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Successful treatment of mandibular central dentinogenic ghost cell tumor with en bloc resection and dental rehabilitation: A case report

Central dentinogenic ghost cell tumor (DGCT) is a rare entity with less than 100 reported cases thus far.[1] The lack of surgical protocol and its high recurrence rate also hinders its successful management. We reported a case of central DGCT with at least 7 years of history, and its successful management.

A 47-year-old Taiwanese male came to our outpatient department for a painless swelling on his right mandibular middle buccal gingiva for at least a year. His medical history revealed a painless right submandibular swelling 7 years ago noticed by the otolaryngologist. Clinical examination revealed a rubbery, dome-shaped soft tissue bulging mass over the right mandibular buccal vestibule at the premolar area, about 10 × 10 × 5 mm in size. The tooth 43 was distobuccally displaced; the tooth 44 was non-vital (Fig. 1A). Radiographic examination showed a heterogeneous, irregular radiopaque aggregate with no demarcated border between the roots of the teeth 42 and 43, in a background of widespread radiolucent lesion (Fig. 1B). The cone-beam computed tomography (CBCT) showed the lesion extending from the tooth 32 to the tooth 46, spanning down to near the lower border of the mandible (Fig. 1C and D). The lesion increased in size comparing to 7 years ago (Fig. 1E), growing from approximately 3200 mm^3 to near 8200 mm^3, with buccal cortical plate perforation at the right anterior to middle mandible. Incisional biopsy was performed. Histopathologic examination of specimen showed a network of ameloblastomatous epithelium with microcysts infiltrating the peripheral connective tissue (Fig. 1F), and groups of ghost cells within the network of ameloblastomatous epithelium (Fig. 1G). The huge segment of dentinoid matrix could also be seen in the specimen (Fig. 1F). These findings are characteristic of DGCT. The en bloc resection of the bone lesion with 10 mm free bone margin and reconstruction with a free fibular flap were performed. After the one-year follow-up with uneventful healing, we performed dental reconstruction using a dental implant-supported overdenture (Fig. 1H and I).

Although central DGCT is a benign tumor, its aggressive nature often poses challenge for the surgeons. It could lead to tooth displacement or resorption, as well as cortical plate expansion or perforation.[1,2] The report indicated that tumor cells can infiltrate the peripheral bone.[3] Furthermore, conservative treatments like curettage frequently resulted in a recurrence, which significantly impacted the patient's quality of life. Among the seven cases reported by Sun et al.,[4] the five cases treated initially with curettage recurred within two years of surgery, with four of them experiencing multiple recurrences.[4] Given its high recurrence rate, the segmental resection with a safety margin of at least 5 mm—similar to that recommended for ameloblastoma—has demonstrated a better prognosis.[4,5]

In our case, the lesion grew 2.5 times in size over the span of 7 years, causing the buccal cortical plate erosion

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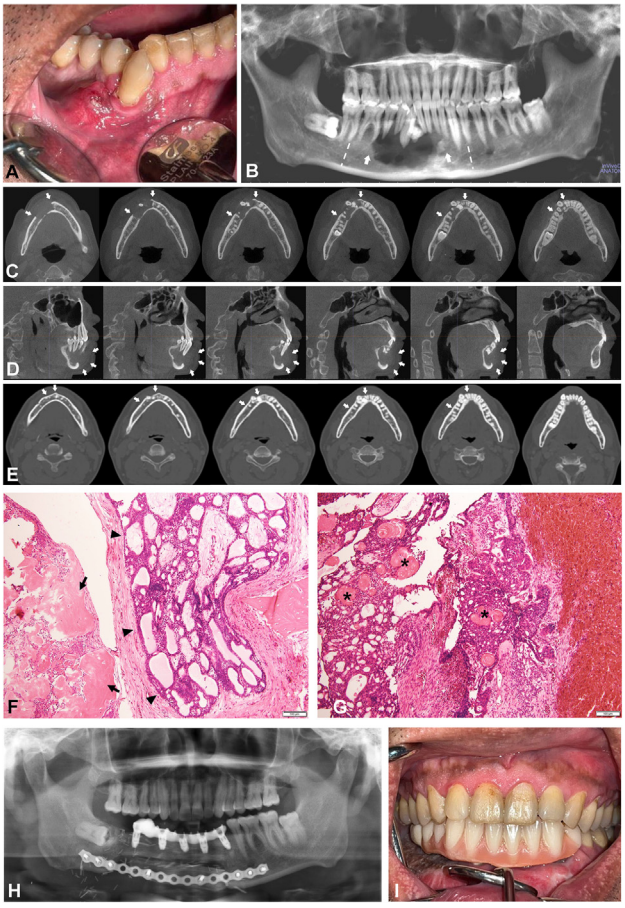

Figure 1 Photographs of a 47-year-old Taiwanese male patient with a right mandibular central dentinogenic ghost cell tumor, depicting clinical findings, historical and current radiographic findings, histopathologic findings, and radiographic along with the clinical follow-up findings. (A) Intraoral examination showing a rubbery, dome-shaped soft tissue bulging mass about 10 × 10 × 5 mm in size over the right mandibular buccal vestibule at the premolar area; the surface was red with a minor ulceration. The mandibular right canine (tooth 43) was buccally displaced; the mandibular right first premolar (tooth 44) was non-vital by the electric pulp testing. (B) Panoramic view of the cone-beam computed tomography (CBCT) showing a heterogeneous, irregular radiopaque aggregate with no demarcated border between the roots of the teeth 42 and 43, in a background of partially ill-defined, irregular radiolucent lesion, spanning from the root apex of the tooth 46 to the root apex of the tooth 41 (pointed out by the white arrowheads), from near the marginal bone of the tooth 44 to near the lower border of the mandible. Roots of the teeth 42 and 43 were displaced to the side of the radiopaque aggregate. There was no sign of inferior border expansion or thinning of the mandible, nor was there bulging of the right inferior alveolar canal. With the span of the tumor considered, we estimated that the en bloc resection by segmental mandibulectomy with the lateral borders encapsulating at least 10 mm of the uninvolved bone (marked by the white dashed lines) would be needed. (C) Axial sections of the CBCT from the level of middle third of the root to the root apex of the tooth 43, taken during the second visit were displayed. They showed the lesion hypodense to the bone marrow growing up to 25.9 × 29.0 × 11.0 mm in 7 years, spanning laterally to the root apex of the tooth 46 and the root apex of the tooth 31 (pointed out by the white arrowheads), while the radiopaque lesion remained the same size. (D) Sagittal sections of the CBCT from the right premolar area to the middle plane were displayed. They showed loss of trabecular bone and buccal cortical plate from the teeth 42 to 45, from the cervical area of the teeth 44 to near the lower border of the mandible. The intrabony part of the lesion has an ill-defined margin, while the perforated part of the lesion was seen separated from the muscle layer by the periosteum (pointed out by the white arrowheads), which likely contributed to its rubbery texture. This further suggested that the en bloc resection by the marginal mandibulectomy including the buccal muscle layer had the chance to fully remove the tumor. (E) Axial sections of head and neck CT taken 7 years prior to the first visit to our OPD, from the similar levels as that of Fig. 1C were displayed. They showed an ill-defined 8.6 × 5.8 × 8.0 mm radiopaque lesion between the teeth 42, 43 and 44 with an irregular 16.6 × 22.4 × 8.6 mm lesion hypodense to the bone marrow surrounding the radiopaque lesion (pointed out by the white arrowheads). The radiolucent lesion spanned from the tooth 45 to the tooth 42, and no further below the root apex of the tooth 43. No overt bony expansion or buccal cortical plate perforation was noted. Compared to the current CBCT image, such lesion had the chance of complete removal through marginal mandibulectomy with far less structure removed. (F) Photomicrographs of specimen showing a network of ameloblastomatous epithelium, with microcysts, infiltrating the peripheral connective tissue (pointed out by the black triangles). The huge segment of dentinoid matrix can also be seen (pointed out by the black arrowheads). (hematoxylin and eosin stain; original magnification; 25 ×) (G) Photomicrographs of specimen showing groups of ghost cell (marked by the black asterisks) within the network of the ameloblastomatous epithelium. (hematoxylin and eosin stain; original magnification; 25 ×) (H) Panoramic radiographs taken at the one-year postoperative follow-up showing the well-reconstructed mandible. Surgical management in this case was the en bloc resection of the mandible with 10 mm free bony margin and 5 mm soft tissue margin, leaving at least one uninvolved anatomic barrier on the tumor specimen. The mandible was reconstructed with the free fibular flap; patient also received the full mouth rehabilitation with dental implant-supported overdenture. There were good osseointegration between the mandible and the graft, with no sign of recurrence. (I) Intraoral examination at the one-year postoperative follow-up showing a well-reconstructed mandible with the fully functional dental implant-supported overdenture.

and soft tissue swelling, affecting 8 teeth. This demonstrated the slow but significant progression of the central DGCT. The partially ill-defined bone margin further highlighted its aggressiveness. Our case is the first case of central DGCT treated with en bloc segmental resection of the mandible, then reconstructed with free fibular flap and dental rehabilitation, which resulted in an ideal outcome. This suggests that such treatment regimen may be optimal for the central DGCT.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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