CASE REPORT

Report of Two Cases of Combined Odontogenic Tumors: Ameloblastoma with Odontogenic Keratocyst and Ameloblastic Fibroma with Calcifying Odontogenic Cyst

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Abstract Combined odontogenic neoplasms have rarely been documented. Such tumors have also been described by other researchers as “hybrid” lesions. The histologic features are often identical to other individually well-established odontogenic neoplasms such as ameloblastoma, adenomatoid odontogenic tumor, ameloblastic fibroma (AF), and ameloblastic fibro-odontoma. Their clinical presentation is variable, ranging from cysts to neoplasms showing varying degrees of aggressive behavior. Most combined tumors contain features of one of the odontogenic tumors in combination with either a calcifying odontogenic cyst (COC) or a calcifying epithelial odontogenic tumor. We present two new cases of combined odontogenic tumors: an ameloblastoma with an odontogenic keratocyst and an AF with COC. Predicting clinical outcome is challenging when a combination tumor is encountered due to the paucity of such lesions. One must understand salient features of these entities and differentiate them from the more common conventional neoplasms to expand classification and provide prognostic criteria.

Keywords Combined odontogenic tumors · Mixed odontogenic tumors · Hybrid odontogenic tumors

Introduction

Combined odontogenic lesions, or hybrid lesions, have rarely been reported within the jaws. Of these, the most commonly reported lesion is the adenomatoid odontogenic tumor (AOT) with calcifying epithelial odontogenic tumor (CEOT) [1–7]. Many consider this lesion a variation of AOT; therefore, we did not include the 23 reported cases in our review [7].

The University of Florida’s Biopsy Service received only two combined lesions among the 72,171 specimens submitted from 2004–2012. A thorough search of the literature revealed six previously reported cases of calcifying odontogenic cyst (COC) combined with ameloblastic fibroma (AF). To our knowledge, no combined lesion with distinct features of both ameloblastoma and odontogenic keratocyst (OKC; also referred to as keratocystic odontogenic tumor, or KOT) have been reported in the English language literature. We present two additional rare cases of combined odontogenic lesions and briefly review previous cases in the literature.

Case One

Our biopsy service received an excisional biopsy specimen from a 17 year old female patient with a bony impacted left molar without accompanying radiographic imaging. Microscopic examination of representative sections revealed cystic lining which was eight to ten cells thick and surfaced by corrugated parakeratin. The basal cell layer showed peripheral palisading. Also seen in the specimen were islands of odontogenic epithelium resembling the enamel organ; these islands exhibited peripheral palisading with reverse polarity and apical vacuolization (Figs. 1, 2). This case was diagnosed as combined odontogenic keratocyst with ameloblastoma.

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Fig. 1 The yellow arrow shows classic areas of OKC, while the two blue arrows show ameloblastomatous changes

Case Two

A 10 year old white male was referred by his general dentist in 2005 to an oral surgeon for evaluation of a radiolucency noted in the left mandible. Left facial swelling was noted on clinical examination and the patient reported minor pain. A panoramic radiograph revealed a large multilocular radiolucency of the left angle and ramus of the mandible, distal to impacted tooth #19 (Fig. 3). Microscopic examination of the incisional specimen revealed a cystic odontogenic neoplasm composed of multiple layers of squamous epithelial cells. Aggregates of eosinophilic, keratinaceous material devoid of nuclei interpreted as ghost cells were noted within the lining epithelium. The supporting connective tissue contained numerous strands and clusters of odontogenic epithelial cells. Also noted was an aggregate of delicate, basophilic to

Fig. 3 A large multilocular lesion of the left mandible, involving the majority of the ramus and extending to the distal aspect of impacted tooth #19

myxoid dental papilla-like connective tissue interspersed by islands and strands of odontogenic epithelium. Many of the odontogenic islands were surrounded by an amorphous, eosinophilic product; however, no calcifications or dental hard tissue structures were present (Fig. 4). The diagnosis given was a combined calcifying odontogenic cyst with ameloblastic fibroma.

Discussion

A review of the English language literature revealed 19 examples of hybrid odontogenic lesions including six cases of COC with AF. To our knowledge, no lesions similar to

Fig. 2 **a** High power view of the OKC, showing a cystic lining which is 8–10 cell layers, basal palisading and surface corrugated parakeratin. **b** high power view of the ameloblastoma, showing peripheral palisading with reverse polarity and apical vacuolization

Fig. 4 Calcifying odontogenic cyst (a) in combination with ameloblastic fibroma (b)

Table 1 Previously identified cases of hybrid odontogenic lesions in the literature

Entity	Author	Location	Sex	Age (years)	Treatment	Follow up (years)
AF, COC	14	R mand body	F	6	Enucleation	1.67, NR
	14	L max 14–16	M	13	Compete excision	NS
	14	R mand 32	M	22	Curettage	NS
	15	L mand	M	17	Resection	NS
	16	NS	NS	NS	NS	NS
	10	R max	F	22	Compete excision	1, NR
AFO, AOT, COC	17	Decid L 2nd molar—L lateral inc	M	7	Enucleation	NS, NR
AFO, COC	18	NS	NS	NS	NS	NS
	18	NS	NS	NS	NS	NS
	19	L post mand	M	23	Resection	NS
Amelo, GOC	20	R mand canine and premolars	F	45	NS	NS
	12	Max 1–10	M	65	Resection	2.75, NR
Amelo, COC	9	Mand 20–29	M	35	NS	5, NR
Amelo, CEOT	13	R post max	M	53	Enucleation	5, NR
AOT, Amelo	21	Mand 19–28	M	40	NS	NS
	22	Anterior mandible	NS	NS	NS	NS
	22	Anterior mandible	NS	NS	NS	NS
AOT, Amelo, COC	11	L 1st molar—R 2nd premolar	F	64	Resection	3, NR
AOT, COC	23	Anterior mandible, R-L premolar	M	35	Enucleation	1.5, NR

AF meloblastic fibroma, AFO ameloblastic fibro-odontoma, Amelo Ameloblastoma, AOT adenomatoid odontogenic tumor, CEOT calcifying epithelial odontogenic tumor, COC calcifying odontogenic cyst, GOC glandular odontogenic cyst, M male, F female, NS not stated, L left, R right, NR no recurrence

our ameloblastoma with OKC/KOT have been reported; however, in his comprehensive 1977 article, Brannon described two OKCs with ameloblastomatous change consisting of reverse polarity and apical vacuolization [8]. We have compiled the reported cases, excluding ours, into a table which serves as an updated version similar to Lin et al.'s (Table 1) [9–23]. Information in the table includes location, sex, age, treatment, and any follow-up data. The

most common location these lesions are found is the posterior mandible. Of the 14 cases reporting sex, ten were male compared to four female patients. The median age of patients was 29, with a range of 6–64 years. The rarity of combined odontogenic lesions prevents adequate data reporting regarding treatment planning; however, the treatment is usually dictated by the more aggressive of the involved neoplasms [13]. Of the eight follow-up cases

reported, no lesion had recurred. Six of the cases were treated conservatively, including two AF with COCs, AFO with both AOT and COC, ameloblastoma with CEOT, and AOT with COC. Two were treated with resection, including an ameloblastoma with glandular odontogenic cyst (GOC), and an ameloblastoma with both an AOT and COC. Median follow-up time for these patients was 2.75 years with a range from 1.5 to 5 years. Unfortunately, no follow-up data is available for our cases.

Many authors believe that hybrid odontogenic lesions are not a result of collision between two distinct entities but rather due to the pluripotentiality of the odontogenic epithelium with both lesions likely developing from a common source or ameloblastomatous change in an existing odontogenic cyst [10–12]. We feel this is likely of the ameloblastoma with COC; however, the ameloblastoma with OKC appears to be two distinct entities arising nearly in the same location.

Conclusion

We present two unusual odontogenic lesions with distinct overlapping and combination of histologic features of a combined odontogenic keratocyst with ameloblastoma and a combined calcifying odontogenic cyst with ameloblastic fibroma. In addition, we reviewed the literature and identified 19 previously reported cases of odontogenic lesions exhibiting a combination of histologic features. As more of these entities are reported, pathologists will be better able to recognize these lesions. Due to the paucity of cases, the prognosis, clinical behavior and appropriate treatment of such cases is largely unknown. We hope that as more cases are reported in the literature, a better understanding of combined odontogenic lesions will be obtained.

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