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Granular cell tumour developing in the background of a previous mandibular giant cell lesion: Case report



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ABSTRACT

INTRODUCTION: Granular cell tumours of the mandible are very rare. We present a unique case which has developed at the site of a previous giant cell lesion.

PRESENTATION: 51 year old Caucasian lady had excision of a recurrent giant cell lesion of the anterior mandible. Follow up showed evidence of radiographic recurrence. However, further biopsies from the same site showed granular cell tumour with soft tissues extension. The patient remains well on long term follow up with no evidence of recurrence.

DISCUSSION: This case is unique because the granular cell tumour has evolved from the site of a recurrent giant cell lesion. Conservative surgical excision was an adequate treatment option.

CONCLUSION: Within the limitations of our case study, a correlation between granular cell tumour and giant cell lesion is possible. However, more research is needed to prove this.

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1. Case report

A 51 year old female had a central giant cell lesion removed from her mandible 12 years before presentation but unfortunately the old records were not available for reference. She was referred by her dentist to our oral and maxillofacial surgery unit for possible recurrence of the lesion. The patient was asymptomatic apart from slight altered sensations in her left lower lip and chin which has been there since her operation 12 years previously. There was no local teeth mobility but there was tilting of the roots. There was also expansion buccally. Orthopantomogram showed an irregular well defined 4.5 cm × 2.5 cm radiolucency extending from the lower left second premolar to the lower right canine roots (Fig. 1). The roots of the lower left canine, first and second premolars were tilted distally while the root of the lower left second incisor was tilted mesially.

The patient underwent incisional biopsy of the anterior part of the bony lesion. Histology confirmed a recurrent giant cell granuloma (Fig. 2). She subsequently underwent surgical curettage of the entire bony lesion under general anaesthesia.

The patient was followed up clinically and radiographically. Serial Orthopantomograms showed shrinkage of the lesion with less tilting of the involved roots. Blood tests for corrected Calcium, Albumin and Phosphate were all normal.

The patient had a follow up CBCT scan at 18 months interval which showed two residual isolated areas of radiolucency in the mandible of approximately 1.0 cm in diameter: one in the lower central incisors area, the other in the lower left second premolar area. She underwent further curettage of the two residual lesions.

Histology of the specimens confirmed a recurrent giant cell lesion in the lower left premolar region. However, Histology of the second lesion in the lower right incisor region was reported as a granular cell tumour (Fig. 3).

Histopathology demonstrated cells that were polygonal shaped with oesinophilic cytoplasm, strongly positive on PAS staining. The nuclei were ovoid with inconspicuous nucleoli. The cells had a syncytial arrangement. Where present, the stroma was densely fibrous. Only occasional inflammatory cells were noted. The cells were strongly positive for CD68 immunostain.

At 4 weeks follow up, the patient had developed some granulation type tissue in the lower labial mucosa opposite the anterior surgery site. This new mucosal lesion was excised under local anaesthetic and demonstrated a recurrent granular cell tumour on histopathology (Fig. 4). The neoplastic cells were highlighted by

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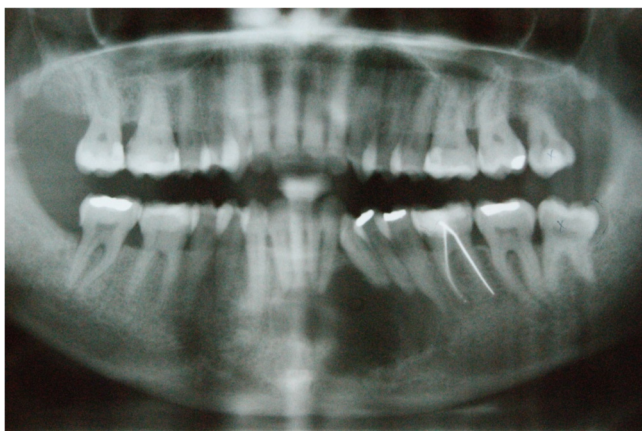


Fig. 1. Radiograph at presentation.

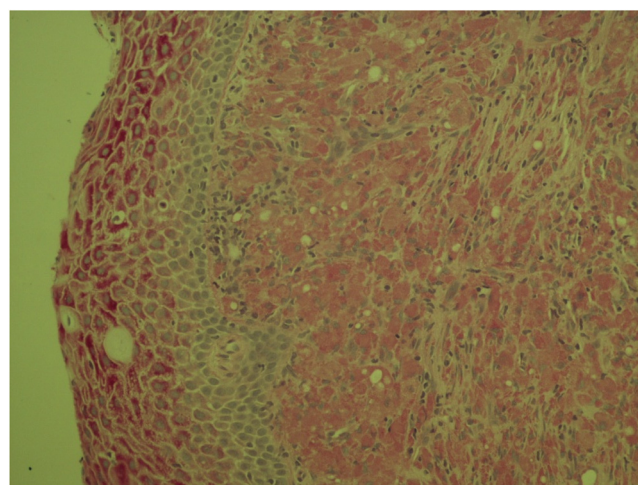


Fig. 4. Recurrent granular cell tumour.

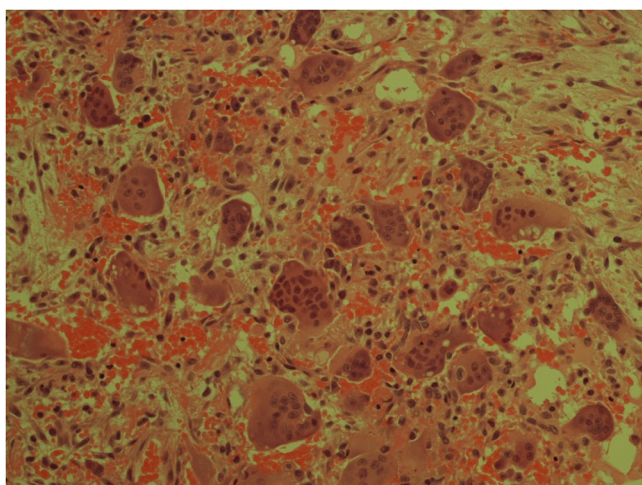


Fig. 2. Giant cell lesion.

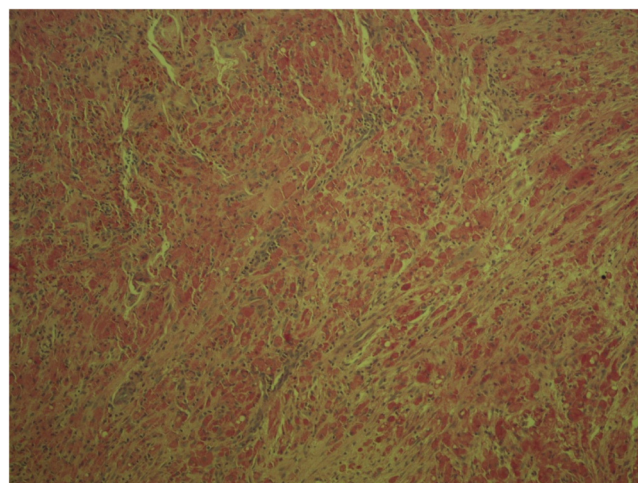


Fig. 5. Granular cell tumour.

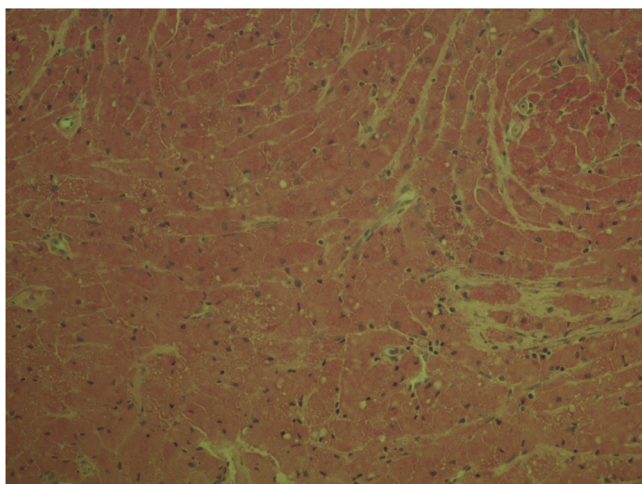


Fig. 3. Granular cell tumour.



Fig. 6. (One year follow up radiograph since the last surgery).

CD68 and S100. There was no evidence of atypia or malignancy. The tumour was incompletely excised.

A further CBCT scan was carried out demonstrating no bony infill of the lesion of the anterior mandible and a possible increase in size. The lesion in the lower left premolar region had shrunk in size and there was good bony infilling.

Further curettage of the lesion in the anterior mandible with removal of the overlying soft tissues and periosteum was carried out. Histology demonstrated features of fibrosis only in the bony cavity, and features of granular cell tumour in the soft tissues specimen (Fig. 5). Again there was no giant cell lesion.

The patient remains well on follow up. A year later orthopantomogram showed very good bony fill of the lesions (Fig. 6).

2. Discussion

Granular cell tumours (GCT) are rare in the mouth [1–7]. The first GCT described was the congenital granular cell epulis (CGCE) of the newborn in 1871 [8]. Abrikossov described the first adult soft tissues GCT in 1926 [9]. It was not until 1962 when Couch and associates [10] described two cases of central GCT.

There is a slight variation in age group incidence when the GCT occurs in the jaw or in the soft tissues. In a series of 18 GCT of adults and 7 cases of CGCE, Dan et al. [1] found that 72.2% of adults involved were females, with an average age in the fourth decade and mainly affecting the tongue, followed by the skin and subcutaneous tissues of the head and neck region. All CGCE patients were females who had the lesions in the anterior maxillary ridge [1].

Carol et al. [16] found a 2:1 female: male incidence ratio in adult GCT with the tongue affected in two thirds of cases, followed by the buccal mucosa, lips and soft palate. Central GCT occur most commonly in the fifth decade and in females predominantly in the posterior part of the mandible [3,4,15,18,20]. However, in our case the GCT was found in the anterior mandible.

It has been suggested that there is a 1–2% rate of malignant transformation in all GCT [11]. Almost all of the reports of malignant variants in the literature concern tumours outside the mouth. However, Krishnamurthy et al. reported a malignant GCT of the tongue [12]. Also there have been reports of SCC of the tongue coexisting with GCT [13,14].

Piattelli et al. described one case of malignant central GCT in the maxilla [19].

GCT in the soft tissues present usually as a solitary, firm, painless lumps. However, multiple synchronous lesions have been described in the literature [17]. Local excision is the treatment of choice although there have been reports of recurrence [1]. In our case, the GCT recurred because it was incompletely excised.

Central GCT most commonly present as a swelling and expansion of the affected area [4,15,20]. The lesion has a well defined radiolucency with or without sclerotic borders [3,4,15,18,20]. Curettage and surgical excision has been described as an effective treatment option with no reported recurrence [3,4,15,18,20]. However, One case in the literature showed recurrence 13 years after it was initially curetted with no extraction of the related teeth [21]. These results are in line with the findings in our case. Curettage of the bony cavity was successful as the final bony biopsy result showed fibrosis only with no recurrence. However the residual soft tissue GCT opposite the bony lesion recurred and needed a second procedure.

Under the light microscope, GCT share a common morphological picture, they are composed of round to polyhedral cells with small round or oval nuclei. The cells cytoplasm contain eosinophilic lysosomes. On the other hand, immunostaining shows variations between CGCE, adult GCT and central GCT [1,3,5,16,22]. CGCE and central GCT share negative reaction to S-100 marker [1,3,16,22,23]. However, there are reports of a positive reaction of the central GCT to the CD68 marker [24] which has been supported in our case. This marker indicates a possible histiocyte origin.

The only S-100 positive GCT is the adult type [1,5,16,22,23]. This has led to the neurogenic theory for the origin of this type of lesion in contrast to the old myogenic theory first advocated by Abrikossov [9] which is based on light microscopy observations.

To add to the uncertainty of histogenesis, some authors believe that demonstration of these marker proteins by immunostaining is a measure of gene expression and cytodifferentiation rather than an indicator of cellular origin [16].

The GCT in our case has developed in a site of a previous giant cell lesion. This raises a question whether this was a mere coincidence

or if the giant cell lesion was a precursor. Further research is needed prior to any solid conclusions.

3. Method

This work has been reported in line with the SCARE criteria [25].

Conflict of interest

The authors have no conflict of interests.

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This study received no funds from anybody.

Ethical standards

The manuscript does not contain clinical studies or identifiable patient data.

Consent

No identifiable patient information is included.

Authors contribution

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References

- [1] D.M. Fliss, M. Puterman, H. Zirklin, A. Leiberman, Granular cell lesions in head and neck: a clinicopathological study, *J. Surg. Oncol.* 42 (November (3)) (1989) 154–160.
- [2] P.J. Lamey, J.S. Rennie, J. James, Multiple granular cell tumors of the palate, *Int. J. Oral Maxillofac. Surg.* 16 (April (2)) (1987) 236–238.
- [3] L. Ardekian, R. Manor, R. Gaspar, D. Laufer, Central granular cell odontogenic tumour: case report and review of literature, *J. Oral Maxillofac. Surg.* 56 (November (11)) (1998) 1343–1345.
- [4] D.K. White, S.Y. Chen, K.S. Hartman, A.S. Miller, L.F. Gomez, Central granular-cell tumour of the jaws (the so-call granular-cell ameloblastic fibroma), *Oral Surg. Oral Med. Oral Pathol.* 45 (3) (1978) 396–405.
- [5] J.A. Garlick, D. Dayan, A. Buchner, A desmoplastic granular cell tumour of the oral cavity: report of a case, *Br. J. Oral Maxillofac. Surg.* 30 (April (2)) (1992) 119–121.
- [6] N.C. Sena Costa, F. Bertini, Y.R. Carvalho, J.D. Almeida, A.S. Rodrigues Cavalcante, Granular cell tumour presenting as a tongue nodule: two case reports, *J. Med. Case Rep.* 6 (February (10)) (2012) 56, <http://dx.doi.org/10.1186/1752-1947-6-56>.
- [7] J.A. Regezi, J.G. Batsakis, R.M. Courtney, Granular cell tumors of the head and neck, *J. Oral Surg.* 37 (1979) 402–406.
- [8] Rajesh Kumar, Shradha Jaiswal, Ankit Singhal, Ridhi Garg, Congenital granular cell lesion: a rare tumour of new born, *J. Oral Maxillofac. Pathol.* 17 (September–December (3)) (2013) 440–442.
- [9] A.I. Abrikossov, Über myome ausgehlichen von der quergestreiften willku muskulatur, *Virchows Arch. (Pathol. Anat.)* 260 (1926) 215–233.
- [10] D.R. Couch, E.E. Morris, F. Vellios, Granular cell ameloblastic fibroma. Report of 2 cases in adults, with observations on its similarity to congenital epulis, *Am. J. Clin. Pathol.* 37 (1962) 398–404.
- [11] Barry Rose, George S. Tamvakopoulos, Eric Yeung, et al., Granular cell tumours: a rare entity in the musculoskeletal system, *Sarcoma* 2009 (2009) 4, Article ID 765927.
- [12] Arvind Krishnamurthy, Rinku George, Urmila Majhi, Urmila majhi malignant granular cell tumor of the tongue: a clinico-pathological challenge, *Indian J. Surg. Oncol.* 5 (March (1)) (2014) 71–74.

- [13] Hee Young Son, Jin Pyeong Kim, Gyung Hyuck Ko, Eun Jae Lee, Seung Hoon Woo, Lingual squamous cell carcinoma surrounded by granular cell tumour, *Chonnam Med. J.* 48 (April (1)) (2012) 65–68.
- [14] R. Caltabiano, A. Cappellani, M. Di Vita, S. Lanzafame, The unique simultaneous occurrence of a squamous cell carcinoma and a granular cell tumour of the tongue at the same site: a histological and immunohistochemical study, *J. Craniofac. Surg.* 19 (2008) 1691–1694.
- [15] L. Ardekian, R. Manor, R. Gaspar, D. Laufer, Central granular cell odontogenic tumour: case report and review of literature, *J. Oral Maxillofac. Surg.* 56 (11) (1998) 1343–1345.
- [16] C.M. Stewart, R.E. Watson, L.R. Eversole, W. Fischlschweiger, A.S. Leider, Oral granular cell tumors: a clinicopathologic and immunocytochemical study, *Oral Surg. Oral Med. Oral Pathol.* 65 (April (4)) (1988) 427–435.
- [17] M.L. Goodstein, D.W. Eisele, V.J. Hyams, H.K. Kashima, Multiple synchronous granular cell tumors of the upper aerodigestive tract, *Otolaryngol. Head Neck Surg.* 103 (October (4)) (1990) 664–668.
- [18] S.D. Vincent, H.L. Hammond, G.L. Ellis, J.P. Juhlin, Central granular cell odontogenic fibroma, *Oral Surg. Oral Med. Oral Pathol.* 63 (June (6)) (1987) 715–721.
- [19] A. Piattelli, C. Rubini, G. Goteri, M. Fioroni, E. Maiorano, Central granular cell odontogenic tumour: report of the first malignant case and review of the literature, *Oral Oncol.* 39 (January (1)) (2003) 78–82.
- [20] Sachin C. Sarode, Gargi S. Sarode, Kedar Vaidya, Central granular cell odontogenic tumour: a systematic review, *J. Oral Pathol. Med.* 43 (2014) 167–176.
- [21] R.B. Brannon, R.K. Goode, L.R. Eversole, R.F. Carr, The central granular cell odontogenic tumour: report of 5 new cases, *Oral Surg. Oral Med. Oral Pathol. Oral Radiol. Endod.* 94 (2002) 614–621.
- [22] R. Mirchandani, J.J. Sciubba, R. Mir, Granular cell lesions of the jaws and oral cavity: a clinicopathologic, immunohistochemical, and ultrastructural study, *J. Oral Maxillofac. Surg.* 47 (1989) 1248–1255.
- [23] Luis M. Junquera, Juan C. de Vicente, Jose A. Vega, Jose L. Losa, Jose M. Albertos, Juan S. Lopez-Arranz, Granular-cell tumours: an immunohistochemical study, *Br. J. Oral Maxillofac. Surg.* 35 (1997) 180–184.
- [24] N. Calvo, D. Alonso, M. Prieto, L. Junquera, Central odontogenic fibroma granular cell variant: a case report and review of the literature, *J. Oral Maxillofac. Surg.* 60 (2002) 1192–1194.
- [25] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. Orgill, The SCARE Group, The SCARE Statement: Consensus-based surgical case report guidelines, *Int. J. Surg.* (2016), article in press.

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