Case Report

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A recurrence odontogenic keratocyst formation of mandible with distinctive features: a case report

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Introduction and importance: Odontogenic keratocyst (OKC) is a distinctive form of developmental odontogenic cyst that deserves special consideration because of its specific clinical behaviour and histopathologic features. The clinical and radiographic features of OKC are indefinite; while some may be associated with pain, swelling or drainage, most of them are asymptomatic. This case reports rare radiographic and histopathological features of recurrence OKC.

Case presentation: A 47-years-old male patient presented with a main complaint of a painful mass in the oral cavity with a history of previous lesions that occurred in the posterior portion of the mandible related to extraction of impacted third molar. The oral examination revealed a swelling in the molar region of the right mandible with lingual plate expansion. The radiographic and histopathologic were consistent with the diagnosis of OKC. Consequently, the lesion was surgically removed, and no clinical or radiological recurrence was observed during the 8-month postoperative follow-up.

Clinical discussion This case explained the clinical differences between OKC and other lesions and highlights the distinctive radiologic and microscopic features that a conflict with previous studies concerning the symptoms that may related to naevoid basal cell carcinoma syndrome, and revealed the proper treatment depending on the recurrence appearance and the treatment methods that used previously.

Conclusions This case highlights a rare multilocular appearance of recurrent OKC in the mandible with no naevoid basal cell carcinoma syndrome related, supports the marginal resection as an effective procedure in the management of recurrent OKCs cases.

Key Words case report, odonotgenic keratocyst, oral and maxillofacial surgery

Introduction

Odontogenic keratocyst (OKC) is a distinctive form of developmental odontogenic cyst that has specific clinical behaviour and histopathologic features[1]. The frequency of OKC has been reported to vary from 3 to 11% of odontogenic cysts[2]. They most commonly occur in the second and third decades of life and show a slight predilection for males (males to female ratio 1.3:1)[1]. In 1962, the histological criteria and the specific clinical behaviour were established for this lesion, which was different from the other jaw cysts[3]. In 2005, the WHO reclassified OKC to keratocystic odontogenic tumour (KCOT) based on its clinical behaviours, including potential aggression, infiltrative growth, and a high rate of recurrence up to 62.5%[4]. However, the WHO reclassified it as OKC in 2017 because of insufficient evidence to support its neoplastic origin[5].

HIGHLIGHTS

- This case represents distinctive radiological and histopathological features of recurrence odontogenic keratocyst (OKC) that could lead to misdiagnosing.
- This OKC case reports no naevoid basal cell carcinoma syndrome (NBCCS) related.
- This case supports the radical resection as a proper management of extensive OKCs.

The diagnosis of keratocyst is purely histopathological. It has a thin, friable wall, typically with minimal inflammation. The lining is stratified squamous epithelium, 6–8 cells thick. There is a flat epithelium connective tissue interface. The basal cell layer is palisaded, hyperchromatic, cuboidal or columnar epithelial cells. Luminal cells are flattened and parakeratotic in a wavy or corrugated appearance. Small satellite cysts, cords or islands of odontogenic epithelium may be seen in fibrous walls[6].

The clinical and radiographic features of OKC are indefinite; while some may be associated with pain, swelling or drainage, most of them are asymptomatic. OKC's commonly occur in the tooth-bearing areas (82%), and some of the cases show an

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association with at least one impacted tooth (27% in mandibular third molar)[1]. "Conservative" treatment usually includes enucleation and/or marsupialization, while "aggressive" treatment includes enucleation associated with adjunct therapies or resection[7]. This case reports rare radiographic and histopathological features of recurrence OKC associated with the previous extraction of the impacted third molar in the mandible. The work has been reported in line with the SCARE 2023 criteria[8].

Presentation of case

A 47-years-old male patient was referred to the Department of Oral and Maxillofacial Surgery in March, 2023 with a chief complaint of a painful mass in the oral cavity and jaw, along with swelling and gradual increase. It was first noticed spontaneously during a panoramic radiograph 6 months ago, and the patient reported a previous lesion that occurred in the same region 4 years earlier with similar symptoms after extraction of the impacted third molar and was diagnosed and treated as an OKC. No significant or serious symptoms in the patient's medical history were reported.

The intraoral features revealed a tumefaction measuring 3×4 cm in size in the posterior portion of the right mandible involving the molar region with lingual plate expansion. The overlying mucosa was normal with no drainage or teeth displacement, but a slight paraesthesia was reported. The extraoral examination showed right facial swelling along with an expanded mandibular right buccal vestibule. The patient presented no skin lesions or X-Ray features suggestive of naevoid basal cell carcinoma syndrome (NBCCS) (Figure 1). Laboratory tests and blood investigations were also within the normal limits.

The panoramic radiograph displayed a periosteal reaction in the lower border of the right side of the mandible with a bubble soap appearance (Figure 2). Furthermore, cone beam computed tomography (CBCT) scan showed a large, expansile, well-defined, corticated border and radiolucent entity in the alveolar bone of the right mandibular angle involving ramus and body and

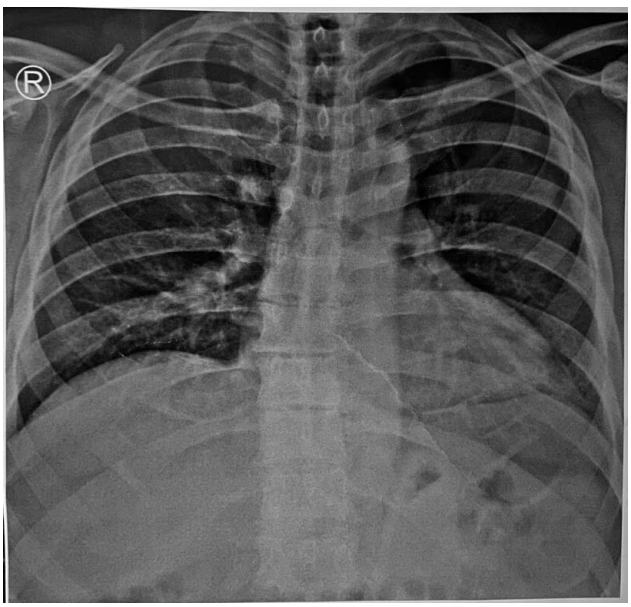

Figure 1. A chest X-ray shows no abnormalities related to naevoid basal cell carcinoma syndrome.

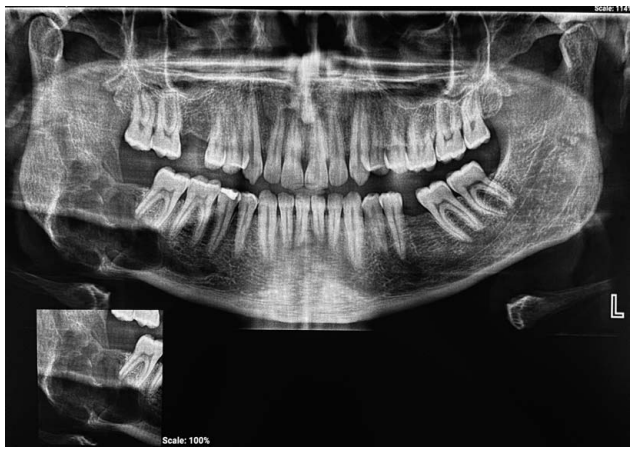

Figure 2. A panoramic radiograph shows the bubble soap appearance of lesion.

associated with the distal root of second molar including root resorption. Localized destruction of lingual and buccal cortical plates is seen with the expansion of the lingual cortex (Figure 3).

An incisional biopsy procedure was performed, and the fragments were sent to the histopathological department. Cheesy materials were seen within the cavity of the lesion, showing multiple dark brown pieces of tissue. Microscopic examination of serial sections demonstrated the lining of a cyst in scattered tiny fragments of stratified squamous epithelium, which is more than 10 layers in thickness, and the basal cell layer shows vague palisading of nuclei and a wavy eosinophilic layer was noticed covering the prickle layer. The rest of the biopsy showed cortical lamellar bone and fibrous connective tissue. These features were consistent with the diagnosis of OKC (Figure 4).

Depending on the clinical and radiographic features of the case we present, radical resection was used as a treatment method to remove the lesion due to its recurrence appearance and aggressive behaviour. The lesion was removed en-bloc with a bony security margin of 1 cm to eliminate the peripheral margins of the lesion, extending into the mandibular ramus to tooth 45; the teeth 46 and 47 were involved (Figure 5). Reconstruction plate was applied in the region of excision concerning patient's perspective and the aim of achieving the functional and aesthetic recovery to maintain soft tissue coverage and in order to improve the patient's postoperative quality of life (Figure 6). However, no evidence of clinical or radiological recurrence was observed during postoperative follow-up for 8 months (Figure 7). Additionally, team care ensured to patient the accessibility to providers across the continuum post-operatively by educating the patient to understand the applied operative procedure and all expected postoperative complications.

Discussion

This case is a type of recurrence case that reports a mandibular OKC affecting the right mandibular angle (the body and ramus are involved), with swelling in the body of the mandible, and pain and paraesthesia as the main clinical features. OKC is defined as a benign jaw neoplasm with a potential for aggressive and infiltrative behaviour that originates from the dental lamina remnants or from the basal cells of overlying epithelium[9]. Its clinical presentation is usually asymptomatic but could be associated

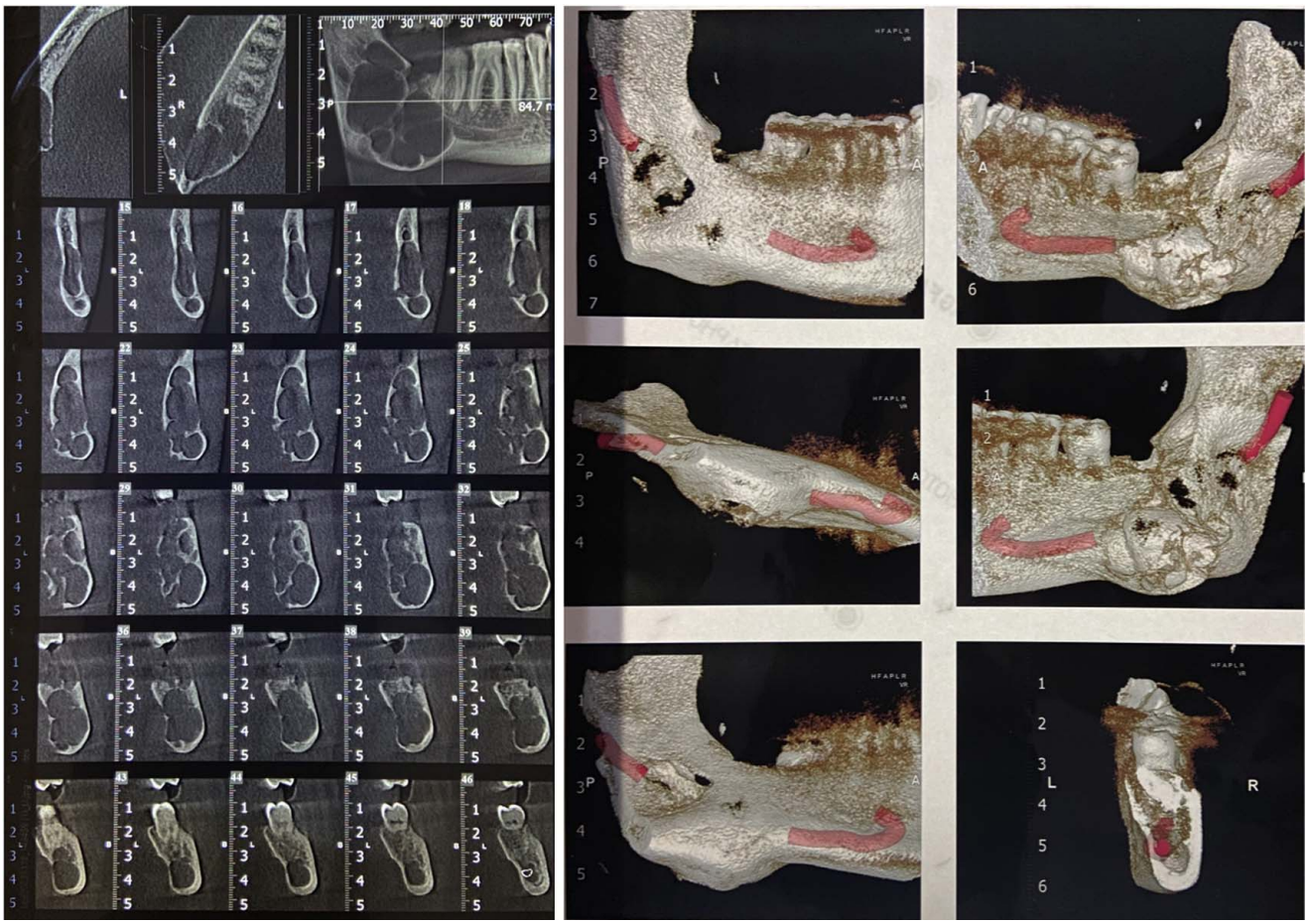

Figure 3. Cone beam computed tomography scan of the right mandibular angle shows the expansion of lingual cortex.

with pain, swelling, drainage and displacement of teeth, and It often has been involved with impacted teeth[10].

This case reports a recurrence OKC in 47-years-old male secondary to an impacted third molar that was extracted previously. OKC is represented as a radiolucent formation with sclerotic borders[11]. In this case, the intraoral examination and radiographic features showed a huge mass with well-defined, corticated borders but no drainage or any teeth displacement.

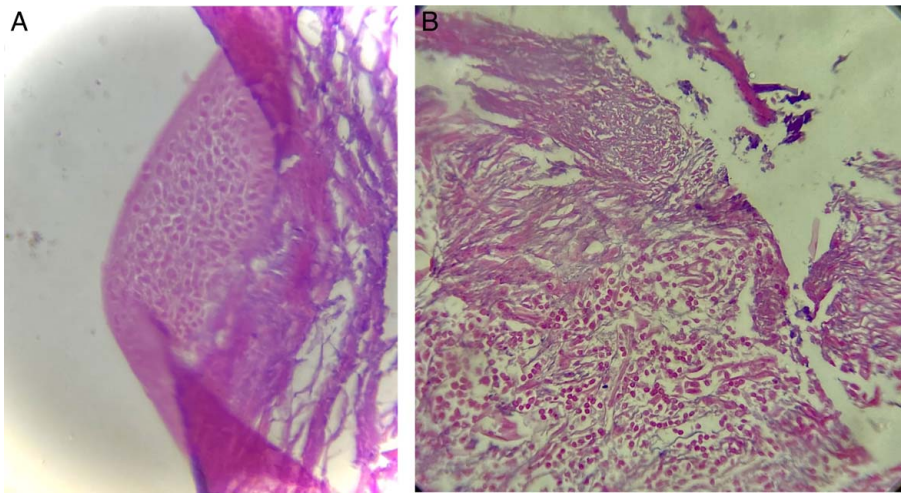

Figure 4. Microscopic features shows (A) stratified squamous epithelium which is more than 10 layers in thickness, and (B) cortical lamellar bone and fibrous connective tissue.

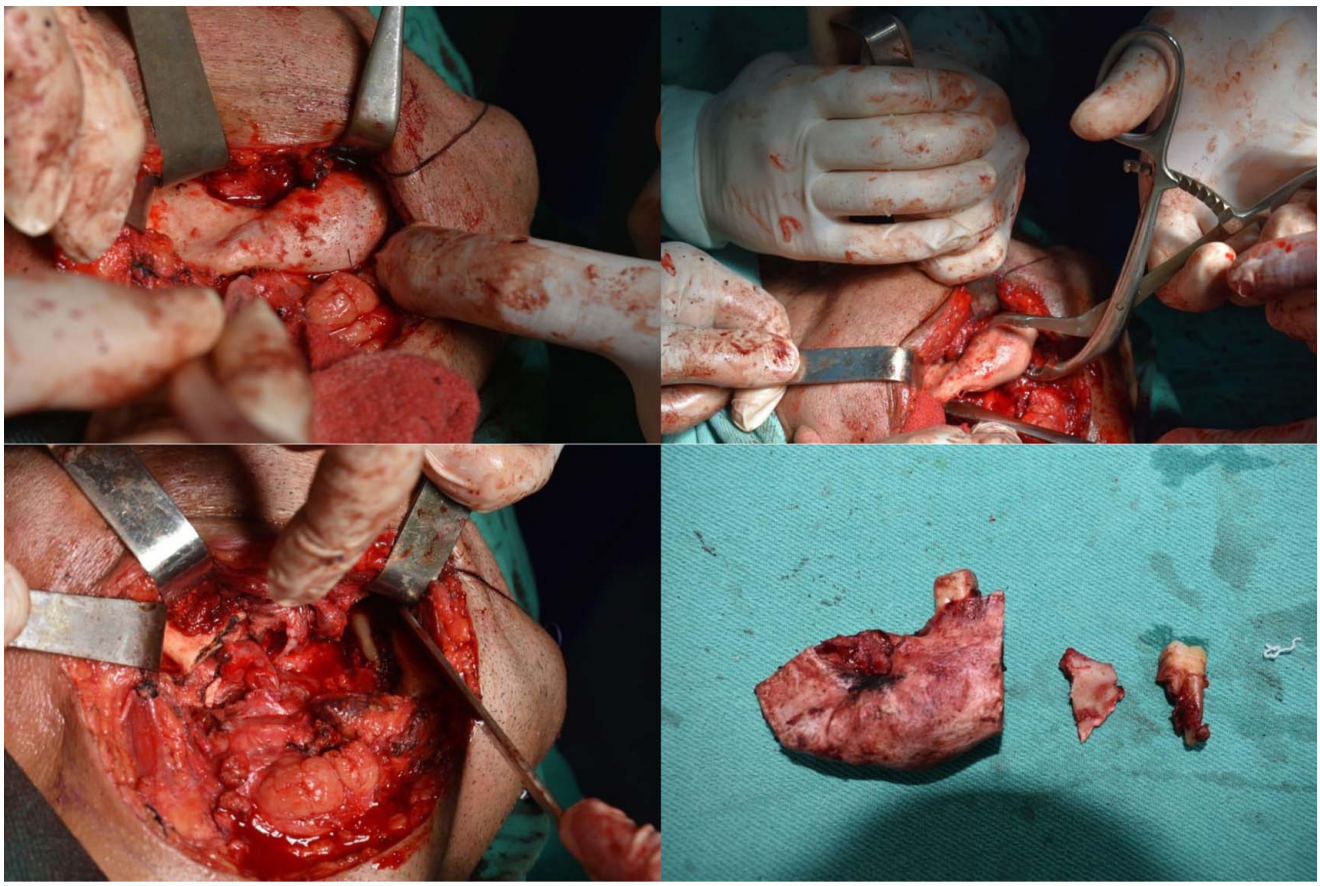

Figure 5. Intraoral operative features while the lesion was removed by marginal resection and en-bloc techniques.

Moreover, the radiographic aspects pretend to be unilocular or multilocular entities, and the multilocular type is often associated with basal cell naevus syndrome (NBCCS) or Gorlin–Goltz syndrome[12]. The medical history and X-Ray films of our patient revealed no NBCCS related to the lesion.

A previous study demonstrated that OKC tends to be in unilocular shape[5]. Also, the multilocular appearance of OKC was found in the maxilla more than in the mandible[2]. In addition, resorption of the roots of adjacent erupted teeth of the lesion is also uncommon[6,13]. However, the CBCT manifestations of the right mandibular angle in the present case showed a multilocular entity in the alveolar bone associated with the distal root of tooth 47, followed by root resorption.

Bone expansion is poorly observed in OKC cases due to its antero-posterior growth within the bone marrow cavity[13]. Despite this, radiographic findings of this case illustrated a wide expansion with bone perforation of the lingual cortex. Notably, it's unusual to see the perforating factor in the recurrence case of OKC[2].

In terms of differential diagnosis, when an OKC is associated with an impacted tooth, it may simulate a dentigerous cyst. Similarly, when an OKC is multilocular and located in the posterior sextant or the ramus of the mandible, it may mimic an ameloblastoma. As a result, dentigerous cysts and ameloblastoma are considered the most common odontogenic lesions in the differential diagnosis of an OKC[14]. The eligibility of the OKCs was determined by OKC histological diagnosis confirmation according to the WHO recommendation[15]. Histologically, OKC is composed of uninflamed fibrous walls, lined by a stratified squamous epithelium, which is 5–8 layers thick with a palisaded hyperchromatic basal cell layer and "corrugated" parakeratotic epithelial cells on the luminal surface[2,16,17]. In this case, the histological features showed the lining of a cyst in scattered tiny fragments of stratified squamous epithelium, but, distinctively, it is more than 10 cell layers in thickness, and the basal cell layer shows vague palisading of nuclei.

In the case of OKC, there are two methods of treatment, one conservative and the other aggressive[18]. The conservative method involves enucleation with or without curettage, decompression and marsupialization[18,19]. Aggressive methods include peripheral osteotomy, cryotherapy (with liquid nitrogen) and application of Carnoy's solution[18,19]. Marsupialization and decompression have been shown to be effective in the management of extensive OKCs with a lower recurrence rate when compared to enucleation alone[20,21]. Chemical cautery with the application of Carnoy's solution has been shown to reduce the recurrence rate; however, it has been banned in some countries due to its suspected carcinogenic effects[22]. Bushabu *et al.*[23] confirmed that multilocular appearance, large OKCs (> 5 cm), multiple recurrent OKCs with or without cortical perforation, and malignant transformation were the main indications of OKC for radical resection. Therefore, following the clinical and radiographic findings of the present case, such as recurrent perforation and multilocular appearance, surgical procedure using enucleation with radical resection was selected as a treatment method.

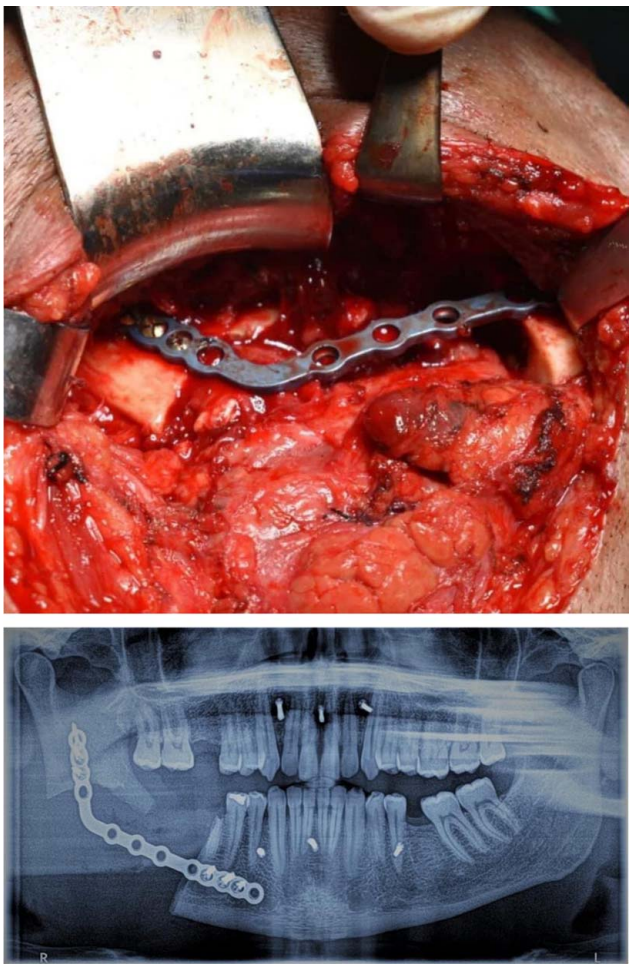

Figure 6. Radiographic and intraoral operative features after applying reconstruction plate.

Conclusions

This case highlights a rare multilocular appearance of recurrent OKC in the mandible with no naevoid basal cell carcinoma syndrome related. It also indicates the hiostopathologic changes that may accompany the recurrence of OKC cases.

Accordingly, our case supports the radical or marginal resection as an effective procedure in the management of extensive, recurrent OKCs cases.

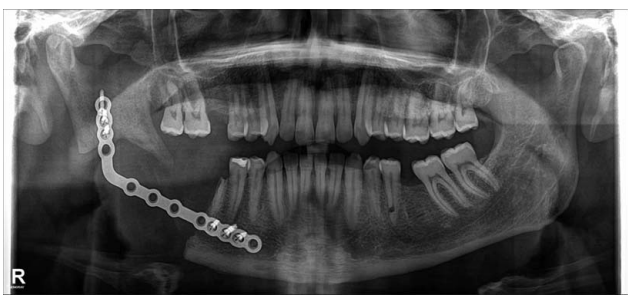

Figure 7. Panoramic radiograph shows no evidence of clinical or radiological recurrence during postoperative follow-up for 8 months.

Ethical approval

Ethical approval is not applicable. The case report is not containing any personal information. The ethical approval is obligatory for research that involve human or animal experiments, so there is no institution that waived ethical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

M.A.: drafting the article. R.S.: histopathological examination. A.A., K.A., A.K.: supervision.

Conflicts of interest disclosure

There are no conflicts of interest.

Research registration unique identifying number (UIN)

This is not an original research project involving human participants in an interventional or observational study but a case report; this registration was not required.

Guarantor

Abdul-Karim Khalil.

Data availability statement

The data are available for sharing.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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