CASE REPORT

WILEY

Prosthodontic restoration of a COVID-19 associated mucormycosis defect: A clinical report

Ioli Ioanna Artopoulou¹ Christos Perisanidis²

¹Department of Prosthodontics, National and Kapodistrian University of Athens, School of Dentistry, Athens, Greece

²Clinical Fellow, Department of Oral and Maxillofacial Surgery, National and Kapodistrian University of Athens, "Evaggelismos" General Hospital, Athens, Greece

Correspondence

Ioli Ioanna Artopoulou, National and Kapodistrian University of Athens, School of Dentistry, Dept. of Prosthodontics, 2 Thivon str., 11 527, Goudi, Athens, Greece. Email:iartopoulou@gmail.com

Ioli Ioanna Artopoulou¹ Evangelos Kalfarentzos² Gregory Polyzois¹

Abstract

Introduction: Coronavirus Disease 2019 (COVID-19) pandemic brought the clinical practice of dentistry and prosthodontics at the threshold of a new era, due to the increasing prevalence of a relative rare condition normally affecting the immunocompromised patients.

Aim: The aim of this clinical case report is to briefly describe the etiopathogenesis, the surgical and prosthodontic management of this evolving medical condition emphasizing the emerging role of the maxillofacial prosthodontist in restoring the patients' well-being.

Methods and Results: The surgical and prosthodontic rehabilitation of a COVID-19 infected patient with a mucormycotic lesion of the maxilla will be presented.

Conclusion: The role of the maxillofacial prosthodontist is important in alleviating this severe COVID-19 associated morbidity.

K E Y W O R D S COVID-19, mucormycosis, prosthodontic rehabilitation

1 | INTRODUCTION

Coronavirus Disease 2019 (COVID-19) pandemic is associated with the current severe acute respiratory syndrome virus 2 (SARS-CoV-2) that ranges from asymptomatic to sever disease necessitating admission in an intensive care unit (ICU).^{1,2} Since the day of this report, it has been responsible for more than 545 million infections worldwide and over 6 million deaths.³ COVID-19 infection has been associated with emerging cases of mucormycosis; an invasive fungal infection of the rhino-orbital and rhino-cerebral region.^{4,5}

Mucormycosis is typically affecting immunocompromised patients with poorly controlled diabetes mellitus being an independent risk factor.^{6,7} Diabetes mellitus, even well-controlled, that is associated with increased morbidity and mortality due to COVID-19 infection, appeared to be the predominant risk factor for COVID-19 related mucormycosis, since predisposes the patients to diabetic ketoacidosis.^{8,9} Additionally, corticosteroid therapy often administered to control the severity of COVID-19 is a significant well-known risk factor for opportunistic fungal infections increasing therefore, the incidence for mucormycosis.¹⁰ Furthermore, severe COVID-19 described cytokine storm syndrome¹¹ resulting in high ferritin levels and iron overload,⁸ along with severe COVID-19 related "endothelialitis" are critical risk factors for angioinvasive mucormycosis.¹² COVID-19 related mucormycosis

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.

is a well-documented entity worldwide, with clinical presentation consistent with the typical diabetes mellitus related mucormycosis. It has been described in many countries, with most of the cases being reported in India.^{7,13} COVID-19 associated mucormycosis a rapidly progressing, deteriorating, and potentially life-threatening condition that usually occurs 0-90 days (Median = 10 days) after COVID-19 diagnosis.¹⁴ Since the prevalence of diabetes mellitus is dramatically increasing in low- and middleincome countries, and the world continues to strive against the COVD-19 pandemic, the associated number of cases of maxillofacial mucormycosis is anticipated to increase presenting therefore, significant diagnostic and therapeutic challenges for the oral and maxillofacial surgeon and the maxillofacial prosthodontist.¹⁵ There are several case reports in the literature, mainly from India, presenting the prosthodontic rehabilitation of post-COVID-19 mucormycosis maxillary defects with interim obturator prostheses.¹⁶⁻²¹ The aim of this clinical report is to present the surgical and prosthodontic management of a challenging COVID-19 related mucormycosis maxillectomy defect, raise awareness regarding this emerging during the pandemic clinical condition, and highlight the role of the maxillofacial prosthodontist in alleviating this severe COVID-19 associated morbidity by restoring patient's wellbeing.

2 | CLINICAL REPORT

A 53-year-old man presented to the Department of Oral and Maxillofacial Surgery, National and Kapodistrian University of Athens, "Evaggelismos" General Hospital, with a chief complaint of persistent pain of the maxilla accompanied by teeth loosening. His medical history was significant for diabetes mellitus and hyperlipidemia controlled by Amaryl (Sanofi US; Bridgewater, New Jersey, USA), Lipidil (Abbott Laboratories; Abbott Park, Iliniois, USA), Glucophage (Merck; Rahway, New Jersey, USA), and Xanax (Pfizer Inc., New York, New York, USA). He reported odynophagia, trismus, mobility of the maxillary teeth, and denied any further head and neck symptoms. He also denied using alcohol and tobacco. He also reported a history of COVID-19 infection 3 months ago and hospitalization due to pneumonia. The patient was admitted to the ICU unit for 10 days and given IV infusion Tocilizumab (800 mg/day in 100 ml saline), IV Dexamethasone (6 mg/day), IV Moxifloxacin (400 mg/day), IV Piperacillin/Tazobactam ($4.5g \times 3/day$), and Ing. Fondaparinux (2.5 mg/day).

A baseline physical examination of the head and neck area revealed acute infection of the middle third of the face and showed significant swelling of the right cheek,



FIGURE 1 Radiographic images of the extensive mucormycotic lesions.

the right infraorbital area, and the upper lip, with skin redness and tenderness. Intraoral examination revealed the presence of all mandibular and maxillary teeth, chronic generalized periodontitis, severe mobility of all the maxillary teeth, diffuse swelling of both maxillary alveolar processes and significant pus discharge through the gingival sulcus of all maxillary teeth. The palatal mucosa was intact and upon palpation the underline bone was soft and irregular throughout the maxilla and the anterior wall of both maxillary sinuses. Further radiographic examination showed destruction of the hard palate and the maxillary bony walls with bony erosions extending to the paranasal sinuses with right orbital floor and right zygomatic bone involvement. Significant mucosal thickening was noted in paranasal sinuses, ethmoid air cells, frontal sinus, and sphenoidal sinus consistent with inflammatory sinonasal disease (Figure 1). Endoscopic biopsy indicated fungal elements in the paranasal sinuses, antifungal therapy was initiated (Ing. Amphotericin B 300 mg/day), and surgical excision was planned. The surgical excision was performed intraorally and involved: bilateral subtotal maxillectomy with aggressive debridement of the maxillary sinuses, endoscopic debridement of the ethmoid sinus, and resection of the involved part of the right zygomatic bone. Histopathologic evaluation of the resected specimens identified the presence of



FIGURE 2 Primary surgical closure leaving two oro-antral openings.

rhizoids and sporangium of Rhizopus microspores and confirmed the initial diagnosis of mucormycosis induced osteomyelitis. Due to reported side effect the anti-fungal therapy scheme was substituted with Ing. Isavuconazole 200 mg/day. The palatal soft tissue was preserved following debridement and was used for primary closure with a palatal flap. Two oro-antral openings (0.5×2 cm) were created by the surgeon bilaterally in the buccal vestibules in the region of the second maxillary premolars, to improve the retention and the stability of the final prosthetic outcome (Figure 2).

The prosthetic rehabilitation treatment plan included the fabrication of a definitive obturator prosthesis with two acrylic projections that would alleviate speech and swallowing and facilitate monitoring of the affected area during the necessary long-term follow-up for possible fungal recurrence, until microvascular surgery with free osteomyocutaneous flap reconstruction will provide the necessary bony support for implant placement and rehabilitation with implant supported prosthesis. The two oro-antral openings were packed with vaseline-soaked gauze, and preliminary impressions were made using irreversible hydrocolloid material (BluePrint X-creme; Dentsply Int, York, Pennsylvania, USA). A maxillary custom tray was fabricated, and the final impression was made by modeling impression compound (Green stick impression compound; Kerr Corp., Brea, California, USA) that was used to border mold the periphery of the tray, as well as the two oro-antral openings and polysulfide impression material (Permlastic; Kerr Corp). A maxillary record base was fabricated and the following clinical (vertical and centric relation records, teeth set up and tryin) and laboratory steps were performed following basic removable prosthodontic principles. Care was taken for proper anterior teeth selection and positioning to accommodate for maximum esthetic restoration of the collapsed upper lip without dislodging the prosthesis. The maxillary



FIGURE 3 Definitive obturator prosthesis with acrylic projections.



FIGURE 4 Obturator prosthesis in occlusion with the lower natural dentition.

obturator prosthesis was processed, placed intraorally, and necessary adjustments were made. To properly add the maxillary sinus projections on the buccal flanges, a functional impression that recorded the contours of the oro-antral openings with a resilient acrylic relining material (Trusoft; Harry J. Bosworth Co., Skokie, Illinois, USA) was made. Articulation, resonance, and swallowing were evaluated clinically with speech and swallowing exercises that assessed nasal emission and food regurgitation. After the necessary follow up adjustments were made, the buccal flange projections were converted to heat polymerized acrylic resin (Lucitone 199; Dentsply Sirona, Charlotte, North Carolina, USA), and the final prosthesis was delivered to the patient (Figure 3, Figure 4, and Figure 5). As a standard of care procedure the patient was asked to complete the Obturator Functioning Scale (OFS) questionnaire and the Distress Thermometer (DT). OFS is a scale used to evaluate the perceived functioning and effectiveness of the complete denture obturator prosthesis and was forward



FIGURE 5 The prosthesis delivered to the patient.

and backward translated from English to Greek.^{22,23} DT is a visual analogue scale used by the National Comprehensive Cancer Network to screen distress in cancer patients. It is available in Greek and was used to assess physical, emotional, social, and practical concerns related to the patient's condition and impact his overall well-being. The patient was monitored, and necessary adjustments were made. He tolerated the prosthesis well and was scheduled to return to the clinic in 6 months for follow-up.

3 | DISCUSSION

During the ongoing COVID-19 pandemic, the medical world is confronted not only with the life-threatening infectious disease, but also with several, sometimes severe and unexpected, morbidities. Mucormycosis of the maxillofacial region, an otherwise rare condition, became a critical concern in the COVID-19 period with significantly rising prevalence. Raising awareness among medical and dental professionals for prompt identification of risk factors, implementation of preventive measures, early recognition of suspicious signs and symptoms, prompt treatment initiation, and guidance for proper management and rehabilitation of this emerging challenging clinical condition is important.

Consequently, a crucial demand for maxillofacial prosthetic rehabilitation arises in patients with

mucormycosis-related acquired maxillofacial defects to restore function and esthetics, preserving therefore, the patient's well-being. Usually as stated in relevant literature mucormycosis-related maxillectomy defects are left open without any surgical reconstruction since this facilitates monitoring by direct visual inspection for possible fungal recurrence.^{15–21} In addition, from a prosthodontic standpoint, when the defect is unrepaired, the resulting prosthesis has enhanced stability and retention, since the impression material more efficiently engages the remaining tissue undercuts. However, in the present clinical report, and due to the patient's young age and increased anxiety regarding post-operative speech alteration and psychosocial restrictions, the surgeon decided to primarily reconstruct the maxillary defect leaving only two small oro-antral defects, an approach consistent with two previous case reports presenting the prosthodontic rehabilitation of maxillary mucormycosis related defects that were not however related to COVID-19 infection.^{24,25} The meticulous capturing of the anatomy of these defects was significantly important for improved stability and retention of the prosthesis. The resulting prosthesis addressed the deriving functional disabilities, restored swallowing, diminished nasal regurgitation of food and liquids, and improved speech intelligibility, contributing to post-operative distress management, preserving therefore, the patient's well-being.

4 | CONCLUSIONS

COVID-19 with its concurrent morbidities might affect oral and maxillofacial structures resulting in significant intraoral defects and facial deformities. The role of the maxillofacial prosthodontist remains crucial in alleviating oral functions and esthetics, while restoring the well-being of the affected individuals.

ORCID

Ioli Ioanna Artopoulou D https://orcid.org/0000-0003-1711-3143

REFERENCES

- 1. Zhou F, Yu T, Du R, et al. Clinical course and risk factors for mortality of adult inpatients with COVID-19 in Wuhan, China: a retrospective cohort study. *Lancet*. 2020;6736:1-9.
- Dhama K, Khan S, Tiwari R, et al. Coronavirus disease 2019– COVID-19. Clin Microbiol Rev. 2020;33:1-48.
- Worldometer. Coronavirus case. Worldometer; 2022. [Internet]. Accessed November 19, 2022. https://www.worldometers.info/ coronavirus/
- Verma DK, Bali RK. COVID-19 and mucormycosis of the craniofacial skeleton: causal, contributory or coincidental? *J Oral Maxillofac Surg.* 2021;20:165-166.

- Al-Tawfiq JA, Alhumaid S, Alshukairi AN, et al. COVID-19 and mucormycosis superinfection: the perfect storm. *Infection*. 2021;49:833-853.
- 6. Farmakiotis D, Kontoyiannis DP. Mucormycoses. *Infect Dis Clin NAm.* 2016;30:143-163.
- Skiada A, Pavleas I, Drogari-Apiranthitou M. Epidemiology and diagnosis of mucormycosis: an update. *J Fungi.* 2020;6: 265-301.
- 8. John TM, Jacob CN, Kontoyiannis DP. When uncontrolled diabetes mellitus and severe COVID-19 converge: the perfect storm for mucormycosis. *J Fungi*. 2021;7:298-304.
- 9. Singh AK, Gupta R, Ghosh A, Misra A. Diabetes in COVID-19: prevalence, pathophysiology, prognosis and practical considerations. *Diabetes Metab Syndr*. 2020;14:303-310.
- Lionakis MS, Kontoyiannis DP. Glucocorticoids and invasive fungal infections. *Lancet*. 2003;362:1828-1838.
- 11. Chen LYC, Quach TTT. COVID-19 cytokine storm syndrome: a threshold concept. *Lancet Microbe*. 2021;2:49-50.
- 12. Ibrahim AS, Spellberg B, Walsh TJ, Kontoyiannis DP. Pathogenesis of mucormycosis. *Clin Infect Dis.* 2012;54:S16-S22.
- 13. Prakash H, Chakrabarti A. Global epidemiology of mucormycosis. *J Fungi*. 2019;26:1-19.
- Hoenigl M, Seidel D, Carvalho A, et al. The emergence of COVID-19 associated mucormycosis: a review of cases from 18 countries. *Lancet Microbe*. 2022;3:543-552.
- 15. Ali IE, Chugh A, Cheewin T, Hattori M, Sumita YI. The rising challenge of mucormycosis for maxillofacial prosthodontists in the Covid-19 pandemic: a literature review. *J Prosthodont Res.* 2022;66:395-401.
- Rodrigues SJ, Diaz EAI, Shetty SS, de Arruda AAJ. Resurgence of mucormycosis in India: a dentist's perspective. *Spec Care Dent*. 2022;42:200-202.
- Ravi MB, Srinivas S, Silina E, Sengupta S, Tekwani T, Achar RP. Prosthetic rehabilitation of rhino orbital mucormycosis associated with COVID-19: a case series. *Clin Cosmet Investig Dent*. 2022;14:1-10.

- Mahajan K, Das M, Kumar GA. Post surgical immediate prosthetic reconstruction in patients with rhinocerebral mucormycosis: case series. J Family Med Prim Care. 2022;11:379-385.
- Garde J, Khan A, Dhande S, Garde D, Muglikar S. Restoring a smile post Covid-19 associated mucormycosis: a case report. J Dental Sci 2021;6:1-8.
- 20. Chincholikar S, Lalwani G. Rehabilitation of post Covid mucormycosis maxillectomy defect with hollow obturator: a case series. *IP Ann prosthodont restor dent*. 2022;8:38-41.
- Aparna G, Alexander A, Sivaraman G, Kalaiarasi R. Temporary obturator using high-density polyurethane foam following maxillectomy during the coronavirus disease 2019 pandemic. J Laryngol Otol. 2022;136:173-175.
- Kornblith AB, Zlotolow IM, Gooen J, et al. Quality of life of maxillectomy patients using obturator prosthesis. *Head Neck*. 1996;18:323-334.
- NCCN Distress Thermometer. Accessed November 19, 2022. http://ncnn.org/professional/physician-gls/pdf/distress-toolgreek.pdf
- 24. Inbarajan A, Natarajan S, Thirumalai Thangarajan S, et al. Impact of prosthodontic treatment on the oral health-related quality of life in mucormycosis patient: a case report. *Cureus*. 2018;10:1-8.
- 25. Shah RJ, Katyayan MK, Katyayan PA, Chauhan V. Prosthetic rehabilitation of acquired maxillary defects secondary to mucormycosis: clinical cases. *J Contemp Dent Pract*. 2014;15:242-249.

How to cite this article: Artopoulou II, Kalfarentzos E, Polyzois G, Perisanidis C. Prosthodontic restoration of a COVID-19 associated mucormycosis defect: A clinical report. *Spec Care Dentist.* 2022;1-5. https://doi.org/10.1111/scd.12809