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Original Article

A retrospective cone beam computed tomography analysis of cemento-osseous dysplasia

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KEYWORDS
Cemento-osseous dysplasia;
Cone beam computed tomography;
Florid;
Subtype

Abstract *Background/purpose:* Radiological examination is indispensable in the diagnosis and follow-up of cemento-osseous dysplasia (COD). The aim of this retrospective study was to describe a series of COD cases, identify the frequencies of COD subtypes, and investigate the demographic and radiological characteristics in relation to subtypes.
Materials and methods: Cone beam computed tomography (CBCT) images/reports of patients with a diagnosis of COD were included in the study. The data collected included information on the age, sex, subtype of COD, location of COD, and region involved. Information regarding the internal density, effects on surrounding structures, and presence of concomitant lesions was also collected. The data obtained were evaluated statistically.
Results: The study group included CBCT images of 142 patients (130 females (91.5%) and 12 males (8.5%)) with a mean age of 46.97 ± 10.57 years. The mandible was involved in almost all cases (99.3%). The most common subtype was florid COD (51.4%) and lesions with hyperdense internal density (81.7%) were more commonly observed. Cortical thinning (78.2%) was a prominent feature. The frequency of root resorption in periapical COD cases (57.1%) was observed to be significantly higher ($p < 0.05$). All hypercementosis cases were associated with florid subtype ($p < 0.05$). In a minority of cases (6.3%), the lesions were associated with bone cysts and osteomyelitis.
Conclusion: CBCT images clearly demonstrated the effect of COD lesions on surrounding structures. CBCT is an appropriate imaging modality for the diagnosis and follow-up of COD which is the most common fibro-osseous lesion in clinical practice.

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Introduction

Cemento-osseous dysplasia (COD), characterized by the replacement of normal bone with fibrous tissue containing mineralized substances consisting of bone, cementum, or both, belongs to the group of fibro-osseous lesions of the jaws.[1–4] COD was reported as the most common fibro-osseous lesion of the jaws in many studies.[5–9] Although the aetiology and pathogenesis remain unknown, the lesions are assumed to originate from the periodontal ligament due to their proximity to the teeth and the formation of cementum-like calcifications and may be the result of an idiopathic/reactive response caused by a disturbance in bone metabolism.[5,6,10–12]

COD predominantly affects women of African and Asian descent, especially in the fourth and fifth decades of life, so hormonal and/or genetic factors are also thought to play a role.[1,7,8,13,14] In the latest classification of the World Health Organization (WHO), COD is divided into three subtypes as periapical, focal, and florid depending on the location and the number of areas involved.[1] The lesions are usually asymptomatic, particularly localized in the mandible, in the apical region of vital teeth or on the edentulous alveolar crest, and most cases are detected incidentally on routine dental radiographs requested for other reasons.[5,11,15]

COD exhibits different radiological characteristics depending on the development stage of the lesion. The early stage immature lesions present a radiolucent/hypodense appearance due to bone resorption and well-vascularized connective tissue deposition, while lesions at the intermediate and mature stages demonstrate mixed and radiopaque/hyperdense appearance, respectively, with a well-defined radiolucent rim.[5,15–18] The differential diagnosis may vary from cystic or inflammatory periodontal lesions to odontogenic tumours depending on the stage.[3,10,14–16,18–20] Care should be paid to avoid misdiagnosis and unnecessary dental procedures such as endodontic treatment/re-treatment, apical resection, and extraction because the lesions are often self-limiting and only radiological follow-up is sufficient.[3,5,14–16,18,20,21]

Although "cemento-osseous dysplasia" is a histopathological term, diagnosis is usually based on the combined evaluation of demographic, clinical, radiological, and follow-up information.[3,7,12,16,17] Invasive dental procedures such as biopsy are contraindicated in these lesions that are susceptible to infections due to cementum-like deposition and decreased local vascularization.[16,17,22] A thorough knowledge of the radiological manifestations is of great importance in the evaluation of COD lesions.[11,13,16]

Recently, cone beam computed tomography (CBCT) has been shown to be a valuable tool in the diagnosis and evaluation of COD lesions, however the number of studies comprising a significant number of COD cases performed using CBCT images is scarce in the literature.[2–4] Based on this information, the aim of this CBCT-based retrospective study was to describe a series of COD cases, to determine the relative frequencies of COD subtypes, and to investigate the demographic and radiological characteristics in relation to subtypes.

Materials and methods

Study design and sample

The design of this retrospective CBCT study was reviewed and approved by the Research Ethics Committee of Marmara University Faculty of Dentistry (protocol number: 2019—316).

A total of 12,670 CBCT images and reports for the period between 2013 and 2019 available in the archives of the Oral and Maxillofacial Radiology Department of Faculty of Dentistry in Marmara University (Istanbul, Turkey) were examined retrospectively. The CBCT scans of patients diagnosed with COD were included in the study and assigned for further analysis. The diagnosis of COD was based on the combined assessment of clinical, radiological, and if available histopathological data. Any missing demographic and clinical data resulted in exclusion.

All CBCT scans had been obtained using a Planmeca ProMax® 3D Mid CBCT unit (Planmeca Oy, Helsinki, Finland) operating at 90 kVp and 10 mA with a field of view dimension of 9×16 cm and a voxel size of 0.2 mm. The images were generated in the digital imaging and communications in medicine (DICOM) format, processed by Planmeca Romexis® software (Planmeca Oy), and analysed in multiplanar reconstructions (coronal, sagittal, axial, cross-sectional, and panoramic).

Data collection and analysis

The data collected from the electronic patient database and Planmeca Romexis® (Planmeca Oy) database included information on the patient age at the time of the CBCT examination, sex, COD subtype (periapical, focal or florid), location of COD (maxilla, mandible or both), region involved (anterior, posterior or both), number of teeth/quadrants involved, and size of the lesion. In addition, information regarding the internal structure (hypodense, mixed or hyperdense), effects on surrounding structures (cortical bone, maxillary sinus, mandibular canal, mental foramen, nasopalatine canal, incisal foramen, nasal fossa, and teeth), and presence of concomitant lesions (cyst or infection) was also collected for each patient.

Following the latest odontogenic tumour classification of the WHO, the COD subtypes were identified as periapical, focal, and florid.[1] Lesions limited to the apical region of the mandibular anterior teeth were classified as periapical, those associated with a single posterior tooth as focal, and those affecting multiple quadrants as florid.

The number of involved teeth was recorded only for periapical, the number of quadrants only for florid, and the size of the lesion only for focal subtype. Teeth associated with the COD lesions were evaluated for the presence of endodontic treatment, root resorption, displacement, and hypercementosis. Root resorptions in endodontically treated teeth were excluded since it was unclear if the origin of the resorption was inflammation or dysplasia.[2,4]

Observer

The CBCT images belonging to patients with COD were selected by an oral and maxillofacial radiologist with 20 years of experience (BG). All assessments in CBCT images belonging to cases diagnosed as COD were performed simultaneously by two observers (BG&MPA) with inbuilt software using a Dell Precision 5520 mobile workstation with a 15.6 inch screen having a resolution of 1920 × 1080 pixels (Dell, Round Rock, TX, USA) in a dark quiet room.

Statistical analysis

Statistical analysis was performed using the IBM® SPSS Statistics 22 software (IBM SPSS, Turkey). Kolmogorov—Smirnov and Shapiro Wilks tests were used to verify the normal distribution in the sample. In addition to descriptive statistics, quantitative data were compared using one-way ANOVA test. For the comparison of qualitative data, chi-square, Fisher's exact, and Fisher Freeman Halton tests were used. The significance level was set at $p < 0.05$.

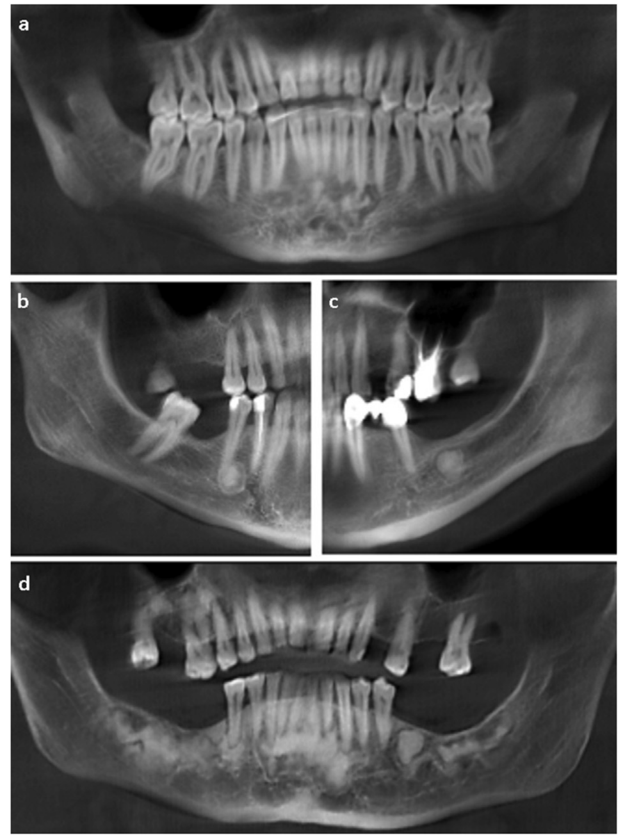

Figure 1 Panoramic reconstruction cone beam computed tomography images of **a.** periapical, **b.** focal (single posterior tooth), **c.** focal (single posterior edentulous alveolar crest), and **d.** florid cemento-osseous dysplasia cases.

Results

Out of a total of 12,670 CBCT scans, 142 COD cases were detected and re-evaluated (1.1%). The mean age of the sample was 46.97 ± 10.57 years (range 24—75). Most of the patients were female (n = 130, 91.5%) aged between 24 and 75 years (46.85 ± 10.70 years). COD was most commonly found in the mid-age population, with peak prevalence in the fifth decade.

From a total of 142 cases, the most common COD subtype was florid (n = 73, 51.4%), followed by focal (n = 55, 38.7%), and periapical (n = 14, 9.9%) (Fig. 1). The difference between the mean ages in COD subtypes, and the distribution of COD subtypes regarding the age groups were not found to be significant ($p > 0.05$). For all COD subtypes, a female predominance was observed ($p < 0.05$). The rate of florid subtype in females (53.1%) and periapical subtype (33.3%) in males was detected to be significantly higher ($p < 0.05$). Demographic characteristics in relation to COD subtypes are further detailed in Table 1.

Table 2 provides the detailed data regarding the radiological characteristics in relation to COD subtypes. The mandible was involved in almost all COD cases (n = 141, 99.3%). Within the mandible, the posterior region was the most prevalent site (n = 125, 88%). COD in the late stage (81.7%) was more commonly observed than the intermediate (48.6%) and the early (9.2%) stages. Significant differences in terms of internal density in relation to the subtypes were detected. The incidence of hypodense internal density (0%) was found to be significantly lower in the focal COD ($p < 0.05$), and mixed internal density (72.6%) was detected to be significantly higher in the florid COD ($p < 0.05$).

The cortical bone was frequently affected by COD and cortical thinning (78.2%) was a prominent feature of all subtypes (Fig. 2). The rates of cortical thinning, expansion, and perforation in focal COD cases were significantly lower in comparison to the periapical and florid subtypes ($p < 0.05$). The floor of the maxillary sinus (n = 10, %13.7) and the mandibular canal (n = 12, 16.4%) were involved more frequently in florid subtype ($p < 0.05$).

Root resorption associated with COD was detected in 23 of the cases (16.2%). The frequency of root resorption in periapical COD cases (57.1%) was observed to be significantly higher in comparison to the focal (3.6%) and the florid COD cases (17.8%) ($p < 0.05$). All hypercementosis cases were associated with florid subtype ($p < 0.05$).

The lesions were associated with bone cysts in 8 of the cases (5.6%), all of which were florid subtype ($p < 0.05$). Moreover, only 1 case (0.7%), which was again the florid subtype, was associated with infection (Fig. 3).

Table 1 Demographic characteristics of cemento-osseous dysplasia cases in relation to subtypes.

		Total	Cemento-osseous Dysplasia Subtype			p
			Periapical	Focal	Florid	
		n (%)	n (%)	n (%)	n (%)	
Age $_{Mean \pm SD}$		46.97 ± 10.57	45.14 ± 10.36	46.84 ± 10.69	47.42 ± 10.63	[a]0.758
Age group	24—39	39 (%27.5)	4 (%10.3)	17 (%43.6)	18 (%46.2)	[b]0.800
	40—49	47 (%33.1)	4 (%8.5)	20 (%42.6)	23 (%48.9)	
	50—59	37 (%26.1)	4 (%10.8)	10 (%27)	23 (%62.2)	
	60+	19 (%13.4)	2 (%10.5)	8 (%42.1)	9 (%47.4)	
Sex	Female	130 (%91.5)	10 (%7.7)	51 (%39.2)	69 (%53.1)	[c]0.032*
	Male	12 (%8.5)	4 (%33.3)	4 (%33.3)	4 (%33.3)	

*$p < 0.05$.
[a] One-way ANOVA test.
[b] Chi-square test.
[c] Fisher Freeman Halton test.

Table 2 Radiological characteristics of cemento-osseous dysplasia in relation to subtypes.

		Total	Cemento-osseous Dysplasia Subtype			p
			Periapical	Focal	Florid	
		n (%)	n (%)	n (%)	n (%)	
Jaw	Mandible	117 (%82.4)	14 (%100)	54 (%98.2)	49 (%67.1)	[a]0.001*
	Maxilla	1 (%0.7)	0 (%0)	1 (%1.8)	0 (%0)	
	Mandible + Maxilla	24 (%16.9)	0 (%0)	0 (%0)	24 (%32.9)	
Region	Anterior mandible	48 (%33.8)	14 (%100)	0 (%0)	34 (%46.6)	[b]0.001*
	Posterior mandible	125 (%88)	0 (%0)	54 (%98.2)	71 (%97.3)	[b]0.001*
	Anterior maxilla	10 (%7)	0 (%0)	0 (%0)	10 (%13.7)	[c]0.005*
	Posterior maxilla	21 (%14.8)	0 (%0)	1 (%4.8)	20 (%27.4)	[b]0.001*
Internal density	Hypodense	13 (%9.2)	2 (%14.3)	0 (%0)	11 (%15.1)	[b]0.011*
	Mixed	69 (%48.6)	6 (%42.9)	10 (%18.2)	53 (%72.6)	[b]0.001*
	Hyperdense	116 (%81.7)	11 (%78.6)	45 (%81.8)	60 (%82.2)	[b]0.949
Effect on cortical bone	Intact	28 (%19.7)	0 (%0)	20 (%36.4)	8 (%11)	[b]0.001*
	Thinning	111 (%78.2)	14 (%100)	32 (%58.2)	65 (%89)	[b]0.001*
	Expansion	59 (%41.5)	10 (%71.4)	13 (%23.6)	36 (%49.3)	[b]0.001*
	Perforation	49 (%34.5)	9 (%64.3)	10 (%18.2)	30 (%41.1)	[b]0.001*
Effect on anatomical structures	Maxillary sinus	11 (%7.7)	0 (%0)	1 (%1.8)	10 (%13.7)	[c]0.034*
	Mandibular canal	13 (%9.2)	0 (%0)	1 (%1.8)	12 (%16.4)	[b]0.008*
Effect on teeth	Root resorption	23 (%16.2)	8 (%57.1)	2 (%3.6)	13 (%17.8)	[b]0.001*
	Endodontic treatment	15 (%10.6)	0 (%0)	4 (%7.3)	11 (%15.1)	[b]0.146
	Hypercementosis	17 (%12)	0 (%0)	0 (%0)	17 (%23.3)	[b]0.001*
Complication	None	133 (%93.7)	14 (%100)	55 (%100)	64 (%87.7)	[a]0.023*
	Cyst	8 (%5.6)	0 (%0)	0 (%0)	8 (%11)	
	Infection	1 (%0.7)	0 (%0)	0 (%0)	1 (%1.4)	

*$p < 0.05$.
[a] Fisher's Exact test.
[b] Chi-square test.
[c] Fisher Freeman Halton test.

For periapical COD cases, 43 teeth were involved in lesions and the mean number of teeth in each case was 3.07 ± 1.44 (range 1—5). For focal COD cases, 18 were in relation to teeth (32.7%) and 37 were in edentulous areas (67.3%). The size of the focal COD lesions ranged between 5 and 30 mm (11.98 ± 4.84, median 10 mm). Florid COD cases comprised 51 two-quadrant, 11 three-quadrant, and 11 four-quadrant involvement.

Discussion

This study depicts the demographic, clinical, and radiological characteristics of a patient series with COD, but may not be reflecting the prevalence and features of COD in the entire Turkish population. Although our faculty is an important reference centre for the diagnosis and management of oral and maxillofacial lesions and attracts patient

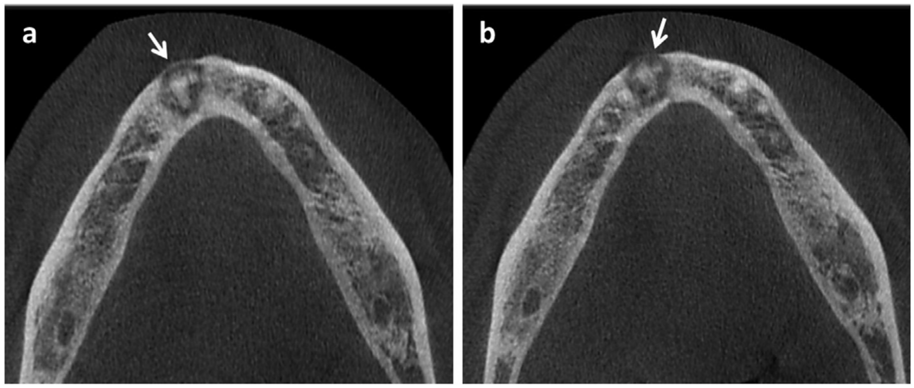

Figure 2 Two different axial cone beam computed tomography slices of the same periapical cemento-osseous dysplasia case showing the effects on cortical bone: **a.** thinning + expansion and **b.** perforation (white arrows).

Figure 3 Panoramic reconstruction and cross-sectional cone beam computed tomography images of two different florid cemento-osseous dysplasia cases with **a.** cystic lesion exhibiting a well-defined hypodense area with cortical involvement and **b.** inflammatory lesion exhibiting hypodense area with a hyperdense area resembling bone sequestration and also hypercementosis.

populations from various parts of the city and its surroundings, the study sample may not represent a random sample of the Turkish population. However, the findings of this study may provide important information and perspectives on the subject under investigation and may guide future research.

There are relatively few similar studies comprising exclusively a significant number of COD cases (Table 3).[2–4,10,13,16,17,22,23] In the literature, the inconsistent nomenclature and descriptions of COD and its subtypes have led to confusion.[3] In most of the earlier studies, COD subtyping was not performed or a specific type of COD was evaluated.[21,24–26] Some studies incorporated COD into the spectrum of lesions formerly referred to as "cementomas",[27,28] whereas others studied COD under the topic of fibro-osseous lesions along with ossifying fibroma and fibrous dysplasia.[7–9,11,29–32] Furthermore, conflicting conclusions regarding the demographic, clinical, and radiological profile of COD were reached. The available data in literature may vary due to the population studied, the origin of the cases (pathology laboratories, oral and maxillofacial radiology units), the classification used, and the design of study performed.[6,14,16]

In previous similar studies, it was reported that the mean age at the time of diagnosis ranged between 40.2 and 55.9 years, most patients with COD were females ranging from 82.3% to 96% of the population studied, and the mandible was affected in 81.4–100% of the individuals (Table 3).[2–4,10,13,16,17,22,23] Consistent with the previous studies, the mean age of the sample was 46.97 years, most of the patients with COD were female (91.5%), and the mandible was affected in 99.3% of the cases in the present study. These findings support studies that pointed to a complex interaction between genetic, hormonal, and environmental factors in the development of this disease[16] and that its origin may be related to the periodontal ligament.[19,25]

In retrospective studies conducted in various countries, a consensus about the most prevalent subtype could not be reached (Table 3).[2–4,10,13,16,17,22,23] Florid COD was the

Table 3 Previous similar studies listed in chronological order with reference number, author(s), country, number of cases, selection criteria of cases, mean age, and most commonly encountered sex/location/subtype.

Author(s) (year)	Country	Cases (n)	Selection of cases	Mean age (years)	Sex (%)	Location (%)	Subtype (%)
Kawai et al. (1999)[10]	Japan	54	histopathology records, clinical records, and radiographs (periapical, occlusal, OPTG, posteroanterior skull radiographs)	50.8	female (91%)	(–)	multiplex (67%)[a]
Cho et al. (2007)[13]	Korea	33	clinical records and radiographs (OPTG)	55.9	female (84.8%)	mandible (100%)	focal (51.5%)
Alsufyani&Lam (2011)[16]	Canada	118	charts and radiographic reports (OPTG, occlusal, periapical, CT, MRI)	44.3 for females 40.1 for males[b]	female (82.9%)	mandible only (81.4)	periapical (78.8%)[c]
Owosho et al. (2013)[17]	USA	35	histopathology records, clinical records, and radiographs (periapical, full-mouth series, OPTG)	53.9	female (94.3%)	mandible (100%)	florid (48.6%)
Cavalcanti et al. (2018)[2]	Brazil	82	CBCT	49.8	female (86.6%)	mandible (91.5%)	periapical (57.3%)
Oh et al. (2019)[3]	Australia	62	CBCT	40.2	female (82.3%)	mandible (100%)	focal (91.9%)[d]
Benaessa et al. (2019)[23]	South Africa	133	histopathology records, clinical records, and radiographs (OPTG)	53.4	female (94.7%)	mandible (95.5%)	florid (69.9%)
Olgac et al. (2020)[22]	Turkey	135	histopathology records, medical records, and radiographs (–)	40.6	female (88.9%)	mandible (97%)	focal (61.5%)
Kato et al. (2020)[4]	Brazil	60	CBCT	46.57	female (96.6%)	mandible (100%)	florid (80%)
Present study Gumru et al.	Turkey	142	CBCT	46.97	female (91.5%)	mandible (99.3%)	florid (51.4%)

OPTG: orthopantomograph; CT: computed tomography; MRI: magnetic resonance imaging; CBCT: cone beam computed tomography; (-) not provided.
[a] Florid + periapical subtypes classified as multiplex.
[b] Overall mean age not provided.
[c] Focal + periapical subtypes classified as periapical.
[d] Focal + periapical subtypes were classified as focal.

most common subtype (51.4%) in the present study, however its frequency was lower compared to the studies conducted in South Africa and Brazil reporting a frequency of 69.9% and 80%, respectively.[4,23] This finding may reflect the fact that florid subtype is more frequently characterized by mixed or hyperdense internal density, greater bone involvement, and pathognomonic features compared to other subtypes. Another reason may be that the florid COD cases more often become symptomatic resulting in patient complaints and admission.[4,9] In addition, considering that some cases of focal subtype were reported to progress to florid subtype,[33] available data may suggest that the COD cases were diagnosed quite late.[9]

Although the use of two-dimensional imaging modalities such as panoramic and periapical radiographs may usually be sufficient in most COD lesions, the information they provide regarding the extent of the lesions and relationship with surrounding structures is limited, and COD lesions were reported to reveal minimal effects on adjacent structures in studies conducted by means of these techniques.[2–4,10,16] In COD cases, CBCT evaluations, allowing for three-dimensional imaging with minimal distortion, high spatial resolution, and no overlapping, have increased recently and are indicated primarily not for diagnostic confirmation but rather to assess the involvement of adjacent structures.[4]

Radiological appearance of COD may range from hypodense to mixed or hyperdense areas on CBCT images due to the maturation level and mineralized tissue deposition over time.[2] Since COD lesions in the early stages resemble periapical inflammation, their distinction from chronic periapical lesions is difficult.[15,34] Previous studies reported majority of the COD lesions to present mixed internal density.[2,4,16] This finding was attributed to the patients seeking care during the progression of the lesion and the fact that this maturation phase was quite characteristic for the recognition of radiological findings.[2] Nevertheless, in our study, most of the COD lesions were detected to be hyperdense (81.7%) indicating the greater calcification of neoformed tissue and unfortunately late detection of lesions.

In the current study, the relationship of COD with adjacent structures was investigated by means of multiplanar CBCT reconstructions. In all COD subtypes, similar to that reported in the previous limited number of studies performed using CBCT images, cortical bone was detected to be the most affected structure. The most common damage in the cortical bone was thinning, followed by expansion, and perforation.[2–4] However, the figures reported by Cavalcanti et al.[2] are lower than the current study, which may be attributed to the difference in the most prevalent COD subtypes. In florid COD, usually more damage occurs in the cortical bone compared to the other subtypes.[35,36]

Another effect of COD on the adjacent structures is root resorption, which is considered important in evaluating the severity of damage.[4] Root resorptions were detected to be rare in COD patients undergoing two-dimensional imaging,[16] however higher in those submitted to CBCT evaluation.[2,4] Even when the endodontically treated teeth were excluded in this study, a number of teeth with root resorption associated with COD lesions were identified (16.2% of cases). Hypercementosis, hyperdense areas of the lesions attached to roots, was reported to be observed in 9% and 17% of the COD cases,[10,16] and in 8.2% of the teeth.[4] Similarly, this figure was 12% of the cases in our study.

COD lesions usually remain asymptomatic unless secondarily infected, and ideally the diagnosis should be based on a combination of clinical and radiological assessments, and biopsy is not recommended except for a limited number of cases.[9,11,12,16,22,23] In the current study, most cases were asymptomatic and the lesions were noticed during routine dental examinations with the exception of only one case of infected COD. Infection occurring within a COD lesion, namely osteomyelitis, is the major complication that may be encountered.[37] Therefore, any surgical intervention, including those for diagnostic purposes such as biopsy, is usually not recommended due to the infection risk.[19,37] Simple and aneurysmal bone cysts and non-specific cystic degenerations have also been associated with florid, and less frequently with periapical COD.[2,16,38–40] In our sample, 8 cases of florid COD with bone cysts were identified, however detailed information regarding the type of cysts were not available in the electronic patient database which can be considered as another limitation of this study. The frequency of osteomyelitis and bone cysts, as a complication of COD, varied in different studies, and it should not be surprising that the frequency was found to be high in studies where the case selection was based on histopathological diagnosis.[22,23,31]

In conclusion, florid COD was the most common subtype indicating that COD lesions are usually detected in the late period after causing significant damage in the surrounding structures. Three-dimensional CBCT images provide information related to the extent of involvement and better understanding the effects on surrounding structures, and are useful in the diagnosis and follow-up of COD cases.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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