Characterizing Restorative Dental Treatments of Sjögren's Syndrome Patients Using Electronic Dental Records Data

Zasim Siddiqui[a], Yue Wang[a], Payal Makkad[a], Thankam Thyvalikakath[a,b]

[a] Dental Informatics Core, Indiana University School of Dentistry, Indianapolis, Indiana, USA
[b] Center for Biomedical Informatics, Regenstrief Institute, Indianapolis, Indiana, USA

Abstract

Scant knowledge exists on the type of restorative treatments Sjögren's syndrome patients (SSP) receive in spite of their high dental disease burden due to hyposalivation. Increased adoption of electronic dental records (EDR) could help in leveraging information from these records to assess dental treatment outcomes in SSP. In this study, we evaluated the feasibility of using EDR to characterize the dental treatments SSP received and assess the longevity of implants in these patients. We identified 180 SSP in ten years of patients' data at the Indiana University School of Dentistry clinics. A total of 104 (57.77%) patients received restorative or endodontic treatments. Eleven patients received 23 implants with a survival rate of 87% at 40 months follow-up. We conclude that EDR data could be used for characterizing the treatments received by SSP and for assessing treatment outcomes.

Keywords

Dental Records; Sjögren's Syndrome; Dental Implants

Introduction

Sjögren's syndrome (SS) is a chronic autoimmune disorder of exocrine glands, particularly salivary and lacrimal gland, characterized by lymphocytic infiltration of affected gland resulting in the dryness of the mouth and eyes [13]. It is the second most common autoimmune connective tissue disease affecting up to 3.1 million Americans, with approximately 1 in every 70 people affected by primary SS [17; 23; 40]. It is common among middle aged people, with a high prevalence in females (female: male 9:1) [6]. It can occur alone as primary SS or in conjunction with other connective tissue diseases as secondary SS, such as rheumatoid arthritis (RA), systemic lupus erythematous, and systemic sclerosis [13; 16; 37]. Both primary and secondary SS have similar pathophysiology, signs, and symptoms [7]. The exact etiopathogenesis of SS is unclear and considered to be multifactorial. However, its etiology has been associated with endocrine, genetic and viral factors and alteration in the regulation of cell apoptosis [20; 24; 28]. At present, SS is an incurable disease with symptomatic management options. However recent evidence showed effectiveness of early immunomodulation in limiting disease progression among SS patients [33].

SS patients experience a high caries risk due to reduced salivary flow leading to premature tooth loss despite maintaining good oral health, visiting dentists more frequently, using fluoridated toothpaste, and having more awareness about their disease and oral health [3; 4; 10; 31; 38]. Hyposalivation and early loss of teeth significantly interfere with the individual's normal oral functions such as speaking, chewing, and swallowing thereby compromising their physical, social and emotional quality of life [5; 15; 26; 29]. These patients often require costly, early life restorative treatments to maintain normal oral functions due to tooth loss [8; 38]. Despite their huge dental disease burden, limited studies exist characterizing SS patients' oral health and dental treatments.

To date, most of the knowledge on SS treatments comes through surveys and interviews on patients' experiences and challenges with maintaining good oral health and receiving dental treatments [2; 10]. Results from these studies report SS patients have difficulties with maintaining good oral health, high caries risk and incompatibility with tooth/tissue supported prosthesis due to mucosal dryness. These challenges with maintaining good oral health highlight the need for clinical research. Very few clinical studies have investigated SS patients' oral health or outcomes from dental treatments received. Lately, implant retained prosthesis are heralded as the treatment of choice to replace lost teeth in patients with SS. However, implant success are inconclusive as they are either case reports or studies with small sample size [9; 19; 30].

The historic use of paper-based records in dental clinics makes retrospective studies of SS difficult. Also, challenges with identifying and confirming SS diagnosis and associated comorbidities were a major barrier in performing clinical studies. The increased adoption of EDR in dental practices offers an opportunity to study the outcomes of various dental treatments among SS patients using EDR data [34].

Studies have shown increased adoption of EDR in both private and academic dental settings in United States and this trend is expected to continue in the future [32; 34; 35]. This trend echoes with the EDR adoption and computer usage in dental practices in other countries including Canada, China, UK, and Brazil [1; 12; 18; 21]. Most countries showing high adoption of EDR used it not only for administrative purposes but also for patient care documentation at the point of care. However, few countries have also shown to be using EDR and computers in dental offices mainly for administrative purposes [1; 12]. In US academic settings, more than 90% of the dental schools document patient care using EDR [32].

Approximately 76% of US independent and group practices use EDR for patient care documentation in 2013 [34] compared to 48% of physician offices having a basic Electronic Health Record in 2014 [14]. Thus, EDRs are a potential data source for clinical research. In this study, we demonstrate the use of EDR data in evaluating dental treatment procedures and outcome for SS patients.

The objective of this study was to determine the feasibility of using EDR to characterize the restorative and endodontic dental treatments for SS and assess the longevity of dental implants placed in patients with SS at the Indiana University School of Dentistry clinics. The long-term objective of our

research is to advance our knowledge of SS and to develop best practice guidelines toward improved oral health and quality of life.

Methods

This study was approved by IRB 1611054551. We retrieved a limited data set of patients seen between January 1, 2005 and October 31, 2016 by performing keyword search for the term "Sjogren" in the EDR. We identified 270 records that contained the term "Sjogren" in the medical history forms, progress/clinical notes, specialty and medical consultation forms, caries risk assessment, and management forms. We used keyword search to identify patients diagnosed with SS as patient's medical and medication histories are typically documented in free-text format or within progress notes.

Two trained dental researchers manually reviewed the clinical notes to identify patients who reported having SS. Unambiguous records stating patients diagnosed with SS were included whereas records only mentioning "Sjogren" as a suspected disease, differential diagnosis, or family history etc. were excluded. Disagreements between researchers were discussed and resolved through consensus.

Next, we retrieved the treatment history of these patients using Current Dental Terminology (CDT) [27; 36] codes that are routinely used to document dental procedures performed in dental practices. We identified the CDT codes related to restorative and endodontic treatment procedures and grouped them into five major treatment types (Table 1). Treatment types were further divided into treatment procedures based on the types of materials (resin-based composite, amalgam), location (maxillary, mandibular), and extent (partial, complete) (Table 2). Each treatment procedure included CDT codes representing that procedure type. For instance, Resin-based composite contains codes: D2330 - D2335 and D2390 - D2394. Amalgam restoration contains codes: D2140, D2150, D2160, and D2161.

We performed descriptive statistics on demographics and treatments. Life tables were constructed to assess survival rates of implant procedures. Two researchers also manually reviewed clinical notes of failed implant records to detect reasons for implant failure.

Results

A total of 180/270 patients were identified with SS. Among them, 165 (91.6%) were female, 11 (6.66%) male, and four patients (2.22%) did not report their gender. These patients had a mean age of 63.75 years (standard error: 1.06 years) with 160 (88.89%) of them being 45 years or older. Among the patients who reported ethnicity, 100 (55.56%) were Caucasian and 13 (7.2%) were African Americans. 61 (33.89%) patients did not report their ethnicity. Only 75 (41.66 %) patients had dental insurance. 120 patients (67%) had a follow-up visit of more than one year. The average follow-up was 5.23 years (SE: 0.32 years).

Tables 1 and 2 show the distribution of treatment types and treatment procedures, respectively. 104 (57.77%) patients received restorative and/or endodontics treatment. These patients received 1,085 different restorative and/or endodontics treatments while the remaining patients received oral examinations with diagnostic procedures, prophylaxis treatment, periodontal therapy, or surgical treatment including tooth extraction. Most common restorative procedures were resin-based composite and amalgam restorations followed by fixed partial denture procedures. 24 patients received 41

complete or partial dentures and 25 patients received 33 endodontic treatments. The mean patient age for patients receiving partial dentures, complete dentures, and endodontic treatments was 66.87, 62.14, and 58.78 years, respectively.

Table 1– Number of treatments received by SS patients

Treatment Types	Number of patients (%)*	Number of Procedures	Mean Patient age (SE)
Restorative	90 (50)	866	61.22 (0.42)
Fixed	41 (22.8)	122	62.61 (0.80)
Partial denture	24 (13.3)	41	66.87 (1.12)
Denture	11(6.1)	23	62.41 (2.19)
Implants	25 (13.9)	33	58.78 (1.88)

*Number of patients >104 due to multiple treatments.

Table 2 - Distribution of treatment procedures in Sjögren's syndrome patients

Treatment Procedures	Number of procedures (%)
Restorative treatment	
Resin-based composite restoration	654 (60.28)
Amalgam restoration	150 (13.82)
Post and core	61 (5.62)
Inlay and onlay	1 (0.09)
Total	**866 (79.82)**
Fixed partial denture (FPD)	**122 (11.24)**
Denture	
Complete denture - maxillary	14 (1.29)
Complete denture - mandibular	4 (0.37)
Partial denture- maxillary	7 (0.65)
Partial denture -mandibular	15 (1.38)
Overdenture	1 (0.09)
Total	**41 (3.78)**
Implants	
Implants - maxillary	12 (1.11)
Implants - mandibular	11 (1.01)
Total	**23 (2.12)**
Endodontics treatment	
Root canal treatment (RCT)	27 (2.49)
Retreatment of RCT tooth	4 (0.37)
Apicoectomy	1 (0.09)
Therapeutic pulpotomy	1 (0.09)
Total	**33 (3.04)**
Total treatment procedures	**1,085 (100.0)**

Eleven patients received 23 implant treatments, 12 (52.17%) in the maxilla and 11 (47.82%) in the mandible. As shown in Table 3, three implants failed (two in maxilla and one in mandible) making survival rate of approximately 87% during an average follow-up period of 40 months. These 3 implants failed in two patients in the second and fifth months after placement. One implant failed due to osseointegration and the

remaining two were removed due to mobility (lack of osseointegration), erythema of surrounding implant area, vertical bone loss and horizontal ridge deficiency after implant placement. All three implants were lost in the preloading phase. Survival rate of loaded implants was 100%. Table 3 demonstrates the survival rate of implants.

Table 3 - Life table analysis for implants showing the time interval in years, number of implants that existed during these time intervals (N), number of failures (NF), replaced implants (RI), failure rate (Failure%) and survival rate (% S)during this interval, and cumulative survival rate (Cum %%)

Years	N	NF	RI	% Failure	% S	Cum % S
0-1	23	3	0	13.04	86.96	86.96
1-2	18	-	-	0.0	100.0	86.96
2-3	8	-	-	0.0	100.0	86.96
3-4	8	-	-	0.0	100.0	86.96
4-5	8	-	-	0.0	100.0	86.96
5-6	7	-	-	0.0	100.0	86.96
6-7	6	-	-	0.0	100.0	86.96
7-8	5	-	-	0.0	100.0	86.96
>8	5	-	-	0.0	100	86.96

Discussion

In this study, we attempted to assess the feasibility of using EDR data to characterize the dental treatments for SS patients and determine the longevity of dental implants placed. Study results indicate that EDR data could be utilized to characterize the dental treatments in SS patients and assess the effectiveness of these treatments in restoring their oral functions. To the best of our knowledge, this is the first study of dental treatments among SS patients using EDR data. We identified 50% of SS patients received dental restorations using materials such as composite resin and amalgam. Approximately 14% of patients received endodontic treatments to treat infection/disease involving dental pulp. Implants are emerging as a popular alternative to restore lost teeth due to difficulty with tolerating removable denture prosthesis as a result of mucosal dryness. However, we identified only 11 (6.11%) patients who received implant treatment. The high cost and limited coverage of implant treatments under dental insurance in the United States could be reasons for this small number of implant placement. In addition, many of these patients already have high medical expenses due to associated comorbid conditions [38]. We found the average number of dental visits for patients with more than one year of follow-up to be 5.23 visits/year, which is higher than the 4 visits/year reported in a previous study on primary SS patients in the United States [38]. A high number of dental visits with only 41.66% of patients having dental insurance indicated that SS patients incur high dental expenses.

In this study, three implants failed in two patients. The failure rate of approximately 13% on 40 months of follow-up is higher than all the previous studies in SS patients except for the 1999 case review series by Isidor et al. in Denmark, which reported failure rate of 16.7% on 48 months of follow up[9; 11; 19; 22]. Implant failure rate in SS patients is high compared to the 98% success of implants in medically health patients on 10 years of follow-up [22; 25; 39]. Curiously, the

three dental implants that had failed were all during the preloading phase, whether such a trend can be substantiated warrants further investigation. Furthermore, future research in this area is needed to evaluate the impact of different risk factors such as immunosuppressant therapy, associated comorbid conditions and smoking on the implants survival in SS patients.

Several limitations exist within our study. First, SS patients were identified by extracting information from the EDR using the term, "Sjogren". Patients with Sjögren's syndrome whose disease was documented using other lexical variations such as "SjS" would not be identified using our extraction method. Second, Sjogren documentation was based upon patient self-reported data, the reliability of patient self-reported Sjögren's syndrome have not been evaluated. Third, the survival rate of implants within our study was based on 23 implant treatments. The failure rate could be exaggerated due to the limited number of implants.

Future work would be to expand our terminology to include other terms for Sjögren's syndrome. Additionally, we identified the Sjögren's syndrome patient population using information within the EDR. To expand our cohort, we could also use other sources such as medical records for identifying dental patients with Sjögren's syndrome. Furthermore, the accuracy of patient self-reported Sjögren's syndrome needs to be validated with other sources such as medical records.

Conclusion

EDR data could be used for identifying treatments received by SS patients and assessing outcomes. However, further studies are required to evaluate the impact of confounding variables on the outcome of these treatments. Such studies will facilitate developing best practice guidelines to improve oral health among these patients.

Acknowledgements

References

[1]   R. Abramovicz-Finkelsztain, C.G. Barsottini, and H.F. Marin, Electronic Dental Records System Adoption, Stud Health Technol Inform 216 (2015), 17-20.

[2]   K. Albrecht, J. Callhoff, G. Westhoff, T. Dietrich, T. Dorner, and A. Zink, The Prevalence of Dental Implants and Related Factors in Patients with Sjogren Syndrome: Results from a Cohort Study, J Rheumatol 43 (2016), 1380-1385.

[3]   J.C. Atkinson and P.C. Fox, Sjogren's syndrome: oral and dental considerations, J Am Dent Assoc 124 (1993), 74-76, 78-82,84-76.

[4]   M. Baudet-Pommel, E. Albuisson, J.L. Kemeny, F. Falvard,J.M. Ristori, M.P. Fraysse, and B. Sauvezie, Early dental loss in Sjogren's syndrome. Histologic correlates. European Community Study Group on Diagnostic Criteria for Sjogren's Syndrome (EEC COMAC), Oral Surg Oral Med Oral Pathol 78 (1994), 181-186.

[5]   P.P. Binon, Thirteen-year follow-up of a mandibular implant- supported fixed complete denture in a patient with Sjogren's syndrome: a clinical report, J Prosthet Dent 94 (2005), 409-413.

[6]   S.J. Bowman, G.H. Ibrahim, G. Holmes, J. Hamburger, and J.R. Ainsworth, Estimating the prevalence among Caucasian women of primary Sjogren's syndrome in two general practices in Birmingham, UK, Scand J Rheumatol 33 (2004), 39-43.

[7]   A.J. Carr, W.F. Ng, F. Figueiredo, R.I. Macleod, M. Greenwood, and K. Staines, Sjogren's syndrome - an update for dental practitioners, Br Dent J 213 (2012), 353-357.

[8] S. Carsons, A review and update of Sjogren's syndrome: manifestations, diagnosis, and treatment, Am J Manag Care 7 (2001), S433-443.

[9] K. Chochlidakis, C. Ercoli, and S. Elad, Challenges in implant-supported dental treatment in patients with Sjogren's syndrome: A case report and literature review, Quintessence Int 47 (2016), 515-524.

[10] L.B. Christensen, P.E. Petersen, J.J. Thorn, and M. Schiodt, Dental caries and dental health behavior of patients with primary Sjogren syndrome, Acta Odontol Scand 59 (2001), 116-120.

[11] M. de Mendonca Invernici, A. Finger Stadler, G. Vale Nicolau, M.A. Naval Machado, A.A. Soares de Lima, and M. Compagnoni Martins, Management of Sjogren's Syndrome Patient: A Case Report of Prosthetic Rehabilitation with 6-Year Follow-Up, Case Rep Dent 2014 (2014), 761251.

[12] C. Flores-Mir, N.G. Palmer, H.C. Northcott, F. Khurshed, and P.W. Major, Perceptions and attitudes of Canadian dentists toward digital and electronic technologies, J Can Dent Assoc 72 (2006), 243.

[13] R.I. Fox, Sjogren's syndrome, Lancet 366 (2005), 321-331. [14]M.F. Furukawa, J. King, V. Patel, C.J. Hsiao, J. Adler- Milstein, and A.K. Jha, Despite substantial progress In her adoption, health information exchange and patient engagement remain low in office settings, Health Aff (Millwood) 33 (2014), 1672-1679.

[15] M. Gandia, E.M. Morales-Espinoza, R.M. Martin-Gonzalez, S. Retamozo, B. Kostov, R. Belenguer-Prieto, D. Buss, M. Caballero, A. Bove, H. Gueitasi, P. Brito-Zeron, A. Siso- Almirall, M.J. Soto-Cardenas, and M. Ramos-Casals, Factors influencing dry mouth in patients with primary Sjogren syndrome: usefulness of the ESSPRI index, Oral Health Dent Manag 13 (2014), 402-407.

[16] A. Hajiabbasi, I. Shenavar Masooleh, Y. Alizadeh, A.S. Banikarimi, and P. Ghavidel Parsa, Secondary Sjogren's Syndrome in 83 Patients With Rheumatoid Arthritis, Acta Med Iran 54 (2016), 448-453.

[17] C.G. Helmick, D.T. Felson, R.C. Lawrence, S. Gabriel, R. Hirsch, C.K. Kwoh, M.H. Liang, H.M. Kremers, M.D. Mayes, P.A. Merkel, S.R. Pillemer, J.D. Reveille, and J.H. Stone, Estimates of the prevalence of arthritis and other rheumatic conditions in the United States. Part I, Arthritis Rheum 58 (2008), 15-25.

[18] J. Hu, H. Yu, E. Luo, E. Song, X. Xu, H. Tan, and Y. Wang, Are Chinese dentists ready for the computerization of dentistry? A population investigation of China's metropolises, J Am Med Inform Assoc 16 (2009), 409-412.

[19] F. Isidor, K. Brondum, H.J. Hansen, J. Jensen, and S. Sindet- Pedersen, Outcome of treatment with implant-retained dental prostheses in patients with Sjogren syndrome, Int J Oral Maxillofac Implants 14 (1999), 736-743.

[20] M. Ittah, C. Miceli-Richard, J. Eric Gottenberg, F. Lavie, T. Lazure, N. Ba, J. Sellam, C. Lepajolec, and X. Mariette, B cell- activating factor of the tumor necrosis factor family (BAFF) is expressed under stimulation by interferon in salivary gland epithelial cells in primary Sjogren's syndrome, Arthritis Res Ther 8 (2006), R51.

[21] J.H. John, D. Thomas, and D. Richards, Questionnaire survey on the use of computerisation in dental practices across the Thames Valley Region, Br Dent J 195 (2003), 585-590;discussion 579.

[22] A. Korfage, G.M. Raghoebar, S. Arends, P.M. Meiners, A. Visser, F.G. Kroese, H. Bootsma, and A. Vissink, Dental Implants in Patients with Sjogren's Syndrome, Clin Implant Dent Relat Res (2015).

[23] R.C. Lawrence, D.T. Felson, C.G. Helmick, L.M. Arnold, H. Choi, R.A. Deyo, S. Gabriel, R. Hirsch, M.C. Hochberg, G.G. Hunder, J.M. Jordan, J.N. Katz, H.M. Kremers, and F. Wolfe, Estimates of the prevalence of arthritis and other rheumatic conditions in the United States. Part II, Arthritis Rheum 58 (2008), 26-35.

[24] M. Margaix-Munoz, J.V. Bagan, R. Poveda, Y. Jimenez, and G. Sarrion, Sjogren's syndrome of the oral cavity. Review and update, Med Oral Patol Oral Cir Bucal 14 (2009), E325-330.

[25] H.J. Meijer, G.M. Raghoebar, R.H. Batenburg, and A. Vissink, Mandibular overdentures supported by two Branemark, IMZ or ITI implants: a ten-year prospective randomized study, J Clin Periodontol 36 (2009), 799-806.

[26] J.M. Meijer, P.M. Meiners, J.J. Huddleston Slater, F.K. Spijkervet, C.G. Kallenberg, A. Vissink, and H. Bootsma, Health- related quality of life, employment and disability in patients with Sjogren's syndrome, Rheumatology (Oxford) 48 (2009), 1077-1082.

[27] G. Melenyk, The updated ADA CDT manual, J Mich Dent Assoc 90 (2008), 26.

[28] H. Nakamura, A. Kawakami, and K. Eguchi, Mechanisms of autoantibody production and the relationship between autoantibodies and the clinical manifestations in Sjogren's syndrome, Transl Res 148 (2006), 281-288.

[29] J.J. Napenas and T.S. Rouleau, Oral complications of Sjogren's syndrome, Oral Maxillofac Surg Clin North Am 26 (2014), 55-62. [30]A.G. Payne, J.F. Lownie, and W.J. Van Der Linden, Implant-supported prostheses in patients with Sjogren's syndrome: a clinical report on three patients, Int J Oral Maxillofac Implants 12 (1997), 679-685.

Address for correspondence

Zasim Siddiqui, BDS, MS

Indiana University School of Dentistry, 1121 W Michigan Street, Room DS 267, Indianapolis, IN 46202, USA

Email: zasiddiq@iupui.edu Phone: +1-317-274-7130.

Preferred method of contact is through email.