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Enucleation and surgical stent as a treatment strategy for a large unicystic ameloblastoma: Case report and review of literature

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ABSTRACT

INTRODUCTION: Ameloblastoma is a benign neoplasm of odontogenic origin with local invasive characteristics and a high recurrence rate. It compromised 1% of the jaw's cysts and tumors with only 10–15% in children.

PRESENTATION OF CASE: A 14-year-old boy sought treatment for a painless swelling involving the right side of the face started one year ago. The intra-oral examination displayed a firm mass associated with 46, 47 teeth, and the angle of the mandible. The radiographic examination revealed a large well-defined homogeneous radiolucency extending from the 46 region to involve the angle and extending towards the coronoid and condylar processes. An incisional biopsy confirmed the diagnosis of unicystic ameloblastoma. Treatment was planned according to the patient's age: Phase I: Surgical enucleation. Phase II: Construction and insertion of a surgical stent. Phase III: Construction of a transitional acrylic Kennedy class II partial denture. Phase IV: the patient is scheduled for a definitive implant-supported prosthesis at 18 years old.

DISCUSSION: The management of ameloblastoma is influenced by the age of the patient, the extension, duration, and position of the lesion, and the histopathological variants. Several authors recommended enucleation as a conservative treatment approach to eliminate the esthetical, functional, and psychological squeals associated with the radical approach. The use of a surgical stent protects the enucleated cavity and promote tissue healing.

CONCLUSION: Enucleation and subsequent surgical stent not only eliminates the disease, but also preserves the bone structure, prevents the facial disfigurement, and significantly improve the patient's esthetic, mastication, oral health, and quality of life.

1. Introduction

Ameloblastoma is a benign neoplasm of odontogenic origin with locally invasive characteristics and high recurrence rate [1–4]. Although it is slow-growing, it tends to attain a considerably large size causing severe facial disfigurement and functional implications [3,2–4]. It was first reported by Cusack in 1827 and named by Ivy and Churchill in 1930 [5].

It comprises 1% of all cysts and tumors of the jaw and 11–18% of all odontogenic tumors [6]. It is the most prevalent odontogenic tumor in developing countries [7] with a particularly high incidence in Africa and China [8]. Though only 10–15% cases of ameloblastoma occur in children, a higher percentage (25%) has been reported in Africa and Asia [9].

The historical background of ameloblastoma in children was first reported by Small and Waldron in 1955 [10]. They investigated 1036 cases of ameloblastoma, of which only 19 (2%) occurred in patients aged less than 10 years and 75 (9%) in those aged 10–19 years [10].

Ord et al. [11] revealed that the average age of African children with ameloblastoma is 14.7 years, and it shows a slight male predominance (1.4:1). Although the mandible is the most affected jaw, and 85% adult cases occur in the third molar region, African children show a higher incidence (44.2%) of symphysis involvement. Moreover, the multicystic ameloblastoma is more prevalent (59.8%), while the prevalence of unicystic ameloblastoma is only 19.5%.

Though ameloblastoma is believed to originate from the cells of dental lamina [12], several etiological factors including local trauma, inflammation, nutritional deficiency, irritation from extraction, and dental caries have been proposed. It is also proposed

Abbreviation: OPG, Orthopantomogram.

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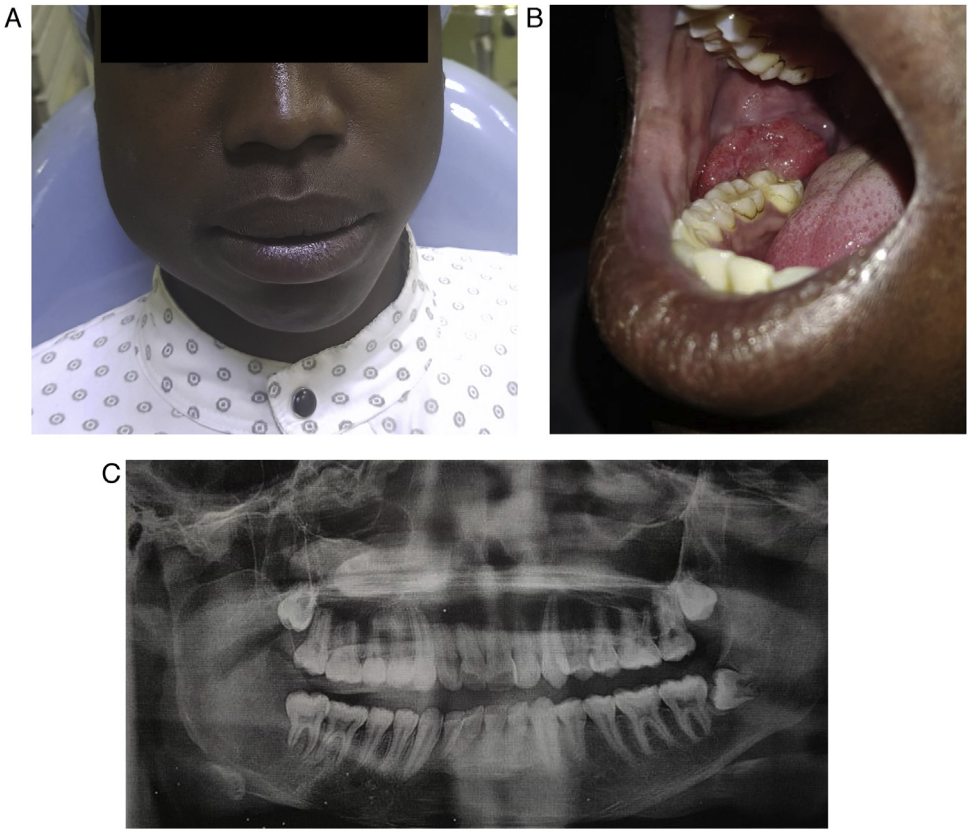

Fig. 1. Patient's clinical presentation.
A. The patient's extra-oral view at the time of presentation shows obvious swallowing of the patient's face's right-hand side, resulting in marked facial asymmetry.
B. The patient's intraoral view shows a painless growth involving the posterior mandible area associated with 46,47teeth and the mandible angle.
C. Panoramic radiograph view showing a well-defined, homogeneous radiolucent lesion extending from the 46 regions toward the coronoid and condylar processes.

to originate from the remnants of odontogenic epithelium and lining of odontogenic cysts [3,13]. A recent theory has also elaborated the role of genetic mutation [3,14,15].

In 2005, the World Health Organization classified ameloblastoma into multicystic (91%), unicystic (6%), peripheral (2%), and desmoplastic (1%) subtypes based on the histopathological features and anatomical location [6,16]. A recent classification has categorized into conventional (85%), unicystic, and peripheral (1%) types [3,17].

The unicystic ameloblastoma is a neoplasm with cystic involvement of the ameloblastic epithelium, and the tumor grows into the lumen and fibrous connective tissue [16] It is classified into luminal, intraluminal, and mural subtypes [3]. It is the second-most common type of ameloblastoma (5–10%), is associated with younger patients and sometimes with unerupted teeth.

The prompt diagnosis and appropriate treatment of ameloblastoma requires the use of orthopantomogram (OPG), cone-beam computed tomography, computed tomography, and/or magnetic resonance imaging [3]. Furthermore, histopathological examination confirms the diagnosis and differentiates among the distinctive subtypes [1–3].

The appropriate management strategy for ameloblastoma remains controversial due to the locally invasive and aggressive nature and high recurrence rate [1–3]. The treatment modalities are dictated by patient age [1,3,4], extension, duration, and location of the tumor [2–4,6], degree of anatomical involvement such as penetration of the cortical bone [3], and histological classification [3,4].

Surgical and non-surgical therapeutic techniques have been proposed [6]. The non-surgical techniques emphasize the use of radiation therapy in inoperable cases such as in medically compromised patients [6,18]. Helical tomotherapy and image-guided or intensity-modulated radiation therapy are the common modes of radiation therapy. Additionally, proton-beam therapy can be used as adjunctive therapy with or without chemotherapy, especially in ameloblastic carcinoma and after multiple postsurgical recurrences [6].

The following two surgical approaches have been advocated: a conservative approach (including marsupialization, enucleation, or enucleation with curettage) and a radical resection approach [1–4]. Although the conservative approach maintains the integrity of the bone and the consequent growth pattern, it is associated with a high recurrence rate [1]. Enucleation is the dissection of an intraosseous cavity [19], having an esthetic priority, as radical treatment may result in significant esthetic, functional, and psychological sequelae that necessitate a reconstruction procedure to restore the patient's quality of life [1]. Gülşen et al. [20] reported a case of a calcifying epithelial odontogenic tumour that was treated successfully with decompression, accompanied with a saline cuff as a tube for irrigation.

A surgical stent is a device used to apply pressure on the soft tissues to facilitate healing and prevent cicatrization or collapse by providing support for the anastomosed structures [21]. It can effectively prevent wound contamination, reduce infection susceptibility, and promote tissue healing, which are of considerable importance when enucleation is performed [22].

This report aims to emphasize the favorable result of enucleation as a treatment modality in a young patient with unicystic ameloblastoma and the utility of surgical stent during the corresponding healing phase. To our knowledge, this is the first case

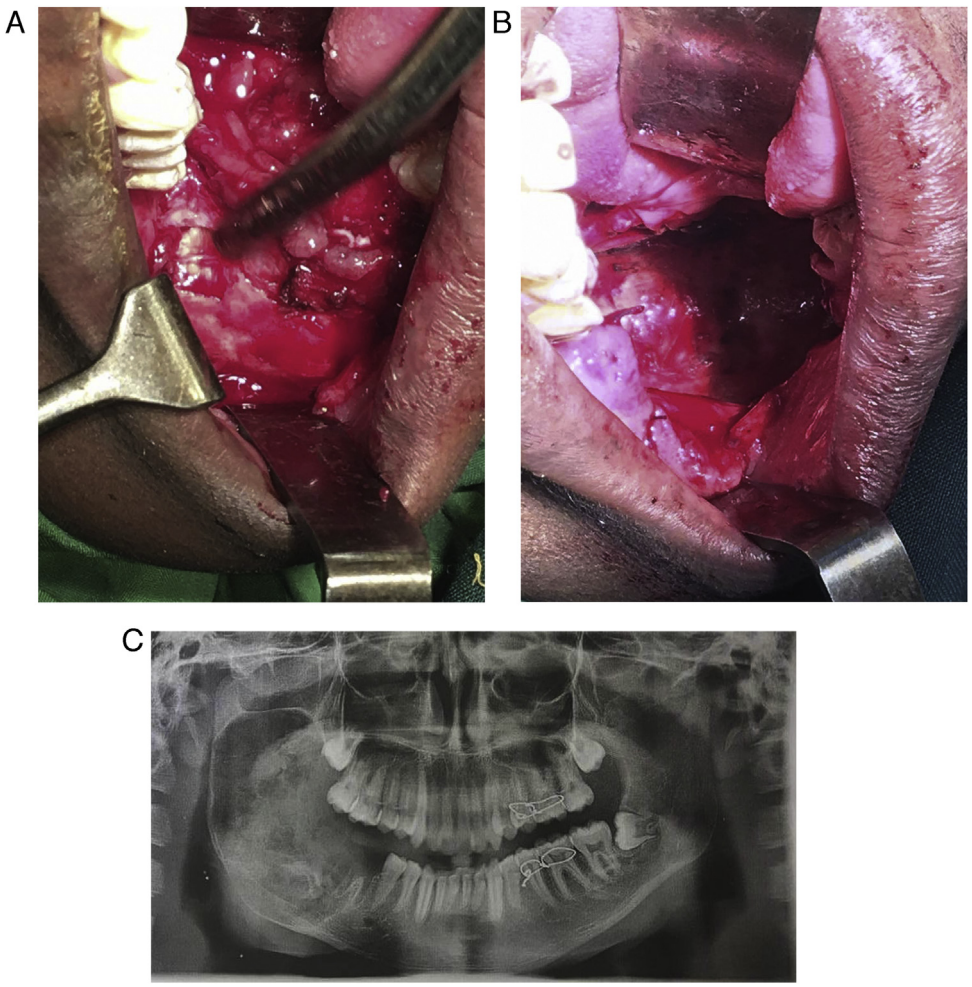

Fig. 2. Surgical treatment total enucleation.
A. Identification of the tumor extension.
B. The intraoral view showing the cavity after complete enucleation of the lesion.
C. Panoramic radiograph after the enucleation showing supporting wire at the area of 25,26 and 35,36, and extraction of 46 and 47 due to association with the lesion.

report to describe the use of a surgical stent in the treatment of ameloblastoma.

2. Presentation of case

A 14-year-old boy presented at our hospital complaining of swelling on the right side of his face (Fig. 1.a). The patient was a non-smoker with no systemic disease and was not taking any medication. Extraoral examination revealed facial asymmetry owing to the presence of a painless mass, which started developing one year ago and involved the right posterior region of the mandible (Fig. 1.b). Intraoral examination displayed a bony mass with egg-shell cracking areas covered with normal mucosa and associated with the mandibular right first and second molars and angle of the mandible. OPG revealed a well-defined, homogeneously radiolucent lesion extending from the right first molar region to the coronoid and condylar processes (Fig. 1.c).

An incisional biopsy was performed, and the histopathological findings were definitive for ameloblastoma. A multidisciplinary team (Maxillofacial Surgeon and Prosthodontists) was formed, and all possible treatment options were discussed with the patient, including surgical enucleation and radical mandibulectomy. The patient demonstrated a high esthetic concern and desired a conservative approach. Hence, the following treatment plan was formulated: Phase I: Enucleation of the tumor. Phase II: Fabrication of a surgical stent. Phase III: Fabrication of a transitional acrylic partial denture. Phase IV: Prosthetic restoration using a definitive implant-supported prosthesis. The ethical approval was obtained from the ethical committee of the Khartoum Dental Teaching Hospital, Federal Ministry of Health, Khartoum, Sudan. The research was registered at the Research Registry with the unique identifying number: researchregistry6210. Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

2.1. Treatment

Surgical enucleation was performed under general anesthesia without primary closure by an oral and maxillofacial surgeon; the first and second molars involved in the tumor were extracted (Fig. 2a–c). Antibiotics and analgesics were prescribed. The patient was referred to the department of prosthodontics for the fabrication of a space-occupying stent. An impression of the defect opening with the surrounding area was recorded using condensation silicone impression material, and a working cast was obtained (Fig. 3a–c). A space-occupying heat-polymerized acrylic stent was fabricated, with three wrought-wire c-clasps on mandibular right second premolar and left first premolar and molar (Fig. 3d–f). The stent was inserted, and complete seal of the defect opening was ensured using pressure indicating paste (Fig. 3g). Oral hygiene

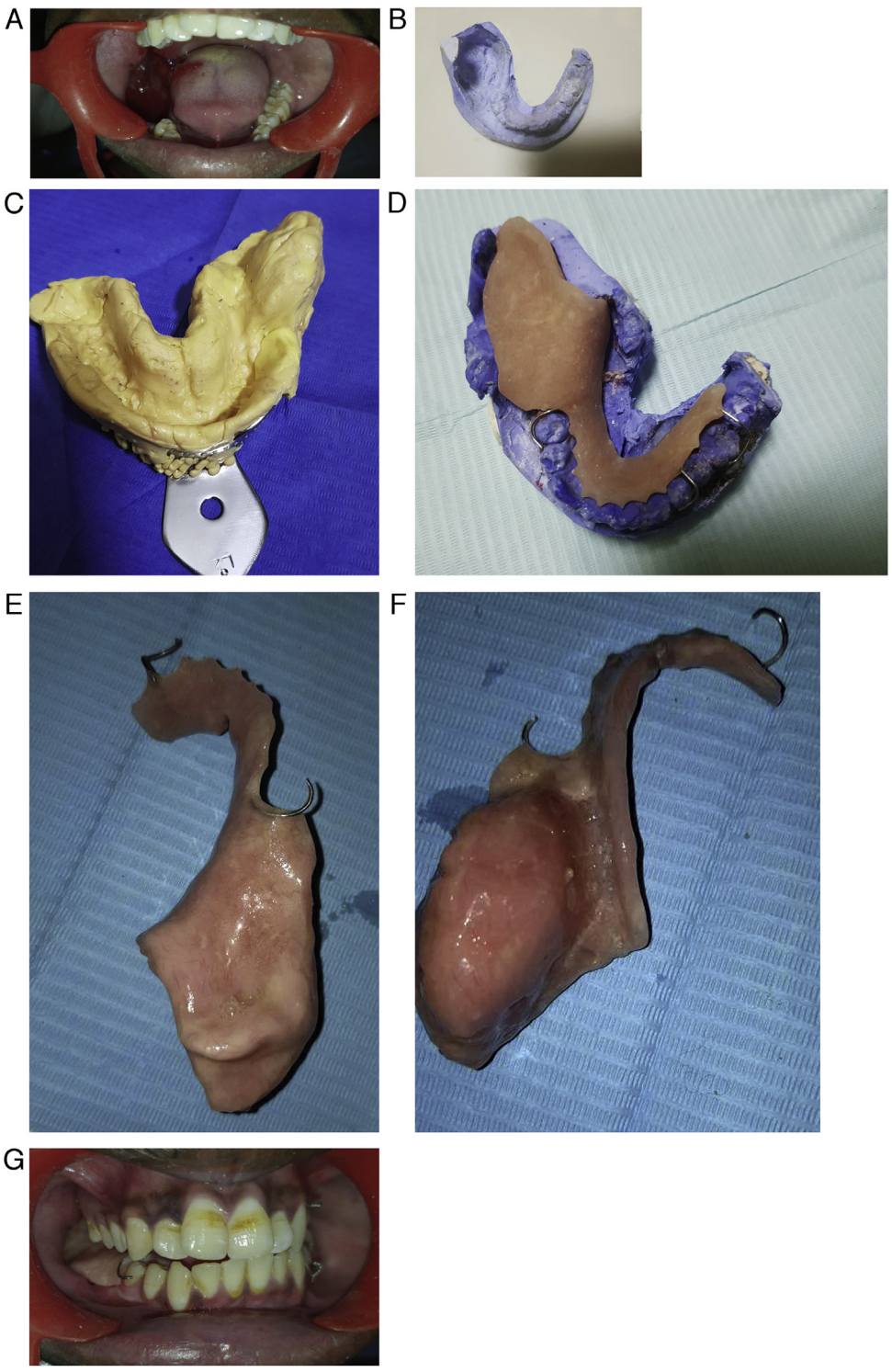

Fig. 3. Stent construction.
A. The intraoral view of the patient showing the enucleated cavity one week after the surgery.
B. A photograph showing a lower impression using condensation silicone impression material and stock tray for the enucleated cavity and the surrounding area.
C. A photograph showing cast obtained from pouring the impression.
D. A photograph showing the processed heat cure acrylic stent on the cast.
E. A photograph showing the polished surface of the stent.
F. A photograph showing the tissue surface of the stent.
G. The intraoral view showing the stent after inserted inside the patients' mouth.

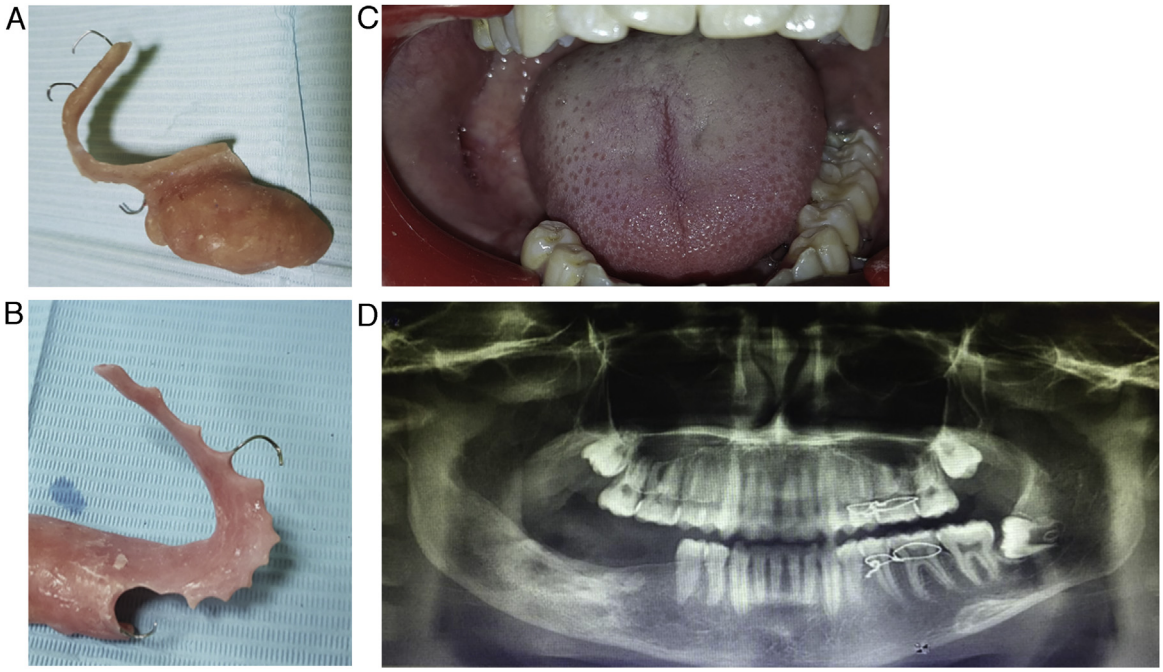

Fig. 4. The adjustment of the Stent during the follow- up visit.
A. A photograph showing the tissue surface of the stent after adjustment in 1 month follow up visit. "note the reduction of the mass".
B. A photograph showing the tissue surface of the stent after adjustment at 8 month follow up visit.
C. The intraoral view of the patient showing complete healing of the defect.
D. Panoramic radiograph after one year showing bone apposition at the defected area.

instructions were provided, and regular follow-up appointments were scheduled at biweekly intervals. At each follow-up appointment, the healing was monitored, and consequently, the tissue surface of the stent was adjusted (Fig. 4a, b).

After eight months, complete healing was noted (Fig. 4c), and OPG displayed bone deposition in the defect without any signs of recurrence (Fig. 4d). A transitional acrylic Kennedy's Class II partial denture was fabricated (Fig. 5a–e). The patient was provided with oral hygiene instructions and scheduled for follow-up appointments at six monthly intervals. Prosthetic restoration with implants is planned when he would be 18 years old. The patient had no complaints during the follow-up period and reported improvements in mastication, speech, and esthetic. After 2 years of follow-up, the patient was highly satisfied with the treatment outcome (Fig. 5f). This case has been reported in line with the SCARE criteria [23].

3. Discussion

Management of ameloblastoma presents a challenge amplified in young patients due to the high local morbidity and recurrence rate [3]. The treatment should aim to eliminate the disease with minimal influence on the patient's oral health, functions, and quality of life [2,3,24–27].

A multidisciplinary team management provides optimum treatment in terms of disease resolution as well as the consequent functional, esthetic, and psychological rehabilitation [2].

The age and sex of our patient match the demographical data reported in the literature [11]. Most ameloblastic lesions (70%–80%) resemble dentigerous cysts radiographically [3,6,10,11]. In this case, histopathological analysis confirmed the diagnosis [3,10,11].

Several factors govern the selection of the optimal treatment modality [1,2,10,11]. The continuous growth and facial bone physiology in children characterized by a higher percentage of cancellous bone, bone turn-over, and periosteal activity affect the management approach considerably [1,11]. In this case, the patient was 14 years old and had high esthetic and functional concerns with a fear of resection sequelae. Thus, the selected treatment was considered as the best treatment option that matched the recommendations of several authors to preserve the patient's oral health, reduce facial disfigurement, and maintain the patient's quality of life [1,3,4,6,19,20,24,27,28].

A surgical stent used to occupy the space created after enucleation prevents fluid and food impaction, protects the underlying tissues, ensures oral hygiene maintenance, eliminates the inconvenient use of a gauze pack, promotes tissue healing, and ensures equal bone deposition and healing from the periphery, i.e., eliminates dead space [21,22]. Thus, the use of a surgical stent revealed significant advantages.

Despite the general agreement for the use of conservative therapy in young patients, [1,2,4,23,25,27] the high recurrence potential of ameloblastoma necessitates a longer and intensive follow-up program, possible only by a satisfactory patient-dentist collaboration [1,2,27]. Hirschhorn et al. [27] recommended a 5- to 20-year follow-up period. The use of a transitional removable prosthesis permits the direct visualization of the margins of the lesion for any evidence of recurrence.

4. Conclusion

The reported treatment approach preserves bone structure, eliminates facial disfigurement, as well as significantly improves the patient's esthetics, masticatory ability, oral health, and quality of life.

Declaration of Competing Interest

The authors report no declarations of interest.

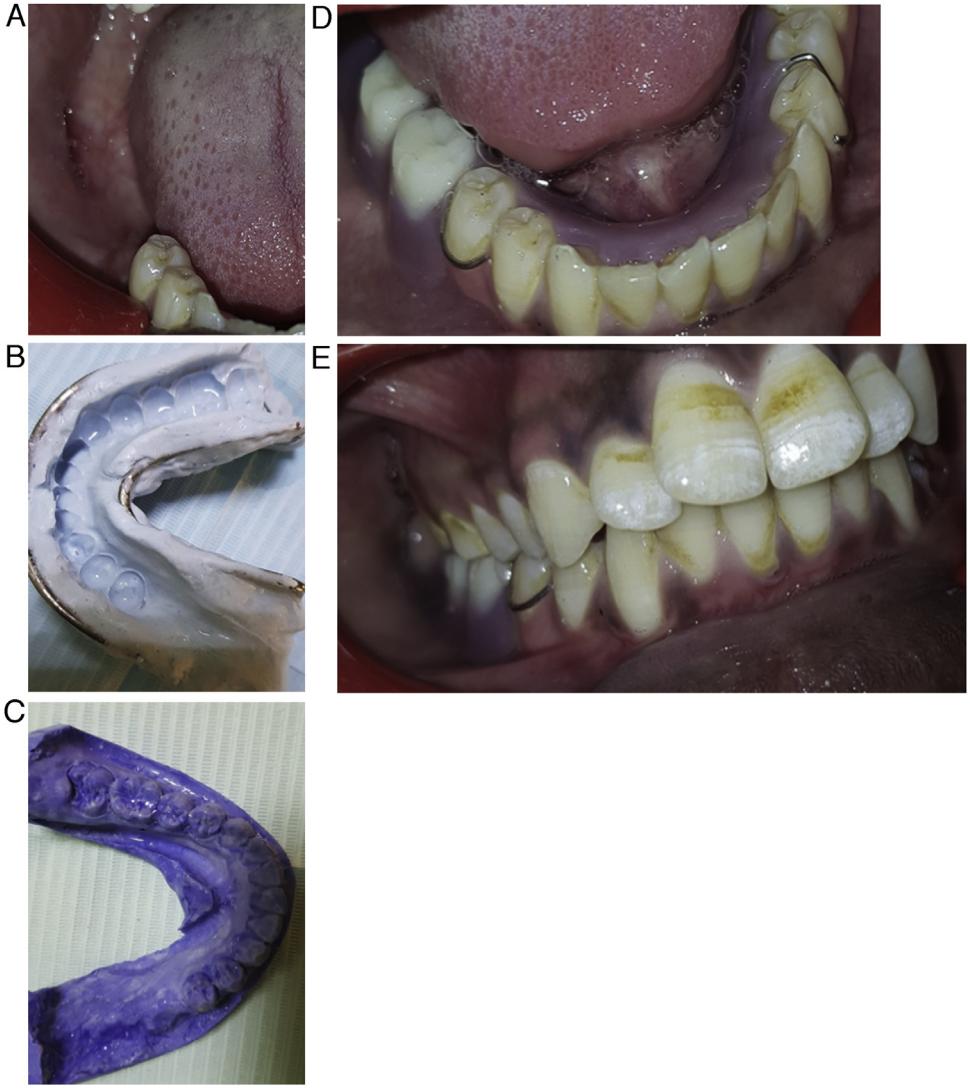

Fig. 5. The post-operative rehabilitation of the patient using transitional acrylic partial denture.
A. The intraoral view of the patient showing Kennedy class II edentulous space.
B. A photograph showing a hydrocolloid primary impression in a stock tray.
C. A photograph showing the primary cast.
D. The intraoral view of the patient showing the inserted acrylic partial denture.
E. The intraoral view of the patient showing the maximum Intercuspation of the patient.
F. The extraoral view of the patient after 2 years of treatment showing significant. improvement in the patient's esthetic with absence of any signs of recurrence of the lesion.

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Ethical approval

The ethical approval was obtained from the ethical committee of the Khartoum Dental Teaching Hospital, Federal Ministry of Health, Khartoum, Sudan. The research was registered at the Research Registry with the unique identifying number: researchregistry6210.

Consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

Authors contribution

Awadalkreem F contributed to the conceptualization, prosthetic treatment of the patient, writing, editing, finalization and submission of the case.

Omer Abdoun involved in intervention/performing the surgery, and supervision of the case.'

Registration of research studies

1. Name of the registry: NA.
2. Unique identifying number or registration ID: researchregistry6210.
3. Hyperlink to your specific registration (must be publicly accessible and will be checked): https://www.researchregistry.com/browse-the-registry#home/.

Guarantor

Fadia Awadalkreem.
Omer Abdoun.

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