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Case Report

Hybrid Central Odontogenic Fibroma/ Central Giant Cell Lesion of the Mandible: A case Report and Review of the Literature

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Abstract

Central odontogenic fibroma (COF) is a rare neoplasm of the jaw. The combination of COF and Central Giant Cell Granuloma (CGCG) is exceedingly rare. To the best of our knowledge just 57 cases of hybrid COF-CGCG have been reported worldwide. Here we report a case of hybrid COF-CGCG in a 31-year-old woman. a well-defined radiolucency in the premolar region of the mandible was seen. Surgical excision was performed, and histologic study shows fibro-cellular tissue containing giant cells, zones of mesenchymal cells proliferation, in a whorled pattern and, multiple cords and small nests of odontogenic epithelium. Also, this report presents a literature review of previously reported cases.

Introduction

Central odontogenic fibroma (COF) is an uncommon controversial lesion of the jaws that comprises less than 5% of all odontogenic tumors [1,2]. This odontogenic lesion has been reported in an age range from 4 to 80 year with a slight female predilection. The mandible is affected in majority of cases but locally, the lesion appears in two different positions in jaws [3]. It occurs anterior to the first molar in maxilla, while half of the mandibular lesions usually are located posterior to the molar region [4]. Odontogenic Fibromas have slow growth rate [5-7]. Smaller lesions are usually asymptomatic [4], while larger ones may be associated with pain, localized boney expansion and root resorption. Less than half of the odontogenic fibromas are related with unerupted or displaced teeth [7]. Radiographically, odontogenic fibroma imitates other jaw lesions and therefore has a "rather nonspecific" radiographic feature [8]. These features vary [7] from well-defined unilocular radiolucent lesions for smaller odontogenic fibromas to multilocular radiolucencies for larger lesions or mixed ground glass defects for some other cases [3-5, 9].

Considerable histopathologic diversity has been observed in lesions reported as central odontogenic fibroma. The World Health Organization (WHO) described two histologic variants for COF: simple type /epithelium-poor type/ WHO type/ complex/ epithelium-rich type. Stellate fibroblasts in a whorled pattern with fine collagen fibrils and small nests or strands of inactive epithelial rests are seen in the simple type [2,10]. A more complex pattern is seen in the epithelium-rich odontogenic fibroma. Cellular fibrous connective tissue with interlacing bundles of collagen fibers and long strands of odontogenic epithelium which may be a significant component are seen [9].Connective tissue pattern varies from densely hyalinized to myxoid [3]. Enucleation and curettage are common treatments of odontogenic fibromas. Lesions respond well to surgical enucleation with few recurrences have been reported [7]. Giant Cell Granuloma Lesion (GCGL) is a non-neoplastic osteolytic bone lesion [10,11]. Mesenchymal spindle cells proliferation, multinucleated giant cells, hemorrhage and hemosiderin deposition are seen in GCGL [1,2]. In 1992, Allen et al reported a hybrid COF-CGCG lesion for the first time [12], then other cases reported by Odell et al and Mosquda et al in 1997 and 1999, respectively [8,13]. All of them described a fibro cellular tissue including odontogenic epithelial strands and nests with distribution of giant cells [2,5]. Previously reported cases including the present case of HCOF-CGCG are summarized in Table 1.

Table 1: R: right, L: left, Ant: anterior, Post: posterior, C: canine, I: incisor, PM: premolar, M: molar, RL radiolucency, URL unilocular radiolucency, MRL multilocular radiolucency, RO radiopaque NA not available, FU follow up

Case	Age (years)	Gender	Location	Associated features	Radiographic findings	Treatment	Recurrence	Ref
1	66	F	R Mandible	RCT tooth	MRL	Curettage	None at 6 month FU	(12)
2	14	F	L Mandible	Vital teeth,	URL,3/5 cm	curettage	None at 48 month FU	(12)
3	30	F	L Mandible	no expansion	MRL 1.5*2.0 cm	Curettage, Curettage of recurrent lesion	Yes, at 14 month FU	(12)
4	5	F	Anterior maxilla	Orthodontic treatment, some expansion	-	Curettage	None	(8)
5	11	M	Post maxilla, extending to antrum	Buccal expansion	URL	Conservative excision of recurrent lesion	Yes, after 36 month	(8)
6	20	F	Mandible (PM-M)	Buccal expansion	URL 1.5*1.0 cm	Curettage	None	(8)
7	21	F	Post Mandible	-	URL 3*2 cm	Curettage	None	(8)
8	22	F	Mandible (PM-M)	Buccal expansion	-	Curettage and extraction of involved tooth	None	(8)
9	39	F	Mandible (PM-M)	Buccal expansion, Cortical perforation	-	Curettage	None	(8)
10	43	F	Mandible	Expansion, teeth mobile	-	Curettage	Yes, after 36 month	(8)
11	50	F	Mandible PM	-	URL	Curettage of recurrent lesion Curettage	None	(8)
12	17	F	R Mandible (C-PM)	-	MRL 2.5*2 cm	Curettage	None at 72 month FU	(13)
13	57	F	Mandible (PM-M)	Buccal expansion	URL 2.0*2.5 cm	Curettage	None at 18 month FU	(5)
14	18	M	Mandible (PM-M)	Buccal expansion	RL	Surgical excision	Lost to FU	(1)
15	20	F	Mandible (PM-M)	-	RL	Surgical excision	None at 117 month FU	(1)
16	50	M	Mandible (PM-M)	-	RL	Surgical excision	None at 28 month FU	(1)
17	73	M	Mandible (PM-M)	-	RL	Surgical excision	None at 43 month FU	(1)
18	15	M	Mandible (PM-M)	-	RL	Surgical excision	None at 76 month FU	(1)

19	59	M	Mandible (PM-M)	-	RL	Surgical excision	None at 39 month FU	(1)
20	25	M	Mandible (PM-M)	-	RL	Surgical excision	Lost to FU	(1)
21	24	F	Mandible (R M-L M)	-	-	Curettage	None at 8 month FU	(14)
22	14	M	L Mandible(M)	Cortical Expansion	URL 4.0*3.2 cm	Surgical excision	None at 16 month FU	(15)
23	14	M	L Mandible (PM-M)	Buccal and Lingual swelling	MRL 4.5*3.0 cm	Surgical excision	None at 24 month FU	(15)
24	42	F	Mandible, body	Buccal expansion	RL	Enucleation \ Curettage	None	(16)
25	27	F	Mandible, Ramus	-	RL	Enucleation \ Curettage	None	(16)
26	75	F	Ant Mandible	Impaction	URL	Curettage	NA	(17)
27	22	F	R Mandible (PM-M)	-	MRL	Surgical excision	NA	(18)
28	22	F	R Mandible (LI-M)	Expansion & swelling	Mostly URL with scalloped edge, with hint of MRL in post area	Surgical excision	None at 24 month FU	(19)
29-35**	~49	5M 2F	Mandible	Lingual & inferior expansion	-	NA	Yes 3 case	(20)
36	12	F	Ant Mandible	-	RL	NA	-	(21)
37	42	F	L Mandible (M)	Asymptomatic	-	Surgical excision	None at 12 month FU	(22)
38	10	M	Ant Mandible (C-I)	Edentulous area	URL 1.9*1.8 cm	Curettage	None at 72 month FU	(9)
39	63	F	L Mandible (M)	Buccal and Lingual expansion, impaction	URL 1.7*1.0 cm	-	Awaiting treatment	(9)
40	62	M	R Mandible (PM)	_ Buccal expansion	URL	Curettage	None at 12 month FU	(9)
41	65	F	L Mandible	Asymptomatic	MRL 0.9*0.8cm	Surgical excision	None	(23)
42	31	F	L Mandible (PM-M)	Cortical expansion, paresthesia	RO	Surgical excision	None at 9 month FU	(24)
43	33	F	L Mandible (PM)	-	URL	NA	None at 12 month FU	(25)

44	12	F	Ant Mandible (C-L)	Bone expansion, Vital teeth Divergence of teeth root	URL	Surgical excision	None at 5 month FU	(26)
45	27	M	L Mandible (M)	Buccal expansion	RL	Enucleation/ Curettage	None	(2)
46	14	M	L Mandible (M)	-	URL	Lesion resection	None at 24 month FU	(27)
47	31	F	R Mandible (PM)	Vital teeth	URL	Surgical excision	None at 6 month	Present case

*This table does not include the cases from Fowler et al (3 cases) and Pontes (7 cases) (2)
**There was not available data for cases presented by Hassan et al
***We complete the Upadyaya et al Table

Case Report

A case of hybrid COF-GCGL was identified from Urmia Oral and Maxillofacial Pathology department, Iran. Excisional biopsy of tumor was obtained and multiple 4 µm thick formalin fixed paraffin embedded sections were stained with hematoxylin and eosin (H & E). A 31-year-old female patient with an asymptomatic radiolucent lesion in the mandibular was referred to Oral and Maxillofacial Department of Surgery. There was no history of discomfort, pain or traumatic injury. Intraoral examination revealed a non-tender mass of right mandibular body. Radiographs of the patient included panoramic, occlusal and periapical views that revealed well circumscribed, unilocular radiolucency (Figure 1 & 2). Histopathologically, zones of myxoid mesenchymal cells proliferation, in a whorled pattern, multiple cords and small nests of odontogenic epithelium were observed as shown in Figure 3.

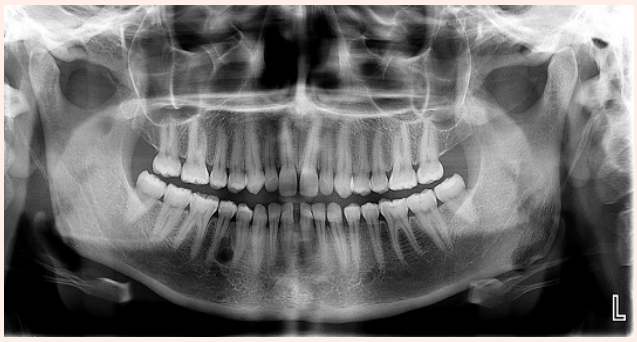

Figure 1: Panoramic radiograph shows an approximately 1 cm radiolucent lesion in the premolar area of right mandibular body.

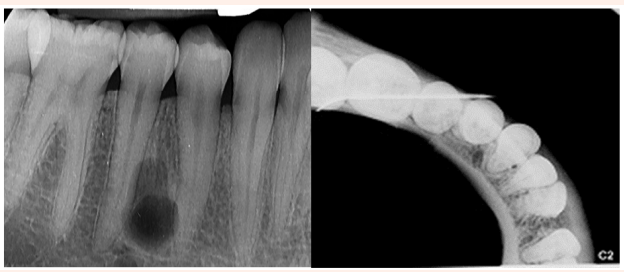

Figure 2: A. Periapical view demonstrates a well-defined radiolucency in the inter-radicular area of right premolars. B. Occlusal view demonstrates no buccal or lingual expansion.

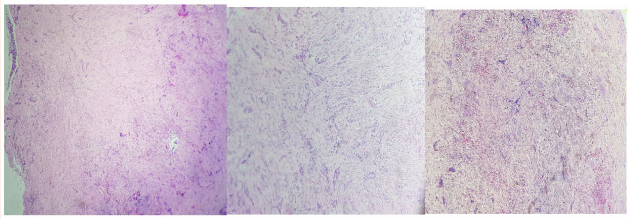

Figure 3: A) Areas of COF and CGCL. Note the close proximity of COF and CGCL (H&E, original magnification ×10) B) Odontogenic epithelial cords with cytoplasmic clearing in close apposition to giant cells (H&E, original magnification ×20) C) numerous multinucleated giant cells in a cellular and hemorrhagic fibrous connective tissue stroma.

Discussion

HCOF-CGCG was first reported by Allen et al in 1992. Also, it can be seen in wide age range, but occurs frequently in second and third decades of life. HCOF-CGCG is a rare hybrid lesion and tends to develop in posterior mandibular area with a female predilection [1,2,5]. It is asymptomatic most of the time, but pain and tooth displacement are found in some cases [13-15,28]. Larger lesions can cross the midline or involve sinuses [8]. According to Taylor et al. study, the lesion is separated from adjacent bone and there is not any encapsulation [13]. Boney cortex thinning and perforation has been reported in some cases [1,8,13]. Radiographically majority of lesions are well-defined unilocular radiolucencies (Table 1). Multilocular lesions and ill-defined borders has been seen in the rest of cases [12]. According to previous documented cases, association with impacted teeth, teeth with root canal therapies or undergoing orthodontic treatment have been reported [12,16].

Three hypotheses have been mentioned to describe the nature of this hybrid lesion:

- The first one suggests "collision tumor" which demonstrates synchronous occurrence of COF in the same position of a CGCG [5,8,12,13].
- The second hypothesis states that the primary CGCG induces COF formation(12) by producing growth factors, cytokines and chemokines [1,8].
- The third one suggests that the primary CGCG is a reparative response to COF with some stimulus like trauma [5,8].

HCOF-CGCG histologically shows densely collagenous to fibro myxoid stroma with several nests of inactive odontogenic epithelium intermixed with plump to flat spindle shaped fibrocellular connective tissue [2]. Also Osteoid deposits, duct like spaces and hyaline basement membrane globules in odontogenic epithelium have been reported [29-31]. present case occurred in a 31-year-old female. If we take into account all data from known cases, 61.7% of total cases occurred in females and 38.7% in males (1.6:1 female to male ratio) which is approximately the same as Upadhyaya report [9]. This is different from female to male ratio previously reported by Tosios et al. [1].

Except two studies, tumors developed in the lower jaw especially at the posterior region. The two mentioned cases occurred in anterior and posterior maxilla [8]. Bony expansion was seen in most of the cases (Table 1), but the present case revealed no buccal or lingual expansion. Also, bony cortex thinning was not present that is in contrast with other reports [2]. Paresthesia is an uncommon finding and was just reported in one case [23]. Also, the lesion size was 1cm that is smaller than other reports [5,8,12,13]. All known cases with the exception of one [24] reveled a radiolucent feature. Similar to previous reports, this case demonstrated a well-defined unilocular radiolucency which was asymptomatic and found accidentally on a routine panoramic radiograph. Curettage was done for 20 cases and 17 cases were treated surgically. recurrence noticed in 6 reports after curettage (12.7%), but there was no evidence of recurrence in 6 months follow-ups (Table 1). Like other reports the lesion had fibro-cellular tissue, myxoid areas, small nests of odontogenic epithelium and multiple cords [1,2,5,9,23]. According to this information HCOF-CGCG has a similar approach to CGCG in development site and behavior [5,9]. Different treatment plans include curettage and excisional surgery. close follow-up is recommended at least 1 year to ensure there is no signs of recurrence [5,9]. It was recommended to rule out the possibility of hyperparathyroidism [23] according to existing of multinucleated giant cells in brown tumors of hyperparathyroidism [9].

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