive	M
11	-	positive	negative	positive	positive	positive	M
12	positive	positive	positive	positive	-	positive	F
13	positive	positive	positive	positive	-	positive	F
14	positive	positive	positive	positive	-	positive	F
15	positive	positive	-	positive	negative	positive	F
16	negative	positive	positive	positive	-	positive	F
17	positive	positive	positive	positive	-	negative	M
18	positive	positive	positive	positive	-	positive	F
19	positive	positive	positive	positive	-	-	F
20	negative	positive	-	positive	positive	positive	F
21	positive	positive	positive	positive	-	-	M
22	positive	positive	positive	positive	--	-	F

- Not recorded or not performed.

1. Primary SS (n=20), secondary SS (n=2), 4 patients no reliable data to allow a distinction between primary and secondary SS.
2. 20 patients diagnosed with primary Sjögren’s syndrome:
 - a. Positive LSGB (n=19) → 80% positive serology (n=16)
 - b. Positive serology (n=16) → 1 negative LSGB
 - c. Negative serology (n=4) → 3 positive LSGB

Table 3. Result of labial salivary gland biopsy per department groups.

	Number of labial biopsies	Number of positive diagnosed SS patients (n=26)*	Number of negative diagnosed SS patients (n=68)
Rheumatology, Internal Medicine and Oral and Maxillofacial Surgery (only by request of the depts. of Rheumatology or Internal Medicine)	62	23 (37%)	39 (63%)
Other departments (not by the request of the depts. of Rheumatology or Internal medicine), e.g. Otolaryngology, Dermatology, Plastic surgery, Ophthalmology	32	3 (9%)	29 (91%)

*The difference in the percentages of positive biopsies between the two different department groups is significant ($p \leq 0.05$).

Discussion and Conclusion

1. The present study has been undertaken for two reasons:
 - 1st: examine in what way labial biopsies taken in our Institution in a 10-year period had contributed to the assessment of a diagnosis of Sjögren’s syndrome
 - 2nd: explore the influence of the recently proposed American College of Rheumatology(ACR,2012) criteria with regard to the role of the LSGB
2. 2% of patients experience long term postoperative complaints of numbness or hyperaesthesia at the site of the biopsy
3. Table 3. It seems advisable to perform LSGBs only after counseling of the patients by the departments of Rheumatology or Internal Medicine.
4. Table 1. high specificity(0.91) and a high sensitivity(0.92) of the LSGB with regard to the presence of SS
5. In postmortem studies older age was associated with high false-positive rates of LSGB. In view of a mean age of 50 years at the time of the biopsy, age does not seem to have a major influence on the present results.
6. In the past, it has been suggested that assessment of the various percentages of immunoglobulins in the plasma cells can be a criteria for the diagnosis of Sjögren’s syndrome. However, no other studies have been published and they have not been incorporated in the AECG classification nor in the ACR classification.
7. American College of Rheumatology(ACR, 2012) criteria, at least 2

Symptoms & signs	Explanation
Serologic findings	positive serum anti-SSA and/or anti-SSB
Ocular signs	ocular staining score >3
<u>Histopathology</u>	<u>Minor salivary glands, lymphocytes foci</u>

No significant difference between the diagnostic value of AECG and ACR

8. ACR classification
 - a. Serologic(+) and ocular test(+) → LSGB redundant
 - b. Serologic(-) or ocular test(-) → LSGB is indicated
 - c. Serologic test and the ocular test should be performed before taking a LSGB

題號	題目
1	<p>何者非 Sjögren's syndrome 常見的症狀?</p> <p>(A) Xerostomia(dry mouth)</p> <p>(B) Xerophthalmia (dry eyes)</p> <p>(C) Parotid gland enlargement</p> <p>(D) Otitis media</p>
<p>答案</p> <p>(D)</p>	<p>出處：Oral and maxillofacial pathology, 3rd 2009</p> <p>Sjögren's syndrome is a chronic, systemic autoimmune disorder involves salivary and lacrimal glands resulting in xerostomia(dry mouth), xerophthalmia (dry eyes)</p> <p>Fig. 11-25 Sjögren's syndrome. Benign lymphoepithelial lesion of the parotid gland.</p>
題號	題目
2	<p>下列何者非 Americam-European Consensus Group (AECG)定義診斷 Sjögren's syndrome 的 criteria 的項目?</p> <p>(A) Oral symptoms</p> <p>(B) Labial salivary gland biopsy</p> <p>(C) Immunohistochemical assessment of immunoglobulins in the plasma cells</p> <p>(D) Ocular signs</p>
<p>答案</p> <p>(C)</p>	<p>出處：Oral and maxillofacial pathology, 3rd 2009</p> <p>P468, Box 11-2</p>