A Novel Approach for Immediate Implant-Based Oral Rehabilitation in a Sjögren's Syndrome Patient Using Virtual Surgical and Prosthetic Planning

Matthijs in 't Veld, MD[1]
Engelbert A.J.M. Schulten, DDS, MD, PhD[1]
Ulf Neveling[2]
Derk H. Jan Jager, DDS, PhD, Mpros, RCSed[1]
Frank K.J. Leusink, DDS, MD, PhD[1]*

Patients with Sjögren syndrome (SS) experience difficulties in wearing conventional dentures. After removal of all teeth, the oral rehabilitation is challenging and time consuming using conventional treatment protocols. Although implant-retained overdentures are beneficial for this specific patient group, the average total oral rehabilitation time (TORT) usually takes at least 9 months and needs to be reduced to increase patients' quality of life (QoL). In this paper, we report on a new treatment concept for immediate implant-based oral rehabilitation in a 77-year-old patient with partial edentulous SS. Because of persistent pain, discomfort, and retention problems with the conventional prosthetic devices, full clearance of the remaining mandibular dentition and immediate oral rehabilitation with an implant-retained overdenture were suggested. The treatment protocol included virtual surgical planning (VSP), combining a guided bone reduction of the mandibular alveolar process, immediate dental implant placement, and restoration using a prefabricated bar and placement of the overdenture.

This method demands the use of ionizing 3D imaging optionally combined with an optical dental scan or a conventional impression. Furthermore, one needs to gain experience using VSP software.

This novel treatment concept for immediate implant-based oral rehabilitation using VSP proved to be feasible and safe in a patient with SS, resulting in a significantly reduced TORT and improved QoL. Further research is needed to what extent this treatment concept could be beneficial to other patient groups, such as patients with head and neck cancer.

Key Words: CAD/CAM, immediate implant placement, oral rehabilitation, Sjögren's disease, VSP

Introduction

Sjögren syndrome (SS) is a chronic and progressive autoimmune disease causing irreversible damage to the exocrine glands and is associated with B and T lymphocyte infiltration of the affected glands.[1,2] Because it mainly affects the lacrimal glands and the salivary glands, the predominant symptoms are dry eyes and xerostomia.[3] Further oral implications of SS are tooth decay, tooth loss, fungal infections, traumatic oral mucosal lesions, dysphagia, dysgeusia, and inflammation of the salivary glands.[4,5] Due to hyposalivation, patients can experience difficulties in eating and speech. These symptoms significantly affect the patients' quality of life (QoL).[6–8] In SS, saliva loses its antimicrobial function and the ability to buffer and lubricate.[3,9] This results in an increase in caries incidence, which ultimately may lead to loss of teeth.[4] Replacement of missing teeth by conventional dentures could be difficult in these patients because of complications such as pain and discomfort from denture irritation and loss of retention.[10,11] Retention of a conventional denture will be limited because denture retention is dependent on the salivary layer between the denture base and the oral tissues.[12] Moreover, because of recurrent oral candidiasis and reduced lubrication by saliva, denture supporting mucosal tissues become fragile and susceptible to traumatic lesions.[13,14] As conventional dentures are poorly tolerated in individuals with SS, dental implant-supported overdentures are commonly used. Dental implants are regarded a viable treatment option for these patients.[15]

Unfortunately, after removal of the remaining teeth, conventional dentures are still needed to bridge the wound healing and implant osseointegration phase before placement of the implant-supported overdenture. This phase is regarded as a major burden to patients with SS. Although implant-retained overdentures are beneficial for this specific patient group, the average total oral rehabilitation time (TORT) usually

[1] Amsterdam UMC and Academic Centre for Dentistry Amsterdam (ACTA), Vrije Universiteit Amsterdam, Cancer Center Amsterdam, Department of Oral and Maxillofacial Surgery/Oral Pathology, Amsterdam, The Netherlands.
[2] Camlog Vertriebs GmbH, DEDICAM Guide Department, Wimsheim, Germany.
* *Corresponding author*, email: f.leusink@amsterdamumc.nl
https://doi.org/10.1563/aaid-joi-D-20-00420

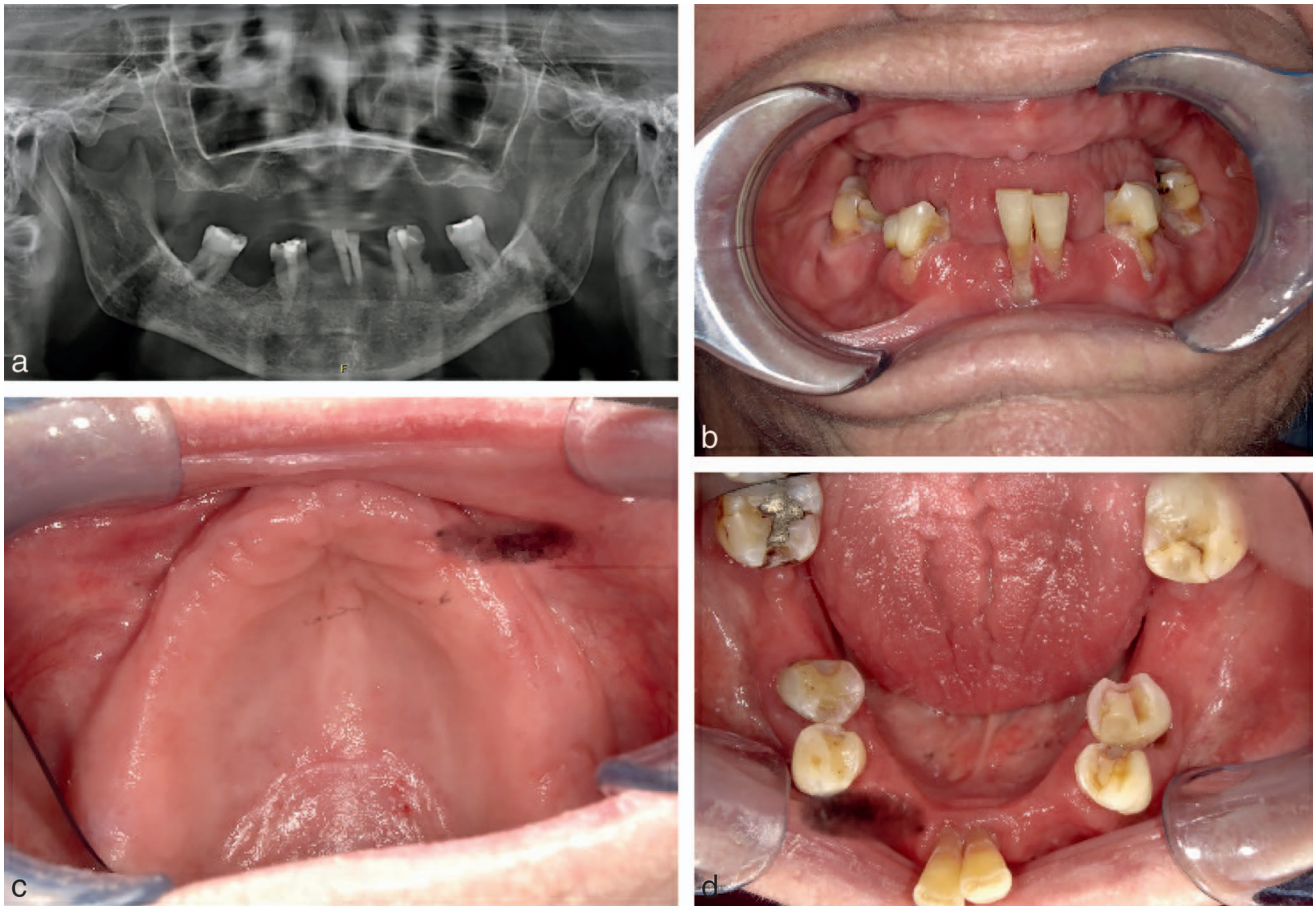

Figure 1. Preoperative images. (a) Panoramic radiograph of the patient. (b) Frontal intraoral view of the edentulous maxilla and partial dentition in the mandible. (c) Occlusal intraoral view of the edentulous maxilla. (d) Occlusal intraoral view of the partial dentition in the mandible.

takes at least 9 months and needs to be shortened to reduce the patients' burden.[16]

One of the major developments in implant surgery over the past decades is the introduction of virtual surgical planning (VSP) techniques. These techniques allow accurate and reliable planning and placement of dental implants. Other benefits of VSP include cost-effectiveness, durability, predictability, and simplicity.[17,18] Using VSP techniques, not only the positions of the dental implants can be planned, also the superstructure (ie, the retention bar) can be prefabricated, which also applies for the overdenture itself. With the use of VSP techniques and prefabrication of the retention bar and overdenture, the oral rehabilitation regarding implant placement and restoration can be performed in one session, which could be beneficial and could help to relieve the discomfort, especially in patients with SS.

The aim of this case report is to describe a novel multidisciplinary approach of immediate oral rehabilitation, including the removal of the remaining dentition, a marginal mandibular resection, dental implant placement, and prosthetic restoration in a patients with SS using VSP. This paper was written according to the CARE statement guidelines.[19]

Case Study

Clinical presentation

In 2019, a 77-year-old female patient was referred to our department by a maxillofacial prosthodontist at the Center for Special Care Dentistry Amsterdam to explore the possibility of an implant-based oral rehabilitation in the lower and upper jaw. Her major complaints consisted of chewing dysfunction and pain related to lack of retention of both upper and lower dentures. In an attempt to improve the chewing function with noninvasive techniques, 3 new upper dentures had been previously fabricated.

Although hyposalivation ($<$0.1 mL/min unstimulated whole mouth salivary secretion), xerostomia, and burning mouth syndrome were initially suspected, serological tests and histopathological examination of a biopsy of the minor salivary glands in the lower lip confirmed the diagnosis of SS, according to the 2002 American-European Consensus Group Criteria.[20]

On oral examination a fully edentulous maxilla with a Cawood and Howell class V atrophy and a partially edentulous mandible was observed. In the remaining dentition (teeth 37, 35, 34, 31, 41, 44, 45, and 47 were present) caries was diagnosed in the premolar 35, and gingival recessions without

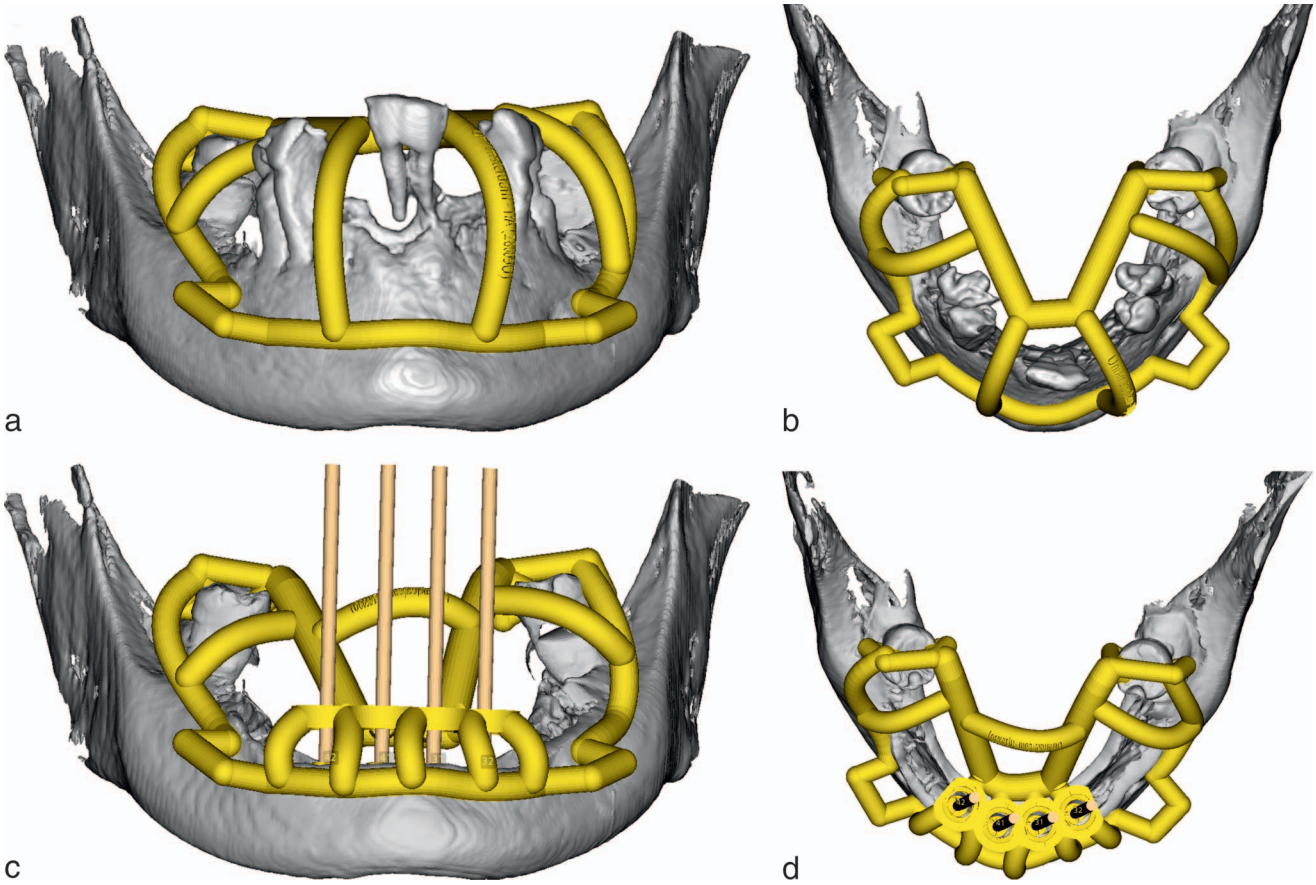

Figure 2. Preoperative virtual planning images. (a) Frontal oblique view of the bone reduction guide. (b) Occlusal view of the bone reduction guide. (c) Frontal oblique view of the drill guide. (d) Occlusal view of the drill guide.

pockets >3 mm were seen around all remaining teeth. The lower incisors 31 and 41 had a mobility grade II according to Miller's Tooth Mobility Index.[21]

The panoramic radiograph did not reveal any other abnormalities (Figure 1a). Cone-beam computed tomography (CBCT) was acquired for VSP purposes. Furthermore, intraoral photographs were made pre-, intra- and postoperatively (Figure 1b–d).

Although an implant-retained upper denture while maintaining the lower dentition could relieve the patient's complaints, we considered the status of the lower dentition in this patient with SS too poor to refrain from dental clearance. Therefore, to reduce the TORT, the following treatment plan was designed together with a maxillofacial prosthodontist: removal of the remaining mandibular dentition, guided vertical reduction of the mandibular alveolar process followed by guided placement of 4 dental implants and immediate restoration by connecting a prefabricated milled retention bar and overdenture. The patient agreed with this treatment plan and signed a written informed consent.

Preoperative planning

The CBCT scan (PaX-Zentith 3D, Vatech Co Ltd, Hwaseong, Korea) was acquired using the following settings: 16 × 14 cm field of view, 360° rotation, 105 kVp, 5.2 mA, 15 seconds scan time, and a radiation dose of 14.02 mSv. The TRIOS (3Shape A/S, Copenhagen, Denmark) intraoral scanner was used to obtain the most accurate 3D image of the dentition. The CBCT and the optical scan were fused to facilitate design of both dental and bony supported guides for accuracy reasons.

All data sets were stored as Digital Images and Communication in Medicine (DICOM) files and were imported into medical image processing software. ProPlan CMF software (Materialise NV, Leuven, Belgium) was used to design the osteotomy plane outlining the necessary amount of bone reduction of the alveolar process to create sufficient intermaxillary space needed for the superstructures (bar and prosthesis).

The guides were designed according to the following requirements: (1) seating should be easy and stable. Therefore, in this case the following landmarks were used: molars 37 and 47, and the mental protuberance of the mandible. (2) Cut-outs should be located around the mental foramen in order to preserve the mental nerve. All DICOM and stereolithography (STL) files were sent to the dental technician (UN) (DEDICAM, Camlog, Wimsheim, Germany) to design the bone reduction and drill guides (Figure 2a–d).

SMOP software (Swissmeda, Zurich, Switzerland) was used to combine and align the DICOM and the STL files for the preplanning. For the design of the bone reduction guide, the DICOM data has been converted into a STL file and reduced to

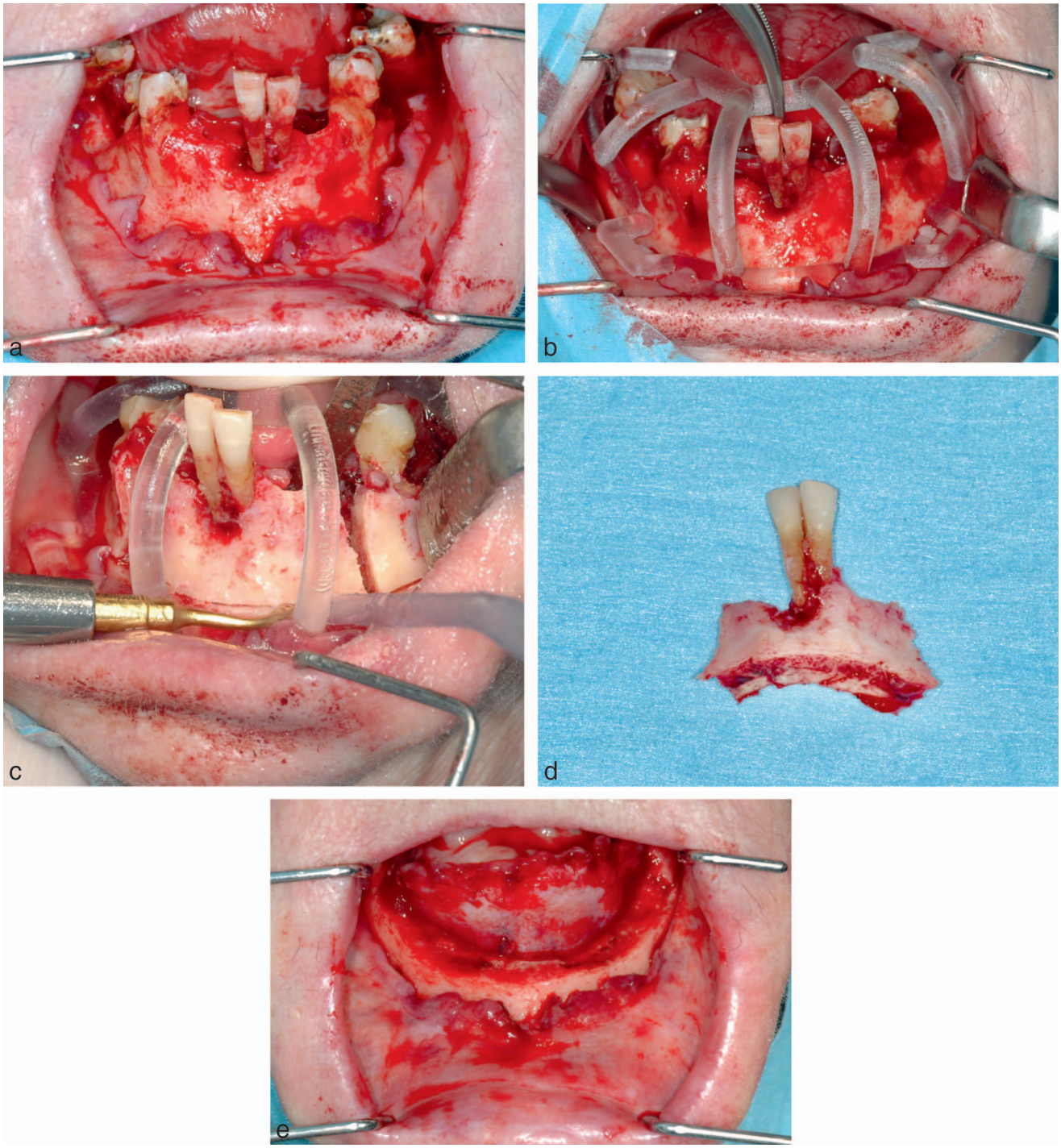

Figure 3. Intraoperative photographs. (a) Frontal view of the mandibular arch after reflection of the mucoperiosteum. (b) Seating of the bone reduction guide. (c) The cutting planes using piezoelectric surgery. (d) En-bloc resection of the teeth in the interforaminal region. (e) The mandibular arch after the marginal mandibulectomy, extraction, and equalization.

the desired level in Exocad software (Exocad DentalCAD, Darmstadt, Germany). This is necessary to have a proper base for the construction of the 2 guides. For the integration of the 2 molars, the STL files of the bone and the model had to be combined into 1 single STL file as scatterings made it impossible to use the bone STL directly. SMOP was also used to position 4 dental implants (∅ 3.8 mm, L 11 mm; Conelog Guided Progressive-line implants, Camlog Biotechnologies AG, Basel, Switzerland) in the interforaminal region of the mandible and to design the drill guide. The center-to-center distance between the dental implants was planned at 10 mm. To preserve the mental nerve, a safety margin of 5 mm between the most lateral implants and the mental foramen had to be maintained. The drill sleeves were positioned in the drill guide

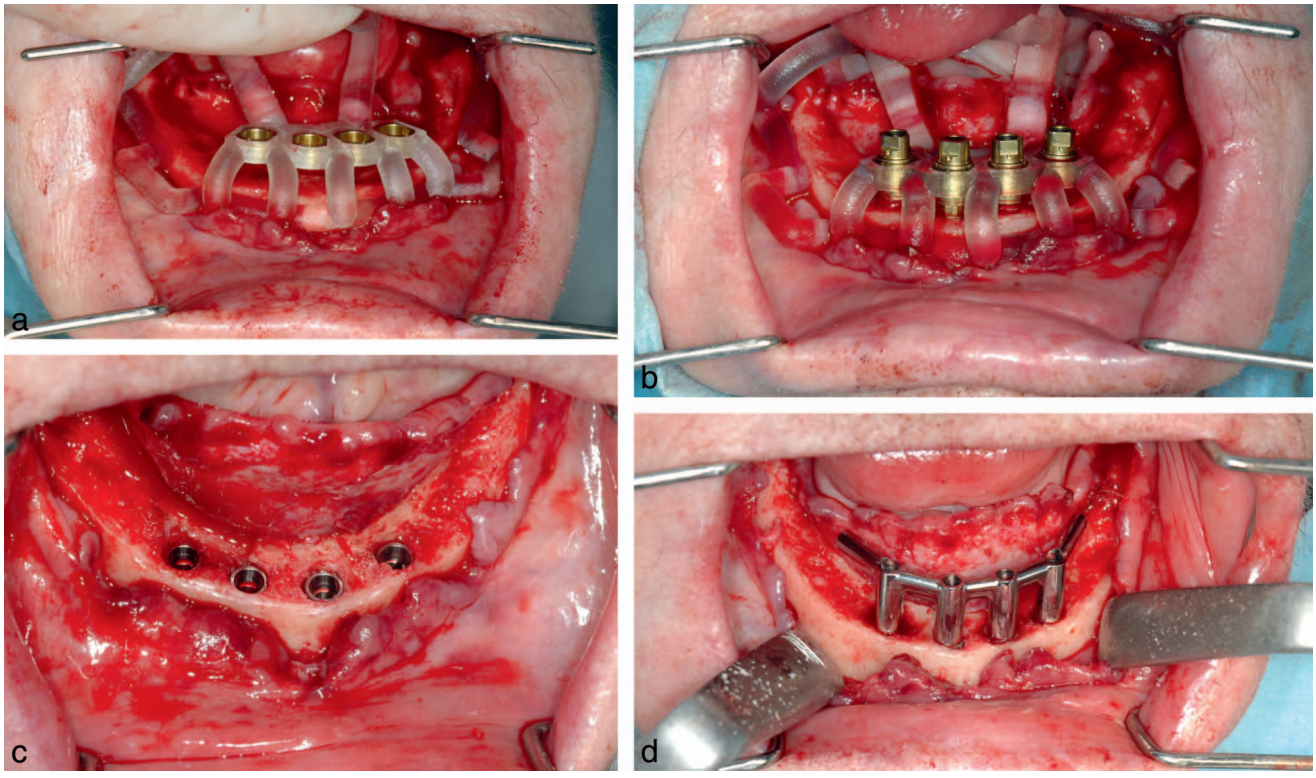

Figure 4. Intraoperative photographs of the dental implant surgery phase. (a) Seating of the drill guide. (b) Guided placement of the 4 implants. (c) Frontal view of the placed implants. (d) Placement of the retention bar.

according to the manufacturer's guideline.[22] To design the retention bar, the case data were exported as STL files and re-imported into Exocad dental CAD software. The final designs were presented and discussed in a video conference call and were produced after approval of the surgeon (FL) and the maxillofacial prosthodontist.

Surgical technique

The following surgical steps were performed under local anaesthesia and perioperative antibiotic prophylaxis (amoxicillin/clavulanic acid 500/125 mg orally, 3 times a day during 1 week, starting 1 day preoperatively): (1) guided reduction of the alveolar process (Figure 3a–e); (2) immediate guided dental implant placement; and (3) immediate restoration of the dental implants by connecting a prefabricated milled retention bar (Figure 4a–d).

A marginal incision was made from molar 37 to 47, followed by elevation of the mucoperiosteum (Figure 3a). Subsequently, the alveolar process was inspected, and the mental nerves were identified. The bone reduction guide was seated on the molars 37 and 47, and the mental protuberance (Figure 3b). Using piezoelectric surgery and the bone reduction guide, the horizontal osteotomy was performed and all teeth in the interforaminal region were removed in 1 piece (Figure 3c and d). The Luer forceps and a burr were used to remove sharp edges at the osteotomy plane (Figure 3e).

The drill guide was seated on the same landmarks as the bone reduction guide (Figures 2c and 4a). The implant osteotomies were carried out using the guided drill protocol according to the manufacturer's guideline.[22] Subsequently, the 4 Conelog Guided Progressive-line implants (Camlog Biotechnologies AG) were manually inserted, using the torque wrench. Primary stability of all 4 implants was achieved measuring torque values higher than 30 Ncm (Figure 4b and c). The prefabricated retention bar was placed and connected to the 4 implants by screw-fixation (Figure 4d), showing a perfect fit.

Finally, primary closure of the mucosa was accomplished (Figure 5a). Although the overdenture was ready to be placed subsequently, a second session for the placement of the denture was scheduled on patient's request. However, due to the COVID-19 regulations the overdenture was placed, after application of a resilient lining material, 6 weeks postoperatively (Figure 5e).

Follow-up

After the procedure, a panoramic radiograph and a lateral cephalogram were made, showing good positioning of the 4 dental implants and confirming the perfect fit of the retention bar on the implants (Figure 5b and c). Recalls were performed after 6 and 8 weeks (Figure 5d and e). No surgical complications were observed during follow-up, and no sensory disturbance of the mental nerves was reported. Using the Oral Health Impact Profile (OHIP-14) questionnaire, the patient reported a total score of 57 two weeks preoperatively and 25 one month after placement of the overdenture. The decreased total score of the OHIP-14 questionnaire indicates an improved chewing function and an increased QoL. The patient was scheduled for her

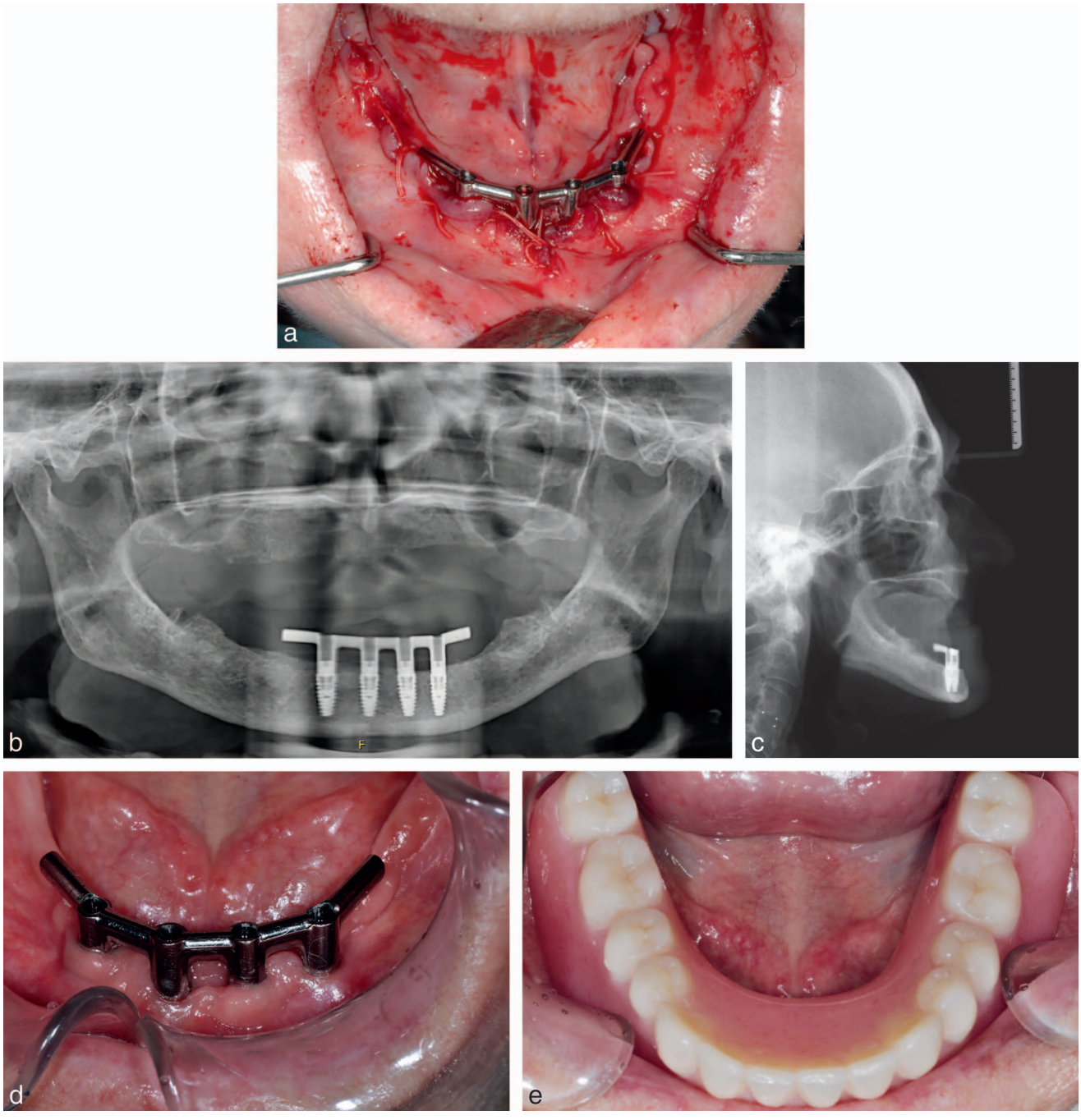

Figure 5. Postoperative clinical and radiographic images. (a) Frontal view of the mandibular arch with the retention bar after primary closure. (b) Panoramic view demonstrating the 4 implants and the retention bar in the interforaminal region. (c) Lateral cephalogram. (d) Frontal view of the mandibular arch with the retention bar. (e) The implant-retained overdenture 6 weeks postoperatively.

regular check-up and oral hygiene appointment 6 months postoperatively.

Discussion

We report on the oral rehabilitation of a patient with SS with an indication for replacement of a desolate dentition by an implant-supported overdenture using a new treatment concept aiming to reduce the TORT. Although our new treatment concept is especially designed for vulnerable patients, for example, SS or patients with head and neck cancer (HNC), it could also be beneficial for patients without such pathology, even though these healthy individuals could also function properly with temporary conventional dentures. Clearly patients with SS cannot function properly with temporary conventional dentures due to pain, lack of lubrication, and

lack of retention of prosthetic devices.[23] The average TORT of a conventional treatment protocol is at least 9 months and this period is regarded as a major burden by patients with SS.

In the last decades dental implants have become standard care in restoring the edentulous jaw.[24] Dental implants are also regarded as a viable treatment option in patients with SS.[25] In a study by Korfage et al,[15] it was found that implants in patients with SS seem to perform comparable with implants in healthy patients. The only difference was that patients with SS appear to have more signs of peri-implant soft tissue infection. In addition, in a systematic review performed by Almeida et al,[26] high survival rates of dental implants in patients with SS were reported, with an average of 93.7% in a mean period of 3.97 years. Chrcanovic et al[27] included a total of 19 studies, and 705 dental implants were followed up for a mean period of 72.5 months. This systematic review reported a failure rate of 4.1% (29 of the 705 implants were lost). However, the success of dental implants in our treatment concept has not been previously reported in the literature.

In patients with HNC with an indication for dental clearance prior to radiotherapy to prevent osteoradionecrosis, this new treatment concept could contribute in 2 different ways: (1) to maintain masticatory and swallowing function; (2) to facilitate patients in maintaining optimal nutritional status.[28–30]

With the development of VSP techniques over the last few years, a reliable and accurate preoperative planning can be made. The surgical guides used in this case were seated on the molars and the mental protuberance. However, if the molars are absent, an alternative could be to seat the guides on other anatomical structures like the alveolar process itself. In such a case, no optical scan of the dentition is needed and the preoperative VSP could be carried out only using CBCT.

Limitations of this treatment concept are as follows: (1) the necessity of the CBCT itself and its corresponding additional radiation dose for the patient. (2) In case of presence of the first and second molars either a conventional impression or an optical scan of the dentition is needed. (3) One needs to gain experience using VSP software (ie, SMOP). Designing the guides in this case took 20 minutes. Unfortunately, immediate functional loading of the implants was intended, but due to minor adjustments to the overdenture and the strict regulations in the COVID-19 pandemic, the prefabricated overdenture could not be placed earlier than 6 weeks postoperatively. However, early functional implant loading was achieved because placement of the overdenture took place within 3 months after implant placement.[31]

This treatment concept incorporates 4 dental implants and a retention bar with distal extensions resulting in an almost fully implant-supported overdenture. It is questionable whether this treatment concept should be used with less than 4 implants. Further research is needed to discover whether this treatment protocol could be performed with less than 4 implants. The osteotomy results in a loss of cortical mandibular bone, which could lead to a lack of primary stability of the dental implants. Therefore, the authors believe that 2 implants might be insufficient to support a fully implant-supported overdenture, especially in this treatment concept based on a vertical guided bone reduction, immediate guided implant placement, early loading and restoration. Additionally, a study including finite-element-analysis comparing 3 versus 4 implants might further elucidate this question. Furthermore, the authors would like to translate this treatment modality in the oral rehabilitation of patients with HNC, especially because this patient group could benefit from this concept as described earlier.

In this patient with SS with an indication for dental clearance, the TORT could be significantly reduced using the new treatment concept described. During follow-up no complications were observed, and the patient reported an improved QoL using the OHIP-14 questionnaire.

Conclusion

To the best of our knowledge, this is the first case report describing a guided vertical bone reduction combined with immediate oral rehabilitation including dental implant placement and restoration in a patient with SS using VSP. We believe this concept definitely reduces the TORT, improves patients' QoL, and could be beneficial to other vulnerable patient groups, such as patients with HNC.

Note

None of the authors have a conflict of interest regarding the techniques and materials described.

References

1. Fox RI. Sjogren's syndrome. *Lancet*. 2005;366(9482):321–331.
2. Reksten TR, Jonsson MV. Sjogren's syndrome: an update on epidemiology and current insights on pathophysiology. *Oral Maxillofac Surg Clin North Am*. 2014;26(1):1–12.
3. Mathews SA, Kurien BT, Scofield RH. Oral manifestations of Sjogren's syndrome. *J Dent Res*. 2008;87(4):308–318.
4. Maarse F, Jager DH, Forouzanfar T, Wolff J, Brand HS. Tooth loss in Sjogren's syndrome patients compared to age and gender matched controls. *Med Oral Patol Oral Cir Bucal*. 2018;23(5):e545–e551.
5. Amerongen AV, Veerman EC. Saliva–the defender of the oral cavity. *Oral Dis*. 2002;8(1):12–22.
6. Enger TB, Palm O, Garen T, Sandvik L, Jensen JL. Oral distress in primary Sjogren's syndrome: implications for health-related quality of life. *Eur J Oral Sci*. 2011;119(6):474–480.
7. Rusthen S, Young A, Herlofson BB, et al. Oral disorders, saliva secretion, and oral health-related quality of life in patients with primary Sjogren's syndrome. *Eur J Oral Sci*. 2017;125(4):265–271.
8. Fernandez-Martinez G, Zamora-Legoff V, Hernandez Molina G. Oral health-related quality of life in primary Sjogren's syndrome. *Reumatol Clin*. 2020;16(2 part 1):92–96.
9. Chaudhury NM, Shirlaw P, Pramanik R, Carpenter GH, Proctor GB. Changes in saliva rheological properties and mucin glycosylation in dry mouth. *J Dent Res*. 2015;94(12):1660–1667.
10. Niedermeier WH, Kramer R. Salivary secretion and denture retention. *J Prosthet Dent*. 1992;67(2):211–216.
11. Guggenheimer J, Moore PA. Xerostomia: etiology, recognition and treatment. *J Am Dent Assoc*. 2003;134(1):61–69; quiz 118–119.
12. de Koomen HA, van Velzen WA. [Saliva and dentures]. *Ned Tijdschr Tandheelkd*. 1992;99(3):97–99.
13. Cannon RD, Chaffin WL. Oral colonization by Candida albicans. *Crit Rev Oral Biol Med*. 1999;10(3):359–383.
14. Ship JA. Diagnosing, managing, and preventing salivary gland disorders. *Oral Dis*. 2002;8(2):77–89.
15. Korfage A, Raghoebar GM, Arends S, et al. Dental implants in

patients with Sjogren's syndrome. *Clin Implant Dent Relat Res*. 2016;18(5): 937–945.

16. Ye M, Liu W, Cheng S, Yan L. Immediate vs conventional loading of mandibular overdentures: a comprehensive systematic review and meta-analysis of randomized controlled trials [published online ahead of print November 18, 2020]. *J Oral Implantol*.

17. Kapos T, Ashy LM, Gallucci GO, Weber HP, Wismeijer D. Computer-aided design and computer-assisted manufacturing in prosthetic implant dentistry. *Int J Oral Maxillofac Implants*. 2009;24(suppl):110–117.

18. Patzelt SB, Spies BC, Kohal RJ. CAD/CAM-fabricated implant-supported restorations: a systematic review. *Clin Oral Implants Res*. 2015; 26(suppl 11):77–85.

19. Gagnier JJ, Kienle G, Altman DG, et al. The CARE guidelines: consensus-based clinical case reporting guideline development. *BMJ Case Rep*. 2013;2013.

20. Vitali C, Bombardieri S, Jonsson R, et al. Classification criteria for Sjogren's syndrome: a revised version of the European criteria proposed by the American-European Consensus Group. *Ann Rheum Dis*. 2002;61(6):554–558.

21. Miller SC. *Textbook of Periodontia (Oral Medicine)*. The Blakiston Co., Philadelphia. 1950.

22. Camlog Biotechnologies. Instructions for Use Guide System for CONELOG® PROGRESSIVE-LINE Implants. 2020.

23. Cassolato SF, Turnbull RS. Xerostomia: clinical aspects and treatment. *Gerodontology*. 2003;20(2):64–77.

24. Feine JS, Carlsson GE, Awad MA, et al. The McGill Consensus Statement on Overdentures. Montreal, Quebec, Canada. May 24–25, 2002. *Int J Prosthodont*. 2002;15(4):413–414.

25. Daneshparvar H, Esfahanizadeh N, Vafadoost R. Dental Implants in Sjogren Syndrome. *Eur J Transl Myol*. 2020;30(2):8811.

26. Almeida D, Vianna K, Arriaga P, Moraschini V. Dental implants in Sjogren's syndrome patients: a systematic review. *PLoS One*. 2017;12(12): e0189507.

27. Chrcanovic BR, Kisch J, Wennerberg A. Dental implants in patients with Sjogren's syndrome: a case series and a systematic review. *Int J Oral Maxillofac Surg*. 2019;48(9):1250–1259.

28. Langendijk JA, Doornaert P, Rietveld DH, Verdonck-de Leeuw IM, Leemans CR, Slotman BJ. A predictive model for swallowing dysfunction after curative radiotherapy in head and neck cancer. *Radiother Oncol*. 2009; 90(2):189–195.

29. Langmore S, Krisciunas GP, Miloro KV, Evans SR, Cheng DM. Does PEG use cause dysphagia in head and neck cancer patients? *Dysphagia*. 2012;27(2):251–259.

30. Kraaijenga SA, van der Molen L, van den Brekel MW, Hilgers FJ. Current assessment and treatment strategies of dysphagia in head and neck cancer patients: a systematic review of the 2012/13 literature. *Curr Opin Support Palliat Care*. 2014;8(2):152–163.

31. Laney WR. Glossary of oral and maxillofacial implants. *Int J Oral Maxillofac Implants*. 2017;32(4):Gi–G200.