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Case report

A peripheral dentinogenic ghost cell tumor: A case report

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ABSTRACT

Dentinogenic ghost cell tumors (DGCTs) are extremely rare, accounting for less than 0.5% of all odontogenic tumors. DGCTs were described initially as solid neoplasms of calcifying odontogenic cysts (COCs). In the 2005 WHO Histological Classification of Odontogenic Tumors, COCs were classified as tumors and renamed as calcifying cystic odontogenic tumors (CCOTs). The term of DGCT was retained and histologically defined as a locally invasive neoplasm characterized by ameloblastoma-like islands of epithelial cells in a mature connective tissue stroma, aberrant keratinization comprised of ghost cells, and an association with dysplastic dentin. DGCTs were subdivided into two variants: intraosseous (central) and extraosseous (peripheral). Generally, peripheral DGCTs occur less frequently than central ones. We report a rare case of a peripheral DGCT in a 60-year-old man with a well-circumscribed nodule in the left canine region of the maxilla. The lesion measured about 11 millimeters in diameter. No bone involvement was observed radiographically. Based on clinical and radiographic findings, the tumor was provisional diagnosed as benign. Consequently, it was enucleated completely, and a histopathological diagnosis of DGCT was made. The patient has remained disease-free for 18 months postoperatively. The biologic behavior of central DCGTs is considered to be more aggressive and require radical treatment. In contrast, peripheral DGCTs have been reported to be non-aggressive, because local bone resorption or recurrences have not yet been reported. There are no differences in the histopathologic features between the central and peripheral variants. Accordingly, accurate preoperative evaluation is crucial to differentiate between these lesions including other odontogenic tumors.

1. Introduction

Dentinogenic ghost cell tumors (DGCTs) were defined histopathologically in the 2005 World Health Organization (WHO) Histological Classification of Odontogenic Tumors as locally invasive neoplasms characterized by ameloblastoma-like islands of epithelial cells in a mature connective tissue stroma, aberrant keratinization comprised of ghost cells, and an association with dysplastic dentin [1]. Due to the lesion's histologic diversity, different authors have used many descriptive terms, i.e., calcifying ghost cell odontogenic tumor [2], keratinizing ameloblastoma [3], cystic calcifying odontogenic tumor [4], peripheral odontogenic tumor with ghost cell keratinization [5], dentinoameloblastoma [6], ameloblastic dentinoma [7], epithelial odontogenic ghost cell tumor [8], and odontogenic ghost cell tumor [9]. In 1981, the term DGCT first was proposed as a neoplastic counterpart of calcifying odontogenic cysts (COCs) [10]. COCs were described primarily as distinct clinicopathologic entities in 1962 [11], when they were characterized as cystic in nature. Certainly, COCs form cystic structures in many cases, but they also have been charac-

Abbreviations: DGCT, dentinogenic ghost cell tumor; COC, calcifying odontogenic cyst; CCOT, calcifying cystic odontogenic tumor.

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terized as solid growths or local infiltrates, which are not often compatible with cysts. Over the years, different authors have proposed several classifications to clarify the nature and classify the different histopathologic features of this lesion [12–15]. Because of identification of extensive diversity in its biologic behavioral and histopathologic features, in 2005 the WHO classification was changed to clearly reflect the tumorous character. COCs were renamed as calcifying cystic odontogenic tumors (CCOTs) and defined as benign cystic neoplasms of odontogenic origin, characterized by an ameloblastoma-like epithelium with ghost cells that may calcify [16]. The designation of DGCT was retained and histopathologically DGCTs were defined as mentioned above. Each lesion exhibits a central or peripheral process. Ameloblastoma-like odontogenic epithelium, ghost cells, and dysplastic dentin are observed in both variants. Ghost cells are large polygonal cells with pale eosinophilic cytoplasm and faint nuclear outlines [17]. CCOTs present normally as slow-growing painless cystic neoplasms. Radiologically, they appear as well-defined radiolucent masses and rarely recur after surgical excision.

DGCTs occur centrally in the jaw and peripherally in the gingival or alveolar mucosa [18]. Central DGCTs are more aggressive than peripheral DGCTs, with a higher rate of recurrence postoperatively [19]. Because peripheral DGCTs occur less frequently than central ones [20,21], little information is available. According to existing reports, peripheral DGCTs normally can be managed by local excision alone; a treatment for this lesion has not been weighed based on preoperative radiographic findings. The purpose of this case report is to describe the surgical clinical management of a peripheral DGCT. The radiographic and histologic features are discussed along with a literature review.

2. Case report

A 60-year-old man was aware of a painless mass in the left maxillary gingiva for 1 year. Because it enlarged before he presented to the dental clinic, he was referred to our hospital for examination. His chief complaint was a painless swelling in the anterior maxillary gingiva. He had hypertension and hyperuricemia. The family history was unremarkable. No cervical lymph adenopathy was noted. The lesion, a semispherical mass with a well-defined border and elastic hardness on palpation, was in the cervicolabial gingiva of the left maxillary canine, measured 11 × 10 mm, and was reddish in color (Fig. 1A). The teeth adjacent to the mass were unaffected with no local displacement, mobility, or pain on percussion. Panoramic radiographic examination did not show bone expansion with extension into soft tissues, a radiolucent or radiopaque appearance in the area of the lesion, or resorption of adjacent teeth (Fig. 1B). Thus, an intraosseous (central) lesion was excluded. Based on the clinical examination, the mass was diagnosed as a maxillary benign tumor and pyogenic granuloma was considered in the differential diagnosis. An excisional biopsy was performed under local anesthesia. The mass, which measured 14 × 12 × 5 mm, was resected easily as a lump along with the periosteum. Histopathological analysis showed a tumoral mass of about 2.5 mm surrounded by fibrous connective tissue stroma (Fig. 2A). The tumor was comprised of eosinophilic dentinoid material (Fig. 2B) and odontogenic epithelial cells resembling those of an ameloblastoma (Fig. 2C). At the periphery of the tumoral islands, the cells were arranged in a palisading fashion with no atypical cells. Several epithelial cells were transforming into ghost cells with granular eosinophilic cytoplasm and a faint nuclear outline (Fig. 2D). The tumor was diagnosed histologically as a peripheral DGCT of the maxillary gingiva based on the presence of ameloblastomatous epithelium with dentinoid material and ghost cells. The lesion appeared to be completely removed. At 18 months postoperatively, there were no signs of a local recurrence.

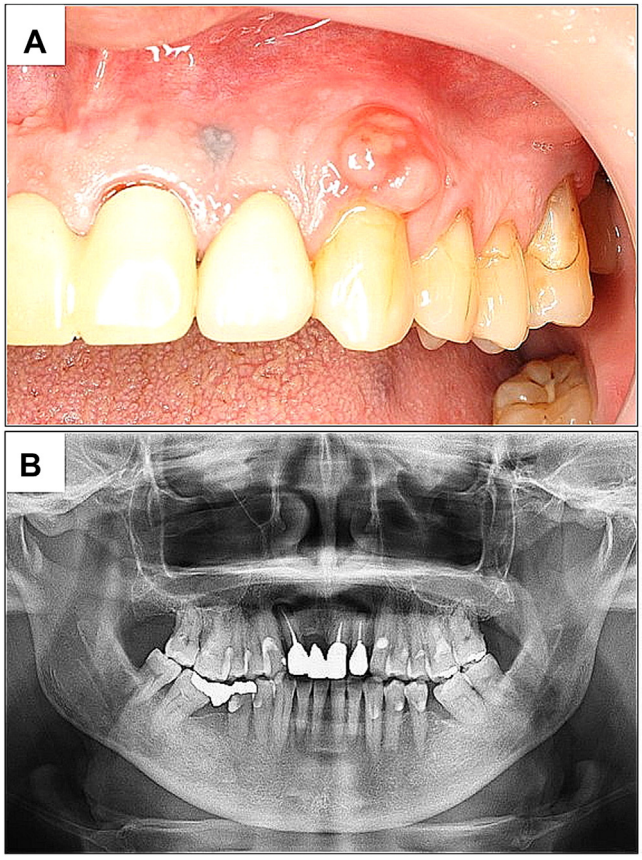

Fig. 1. An intraoral photograph shows a semispherical reddish and nodular lesion with a well-defined border and elastic hardness in the alveolar mucosa of the left maxillary canine (A). A panoramic radiograph shows neither a radiolucent nor radiopaque lesion in the left maxilla. No resorption of teeth adjacent to the lesion is observed (B).

3. Discussion

DGCTs are extremely rare lesions that were formerly considered a solid variant of COCs [10,13,14,22]. COCs account for 1% to 2% of all odontogenic tumors, and only 2% to 14% of COCs are solid tumors [13,14,23]. In 2005 the WHO classified COCs as tumors and termed those CCOTs. Since then, DGCTs have been the solid variants of CCOTs. DGCTs comprise 0.3% to 0.5% of odontogenic tumors [24,25]. Due to their rarity, few reports have provided detailed descriptions of this lesion, and it has not been recognized as an isolated entity [26]. In addition, because many authors have use different terms to describe the lesion, it is relatively difficult to obtain an accurate description. Prætorius et al. reported patient ages ranging from the second to the ninth decades, with a predilection for development in men and no preference for the maxilla or mandible[10]. As with other odontogenic tumors, DGCTs are classified by location as central or peripheral lesions. Peripheral DGCTs occur less often than the central variant and show a predilection for the anterior jaw [1]. Clinically, the tumor is usually asymptomatic [13,27], grows slowly, is a sessile and sometimes pedunculated exophytic nodule, and often varies from 5 to 10 mm in diameter [1]. Radiographs show saucerization of the underlying bone in about 20% of cases [26,28].

To date, only 18 cases of peripheral DGCTs with accompanying clinical and radiographic features have been reported [7,10,29–40] (Table 1). The DGCTs were found in patients over a wide range of ages from 13 to 83 years (mean, 56 years). More than 60% of DGCTs

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