

原文題目(出處)：	A granular-cell odontogenic tumour occurring alongside orofacial granulomatosis: a report of the first case
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內文：

■ Introduction

Granular-cell odontogenic tumours (GCOTs):

- Benign odontogenic neoplasms
- Age, Sex: Most commonly in middle-aged women
- Site: Mandible
- Symptom: Asymptomatic swelling
- Diagnosis: Biopsy and histological examination
- Differential diagnosis: Ameloblastoma, fibrous dysplasia, Peripheral ossifying fibroma
- Treatment: Surgical excision or curettage
- Prognosis: Recurrence and malignant transformation are rare

Orofacial granulomatosis (OFG):

- Age: Children and young adults
- Site: Lips
- Symptom: Asymptomatic enlargement
- Cause: May be genetic, infectious or immune-mediated in origin
- Diagnosis: Clinical presentation, biopsy of the affected mucosa, with histological examination revealing non-caseating granulomas
- Differential diagnosis: Crohn's disease, Angioneurotic oedema, potential dietary allergens
- Treatment: Intralesional corticosteroid injection

■ Case report

Present illness:

A 19-year-old woman of Afro-Caribbean origin presented with a 3-year history of a gradually enlarging hard painless swelling of the right mandible. Recurrent upper lip swelling, occurring at 2-month intervals over the last 5 years, was also reported (Figs 1 and 2). The patient was otherwise fit and well; she was a smoker and consumed a moderate amount of alcohol.



Figure 1 Diffuse swelling to upper lip.



Figure 2 Swelling to right mandible.

Clinical findings:

- Right sided facial asymmetry

- No lymphadenopathy
- Bony expansion of the buccal sulcus extending from the lower right second premolar to the lower right second molar
- Overlying mucosa was normal in appearance
- No altered sensation to the lower lip

X-ray findings:

- Radiolucency extending from the distal aspect of the root of the lower right 2nd premolar to the mesial aspect of the unerupted lower right 3rd molar, with expansion of the mandible in the inferior– superior aspect.
- Honeycomb pattern
- Minimal resorption of the distal root of the 1st molar was evident, with no displacement of the dentition

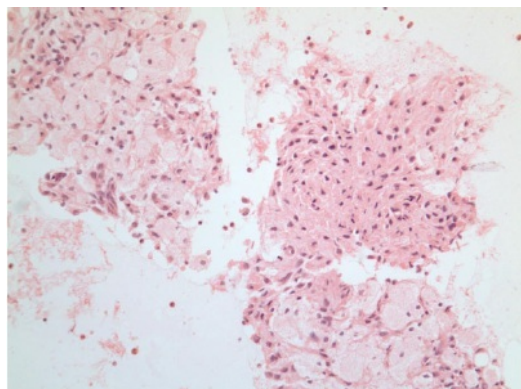


Figure 3 Unilocular radiolucency to right mandible.

Pathological findings:

Incisional biopsy of the right mandibular cystic lesion

- Myxoid, cellular connective tissue comprising spindle cells with eosinophilic cytoplasm and oval nuclei interspersed with large numbers of granular cells displaying pale, eosinophilic granular cytoplasm and eccentric, oval nuclei (Fig. 4).
- There was no cytological atypia or evidence of malignancy.



- Immunohistochemical analysis
- These granular cells showed cytoplasmic positivity for CD68, as one would expect in a case of GCOT (Fig. 5)

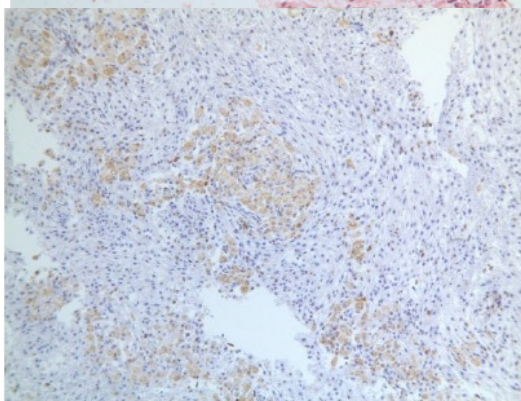


Figure 5 CD68 staining of the granular cells.

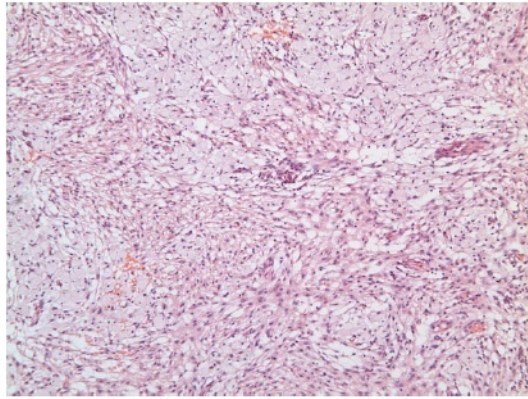


Figure 6 Granular cells in granular-cell odontogenic tumour.

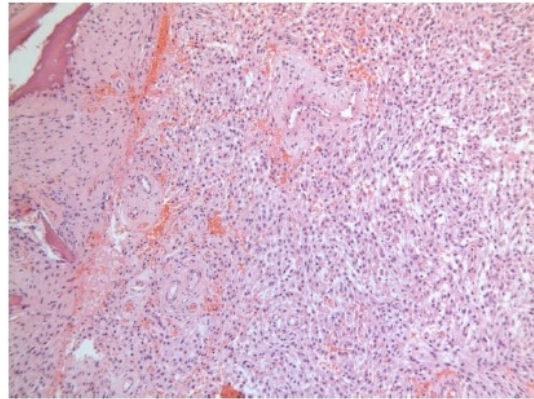


Figure 7 Low-power view of granular-cell odontogenic tumour.

Biopsy of the upper lip

- These appearances are consistent with those of OFG. However, they can also be seen in Crohn’s disease, which needs to be clinically excluded.

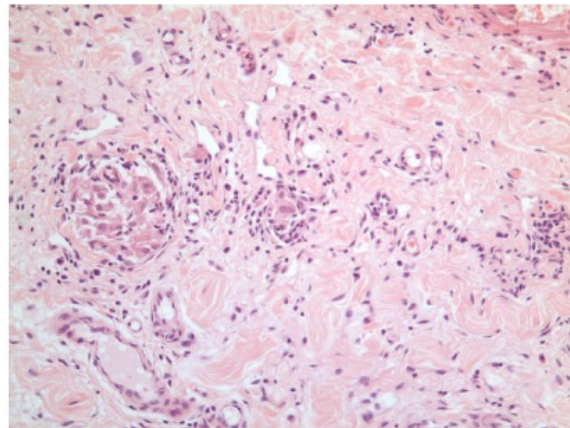


Figure 8 Orofacial granulomatosis. Non-caseating granuloma evident to left of slide. Perilymphatic chronic lymphocytic infiltration also illustrated.

Management

- The GCOT was removed by curettage under general anaesthetic.



Figure 9 Removal of a window of bone to expose the granular-cell odontogenic tumour.

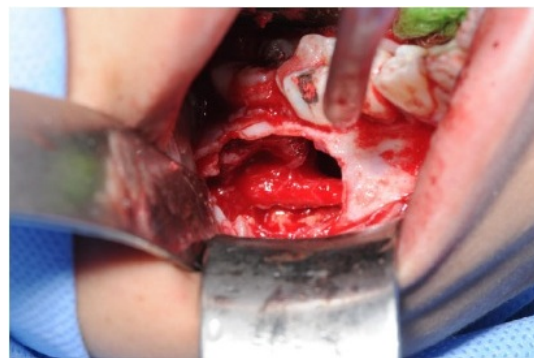


Figure 10 The intact neurovascular bundle.



Figure 11 Post-operative orthopantomogram.

- Enlargement to the upper lip associated with the OFG was reduced by local infiltration of a corticosteroid
- **4 months postoperatively follow up**
 - ✓ The GCOT showed no evidence of recurrence.
 - ✓ There was evidence of recurrent inflammation to the upper lip, and an additional intralesional corticosteroid injection was provided.
- **6 months postoperatively follow up**
 - ✓ No evidence of anaesthesia to the cutaneous distribution of the IAN
 - ✓ All teeth in the quadrant had a positive response to vitality testing.
 - ✓ An OPT radiograph: bony infill to the area of curettage. The abnormal 'honeycomb' appearance of the mandible was observed to extend from the apex of the lower right 2nd premolar to the midline of the lower right 3rd molar.
 - ✓ There remained evidence of expansion of the mandible in the superior-inferior aspect with bowing of the lower border.



Figure 12 Orthopantomogram taken 6 months post-operatively.

■ Discussion

Granular-cell odontogenic tumours (GCOTs):

- Rare benign odontogenic neoplasm with fewer than 40 reported cases.
- Common in female
- High proportion of cases are observed in patients of 60 to 80 y/o
- Asymptomatic buccal expansion of the posterior mandible
- X-ray: unilocular or multilocular RL, RO presentation has been described
- No evidence of displacement of the dentition

- The inferior dental canal was observed to pass through the lesion and to have been preserved intact. Previous cases have illustrated displacement of the inferior dental canal inferiorly.

Orofacial granulomatosis (OFG):

- OFG is an uncommon condition characterised by recurrent swelling of the orofacial region
- Corticosteroids are the mainstay of clinical treatment and are intended to reduce inflammation and lower the incidence of recurrence.

■ **Conclusion**

- The occurrence of a granular-cell odontogenic tumour alongside a presentation of orofacial granulomatosis has not previously been reported.
- Both conditions are known to have an immune-mediated component, a granulomatous reaction can be observed in cases of GCOT. Despite this, the two conditions do not appear to be related.

題號	題目
1	Choose the false statement of Granular-cell odontogenic tumors. (A) Benign odontogenic neoplasms (B) Asymptomatic swelling over mandible (C) Most commonly in young female (D) Treatment involves either surgical excision or curettage
答案(C)	出處：Oral and maxillofacial pathology third edition P729
題號	題目
2	Which statement about Granular-cell odontogenic tumor is wrong (A) Uilocular or multilocular radiolucency lesion was noticed in x-ray findings (B) More than half being older than 40 years of age (C) Would cause of the displacement of the dentition (D) Bone expansion was noticed
答案(C)	出處：Oral and maxillofacial pathology third edition P729