Management of recurrent intraosseous dentinogenic ghost cell tumour: a case report

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ABSTRACT

A dentinogenic ghost cell tumour (DGCT) is a benign, odontogenic neoplasm characterised by ameloblastoma-like epithelium, ghost cells and areas of dysplastic dentine. It typically presents as an asymptomatic swelling in the jaw and may demonstrate locally aggressive behaviour with a tendency for recurrence.

In this case report, a DGCT was diagnosed in a 38-year-old male patient with a long-standing history of recurrent mandibular swelling. The patient initially underwent conservative management including enucleation and decompression; however, he failed to attend multiple follow-up appointments due to complex social factors. The patient re-presented eight years later with a large destructive lesion involving the left mandible. A segmental mandibulectomy and floor of mouth resection was performed, and mandibular continuity was restored using a pre-bent titanium reconstruction plate adapted to a 3D-printed model.

Management of DGCT remains variable in the literature and, to our knowledge, no cases of DGCT have been reported in Australasia. Although conservative options have been described, our experience supports early radical resection with clear margins and structured follow-up to reduce recurrence and improve long-term outcomes.

Introduction

Progressive mandibular swelling and facial asymmetry are common presentations in oral and maxillofacial surgery. These findings are frequently attributed to odontogenic infections, cysts, tumours or fibro-osseous lesions. A rare but distinct entity within this spectrum is the dentinogenic ghost cell tumour (DGCT), a benign yet locally aggressive mixed odontogenic neoplasm of both epithelial and mesenchymal origin, occurring in both intraosseous (central) or extraosseous (peripheral) types. Histologically, a DGCT is characterised by ameloblastoma-like epithelial islands, ghost cells and areas of dysplastic dentine. Clinically, it is more frequently reported in males, typically presenting between the third and fifth decades of life, with no significant predilection for the maxilla or mandible.[1,2] However, given that fewer than 140 cases have been documented in the literature, current epidemiological patterns must be interpreted with caution.[3]

The recurrence of DGCT has been well described in the literature. Depending on clinical and radiological features, wide surgical excision is often recommended, particularly in recurrent or extensive cases.[3] Although DGCTs are typically benign, rare cases of malignant transformation have been reported, underscoring the importance of definitive diagnosis and long-term follow-up.[4]

This article describes a case of recurrent intraosseous DGCT of the mandible in a medically complex patient with poor follow-up over a 13-year period. It highlights the diagnostic, surgical and reconstructive challenges in the management of intraosseous DGCT.

Case description

A 38-year-old male patient was initially referred to Waikato Hospital's Oral and Maxillofacial Surgery clinic in 2008 with a two-year history of progressive swelling of the left cheek and jaw. He reported a recent sudden increase in swelling but denied airway compromise or systemic symptoms. His medical history included morbid obesity (body mass index > 60), obstructive sleep apnoea, eczema and a history of smoking. The patient resided in a rural town about two hours from Waikato Hospital and was unemployed.

Clinical examination revealed expansion of the lower left buccal sulcus with mobility of teeth 34–37 and a soft intraoral swelling with unremarkable overlying mucosa. Computed tomography (CT) and orthopantomogram (OPG) imaging demonstrated a large, well-defined unilocular radiolucency involving the left mandibular body, with inferior displacement of the inferior alveolar nerve and resorption of the roots of teeth 33–37. A diagnosis of a calcifying odontogenic cyst was made following incisional biopsy and aspiration. The lesion was later enucleated via an intraoral approach with preservation of the inferior alveolar and mental nerves, and extraction of teeth 35–38 was performed. The patient subsequently defaulted from follow-up.

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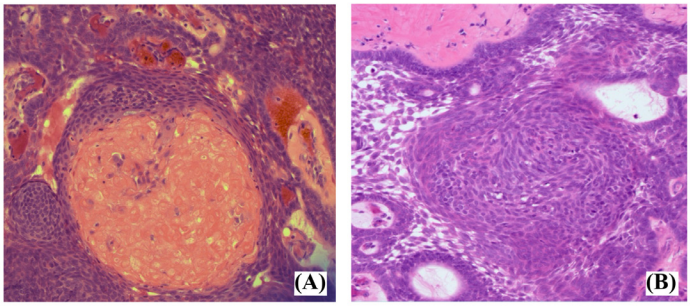


Figure 1. Photomicrographs of the dentinogenic ghost cell tumour pathology stained with Hematoxylin and Eosin. (A) Area of focal ghost cells. (B) Area of stellate reticulum-like cells and palisading ameloblast-like cells. Images were captured at 20x magnification.

He re-presented to the emergency department eight years later in 2016 with further mandibular expansion and resorption of teeth 41–33. CT and OPG imaging demonstrated a large expansile mixed soft tissue/fluid/gas density lesion centred left of the body of the mandible, crossing mental symphysis, with local bone destruction and root resorption. Additionally, soft tissue extension was seen beyond the cortex with associated enlarged local lymph nodes (***Figure 1***).

A decompression tube was placed under local anaesthetic and new biopsies were taken and re-evaluated by oral pathology services at the University of Otago and the University of Sheffield, confirming a diagnosis of intraosseous DGCT. Due to the lesion's locally aggressive behaviour and failure to respond to decompression, surgical resection was recommended; however, this treatment plan was not completed as the patient was further lost to follow-up due to complex social reasons.

The patient then re-presented five years later in 2021 to the emergency department with a one-month history of progressive left submandibular swelling, which had markedly increased in size following the dislodgement of the decompression tube about six weeks before presentation. Clinical examination demonstrated a significant left buccal and submandibular mass, prompting inpatient admission for airway monitoring, further imaging and definitive treatment planning (***Figure 2***).

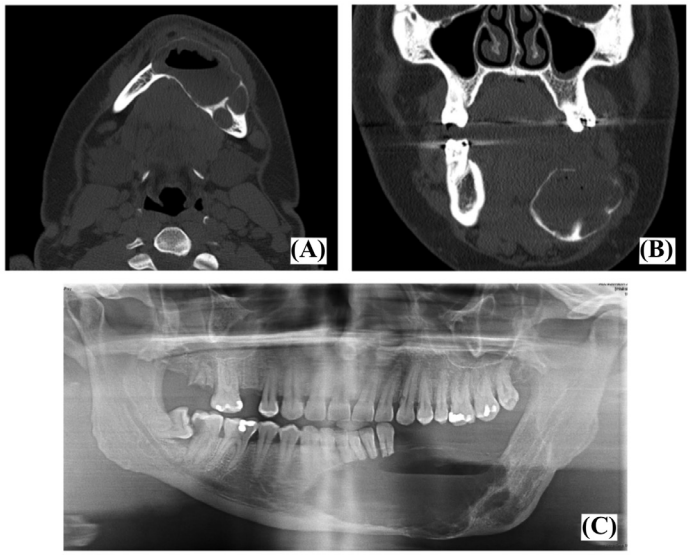


Figure 2. Computed tomography scan in (A) axial and (B) coronal. Orthopantomogram scan in (C), showing a large expansile lesion with soft tissue, fluid and gaseous components involving the left mandible (2016).

Surgical reconstructive planning was complicated by the patient having poor vascular supply to his lower legs and his body habitus rendering any other bony microvascular free flap almost impossible. A virtual surgical plan was carried out for a segmental mandibulectomy and a reconstruction with a 2.4 mm Synthes reconstruction plate pre-bent using an adjusted 3D biomodel following the contour of the unaffected mandible (***Figure 3***). Initially, placement of a patient-specific implant was planned, but due to potential delays and the urgency of treatment required, a biomodel with a pre-bent plate was used.

The patient underwent a subtotal mandibulectomy, bilateral access neck dissection, and extraction of additional teeth. The defect was reconstructed using the pre-bent plate and primary closure of the defect was achieved (***Figure 4***).

Final histopathology confirmed features consistent with a DGCT with areas of secondary aneurysmal bone cyst formation. The patient recovered well post-operatively, with mild oral-phase dysphagia and dysarthria that resolved over time. At ten months after the surgery, there were no clinical or radiographic signs of recurrence, and the patient was satisfied with his functional and aesthetic outcomes (***Figure 5***). Unfortunately, he was subsequently lost to follow-up again and died due to an unrelated issue in 2025.

Discussion

Although a DGCT is classified as a benign odontogenic tumour, it exhibits locally aggressive behaviour characterised by progressive bony expansion, erosion and displacement or resorption of adjacent teeth roots.[4] Intraosseous DGCTs are more frequently encountered than the peripheral variant and tend to demonstrate more aggressive infiltration, cortical perforation and recurrence.[3,5] Clinically, these lesions have no defining pathognomonic features, and radiographically they often present as unilocular or multilocular radiolucencies with radiopaque foci, mimicking other odontogenic lesions.[6,7] Additional non-defining features include root resorption as present in our case.[7,8]



Figure 3. (A) Extraoral photograph following re-presentation to emergency department with left submandibular swelling and intraoral photograph. (B) Same presentation in 2021.

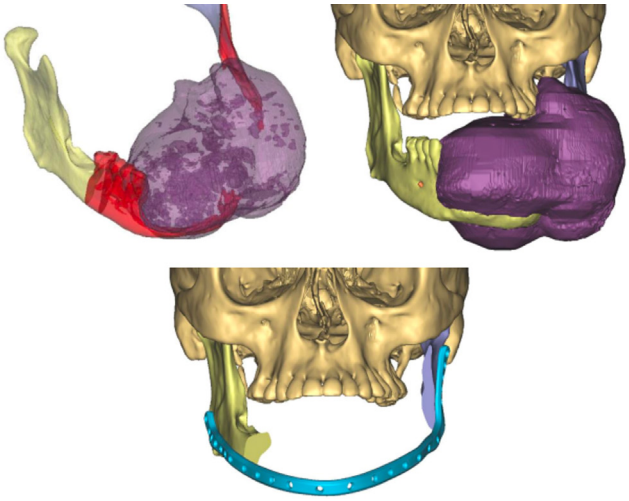


Figure 4. Virtual surgical planning for resection and reconstruction with a pre-bent 2.4 mm reconstruction plate on an adjusted biomodel following the contour of the unaffected mandible.



Figure 5. Post-operative orthopantomogram scan of pre-bent plate in situ following surgery.

The recurrence rate for intraosseous DGCTs has been reported as high as 30.3%, whereas the peripheral variant has a considerable lower rate of 1.9%.[3] The discrepancy can be explained by the infiltrative growth pattern typical of intraosseous DGCT.[9,10] Management strategies include conservative approaches (such as enucleation, curettage and decompression) or more radical approaches (including segmental or marginal resection). Reported recurrence rates after conservative management can reach 73%, compared with about 33% following radical resection with long-term follow-up.[11] Due to the nature of DGCTs, several authors have recommended resection with an adequate safety margin, with reported cases of metastasis to the donor site used for reconstruction following resection of DGCT.[8,12] Regardless, DGCT should not be left untreated due to a malignant potential.[4,13]

When our patient first presented in 2008, the World Health Organization (WHO) had classified the DGCT (in 2005) as a solid variant of the calcifying odontogenic cyst, reflecting a more cystic interpretation of its behaviour.[14] However, in the 2017 and 2022 editions, the WHO reclassified the DGCT as a distinct benign odontogenic tumour with locally aggressive potential.[4,15,16] This redefinition recognised its neoplastic nature, differentiating it from cystic counterparts and emphasising its capacity for recurrence and

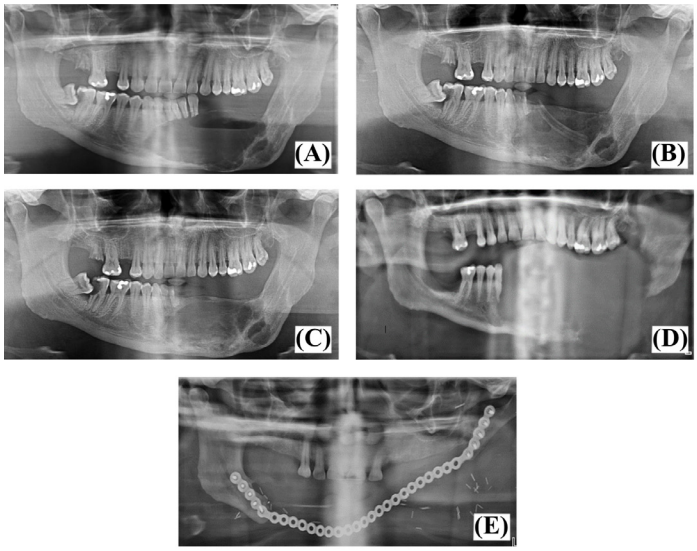


Figure 6. Orthopantomogram scan series of treatment. (A) Presentation in 2016 with resorption in the tooth roots of 41–33. (B) Decompression tubing placed in situ. (C) Lesion four months following decompression therapy. (D) Re-presentation in 2021 with advanced growth of the lesion. (E) Post-operative imaging after resection and reconstruction.

infiltration. In our case, the evolving classification and the recurrence has informed the shift from initial conservative management including enucleation and decompression to definitive surgical resection once the tumour's true biological behaviour became clear. The change in the WHO diagnostic criteria thus had direct implications for both our treatment planning and prognostic expectations.

Although conservative management with long-term monitoring may be considered an appropriate approach for a DGCT, it is important to recognise the practical limitations within our setting alongside no reported cases in Australasia to aid in directing our management. The Oral and Maxillofacial Surgery Department at Waikato Hospital services a large portion of New Zealand's North Island, and many of our patients reside in geographically remote areas.[17] As a result, access to regular follow-up care is often limited, and patient compliance can be challenging, as demonstrated in this case. Therefore, when managing a DGCT, patient-related factors such as geographic accessibility and reliability of follow-up should be weighed alongside clinical and histopathological findings. In certain cases, these considerations may support a more radical surgical approach, even in the absence of aggressive histological features.

Author contributions

Al Balushil L: Data collection, writing original draft, literature review, approved final manuscript.

Luo A: Case identification, writing original draft, data collection, literature review, approved final manuscript.

Bealing M: Writing original draft, literature review, approved final manuscript.

Connor N: Case identification and data collection, literature review.

Singh T: Case identification, provided oversight, expert guidance and final approval of the manuscript.

Conflicts of interest

The authors declare that they have no conflicts of interest.

Patient consent

The patient provided written consent for publication.

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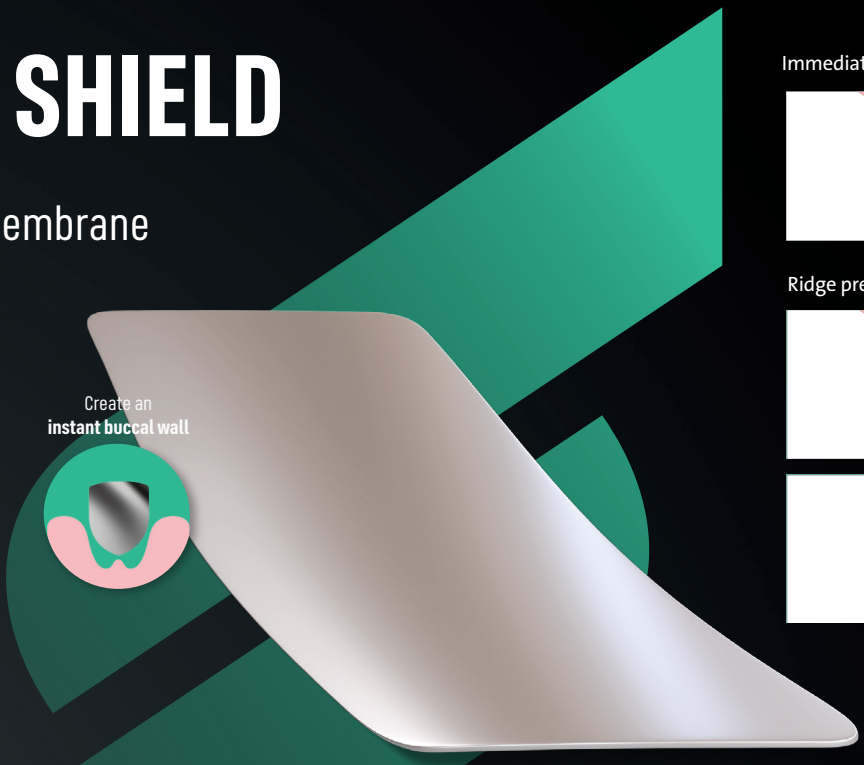
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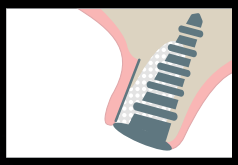
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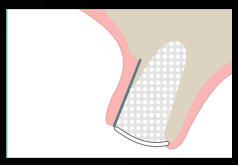
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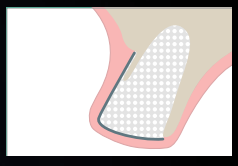


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