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CASE REPORT

Granular cell tumour of the tongue in a 17-year-old orthodontic patient: a case report

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Abstract

To describe the clinical presentation of a granular cell tumour (GCT) in an orthodontic patient, as well as discuss its aetiology and treatment of choice. We present a case of GCT of the tongue in an otherwise healthy 17-year-old male patient along with a brief review of literature on GCTs. The lesion was surgically excised and orthodontic treatment was successfully finalised. Clinically, GCTs are indistinguishable from other benign connective tissue and neural tissue neoplasms and may be found in any site, with cases commonly involving the gastrointestinal system, breast and lung. However, over 50% of cases involve the head and neck, with the tongue being the most frequently involved site (65–85% of oral GCTs). GCTs demonstrate a close anatomical relationship with peripheral nerve fibres and demonstrate the presence of myelin and axon-like structures thus lending credence to their neural origin. The treatment of choice for GCTs is conservative surgical excision. Because GCTs present with a potential for recurrence, follow-up is recommended. While the primary focus of orthodontic treatment is the position of the teeth within the orofacial complex, the health and wellness of the patient and his/her surrounding oral tissues always take precedence. This case demonstrates the importance of routine physical examination of the intraoral and extraoral tissues during routine orthodontic care.

Introduction

In 2011, Moffitt published an investigation of the incidence of pathology noted in the process of obtaining routine 'new patient' diagnostic radiographs[1]. While the incidence of significant pathology in this generally healthy, mostly adolescent, age group was quite small (0.02%), some significant and potentially life-threatening pathologies were observed. This study, while reporting on incidence of radiographic pathology, did not look at the presence of pathology in routine extraoral and intraoral examination. Because the study was survey based, it is likely that it underreports the incidence of pathology found on routine radiographic assessment. In addition, only the opinion and assessment of the orthodontist was assessed, and neither an oral pathologist nor oral and maxillofacial surgeon, both of whom may be more experienced in evaluating radiographs for pathology, were consulted[2]. A more recent publication evaluated a total of 272 cone beam computed tomography films by oral radiologists found an average of over three incidental findings per film[3].

The standard of care for every dentist and dental specialist is to assess not only the radiographs that are taken, but also to conduct an oral exam on each new

patient[4]. Once performed, the exam findings must be reassessed at each subsequent office visit. Because orthodontists see their patients frequently (monthly or alternate months) and generally see a healthy adolescent patient pool, it is possible that over time, the re-examination becomes perfunctory. The following case report illustrates the importance of a thorough assessment as the orthodontist evaluates his or her patients at each visit.

Case report

A 17-year-old male presented to the clinic with a half-cusp Class II Division II malocclusion. He had a palatally impinging overbite with mild maxillary and mandibular crowding (Fig. 1). The temporomandibular joint exam and range of motion exam were performed with normal results. The intraoral/extraoral examination were also normal.

After discussion of his treatment options, it was decided to perform a non-extraction treatment approach to achieve mild maxillary incisor proclination. The family provided their consent and treatment was initiated.

Following 5 months of Class II correction, the patient called the clinic with a complaint of a swelling involving his tongue. He described what could commonly be expected to be an irritation resulting from the Forsus spring. He was provided an immediate appointment to assess his concern. When he came in for observation, he had a swelling on the right lateral aspect of his tongue. Measurements, photographs and a detailed history were taken. Following the visit, an appointment was made with oral and maxillofacial surgery.

At the oral surgery appointment, the now 20-year-old male presented with a 1-month history of an asymptomatic lesion involving the right lateral margin

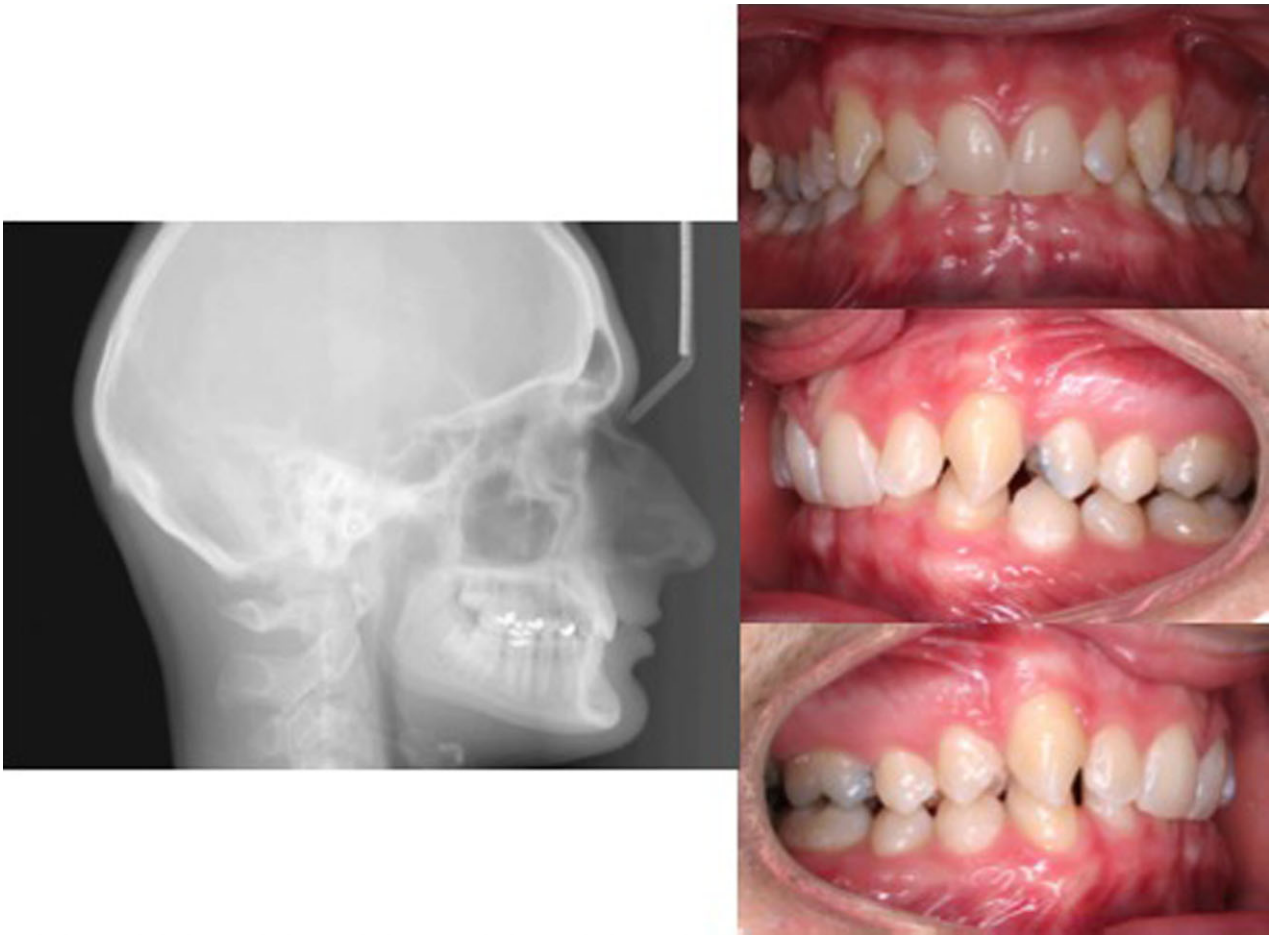

Figure 1 Pretreatment records. The lateral cephalometric radiograph and clinical intraoral photographs depict a 'routine' Class II Division II appearance. The patient has a 100% overbite and approximately one-half cusp Class II relationship of the buccal segment.

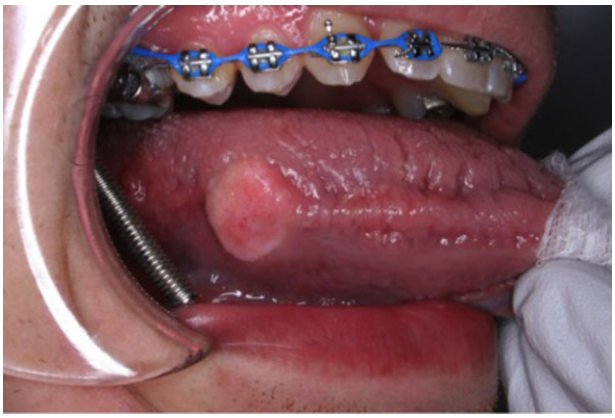

Figure 2 Clinical presentation. A firm light pink submucosal sessile nodule is noted on the right midlateral tongue. Prominent fissuring and a subtle area of benign migratory glossitis are noted anterior to the lesion of interest.

of the tongue (Fig. 2). The patient was 23 months into orthodontic treatment [full fixed edgewise appliances in both arches and Unitek MBT brackets (3M Unitek, Monrovia, CA, USA), along with a fixed functional appliance]. Clinical examination revealed a single, well-circumscribed light pink firm submucosal sessile nodule, measuring 14 × 13 mm, in a background of bilateral benign migratory glossitis and fissuring. After discussion of the differential diagnosis with the family, an incisional biopsy was recommended. The incisional biopsy revealed the classic histopathological features (Fig. 3) of a granular cell tumour (GCT). One month later after follow-up discussion with the family, definitive excision of the lesion was performed (Fig. 4). Histological examination of the excisional biopsy confirmed the initial diagnosis of GCT.

Following the removal of the GCT, the patient was referred back to the orthodontic clinic for completion of his case. The final occlusal result was excellent demonstrating a Class I molar and canine relationship with ideal overbite and overjet (Fig. 5). The patient's recovery from the surgical procedure was uneventful and satisfactory.

Discussion

GCTs are relatively uncommon benign neoplasms, first described by Abrikossoff in 1926. This lesion has also been referenced in the literature as Abrikossoff's tumour, granular cell myoblastoma, granular cell neurofibroma and granular cell schwannoma. GCTs may be found in any site, with cases commonly involving the gastrointestinal system, breast and lung. However, over 50% of cases involve the head and neck. In this area, the tongue is the most frequently involved site accounting for 65–85% of oral GCTs[5–23]. The larynx and lip are the next most commonly affected sites[10]. Other less frequent intraoral locations include the hard palate, buccal mucosa and gingiva. GCTs have also been reported in the parotid gland.

GCTs typically occur in the fourth to sixth decades of life, although cases have been reported in all age groups, ranging from 11 months to 85 years. GCTs are rare in children. The literature indicates a female predilection for GCTs with a 2:1 predominance[7,10,11,13,18,21–23].

GCTs lack encapsulation and tend to extend to the underlying skeletal muscle and peripheral nerve. Clinically, GCTs classically present as benign-appearing, slow-growing, solitary, firm white, pink, or yellowish painless submucosal nodules with a smooth or ulcerated surface. They are rarely larger than 3 cm[24–26]. In 5–15% of cases, they may present as multiple nodules. However, multiple lesions mostly occur in the intradermal and subcutaneous tissue, although rare cases have been documented in the oral mucosa as well[6,7,27–38].

Approximately 2% are classified as malignant based on histopathological presentation. These characteristically present as locally aggressive lesions with distant involvement[6,7,27–38]. Indicators of malignancy include areas of necrosis, haemorrhage, size greater than 4 cm, a high mitotic index, cellular atypia and identification of distant metastasis[10–13,27,29,35,39–41].

At present, the histogenesis of the GCT remains somewhat uncertain[5,12,15,18,23,29,30,42–47]. While originally hypothesised by Abrikosoff to be of muscular origin because of its intimate association with surrounding muscle fibres, the current literature favours neural origin because of strong positive immunohistochemical staining for neural markers such as S-100 protein and neuron-specific enolase[10,23,29,30,45–47]. GCTs demonstrate a close anatomical relationship with peripheral nerve fibres and demonstrate the presence of myelin figures and axon-like structures ultrastructurally, further lending credence to neural origin.

Histologically, GCTs stain intensely eosinophilic and are composed of sheets and nests of large, oval or polygonal cells with abundant granular cytoplasm and small round nuclei that tend to be located centrally. The granular cells often appear intimately associated with muscle and nerve bundles, appearing to emanate from them. Mitotic figures are rarely found. Pseudoepitheliomatous hyperplasia has been reported in the overlying epithelium in up to 87% of cases[23,27,48]. This benign epithelial reaction often mimics the appearance of squamous cell carcinoma and may result in a misdiagnosis, especially when examining

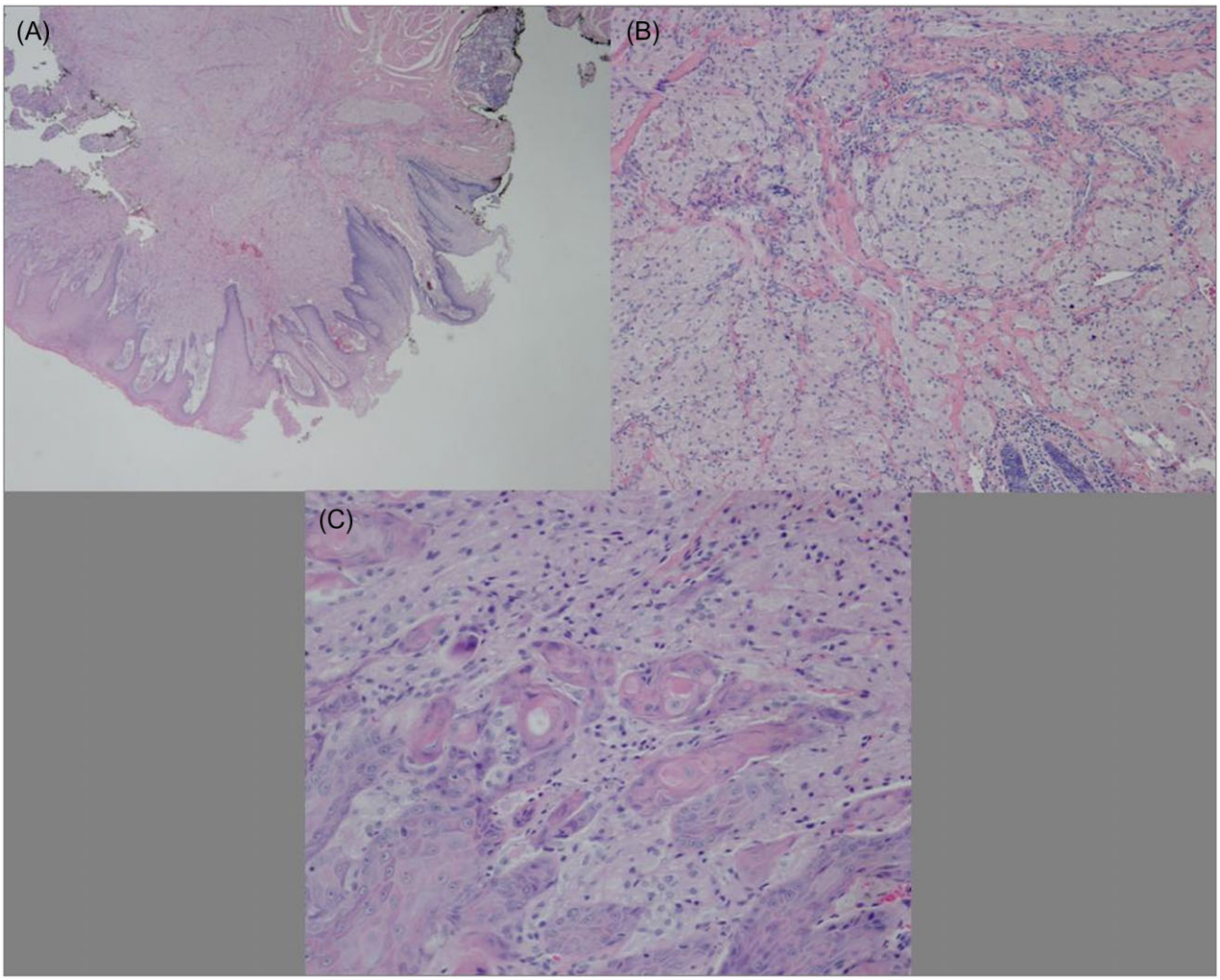

Figure 3 On incisional biopsy, the specimen was noted to consist of a nodule of mucosa surfaced by parakeratinised stratified squamous epithelium (A) demonstrating pseudoepitheliomatous hyperplasia (B) overlying a benign proliferation of large polygonal cells with abundant pale granular eosinophilic cytoplasm (C). The cells are arranged in sheets, nests and cords.

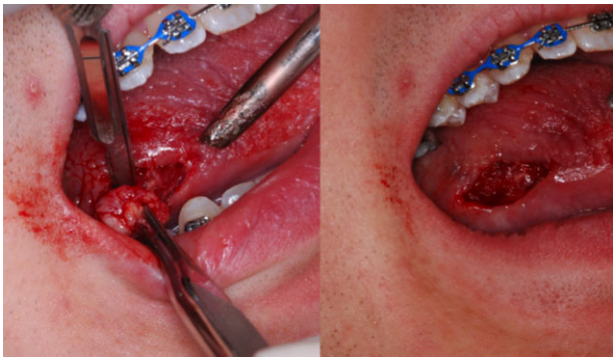

Figure 4 Clinical details of the excisional biopsy.

superficial biopsies. For this reason, a biopsy of sufficient depth to reach the underlying granular cells is essential to avoid a potential misdiagnosis.

Clinically, GCTs are indistinguishable from other benign connective tissue and neural tissue neoplasms. As such, the differential diagnosis typically include fibroma, lipoma, schwannoma, neuroma and neurofibroma[48].

The treatment of choice for GCTs is conservative surgical excision[23,27,30,36,38,48]. Conservative resection of GCTs favours a relatively low recurrence rate of less than 5–10%, with recurrence of the lesion linked to incomplete removal[24,48,49]. Because of the poorly defined margins of a GCT, surgical margins should be completely into the adjacent tissue to ensure complete

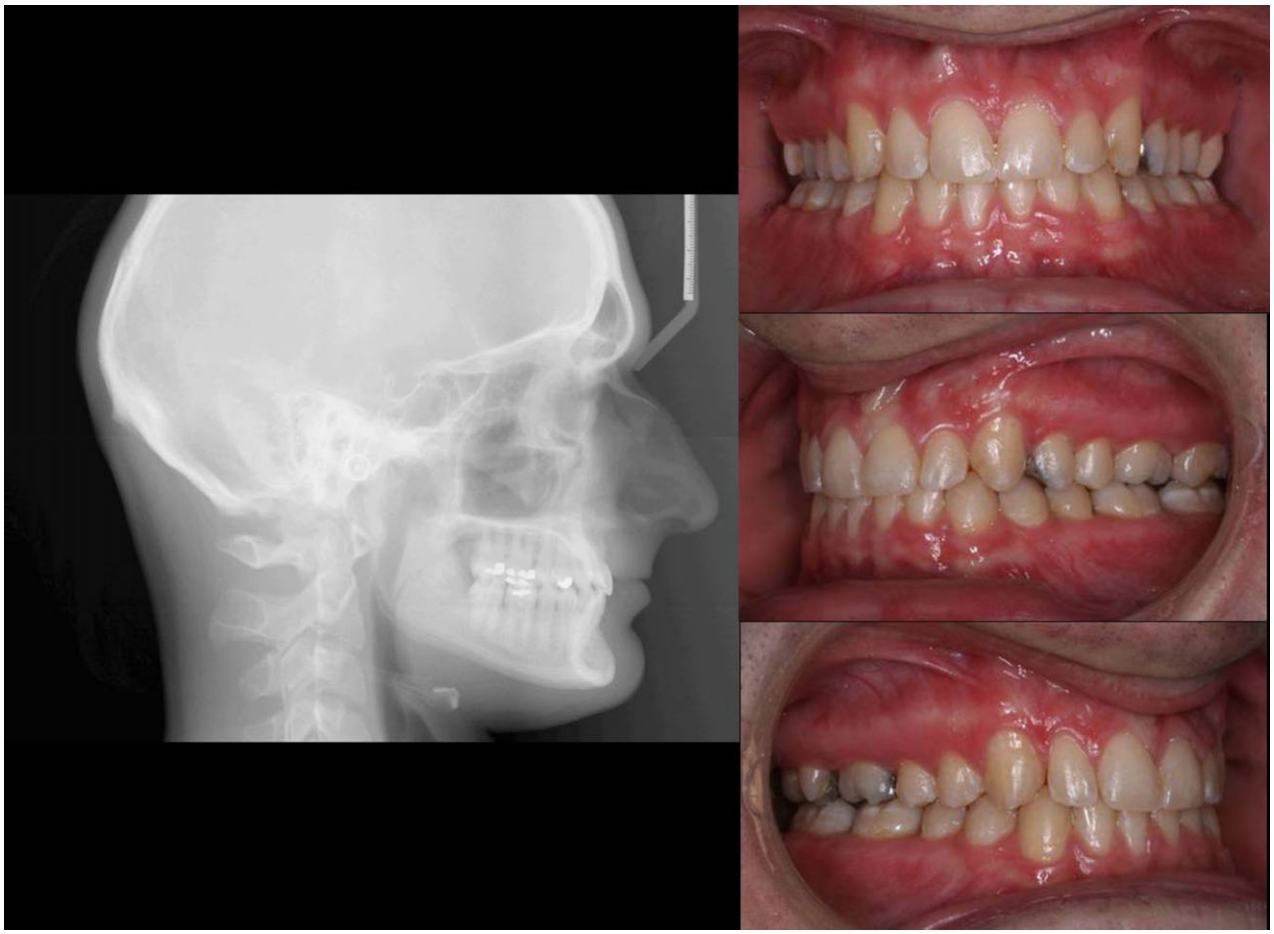

Figure 5 Post-treatment records. The lateral cephalometric radiograph and clinical intraoral photographs depict a properly treated Class I molar and canine relationship. The overbite has been reduced and the patient has a well interdigitated posterior occlusion.

tumour removal. GCTs present with a potential for recurrence; thus, follow-up is recommended[30].

Conclusion

While the primary focus of orthodontic treatment is the position of the teeth within the orofacial complex, the health and wellness of the patient always take precedence. This case demonstrates the importance of routine physical examination of the intraoral and extraoral tissues during routine orthodontic care.

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