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CASE REPORT

Model-assisted marsupialization of a large odontogenic keratocyst in the maxillofacial region using a multicolored 3D-printed model: A novel approach in surgical planning and teaching

Prashanth Konatham Haribabu[1] | **Minaal Verma**[2] | **Akshay Vij**[3]

[1]Specialty Care Unit, Oral & Maxillofacial Surgery, Missouri School of Dentistry & Oral Health, A.T. Still University, Saint Louis, Missouri, USA

[2]Department of Restorative Dentistry & Implant Dentistry, SIU School of Dental Medicine, Alton, Illinois, USA

[3]Specialty Care Unit, Esthetic & Digital Dentistry, Missouri School of Dentistry & Oral Health, A.T. Still University, Saint Louis, Missouri, USA

Correspondence
Akshay Vij, Specialty Care Unit Director of Esthetic & Digital Dentistry, Missouri School of Dentistry & Oral Health, A.T. Still University, Saint Louis, MO, USA.
Email: akshay.vij@gmail.com, akshayvij@atsu.edu

Key Clinical Message

3D printing can improve surgical planning and coordination between the health-care team and serve as a valuable educational tool for students.

Abstract

Although the occurrence of odontogenic keratocysts (OKCs) in the maxillofacial region is not uncommon, their aggressive growth behavior requires advanced surgical techniques for minimal recurrence. The current case report describes the use of a multicolored 3D-printed model as an interactive visual aid for the surgical planning and management of an OKC treated with a minimally invasive surgical decompression technique. A cone-beam computed tomography scan of the patient showed a profound OKC involving the left side of the body of the mandible. A 3D printer was used to print a multicolor resin model of the patient's OKC lesion within the mandible. The printed model was successfully used as a planning tool for surgical intervention (i.e., marsupialization and enucleation) of the OKC. The model was also used as a handheld interactive visual aid for dental students, so they could more effectively understand the anatomical and surgical complexity of the case. The novel use of the multicolor 3D-printed model for treatment of this OKC improved visualization of the lesion during surgical planning and was a valuable teaching tool for educational discussion of this case.

KEYWORDS

3D printing, decompression, marsupialization, multicolored model, odontogenic keratocyst, surgical planning

1 | INTRODUCTION

In 2017, the World Health Organization reclassified keratocystic odontogenic tumors as odontogenic keratocysts (OKCs).[1] These cysts commonly occur in the posterior body and ramus of the mandible. As such, they have the potential to damage bone and adjacent tissues as they expand.[2] Although mostly limited to the bony cortex, a few case reports have reported an OKC in the surrounding soft tissues. In the clinic, patients typically present with pain and a large area of swelling; some discharge may also be evident.[2] In rare instances, the patient may be asymptomatic.[2]

This type of cyst has a high recurrence rate, ranging from 13% to 62%.[3] Therefore, aggressive surgical management strategies are often recommended, such as peripheral ostectomy with chemical cauterization of the marginal bone with Carnoy's solution or en bloc resection of the mandible.[2] However, these procedures may increase morbidity and reduce the patient's quality of life. A more conservative treatment is marsupialization. This decompression technique can be used during the early stages of cyst development as a curative treatment, especially for large unilocular OKC lesions. During this surgical procedure, a window is made in the cortical bone to access the cystic membrane, which is usually composed of parakeratinized tissue, and then the edges of the cyst are sutured to the surrounding oral mucosa.[3] By relieving the pressure from the OKC, subsequent metabolic changes lead to gradual shrinkage of the OKC. This type of cyst is thought to arise through a combination of osmotic pressure and resorption that is compounded by the body's release of prostaglandins and growth factors.[4] Therefore, the marsupialization technique is particularly effective because it decompresses the OKC and relieves the osmotic pressure, while decreasing the released growth factors.[5] As such, changes in environment and cellular metabolism within the OKC are considered contributing factors for reductions in size and volume over time.

Despite obvious benefits for the patient, surgical management of large cysts should be carefully considered to avoid potential damage to vital neural and arterial structures. Further, the complex anatomical structures in the head and neck make excision of lesions in this region particularly challenging. However, emerging 3D image-guided technologies, such as cone-beam computed tomography (CBCT), 3D ultrasound, and magnetic resonance imaging, have been used to help surgeons visually prepare during surgical planning and decision-making that are necessary for a favorable surgical outcome. The current case report describes the use of a multicolored 3D-printed model as an interactive visual aid for the surgical planning and management of an OKC treated with a minimally invasive surgical decompression technique.

2 | CASE PRESENTATION

A 13-year-old African American male presented to a specialty care clinic at a Midwestern dental school with his mother: the patient had been referred to the clinic by his general dentist. The patient had swelling in the lower left side of his jaw and face (Figures 1–3). A thorough extraoral and intraoral evaluation was performed, and a 2D panoramic radiograph was obtained. A large (7 cm × 4 cm) expansile bony lesion (buccolingually) was found on the left body of the mandible, extending below the dental roots and displacing them mesiodistally (Figure 4). The lesion was not tender or pulsatile and was firm on palpation. Tooth #20 was tender on percussion and depressible on evaluation. No exudate, suppuration, or neurosensory changes involving the lingual nerve or inferior alveolar nerve were documented. Intact dental pulp vitality, assessed using warm and cold stimuli, was evident.

FIGURE 1

FIGURE 2

Advanced imaging using a 3D CBCT scan indicated the anterior–posterior and buccolingual expansion and overall extent of the expansile lesion. Imaging also showed the inferior alveolar nerve bundle was displaced to the inferior border of the mandible (Figures 5–10). After obtaining informed consent from the patient and his parent, an incisional biopsy was performed to determine the

FIGURE 3 Extraoral and intraoral swelling on the lower left side of the jaw and face.

FIGURE 4 Panoramic radiograph showing a large radiolucent lesion on the left side of the mandible.

histopathology of the growing lesion. As a result, the bony lesion was diagnosed as an OKC.

The OKC in the current case was large, a decision was made to first debulk or reduce the size of the cystic lesion using marsupialization. Thereafter, the plan involved enucleation with a moderately aggressive peripheral ostectomy. To better visualize and plan this surgical approach, we used a 3D-printed model of the patient's mandible, where the expansile lesion was demarcated with a different color to show the extent of the lesion. This novel approach of creating a printed presurgical model of the pathological lesion allowed us to educate the surgeon, residents, and dental students about navigation around the vital structures close to the lower border of the mandible, such as the neurovascular bundle of the inferior alveolar nerve.

FIGURE 5

FIGURE 6

FIGURE 7

To create the 3D-printed model, the various anatomical structures from the CBCT scan were segmented using the CBCT software (Invivo, Anatomage, Inc.). An STL file was produced that was then printed with a Stratasys J750 printer (Eden Prairie, MN). Advanced multicolor 3D printing was used to create a visual aid that showed the demarcation of borders between specific anatomical structures (Figures 11–13). The OKC lesion was printed in red to provide better contrast with the surrounding tissues and structures. The multicolor model also provided the operating oral surgeon with an accurate visualization of all borders of the OKC and the anatomic changes to the surrounding structures caused by the lesion, visually highlighting

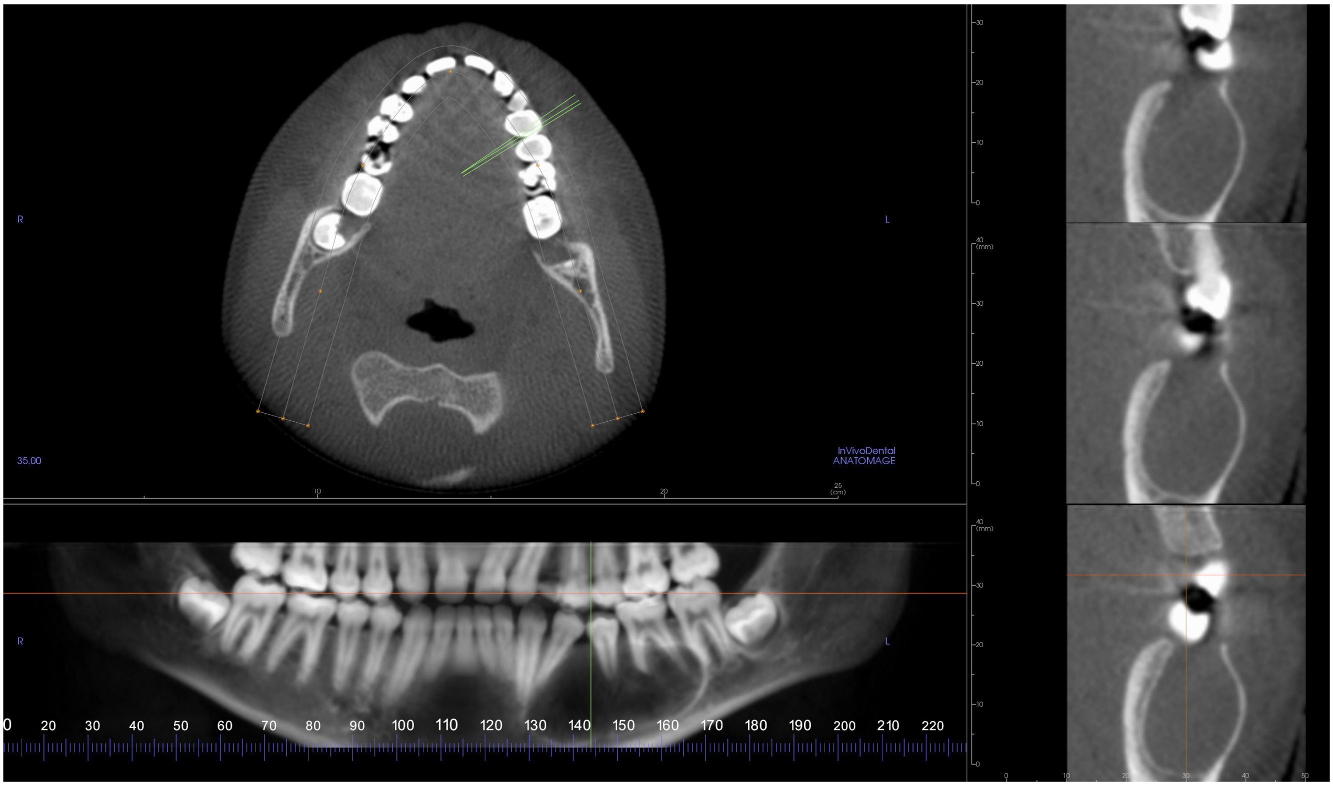

FIGURE 8

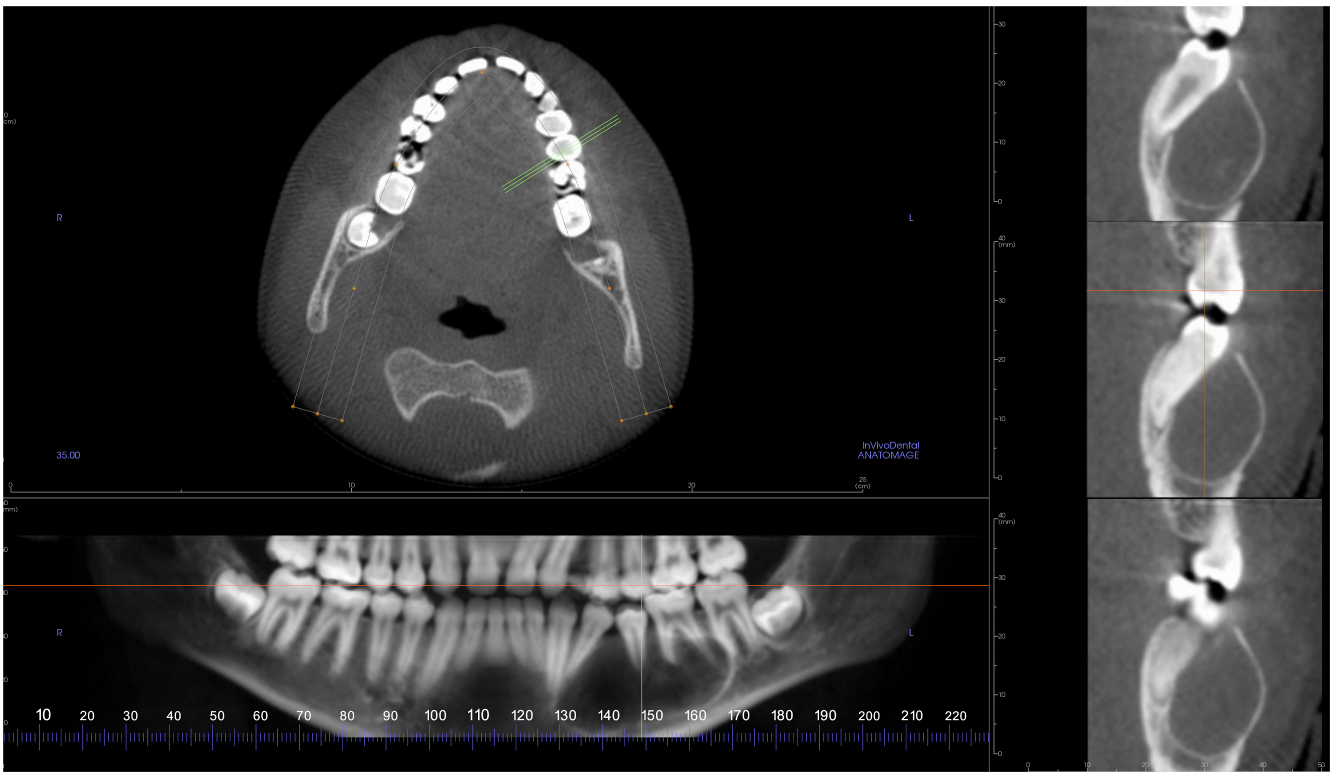

FIGURE 9

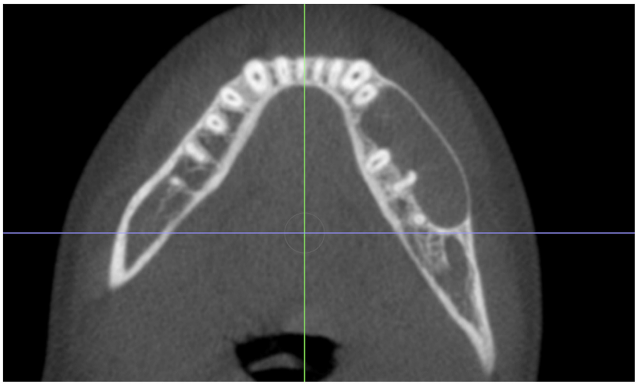

FIGURE 10 CBCT image scans showing the size and extent of the OKC lesion in three dimensions.

FIGURE 11

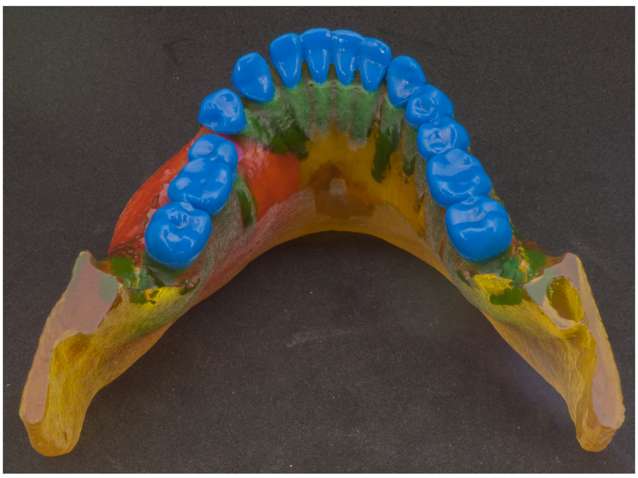

FIGURE 12

the demarcation between pathological and physiological structures.

During the surgical procedure, a lateral (buccal) window was created to access the lesion with minimal morbidity

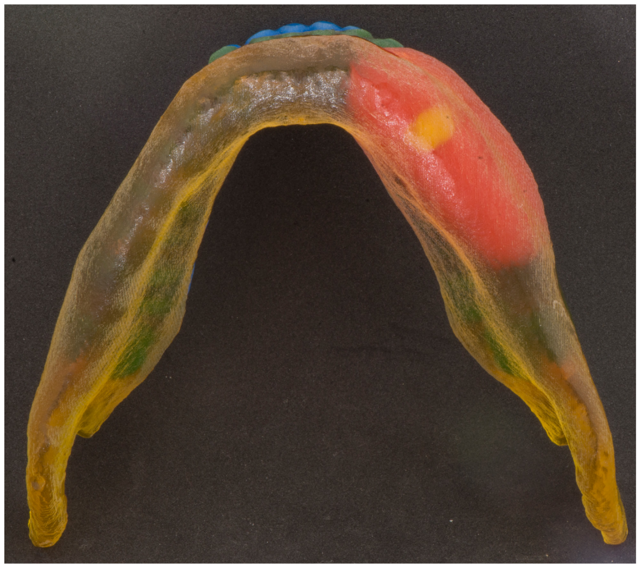

FIGURE 13 Multicolored 3D-printed model to visualize the OKC lesion of the current case.

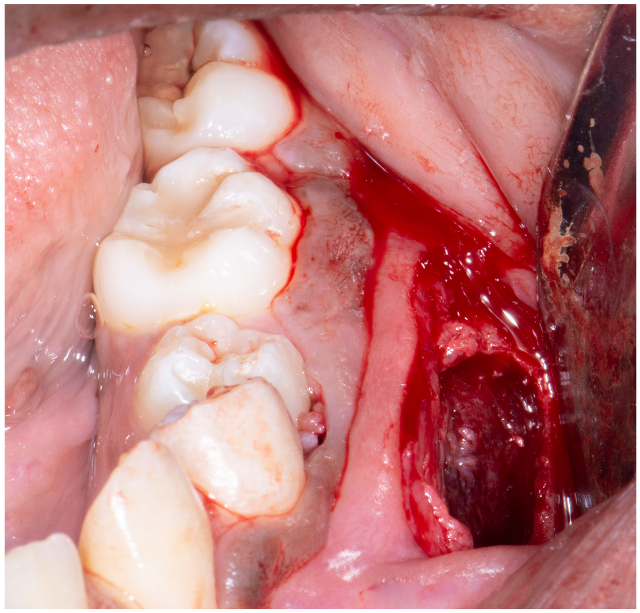

FIGURE 14 Lateral access window.

(Figure 14). An adequately measured nasal trumpet (size 28 [9.3 mm] Robertazzi Nasopharyngeal Airway, Teleflex) was modified, so it could be used as an irrigation device to maintain patency of the cystic cavity and facilitate irrigation (Figure 15). The trumpet was secured into the cystic lining using 3.0 silk sutures (Covidien, Medtronic).

The patient was reevaluated periodically every month for the first 3 months to check the patency of the irrigation trumpet. Thereafter, the patient was evaluated once every 3 months until the cystic lesion had shrunk and there was marked bone deposition and reversal of the expansion of mandibular cortices. At a follow-up appointment 11 months after marsupialization, 2D intraoral and 3D CBCT imaging scans confirmed shrinkage of the previously large OKC lesion and successful deposition of alveolar bone (Figures 16–19). At the same appointment, clinical evaluation indicated that the lower left second premolar (#20) was mobile (Grade II), likely because of severe loss of supporting periodontal bone. Radiographically, remnants of shrunken cystic lining were visible. The risks, benefits, and alternatives of enucleation and the future need for prosthetic replacement were discussed with the patient and his parent. As a result, tooth #20 was extracted, including the fragments of cystic lining adhering to its roots. Careful decortication of surrounding alveolar bone was also performed at that time. At a follow-up appointment 18 months after enucleation, 3D radiographic and intraoral clinical imaging showed uneventful and satisfactory healing (Figures 20–30). For educational and evaluation purposes, a multi-colored 3D-printed model was produced again, using a manufacturing process similar to the pre-operative model, using the postoperative 3D CBCT images (Figures 31–35).

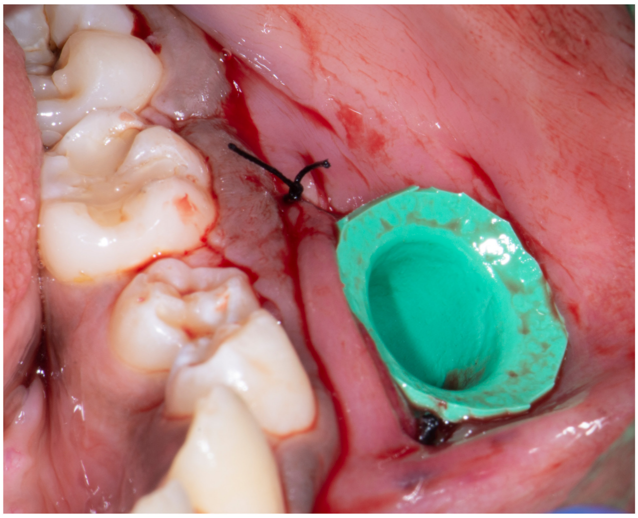

FIGURE 15 A nasal trumpet was secured into place to maintain patency.

3 | DISCUSSION

The current case report described the use of a multi-colored 3D-printed model as an interactive visual aid for the surgical planning and management of an OKC treated with minimally invasive surgical decompression technique. Further, our use of this model was beneficial during treatment, especially since the OKC was a unilocular lesion. The successful outcome of this case suggests that

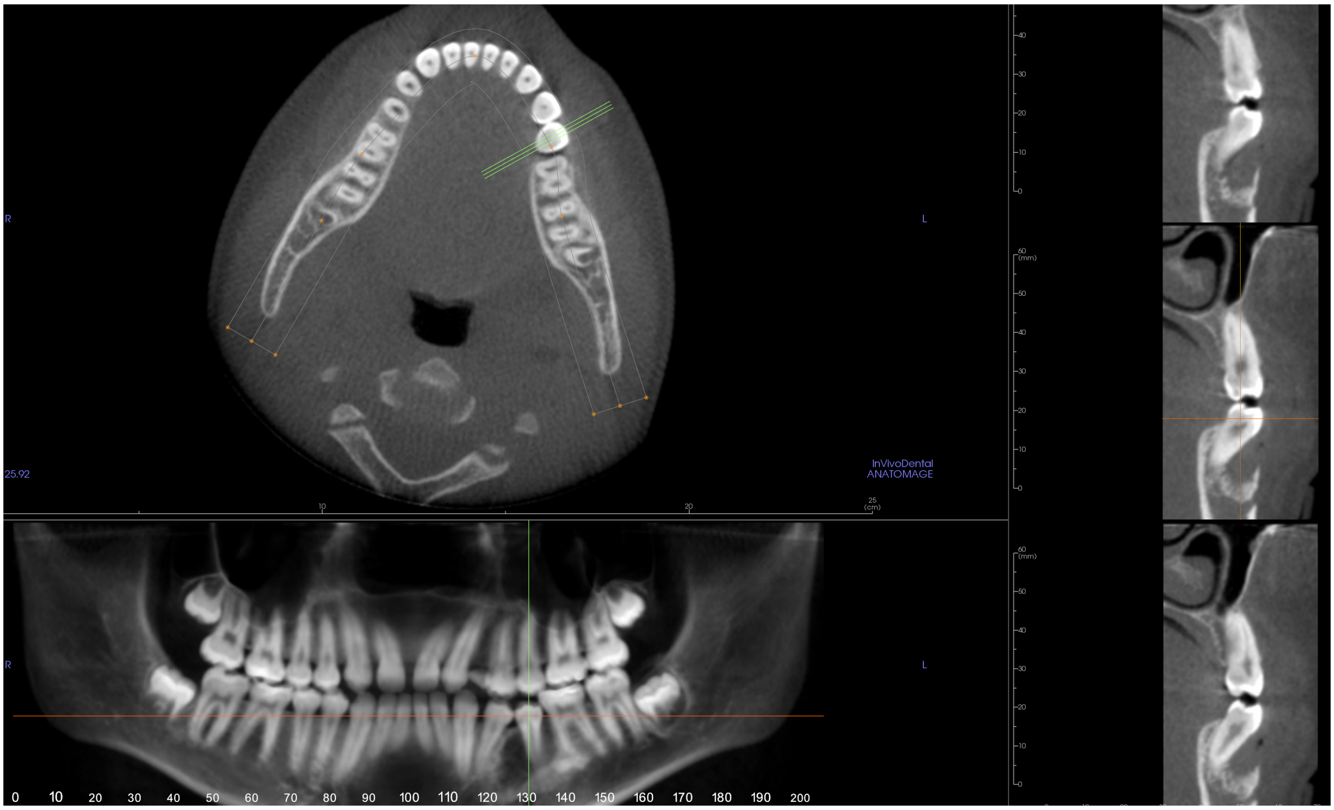

FIGURE 16

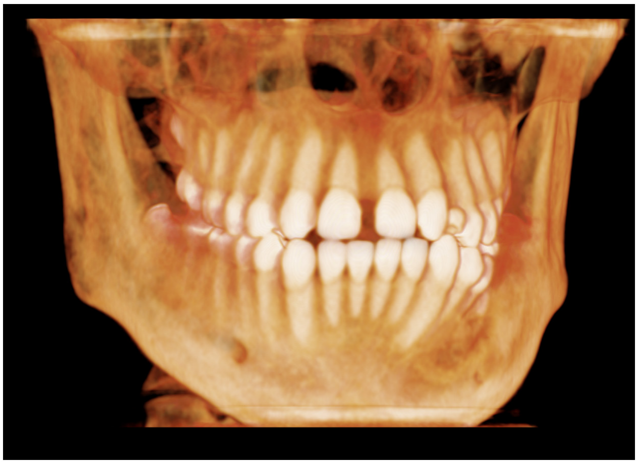

FIGURE 17

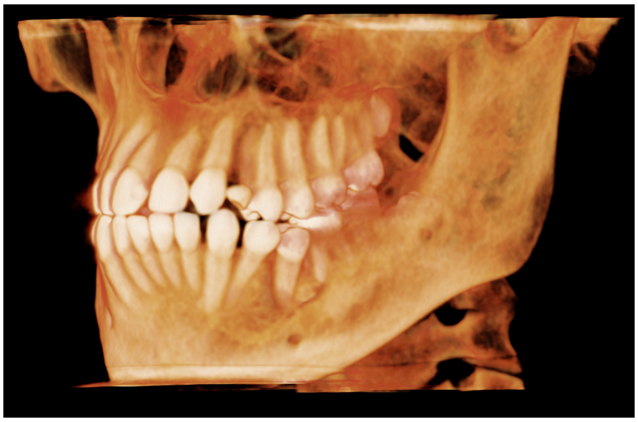

FIGURE 18

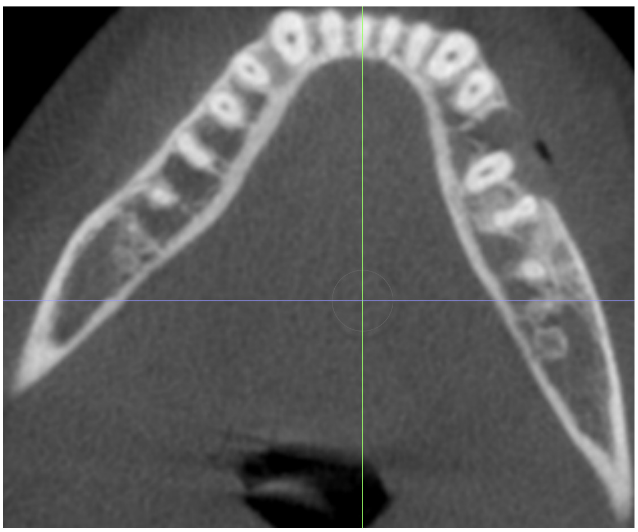

FIGURE 19 Follow-up CBCT scans at 11 months after marsupialization showing the reduction in the size of the OKC lesion.

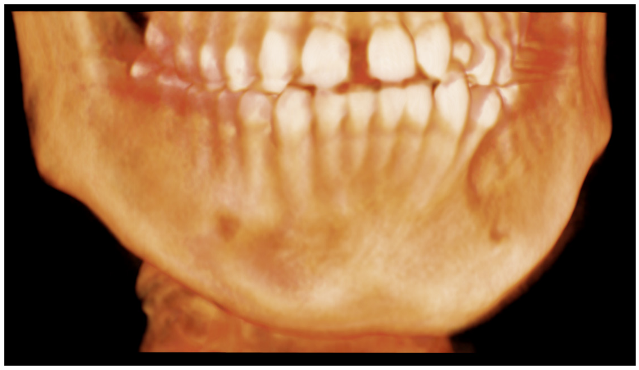

FIGURE 20

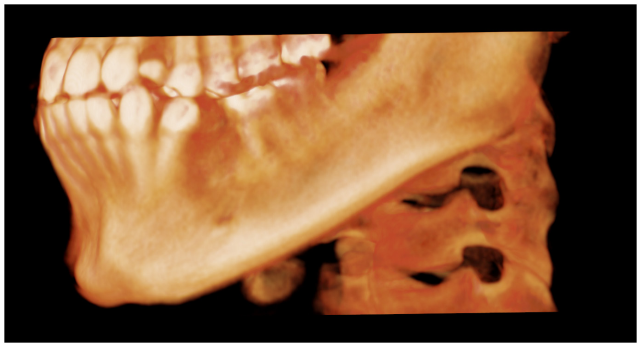

FIGURE 21

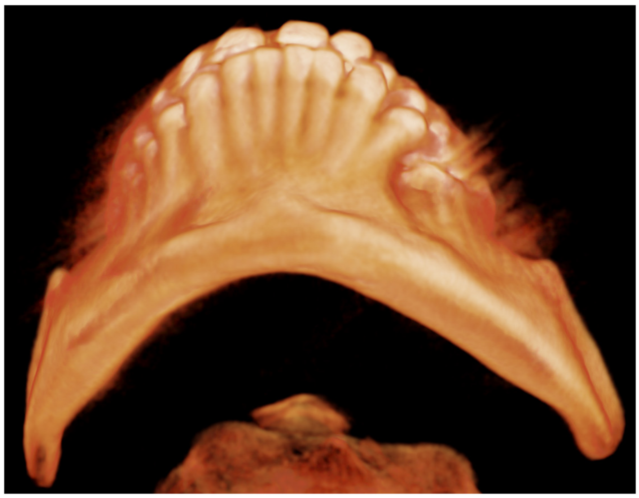

FIGURE 22

multicolored 3D-printed models can effectively support treatment that combines marsupialization and subsequent enucleation. In cases of a multilocular OKC, a 3D-printed stereolithic model would likely be beneficial for surgical planning and procedures. However, a multilocular OKC will likely require a different surgical approach than the one used in the current case report.

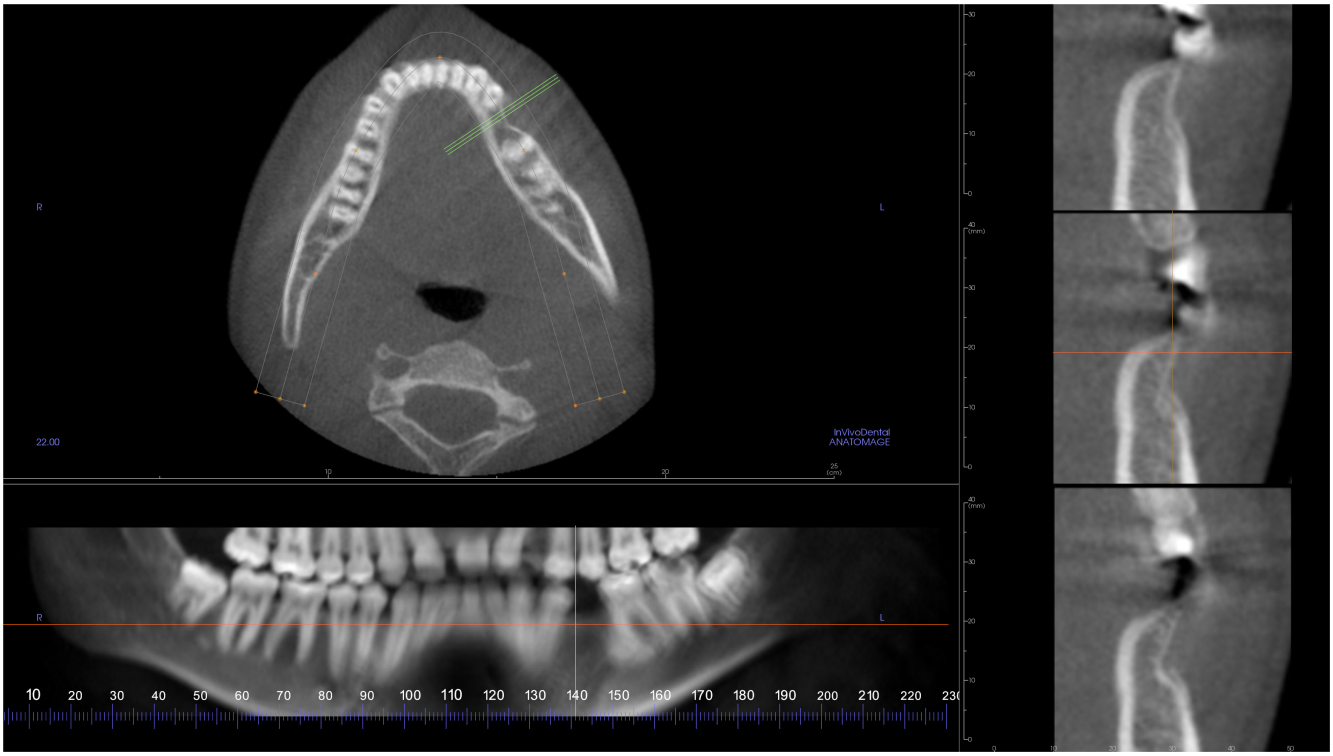

FIGURE 23

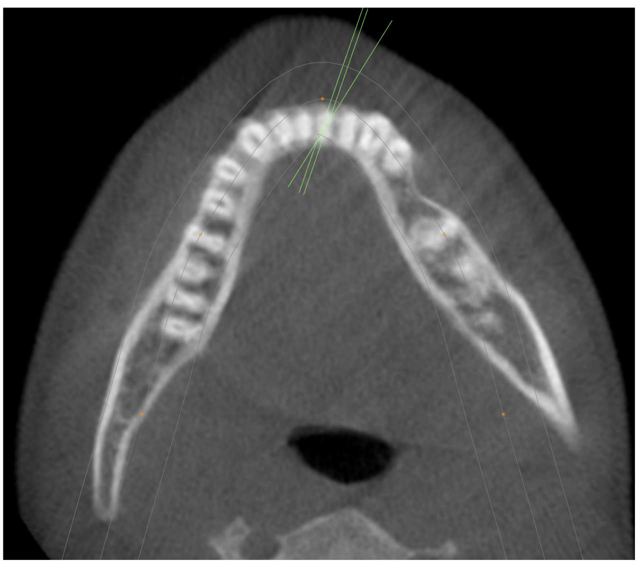

FIGURE 24

By using complex algorithms to layer 2D radiographs into a 3D data set, CBCT scans have over 99% accuracy representing oral structures.[6] For instance, standard deviations range from 12 to 200 μm for full mandibular arch scans and from 7 to 80 μm for a single tooth, which suggests accuracy is better within a narrower field.[6] Because of the processing power of modern computers, CBCT scan data can be digitally printed directly from the data set using STL files.[6] These innovations make surgical planning easier, which can lead to better patient outcomes.

The first step of successful surgical planning is to accurately simulate the patient in a way that is easy to visualize and understand. Therefore, multicolored 3D-printed models, such as that used in the current case report, have key advantages. From a surgical viewpoint, 3D printing and 3D-printed models are valuable for surgeons because they provide a manufactured replica of each patient that illustrates the patient's unique anatomy using a clinically relevant 1:1 ratio of oral structures.[7] When used for surgical planning and procedure simulation before surgery, these models become vital tools. By studying the anatomy preoperatively, clinicians can plan a predictable approach and anticipate challenges with potential solutions, which may reduce operating time, minimize errors, and alleviate mental fatigue of the surgeon.[8] Research suggests that many oral and maxillofacial surgeons believe the use of 3D-printed models greatly enhances their confidence in preoperative planning and choice of appropriate surgical technique.[9]

From an educational viewpoint, 3D-printed models are an important visual aid for patients. They can be used to explain the pathology of the patient's condition and to describe the risks, benefits, and alternatives to surgical intervention, allowing the patient to make more informed decisions about their health and well-being.[9] When used as part of a dental school curriculum, 3D models provide students and residents with a deeper understanding of

FIGURE 25

FIGURE 26

FIGURE 27

FIGURE 28

oral structures through visual and tactile examination which in turn facilitates correlations between theoretical/didactic concepts and clinical scenarios in the absence of a patient. Models can also be used for standardization of teaching and testing, so that all students at undergraduate and postgraduate levels are able to participate equally in learning experiences.[8]

FIGURE 29

FIGURE 30 Clinical and radiographic images showing satisfactory healing 18 months after enucleation.

FIGURE 31

FIGURE 32

FIGURE 33

Despite the many advantages of 3D-printed models for surgical planning, they cannot replace a surgeon's clinical judgment and skill. The success of any surgery depends on the surgeon's knowledge and previous training and experiences. Also, 3D printers may not accurately produce a stereolithographic model that exactly matches the CBCT scan of the patient. When considering the accuracy of a CBCT scan and subsequent 3D-printed model of the scan, reported standard deviations have ranged from 165 to 386 μm.[10] However, research has shown that CBCT scans have clinically acceptable results for presurgical planning and stereolithographic transfer.[10] Therefore, 3D models should be considered as a valuable adjunct for enhancing the efficiency, accuracy, and creativity of oral surgery. Using multicolored 3D-printed models, as in the current case, may be an asset for improving surgical navigability

FIGURE 34

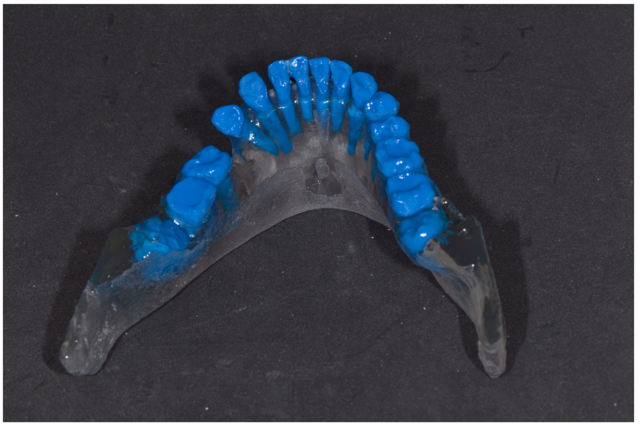

FIGURE 35 Multicolored 3D-printed model to visualize the 2-year postoperative anatomy of the mandible.

around distorted natural anatomy caused by growing lesions. Certainly, their benefits during surgical preparation should not be underestimated.

Several 3D-printing technologies are available for fabricating these models. Some methods use high-energy radiation (laser, ultraviolet, or electron beam) to polymerize or sinter the printing material; others use a process based on extrusion or jetting.[11] Examples of 3D-printing technologies are stereolithography, selective laser sintering, fused deposition modeling, photopolymer jetting, and powder binder printing. In the current case report, we used the photopolymer jetting technology to create our multicolored model since it is a relatively fast method that prints objects with high resolution and quality. For this printing technology, photopolymers and supporting materials are jetted from an inkjet-like printhead on the build platform. When printing is complete, the object is cured and solidified by ultraviolet radiation. When compared with stereolithography and fused deposition modeling printing technologies, photopolymer jetting was a robust method for the fabrication of full-arch dental models and had the highest accuracy.[12] Further, maxillary and mandibular models manufactured with photopolymer jetting were accurate enough to be used for the fabrication of customized patient-specific orthodontic appliances.[12] Therefore, models like the one used in the current case should be considered for a variety of oral health uses because they provide an accurate, individualized model for each patient.

Given its ease of use and demonstrated accuracy, 3D printing is advancing surgical aspects of many oral maxillofacial approaches to treatment. In the current case report, the extent of the OKC lesion was limited to the bone, so we did not incorporate soft tissue in our 3D-printed model. However, representation of soft tissues can easily be included in such models, providing similar visual benefits for surgical planning. For example, flap design and the extent of soft tissue reflection can be provisionalized when incorporated into stereolithographic models. Further, the anatomy of major blood vessels and nerves can also be mapped with multicolor 3D printing of soft tissues. This inclusion of hard and soft tissues in 3D-printed models may be beneficial in clinical and educational settings. When teaching colleagues and students about flap design, the oral surgeon could use a model to illustrate how to avoid damage to the patient's vital structures.

Ideally, future advancements of 3D-printing technologies will be applied to full surgical simulations and, like current 3D-printed models, should provide substantial benefits when compared with benchtop observation and planning. Likewise, these technologies may enhance the training and education of resident surgeons by illustrating the gross anatomy and localized relevant structures of the patient. Because this technology allows for fast and cost-effective mass production of models that simulate individual patients, the need for cadavers in the dental school curriculum may be reduced.

4 | CONCLUSION

In the current case report, a multicolored 3D-printed model was used as an interactive visual aid for the successful surgical planning and management of an OKC treated with minimally invasive surgical decompression technique. By using 3D-printing technology to create a model from the patient's CBCT scan, we were better able to visualize and diagnose the OKC lesion. The incorporation of such models into dental practice may improve surgical planning for oral and dentofacial procedures, especially those involving critical areas near dentoalveolar structures. The novel approach used in the current case also allowed us to educate the surgeon and dental students and residents about navigation around the vital structures close to the lower border of the mandible, such as the neurovascular bundle of the inferior alveolar nerve. Therefore, the use of 3D-printed models should also be considered for educational purposes. Although cysts have previously been treated using a combination of marsupialization and enucleation surgical techniques and 3D printing is increasingly being used in the dental and medical arenas, the use of the combination of these techniques along with pre- and post-3D production of the jaw models makes delivering a high standard of care to patients easier and planning and educating dental specialists easier, more intuitive, and repeatable. For example, this methodology can be incorporated as part of the live interactive teaching armamentarium in the dental school curriculum, and they can be used to provide a platform for treatment discussions that educate the patient and surgical team.

AUTHOR CONTRIBUTIONS

Prashanth Konatham Haribabu: Conceptualization; data curation; investigation; resources; supervision; writing – original draft; writing – review and editing. **Minaal Verma:** Conceptualization; writing – original draft; writing – review and editing. **Akshay Vij:** Conceptualization; data curation; methodology; resources; software; supervision; validation; writing – original draft; writing – review and editing.

CONFLICT OF INTEREST STATEMENT

The authors declare that they have no conflicts of interest related to the contents of this manuscript.

DATA AVAILABILITY STATEMENT

Data sharing not applicable-no new data generated, or the article describes entirely theoretical research.

CONSENT

Written informed consent was obtained from the patient's parent to publish this report in accordance with the journal's patient consent policy.

ORCID

Akshay Vij https://orcid.org/0000-0002-6342-8755

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