

# Implant Treatment in Patients with Sjogren's Syndrome: A Review of the Literature and Two Clinical Case Reports

## Keywords

Sjogren's Syndrome  
Xerostomia  
Implant Survival  
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## ABSTRACT

Implant rehabilitation in patients with Sjogren's Syndrome is reported to offer an improvement in the quality of life, but the scientific evidence on implant survival in patients with Sjogren's Syndrome is scarce. The paper presents a review of the literature on the performance of dental implants in patients with Sjogren's Syndrome and two case reports of patients with Sjogren's Syndrome treated successfully with dental implants. Two female patients suffering from Sjogren's Syndrome were rehabilitated with implant-supported prostheses. After eighteen months and two years respectively, the patients were satisfied with function and aesthetics: restorations were comfortable, stable radiographic bone levels were noted and xerostomia was not reported to affect the function anymore.

## INTRODUCTION

Sjogren's syndrome (SS) is a chronic autoimmune disease affecting the exocrine glands, primarily the lacrimal and salivary glands.<sup>1</sup> SS can be divided into primary, in which ocular and oral manifestations are the only symptoms and secondary in which there is associated autoimmune disease.<sup>2</sup> Genetic and exogenous factors have been considered as triggers of SS, but the aetiology of SS remains unknown.<sup>1,2</sup> Management of SS is challenging since there is no cure and treatment is only palliative.<sup>3</sup>

Xerostomia is the primary oral manifestation of SS which can increase the risk of caries, oral infections and intolerance to removable prosthesis.<sup>3,4,5</sup> In edentulous patients, replacement with a removable denture can cause soreness and discomfort due to xerostomia and friction.<sup>3,4,5</sup> Furthermore, lack of adequate saliva would be detrimental to the retention of the denture.<sup>6</sup>

In partially dentate patients, apart from the above-mentioned problems with a removable option, replacement of missing teeth with a conventional fixed prosthesis is not always feasible depending on the status of the remaining teeth and can lead to increased risk of caries.<sup>4,5</sup> In such cases, implant rehabilitation can be a viable treatment option provided that individualised maintenance will be followed.<sup>7,8,9,10</sup> Although implant supported prostheses are used to improve function and comfort in patients suffering from SS, the scientific evidence on implant survival in patients with SS is scarce.<sup>7,8,9,10</sup>

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The aim of this study is to systematically review the literature on performance of dental implants in patients with SS and present two case reports of patients with SS treated with dental implants.

## LITERATURE REVIEW

### SEARCH STRATEGY:

A PICO type question, “In patients suffering from Sjogren’s Syndrome and treated with dental implants, is there an increased risk of implant failure?” was formulated. The search strategy including a comprehensive and systematic electronic search of both the MEDLINE–Pubmed database and the Cochrane Database was conducted for articles published in English between 1984 and November 2014 in the dental literature. The search strategy was conducted in November 2014 and repeated in April 2015 and was complemented by manual searches of the articles selected from the electronic search. The process was supported by a specialised librarian.

The following key words were used: (Sjögren’s syndrome OR Sjögren OR xerostomia OR dry mouth AND implants (dental OR oral)).The inclusion criteria were SS subjects treated with dental implants, the study reports implant failure, survival or success and the study includes at least 5 subjects and 1year follow-up.

Both the review authors independently assessed the titles and abstracts of the search results. The full text of all studies of possible relevance was obtained for independent assessment of the stated inclusion criteria. The methodological qual-

ity was evaluated using the levels of evidence proposed by the Oxford Centre for Evidence-based Medicine ranging from lowest (level 5, expert opinion without explicit critical appraisal, or based on physiology, bench research, or first principles) to highest (level 1a, systematic reviews with homogeneity of randomized clinical trials).<sup>11</sup>

### SEARCH RESULTS:

In the first original search, 75 articles were identified and after the title search, 6 abstracts were obtained (Isidor *et al* 1999, Spinato *et al* 2010, Binon *et al* 2005, Payne 1997, Borstein *et al* 2009, Candel-Marti *et al* 2011) and the full-text articles were evaluated.<sup>7,8,9,10,12,13</sup> Additional studies were sought by scanning the references cited in the retained papers. In the second original search no additional studies were identified for inclusion.

No clinical trials were identified. One case series study with eight patients<sup>7</sup> and three case reports (level of evidence 5)<sup>8,9,10</sup> were identified in the literature. These case reports were excluded due to the small number of patients or short follow-up period. The two review papers were also excluded since they offered no additional data for inclusion.<sup>12,13</sup>

In the Isidor *et al* study, 8 female patients received 54 machined surface implants (18 in the maxilla, 36 in the mandible). 4 out of 8 patients lost at least 1 implant. 7 implants failed to osseointegrate at the time of the abutment connection (5 in the maxilla and 2 in the mandible) and 2 additional implants were lost in the mandible after the first year of function reporting an implant failure rate of 16.7%.

**Table 1. Studies that were identified reporting implant survival in patients with Sjogren’s syndrome**

	Number of patients	Number of implants	Number of failed implants	Early implant failure	Delayed implant failure
<b>Included Studies</b>					
<b>Isidor et al 1999</b>	8	54 Maxilla: 18 Mandible:36	9 (16.7%)	Maxilla: 5 Mandible:2	Mandible: 2
<b>Excluded Studies</b>					
<b>Payne et al 1997</b>	3	20 Maxilla: 14 Mandible: 6	2 (10%)	Maxilla: 1 Mandible: 1	0
<b>Spinato et al 2010</b>	1	Mandible: 6	0	0	0
<b>Binon 2005</b>	1	Mandible: 6	0	0	0

## CONCLUSIONS:

Implant survival in patients with SS lacks a high level of evidence. Nevertheless, the patients reported dramatic improvement in the quality of life after implant rehabilitation<sup>7,8,9,10</sup> and therefore, offers these patients a possibility to restore oral function and aesthetics.

The following clinical reports present two patients with SS treated with dental implants and the key stages of implant rehabilitation will be discussed.

## CLINICAL REPORT 1

A 51-year-old female suffering from secondary SS presented complaining of soreness and discomfort from her dentures. She reported extraction of her teeth gradually over a 10 year period. She received an acrylic mucosa-borne partial denture after loss of anterior teeth, but reported inability to wear it. Intraoral and radiographic examination confirmed xerostomia, caries in the three remaining maxillary teeth (maxillary right canine, maxillary left canine and maxillary left first premolar), alveolar width deficiency in the edentulous areas and a fully edentulous mandible (Figure 1 and 2).



**Figure 1:** Caries in the maxillary remaining teeth

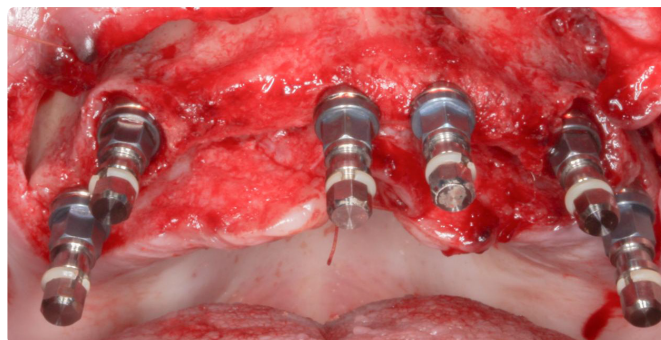


**Figure 2:** Edentulous mandible

The treatment plan was to provide implant retained fixed prostheses to replace the maxillary teeth and an implant retained overdenture to restore the mandible. Implants were more likely to result in an outcome that would restore this patient with a comfortable and functional dentition in comparison to maxillary and mandibular conventional dentures. Radiographic and clinical assessment confirmed that a fixed prosthesis for the maxilla and an overdenture for the mandible were feasible.

The patient received six hydrophilic Straumann® SLActive tissue level implants (Institut Straumann AG, Basel, Switzerland) in the maxilla (bilaterally in the central incisor, canine, and first premolar area) and two intra-foraminal hydrophilic Straumann® SLActive tissue level implants in the mandible.

Implant assessment was based on the SAC assessment tool<sup>14</sup> and preoperative treatment planning involved tooth set ups, diagnostic wax-ups and cone-beam computed tomography with a radiographic stent.<sup>15</sup> Type II implant placement surgery was performed (Figure 3).<sup>16,17,18</sup>



**Figure 3:** Type II implant placement with GBR

After 3 months of uneventful healing period, tooth set ups and full contour wax ups were used to guide the dental technician during the construction of the framework and the definitive restorations.<sup>19,20,21</sup> The maxillary framework was scanned with CS2 scanner and the Cares Visual software (Straumann® Cares® System 8.0, Institut Straumann AG, Basel, Switzerland) using a copy-mill technique and were milled in cobalt-chromium alloy (Coron®).<sup>22,23</sup>

The maxillary framework was designed as screw-retained prosthesis, except in the maxillary right canine area. The screw access in the maxillary right canine was labially positioned and therefore the wax framework in this area was customised to receive an individual cement-retained crown (Figure 4 and 5). GC Initial MC metal ceramic (GC America, Alsip, IL) was applied to the framework to full contour followed by application of pink coloured ceramic to mimic the soft tissues.<sup>24</sup>



**Figure 4:** Labial view of the definitive prosthesis before cementation of the individual crown



**Figure 5:** Occlusal view of the definitive prosthesis before cementation of the individual crown

The definitive prosthesis was screwed and tightened to 35Ncm on each implant and the screw access holes were sealed with composite restorative material. Subsequently, the crown for the maxillary right canine was cemented over the customised abutments using soft temporary cement (Temp-Bond®, Kerr Dental, Orange, CA) (Figure 6).<sup>25,26</sup>



**Figure 6:** Maxillary definitive prosthesis after cementation of the individual crown

In the lower arch, an implant retained overdenture was constructed to restore the edentulous mandible. A cobalt-chrome lingual veneer was incorporated in the overdenture to prevent the incidence of fracture.<sup>27</sup> Individual locator attachments were selected since they are cost-effective and offer similar retention to other type of individual attachments or bars and the complications associated with them can be easily resolved chairside (Figure 7 and 8).<sup>28,29</sup>



**Figure 7:** Locator® abutments in mandible



**Figure 8:** Immediate post-op view of maxillary and mandibular definitive prostheses

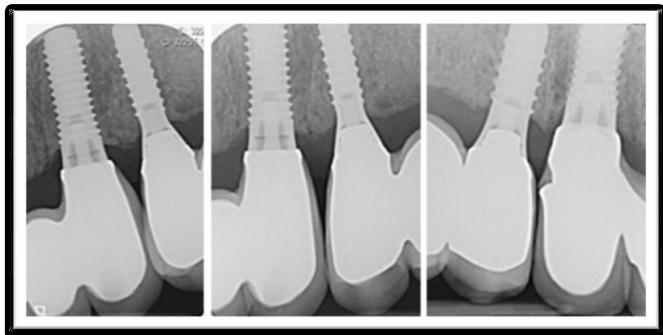
During the 8-week review appointment, peri-implant mucositis was noticed in the maxilla due to inadequate plaque removal (Figure 9). Xerostomia seemed to promote plaque adherence to the prosthesis. Healthy peri-implant tissues were however re-established after individualised maintenance and oral hygiene regime was reinforced (Figure 10).<sup>30</sup> After 18 months the patient was satisfied with function and aesthetics; restorations were comfortable, stable bone levels were noted on radiographs and xerostomia was not reported to affect the function anymore (Figure 11).



**Figure 9:** Peri-mucositis in maxillary implants at 8-week review



**Figure 10:** Healthy peri-implant tissues re-established after individualised maintenance at 6 month review



**Figure 11:** Radiographs at 18-months review showing well maintained bone levels

## CLINICAL REPORT 2

A 53-year-old female with primary SS presented complaining of difficulty in biting with her anterior teeth and dry mouth. She reported that her teeth started to breakdown over a 3-year period. Apart from xerostomia, she experienced altered taste and a high cariogenic diet was used to compensate for that. Intraoral and radiographic examination confirmed xerostomia, tooth surface loss and failing restorations in the mandibular anterior teeth due to caries. The lower right lateral incisor was a retained root (*Figure 12 and 13*).



**Figure 12:** Preoperative view showing caries and failing restorations in mandibular anterior teeth

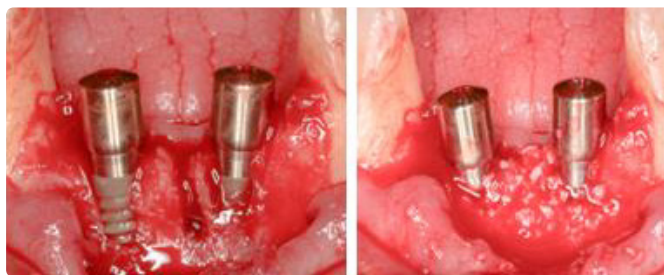


**Figure 13:** Pre-operative radiographs

Due to significantly reduced height of coronal dentine the lower right lateral incisor and the lower central incisors were considered at a high risk of prosthetic failure.<sup>31,32</sup> They were extracted and planned for replacement with an implant-retained prosthesis. Implants were considered as an ideal option considering the width of the edentulous span and the compromised quality of the abutments would not render them successful for a conventional fixed prosthesis. An interim removable denture caused soreness and discomfort and the patient was unable to tolerate it.

The lower left canine was restored since adequate tooth structure was present for a ferrule and a post/crown ration of 1:1.<sup>31,32</sup> The implant assessment and preoperative treatment planning was based on the SAC assessment tool,<sup>14</sup> diagnostic wax-ups and cone-beam computed tomography.<sup>15</sup>

The patient received two hydrophilic Straumann® SLActive tissue level implants in the mandible (right lateral incisor, left central incisor) to provide an implant-supported fixed prosthesis replacing the mandibular central incisors and the mandibular right lateral incisor. Type II implant placement surgery with simultaneous guided bone regeneration was performed to regenerate the missing labial bone and augment ridge contour in the pontic areas with deproteinized bovine bone and porcine collagen membrane (Geistlich BioOss® and BioGide®, Geistlich Pharma AG, Luzern, Switzerland) (*Figure 14*).<sup>16,17,18</sup>



**Figure 14:** Type II implant placement with GBR

After an uneventful healing period of 3 months, full contour wax ups and provisional restorations were used to shape the soft tissues and the emergence profile, determine aesthetics and occlusion to guide the dental technician during construction of the framework and definitive restorations (*Figure 15*).<sup>19,20,21</sup>



**Figure 15:** Metal-acrylic implant provisional bridge

Both abutment screws were emerging through the incisal edges thereby requiring a cement retained partial fixed dental prosthesis. The titanium abutments and the framework were scanned with CS2 scanner and the Cares Visual software using a copy-mill technique and were milled in titanium (Straumann® CARES®, Ti) and zirconium (zerion® HT) alloys respectively.<sup>23</sup> E-max veneering ceramic was applied to the framework to full contour. The abutments were screwed and tightened to 35Ncm on each implant. The labial screw access holes were sealed and the prosthesis was cemented over the abutments using soft temporary cement (TempBond®).<sup>25,26</sup>

At 2-year review the patient was satisfied with the functional and aesthetic outcome; xerostomia not adversely affecting function or comfort anymore and stable bone levels were present radiographically (Figure 16 and 17).



**Figure 16:** Definitive mandibular prosthesis and healthy gingival tissues at 2 year review



**Figure 17:** Radiographs at 2 year review showing stable bone levels

## DISCUSSION

The systematic review revealed that the long-term prognosis of implant survival in patients with SS lacks evidence. Nevertheless, patients reported dramatic improvement in function and comfort after implant rehabilitation<sup>7,8,9,10</sup>. Three cases reports and one case series with eight patients were identified in the literature<sup>7,8,9,10</sup>, and the study with the largest sample

reported 16% implant failure rate.<sup>7</sup> However, the sample was too small to draw meaningful conclusions.<sup>7,33,34</sup> Nonetheless, the high failure rate could be attributed to the machined (smooth) implant surfaces.<sup>35</sup> The mean bone-to-implant apposition is significantly greater in rough surface implants (72.31% +/- 17.76%) when compared to machined surface implants (38.01% +/- 19.32%), regardless of bone quality.<sup>35</sup>

Furthermore, the delayed failures in the Isidor study could be attributed to the reduced manual dexterity of patients preventing them to maintain good oral hygiene since all of them suffered from secondary SS.<sup>8,9,10</sup> The authors did not mention whether the implants failed due to peri-implantitis. Whilst one of our patients, suffering from secondary Sjogren's syndrome, also developed peri-implant mucositis, it was readily resolved after instituting an individualised maintenance programme.

Plaque adherence due to xerostomia and reduced dexterity due to arthritis are important maintenance factors to be considered in patients suffering from SS. Regular review appointments, prosthesis designs which facilitate cleansability, and individualised maintenance programme is necessary to aid plaque control and maintain healthy peri-implant tissues.

## SUMMARY

There is paucity of evidence for success of implants in patients with SS. Improvement in the quality of life is reported, especially when restored with fixed prosthesis. However, the need for regular maintenance in view of plaque adherence due to xerostomia and reduced dexterity due to arthritis is imperative to ensure long-term success.

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## REFERENCES

1. Delaleu N, Jonsson R, Koller MM. Sjögren's syndrome. *Eur J Oral Sci.* 2005;**113**:101-13
2. Vitali C, Bombardieri S, Jonsson R, Moutsopoulos HM, Alexander EL, Carsons SE, Daniels TE, Fox PC, Fox RI, Kassan SS, Pillemer SR, Talal N, Weisman MH; European Study Group on Classification Criteria for Sjögren's Syndrome. Classification criteria for Sjögren's syndrome: a revised version of the European criteria proposed by the American-European Consensus Group. *Ann Rheum Dis.* 2002;**61**:554-8
3. Soto-Rojas AE, Kraus A. The oral side of Sjögren syndrome. Diagnosis and treatment. A review. *Arch Med Res.* 2002;**33**:95-106
4. Kassan SS, Moutsopoulos HM. Clinical manifestations and early diagnosis of Sjögren syndrome. *Arch Intern Med.* 2004;**164**:1275-84
5. Mathews SA, Kurien BT, Scofield RH. Oral manifestations of Sjögren's syndrome. *J Dent Res.* 2008;**87**:308-18
6. Jacobson TE, Krol AJ. A contemporary review of the factors involved in complete denture retention, stability, and support. Part I: retention. *J Prosthet Dent.* 1983;**49**:5-15

7. Isidor F, Brøndum K, Hansen HJ, Jensen J, Sindet-Pedersen S. Outcome of treatment with implant-retained dental prostheses in patients with Sjögren syndrome. *Int J Oral Maxillofac Implants*. 1999;**14**:736-43
8. Spinato S, Soardi CM, Zane AM. A mandibular implant-supported fixed complete dental prosthesis in a patient with Sjogren's syndrome: case report. *Implant Dent*. 2010;**19**:178-83
9. Binon PP. Thirteen-year follow-up of a mandibular implant-supported fixed complete denture in a patient with Sjogren's's syndrome: a clinical report. *J Prosthet Dent*. 2005;**94**:409-13
10. Payne AG, Lownie JF, Van Der Linden WJ. Implant-supported prostheses in patients with Sjögren's syndrome: a clinical report on three patients. *Int J Oral Maxillofac Implants*. 1997;**12**:679-85
11. [http://www.cebm.net/%20levels\\_of\\_evidence.asp](http://www.cebm.net/%20levels_of_evidence.asp)
12. Bornstein MM, Cionca N, Mombelli A. Systemic conditions and treatments as risks for implant therapy. *Int J Oral Maxillofac Implants*. 2009;**24** Suppl:12-27
13. Candel-Marti ME, Ata-Ali J, Peñarrocha-Oltra D, Peñarrocha-Diago M, Bagán JV. Dental implants in patients with oral mucosal alterations: An update. *Med Oral Patol Oral Cir Bucal*. 2011;**16**:787-93
14. Dawson A, Chen S, Buser D, et al. The SAC Classification in implant dentistry. *Quintessence Publishing Go Ltd*; 2009. p. 4-6, 15-17, 21-25
15. Mericske-Stern RD, Taylor TD, Belser U. Management of the edentulous patient. *Clinical Oral Implants Research* 2000;**11**:108-125
16. Buser D, Halbritter S, Hart C, et al. Early implant placement with simultaneous guided bone regeneration following single-tooth extraction in the esthetic zone: 12-month results of a prospective study with 20 consecutive patients. *J Periodontol* 2009;**80**:152-162
17. Hämmerle CH, Chen ST, Wilson TG. Consensus statements and recommended clinical procedures regarding the placement of implants in extraction sockets. *Int J Oral Maxillofac Implants* 2004;**19**:26-28
18. Chen ST, Beagle J, Jensen SS, et al. Consensus statements and recommended clinical procedures regarding surgical techniques. *Int J Oral Maxillofac Implants* 2009;**24**:272-278
19. Jemt T. Restoring the gingival contour by means of provisional resin crowns after single-implant treatment. *Int J Periodontics Restorative Dent*. 1999;**19**:20-29
20. Moscovitch MS, Saba S. The use of a provisional restoration in implant dentistry: a clinical report. *Int J Oral Maxillofac Implants*. 1996;**11**:395-399
21. Lewis S, Parel S, Faulkner R. Provisional implant-supported fixed restorations. *Int J Oral Maxillofac Implants*. 1995;**10**:319-325
22. Abduo J, Lyons K, Bennani V, et al. Fit of screw-retained fixed implant frameworks fabricated by different methods: a systematic review. *Int J Prosthodont* 2011;**24**:207-220
23. Kapos T, Evans C. CAD/CAM technology for implant abutments, crowns, and superstructures. *Int J Oral Maxillofac Implants* 2014;**29**:117-136
24. Salama M, Coachman C, Garber D, et al. Prosthetic gingival reconstruction in the fixed partial restoration. Part 2: diagnosis and treatment planning. *Int J Periodontics Restorative Dent* 2009;**29**:573-581
25. Mehl C, Harder S, Wolfart M, et al. Retrieval of implant-retained crowns following cementation. *Clin Oral Implants Res* 2008;**19**:1304-1311
26. Rosenstiel SF, Land MF, Crispin BJ. Dental luting agents: A review of the current literature. *J Prosthet Dent* 1998;**80**:280-301
27. Salvi GE, Brägger U. Mechanical and technical risks in implant therapy. *Int J Oral Maxillofac Implants*. 2009;**24**:69-85
28. Vere J, Hall D, Patel R, Wragg P. Prosthodontic maintenance requirements of implant-retained overdentures using the locator attachment system. *Int J Prosthodont*. 2012;**25**:392-4
29. Burns DR, Unger JW, Coffey JP, Waldrop TC, Elswick RK Jr. Randomized, prospective, clinical evaluation of prosthodontic modalities for mandibular implant overdenture treatment. *J Prosthet Dent*. 2011;**106**:12-22
30. Lang NP, Berglundh T, Heitz-Mayfield LJ, Pjetursson BE, Salvi GE, Sanz M. Consensus statements and recommended clinical procedures regarding implant survival and complications. *Int J Oral Maxillofac Implants*. 2004;**19**:150-154
31. Sorensen JA, Engelman MJ. Ferrule design and fracture resistance of endodontically treated teeth. *J Prosthet Dent*. 1990;**63**:529-36
32. Sorensen JA, Martinoff JT. Clinically significant factors in dowel design. *J Prosthet Dent*. 1984;**52**:28-35
33. Jung RE, Zembic A, Pjetursson BE, et al. Systematic review of the survival rate and the incidence of biological, technical and aesthetic complications of single crowns on implants reported in longitudinal studies with a mean follow-up of 5 years. *Clin Oral Implants Res* 2012;**23**:2-21
34. Pjetursson BE, Brägger U, Lang NP, et al. Comparison of survival and complication rates of tooth-supported fixed dental prostheses (FDPs) and implant-supported FDPs and single crowns (SCs). *Clin Oral Implants Res* 2007;**18**:97-113
35. Trisi P, et al. Bone-to-implant apposition with machined and MTX microtextured implant surfaces in human sinus grafts. *Int J Periodontics Restorative Dent*. 2003;**23**:427-37