Research Article

Coexisting primary Sjögren's syndrome and type II diabetes: Case report study

Nashwa M. Abd-Allah*, Amal A. Hassan*, Gihan Omar*, Mona Hamdy*, Sahar T. A. Abdelaziz, Waleed M. Abd El Hamid***, Rabab A.S. Mousa******

* Department of Rheumatology and Rehabilitation, Faculty of Medicine, Minia University,
** Department of Ophthalmology, Faculty of Medicine, Minia University,
*** Department of Clinical Pathology, Faculty of Medicine, Minia University,
**** Department of Pathology department, Faculty of Medicine, Minia University, Minia, Egypt.

Abstract
Herein, we describe a 38-year-old female diagnosed with histologically proven of primary Sjögren's syndrome and had type II diabetes. The differential diagnosis may be difficult, but this is not an exceptional case, which highlights the need to critically revise the consideration of diabetes as an exclusion for primary Sjögren's syndrome.
Keywords: Primary Sjögren's syndrome, type II diabetes, sicca symptoms.

Introduction
Sjögren's syndrome (SS) is a systemic autoimmune disease characterized by lymphocytic infiltrates of exocrine glands particularly salivary and lacrimal glands resulting in the clinical hallmarks of SS which are keratoconjuctivitis sicca and xerostomia or sicca complex. The term "keratoconjuctivitis sicca" is derived from Latin and its translation is dryness of the cornea and conjunctiva. Xerostomia refers to the subjective symptoms of dry mouth[1].

There are some reports of pSS associated with diabetes. Diabetic patients usually complaining of sicca symptoms similar to SS make confusion in diagnosis. Type II diabetes has been found to express autoimmune characteristics including the presence of autoantibodies against pancreatic beta cells and self-reactive T cells. The risk of type 2 diabetes after SS is 1.52[2].

Case report:
Thirty eight years old, known diabetic, female patient came to ophthalmology outpatient clinic complaining of foreign body sensation, burning, dryness and diminished lacrimation of both eyes for 8 months duration. She also had dry mouth for 1 year duration with no other sicca or non sicca symptoms.

On examination there were eye lid dermatitis, angular cheilitis, absent salivary pool in the mouth floor and dental caries. Ocular tests revealed Schirmer test < 5 mm, tear film breakup time (TBUT) 5-10 seconds and ocular staining score (OSS) ≥3.

Laboratory investigations showed elevated ESR (38 mm in 1st hour) and hyperglycemia (FBG= 400mg/dl). Serology was negative for rheumatoid factor (RF), antinuclear antibodies (ANA), anti SSA/Ro and anti SSB/La.

Histopathology of minor salivary gland shows a minor salivary glands specimen containing 6 glands with estimated glandular area: 20 mm² exhibiting focal lymphocytic sialadenitis (7 foci) with a focus score > 1, a histologic finding is compatible with SS (Figure 1). These findings combined with OSS ≥ are diagnostic for SS according to American College of Rheumatology (ACR) classification criteria 2012 for SS[3].

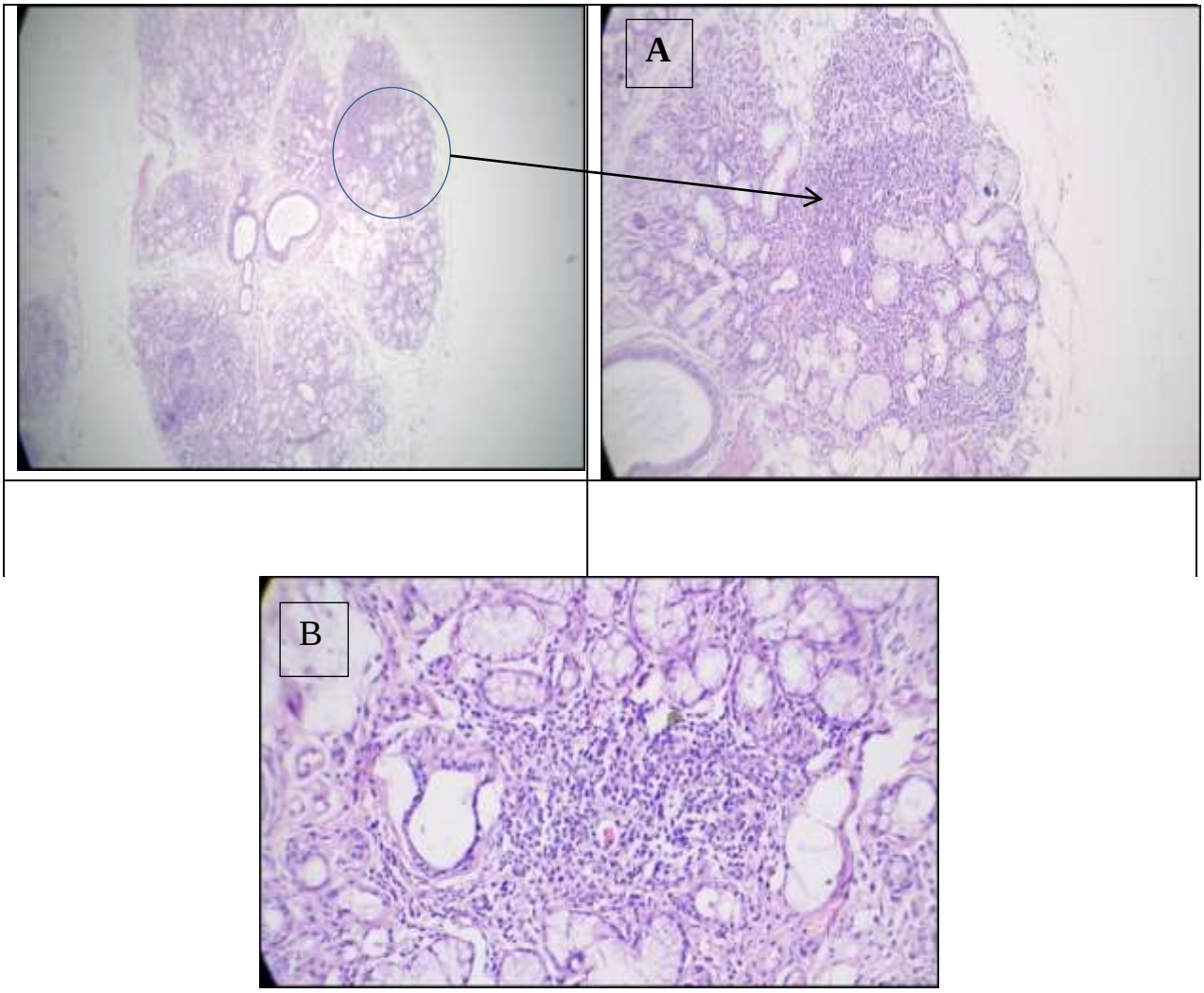

Figure 1. Heamatoxylin and eosin stained labial salivary glands (LSG) exhibting focal lymphocyti sialadenitis (FLS). **(A)** LSG show variously sized lymphocytic foci. The normal appearing acini immediately adjacent to the lymphocytic foci. The entire specimen had a focus score > 1 / 4mm^2 (original magnification × 100). **(B)** LSG show small lymphocytic aggregate that is minimally sized (> 50 cells) for inclusion in a focus score calculation (original magnification × 200).

Discussion

A case report study of Horita et al., reported a case of type 2 diabetes and pSS[4]. He is an old Japanese man aged 73 year and had history of diabetes. He had extra-glandular pulmonary symptoms diagnosed as pleural effusion. Patient's serology showed high ANA titer 1/320 and high titer of RF 1/160. Antibodies to Ro and La were negative. Schirmer test recorded only 1mm/5min. Rose bengal conjunctival staining was positive. Biopsy of the labial salivary gland showed FLS grade 3. Another study of Makimoto et al., reported a diabetic case diagnosed as pSS[5]. The patient is male aged 63 years. Beside sicca symptoms, he had bilateral pleural effusion and hypo-thyroidism. Serology was positive for RF, ANA (titer 1/320), anti Ro but anti La was negative. Biopsy showed atrophy of the salivary glands and infiltrations around the salivary glands consistent with SS.

In conclusion, the presence of diabetes doesn't exclude Sjögren's and SS should take in consideration in diabetic dry eye patients especially those resistant to treatment.

References

1. William E and Clair ST (2013): Sjögren's syndrome. In Kelley's text-book of rheumatology (eds Firestein GS). Elsevier pp 1169-91.
2. Hemminki K, Liu X, Försti A, et al., (2015): Subsequent type 2 diabetes in patients with autoimmune disease. Scientific reports; 5:13871.
3. Shiboski S, Shiboski C, Criswell L, et al., (2012): American College of Rheumatology classification criteria for Sjögren's syndrome: a data-driven, expert consensus approach in the Sjögren's International Collaborative Clinical Alliance cohort. Arthritis care & research; 64(4) 475-487.
4. Horita Y, Miyazaki M, Kadota J-i, et al., (2000): Type II diabetes mellitus and primary Sjögren's syndrome comp-licated by pleural effusion. Internal medicine; 39(11): 979-984.
5. Makimoto G, Asano M, Fujimoto N, et al., (2012): Bilateral pleural effusions as an initial presentation in primary Sjögren's syndrome. Case reports in rheumatology, 2012.