Evaluating the Usefulness of the Minor Salivary Gland Biopsy for the Diagnosis of Sjogren’s Syndrome

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Introduction

Sjögren Syndrome (SS) is a systemic chronic immune mediated inflammatory disorder first described by the Swedish physician Henrik Sjögren in 1933. Most individuals present with evidence of exocrine gland destruction. This results in sicca symptoms including xerophthalmia and xerostomia. Parotid gland enlargement can also be seen. In addition, numerous extraglandular features may develop such as arthralgias, arthritis, Raynaud phenomenon, pulmonary disease, neuropathy, vasculitis, and lymphoma.

The American College of Rheumatology endorses the American-European joint guidelines to establish the diagnosis of Sjögren’s Syndrome. There are six categories: (1) ocular symptoms of dryness, (2) ocular signs such as a positive schirmer’s test, (3) Oral symptoms of dryness, (4) oral signs elicited through salivary flow testing, (5) autoantibody testing, notably SSA and SSB, and (6) a minor salivary gland biopsy suggestive of Sjogren’s. Any of the four of the six criteria establish the diagnosis but either a positive biopsy or positive autoantibodies must be present. In current clinical practice, schirmer’s testing for ocular dryness and salivary flow testing are not commonly performed. The modified clinical diagnosis is based on sicca symptoms and findings, autoantibody testing, and minor salivary gland biopsy.

Although the labial minor salivary gland biopsy is a commonly used diagnostic tool for SS, several studies have questioned the utility based on the invasiveness of the procedure. There exists a wide variation in the rate of positive biopsies. Caporali, in Italy, in the largest retrospective review of these biopsies found a 32% positive rate. Bamba et al at the University of Chicago found a positive rate of 68%. Also, the reproducibility of this study has been questioned. Our data showed that there were no significant differences between the positive and negative biopsy groups in regards to sicca symptoms, joint pain, peripheral neuropathy, or other autoimmune conditions. There was a significantly higher percentage of patients with positive autoantibody tests in the positive biopsy group compared with the negative group. Ocular testing and salivary flow testing were not performed frequently at our institution.

Methods

This is a retrospective study. IRB approval was obtained from the review board prior to this study. The inclusion criteria for this study were simple. They included all minor salivary gland biopsies done to evaluate for Sjogren’s syndrome in the past five years at the Kaiser Oakland, Kaiser Richmond and Kaiser Redwood City medical centers. The patients were identified from the pathology computer logs. The goal of this study is to identify analyzable and clinical variables that may improve the effectiveness of this procedure.

Results

There were 34 total performed with 17 positive and 17 negative biopsies. There were no significant differences in the groups in regards to gender, xerophthalmia, xerostomia, other autoimmune conditions, joint pain, and paresthesia. There was a higher percentage of positive biopsies in patients with positive autoantibody testing. Ocular testing was only performed on two patients. Salivary flow testing was not performed at our institution. The data was then broken down into the three most common clinical pathways to a diagnosis of SS. They are illustrated as pathway #1 with positive sicca symptoms and positive autoantibody testing, pathway #2 with positive sicca symptoms and negative autoantibody testing, and pathway #3 with negative sicca symptoms (but other extraglandular symptoms such as joint pain were present to generate the referral) and positive autoantibody testing.

Discussion

Our data showed that there were no significant differences between the positive and negative biopsy groups based on demographics and clinical signs alone. Dividing them into our three pathways to a diagnosis of SS, there were 13 biopsies performed with a history of positive autoantibodies and positive joint pain. In these situations, the clinical and laboratory data are not conclusive. Therefore, in this study, 13 (38%) of patients underwent biopsies that were probably unnecessary. The usefulness of the biopsy in these situations is questionable given the fact that the other factors have already established a diagnosis. The minor salivary gland biopsy is most useful in the situations where the clinical and laboratory data is questionable. In the situation of positive sicca signs with negative autoantibody testing, the biopsy was positive in 33% of people. In the third group, with a history of extraglandular signs such as joint pain and positive autoantibody testing but an absence of distinct sicca symptom, 43% of the biopsies were positive. In these situations, where the clinical and laboratory data are not conclusive, a minor salivary gland biopsy is useful for establishing the diagnosis. Our data suggest that in the appropriate clinical setting the minor salivary gland biopsy is useful to aid in the diagnosis of Sjogren’s Syndrome as clinical symptoms alone are often not sufficient.

References


