

Cemento-osseous dysplasia: Re-visited

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Abstract

Objectives: *Cemento-osseous dysplasia is a benign fibro-osseous lesion characterised by a radiological presentation that varies as it matures through three distinct stages: early, intermediate and late stages. It encompasses a wide range of radiographic appearances, and its presence in the tooth-bearing regions of the jaws may cause confusion with many other types of entities with radiolucent, mixed or radiopaque appearances. The aim of this paper is to evaluate and review the common radiographic features of cemento-osseous dysplasia on cone-beam computed tomography to allow for better assessment and recognition of this condition.*

Methods: *A series of cone-beam computed tomography datasets with findings of cemento-osseous dysplasia was reviewed using dedicated software. Radiographic features of cemento-osseous dysplasia were recorded.*

Results: *Cemento-osseous dysplasia was more common in females (82.3%) than in males (17.7%). The posterior mandible was the most favoured site (61.3%) for all types of cemento-osseous dysplasia. Cortical expansion (51.6%) and cortical thinning (88.7%) were prevalent radiographic features.*

Conclusions: *Cortical thinning and cortical expansion are not commonly reported features of COD, and our study is the first to present that these features are prevalent regardless of the COD type. It is therefore critical for all dental practitioners to recognise COD and to avoid confusion with other expansile lesions in the head and neck region, such as odontogenic tumours and cysts.*

Keywords: Cemento-Osseous Dysplasia; Cone-Beam Computed Tomography; Fibro-Osseous Lesion; Florid Cemento-Osseous Dysplasia; Focal Cemento-Osseous Dysplasia

Introduction

Cemento-osseous dysplasia (COD) is a cementum or bone producing fibro-osseous lesion confined to the tooth-bearing regions of the jaws. Normal bone architecture is replaced by fibroblasts and collagen fibres with varying amounts of mineralised material. The aetiology of this condition remains unknown but is considered non-neoplastic and possibly derived from reactive or dysplastic changes in the periodontal ligament or medullary bone¹.

Classification

The World Health Organization (WHO) has recently updated the classification system for COD to re-include “cemento” in the nomenclature after it had previously been excluded in favour of the term “osseous dysplasia”.² This re-naming to include “cemento” within the title was to distinguish the lesions as being of odontogenic origin which explains why all COD lesions occur within the tooth-bearing areas in jaws.

Several classifications for COD have been previously proposed and are listed in Table 1. These classifications are largely based on the site and extent of involvement within the jaws. The World Health Organization (WHO) classifies the entity as periapical COD, focal COD and florid COD. Periapical and focal CODs share identical histopathology and clinical profiles apart from their site of involvement in the anterior and posterior jaws, respectively. According to Eversole et al’s classification, both periapical and focal COD are encompassed together by the term focal COD. This paper uses the simplified classification proposed by Eversole et al in 2008: focal COD (FCOD) and florid COD (FL.COD).^{2,3}

Table 1. Proposed classifications of cemento-osseous dysplasia in chronological order.

Year	Classification	Nomenclature of Cemento-osseous Dysplasia
1990	Slootweg and Muller	- Periapical Cemental Dysplasia - Florid Osseous Dysplasia
1992	World Health Organization	- Periapical Cemental Dysplasia (Periapical Fibrous Dysplasia) - Florid Cemento-Osseous Dysplasia (Gigantiform Cementoma, Familial Multiple Cementomas) - Other Cemento-Osseous Dysplasia
1993	Waldron	- Periapical Cemento-Osseous Dysplasia - Focal Cemento-Osseous Dysplasia - Florid Cemento-Osseous Dysplasia
2001	Brannon and Fowler	Osseous Dysplasia (Reactive) - Nonhereditary <ul style="list-style-type: none"> • Periapical • Focal • Florid - Hereditary (Developmental) <ul style="list-style-type: none"> • Familial Gigantiform Cementoma
2004	MacDonald-Jankowski	- Cemento-Ossifying Dysplasia - Periapical Cemental Dysplasia - Florid Cemento-Osseous Dysplasia
2005	World Health Organization	- Periapical Osseous Dysplasia - Focal Osseous Dysplasia - Florid Osseous Dysplasia - Familial Gigantiform Cementoma
2006	Speight and Carlos	- Periapical Osseous Dysplasia - Focal Osseous Dysplasia - Florid Osseous Dysplasia - Familial Gigantiform Cementoma
2008	Eversole et al	- Focal Cemento-Osseous Dysplasia - Florid Cemento-Osseous Dysplasia
2017	World Health Organization	- Periapical Cemento-Osseous Dysplasia - Focal Cemento-Osseous Dysplasia - Florid Cemento-Osseous Dysplasia

These classifications are largely based on the site and extent of involvement within the jaws. The World Health Organization (WHO) classifies the entity as periapical COD, focal COD and florid COD. Periapical and focal CODs share identical histopathology and clinical profiles apart from their site of involvement in the anterior and posterior jaws, respectively. According to Eversole et al's classification, both periapical and focal COD are encompassed together by the term focal COD. This paper uses the simplified classification proposed by Eversole et al in 2008: focal COD (FCOD) and florid COD (FL.COD)^{2,3}.

Focal Cemento-osseous Dysplasia

Focal COD displays a discrete, single site of involvement in any region of the alveolar process. It occurs predominantly in the posterior mandible associated with vital teeth; the maxilla is rarely affected. Lesions are usually asymptomatic and display limited growth potential, rarely exceeding 2 cm in size and seldom causing cortical expansion³.

The radiological presentation of FCOD varies as it matures through three distinct phases: early, intermediate and late stages^{3,4}. The early stage typically presents as a well-defined radiolucency associated with the apices of mandibular teeth. Tooth displacement or resorption is seldom noted. This stage can resemble inflammatory lesions of endodontic origin, and therefore pulpal vitality testing is essential to differentiate the entities and to avoid misdiagnosis⁵. The intermediate stage presents as a mixed radiolucent-opaque pattern with a well-defined radiolucent rim and internal radiopaque calcific depositions². The late stage presents as a diffuse radiopacity, often with ill-defined borders³.

Florid Cemento-osseous Dysplasia

Multiple COD lesions involving two or more quadrants of the jaw are termed florid cemento-osseous dysplasia (FL.COD). In most cases, the lesion extends bilaterally along the mandible and may or may not show concomitant maxillary involvement.

The radiological presentation of FL.COD is characterised by multiple confluent and non-expansile radiopacities with circumferential radiolucencies most commonly seen in the mandibular premolar and molar regions³. Florid COD also progresses through the three distinct stages radiographically; early, intermediate and late. In most cases, FL.COD tends to present extensively in the jaws despite having a similar microscopic appearance to FCOD^{2,6}.

Histopathology

Diagnosis of COD is usually based on radiographic and clinical features without need for biopsy, unless lesions are atypical in presentation. All variants of COD display similar microscopic features, comprising of fibrous stroma with associated calcific products, including woven or lamellar bone, osteoid and cementum-like material. It is uncommon to note osteoblastic rimming. The tissue is generally vascular in its early stages and unencapsulated. With increasing maturity, the lesions become denser, less cellular and less vascular².

Due to the widely varying radiographic features of COD as they progress through the different stages of maturity, they may be confused with other radiolucent, mixed or radiopaque entities occurring in the jaws. With the increasing use of cone-beam computed tomography (CBCT) in dental practice, it is important for clinicians to be familiar with the features of COD in order to recognise the condition and avoid misdiagnosis. This study aims to evaluate and review the common radiographic features of COD on CBCT.

Materials and Methods

Dental CBCT scans of Australian patients acquired in a radiology clinic from the year 2008 to 2018 were evaluated retrospectively using the Invivo6 software program (Anatomage, San Jose, CA, USA). Patient data was anonymised prior to examination. Patient age and gender were recorded separately.

A total of 62 CBCT scans with the incidental finding of COD were carefully selected by a dento-maxillofacial radiologist. All cases of COD located adjacent to endodontically treated teeth or carious teeth were excluded to avoid the possibility of the misdiagnosis as an inflammatory lesion. Early-stage (well-defined radiolucency) COD cases were also excluded for the same reason.

The selected CBCT scans were examined by dental practitioners in the dento-maxillofacial radiology department at an Australian university. Each scan was assessed for the presence of expansion and thinning of the adjacent cortices using orthogonal and other reconstructed planes. Classification based on Eversole et al's 2008 classification, degree of maturity (intermediate and late stages), associated regions (maxilla, mandible and both jaws), tooth displacement, root resorption and fusion between the lesion and the associated teeth were also recorded [3].

All scans were then re-evaluated by two experienced private Australian dento-maxillofacial radiologists. This research has an ethics approval from an Australian university ethics committee.

Data Analysis

The statistical analysis of data was performed using the JASP software program.

Results

Demographics

In this study, out of the 62 patients, 11 (17.7%) were male and 51 (82.3%) were female, aged between 12 and 79 years (mean age SD: 40.2 14.7 years). Distribution of the demographic features in COD is further detailed in Table 2. No FL.COD was detected in the male population.

Table 2. Demographic features of cemento-osseous dysplasia.

Demographics	Cemento-osseous Dysplasia		
	Focal		Florid
	Unilocular	Multilocular	
Total (n=62)	29 (46.8%)	28 (45.1%)	5 (8.1%)
Sex			
Male (n= 11)	3	8	0
Female (n= 51)	26	20	5
Age (years)			
Mean \pm SD	35.7 \pm 15.8	44.0 \pm 13.2	44.8 \pm 6.9
Range	12-65	15-79	33-51

Figure 1. Histogram showing age distribution of cemento-osseous dysplasia.

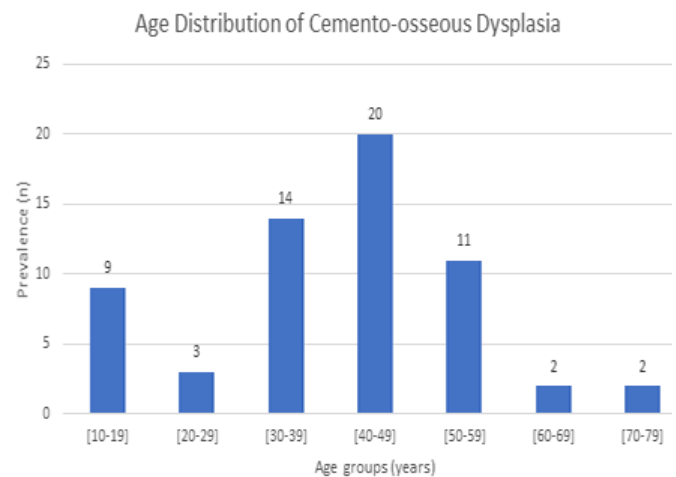


Figure 1 shows the age distribution in all types of COD. Cemento-osseous dysplasia was most commonly found in the mid-age population (30-59 years), with peak prevalence in the 40-49 years age group.

Radiographic Features

Cemento-osseous dysplasia was most commonly found in the mandible (93.5%). Within the mandible, the posterior region – distinguished by the involvement of posterior dentition – was the most prevalent site for all types of COD

Table 3. Distribution of cemento-osseous dysplasia in the jaw based on classification.

Region / Location	Cemento-osseous Dysplasia			
	Focal (unilocular)	Focal (multilocular)	Florid	Total
Total	29 (46.8%)	28 (45.1%)	5 (8.1%)	62 (100%)
Region				
Mandible	29	24	5	58 (93.5%)
Anterior	7	3	0	10 (16.1%)
Posterior	22	15	1	38 (61.3%)
Anterior & Posterior		6	4	10 (16.1%)
Maxilla	-	-	-	-
Both jaws		4		4 (6.5%)
Anterior		1	-	1 (1.6%)
Posterior	-	1	-	1 (1.6%)
Anterior & Posterior	-	2	-	2 (3.2%)

There were no maxilla-only cases of COD found in this study. There were three cases of COD observed in edentulous regions.

Focal COD is divided into unilocular FCOD and multilocular FCOD according to their peripheral morphology

Figure 2. Cone-beam computed tomography images demonstrating unilocular focal cemento-osseous dysplasia associated with the 33 root apex. Cortical thinning in relation to the lesion is also demonstrated.

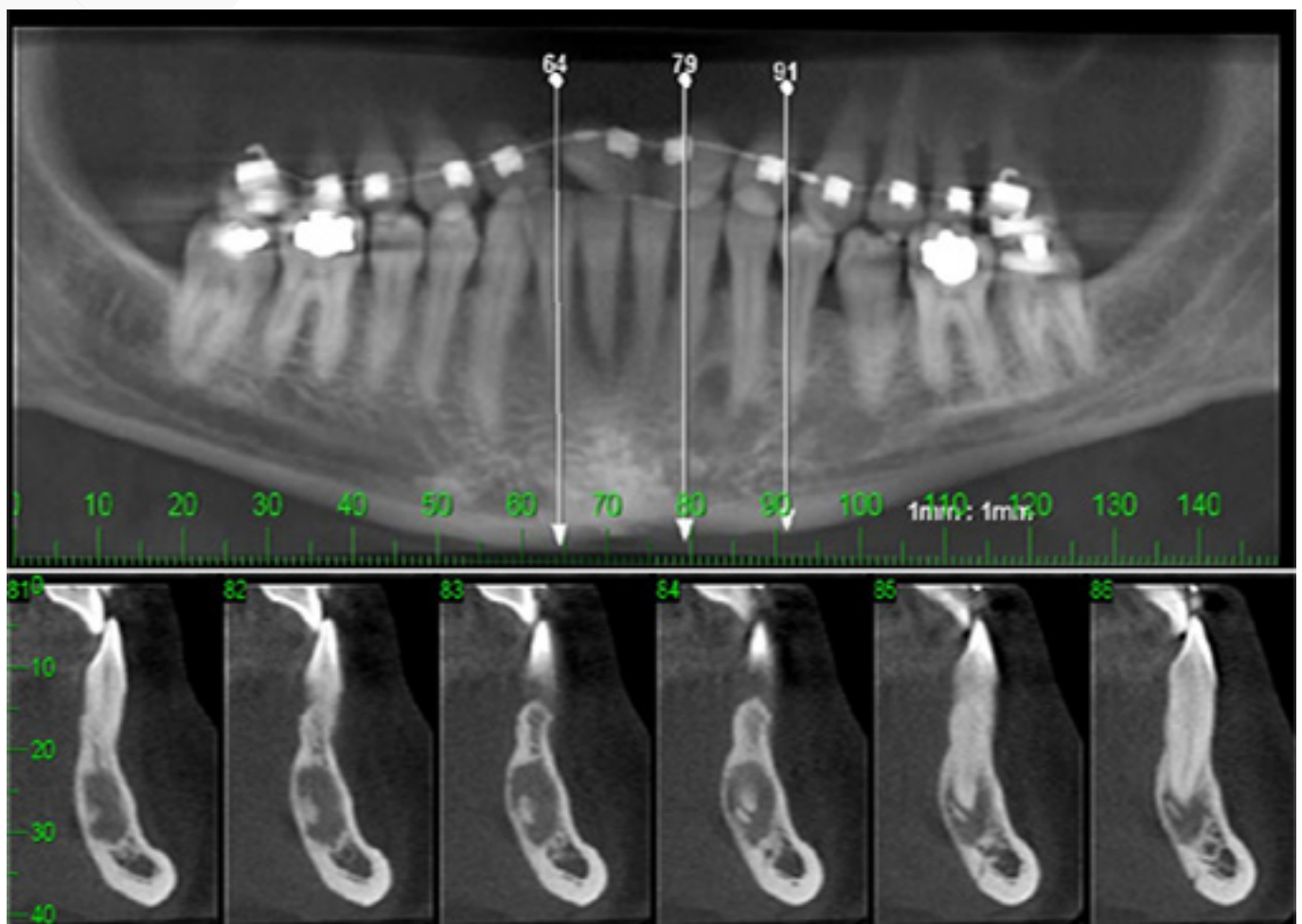


Figure 3. Multilocular focal cemento-osseous dysplasia in the right posterior mandible. Multiple internal calcifications are also noted (intermediate stage with heterogenous pattern).

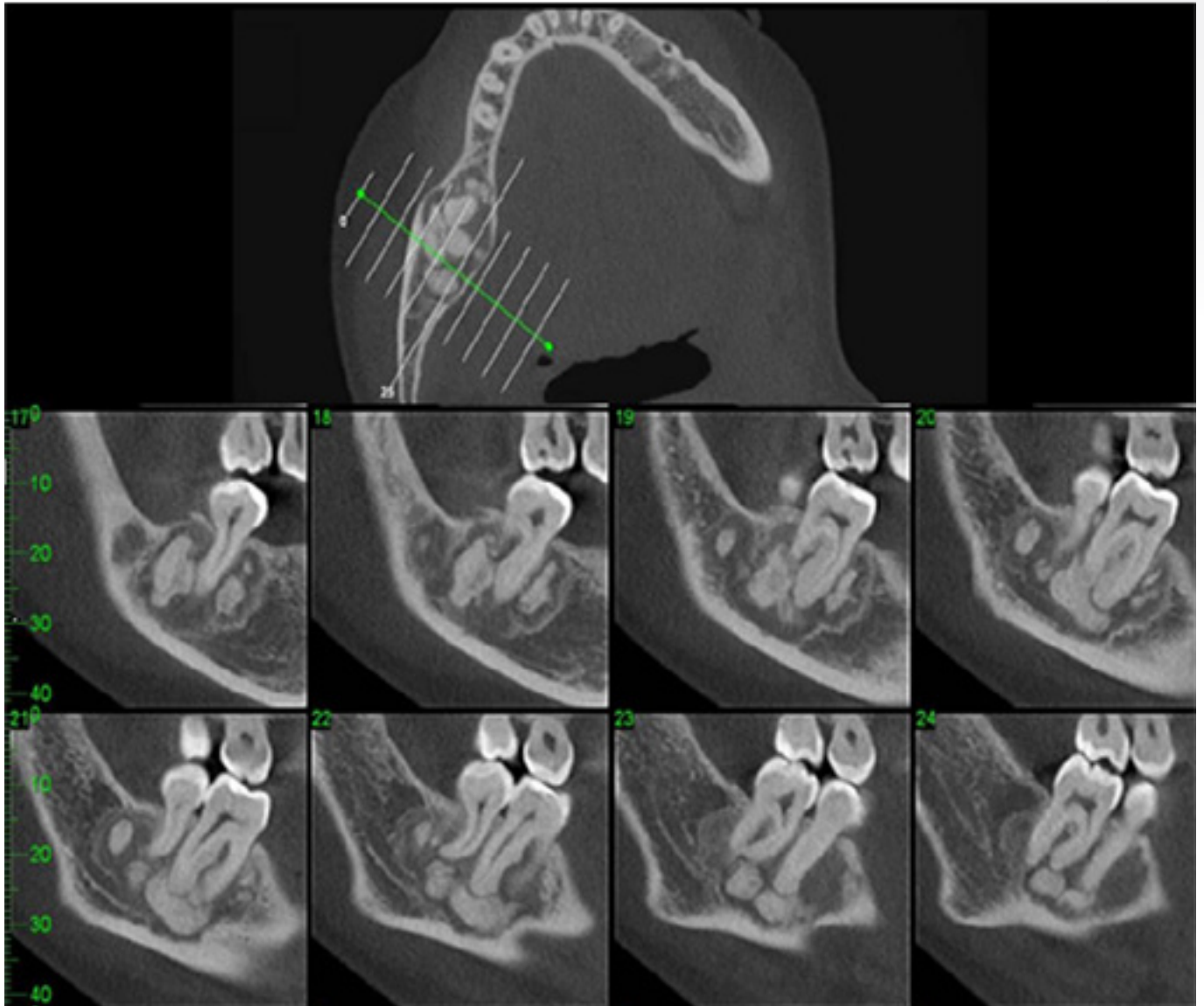


Table 4. Radiographic features of cemento-osseous dysplasia.

Radiographic Features	Cemento-osseous Dysplasia			
	Focal (unilocular)	Focal (multilocular)	Florid	Total
Total	29 (46.8%)	28 (45.1%)	5 (8.1%)	62 (100%)
Cortical Expansion				
Present	14 (22.6%)	14 (22.6%)	4 (6.5%)	32 (51.6%)
Absent	15 (24.2%)	14 (22.6%)	1 (1.6%)	30 (48.4%)
Cortical Thinning				
Present	25 (40.3%)	25 (40.3%)	5 (8.1%)	55 (88.7%)
Absent	4 (6.5%)	3 (4.8%)	-	7 (11.3%)
Maturity				
Intermediate – heterogenous calcification	11 (17.7%)	9 (14.5%)	3 (4.8%)	23 (37.1%)
Intermediate – homogenous calcification	12 (19.4%)	14 (22.6%)	2	28 (45.2%)
Late (complete calcification)	6 (9.7%)	5 (8.1%)	-	11 (17.7%)
Tooth Displacement	-	-	-	-
Root resorption	-	-	-	-
Fusion				
Present	1 (1.6%)	4 (6.5%)	-	5 (8.1%)
Absent	28 (45.1%)	24 (38.7%)	5 (8.1%)	57 (91.9%)

Table 4 presents the data referring to different radiographic features of COD for each classification.

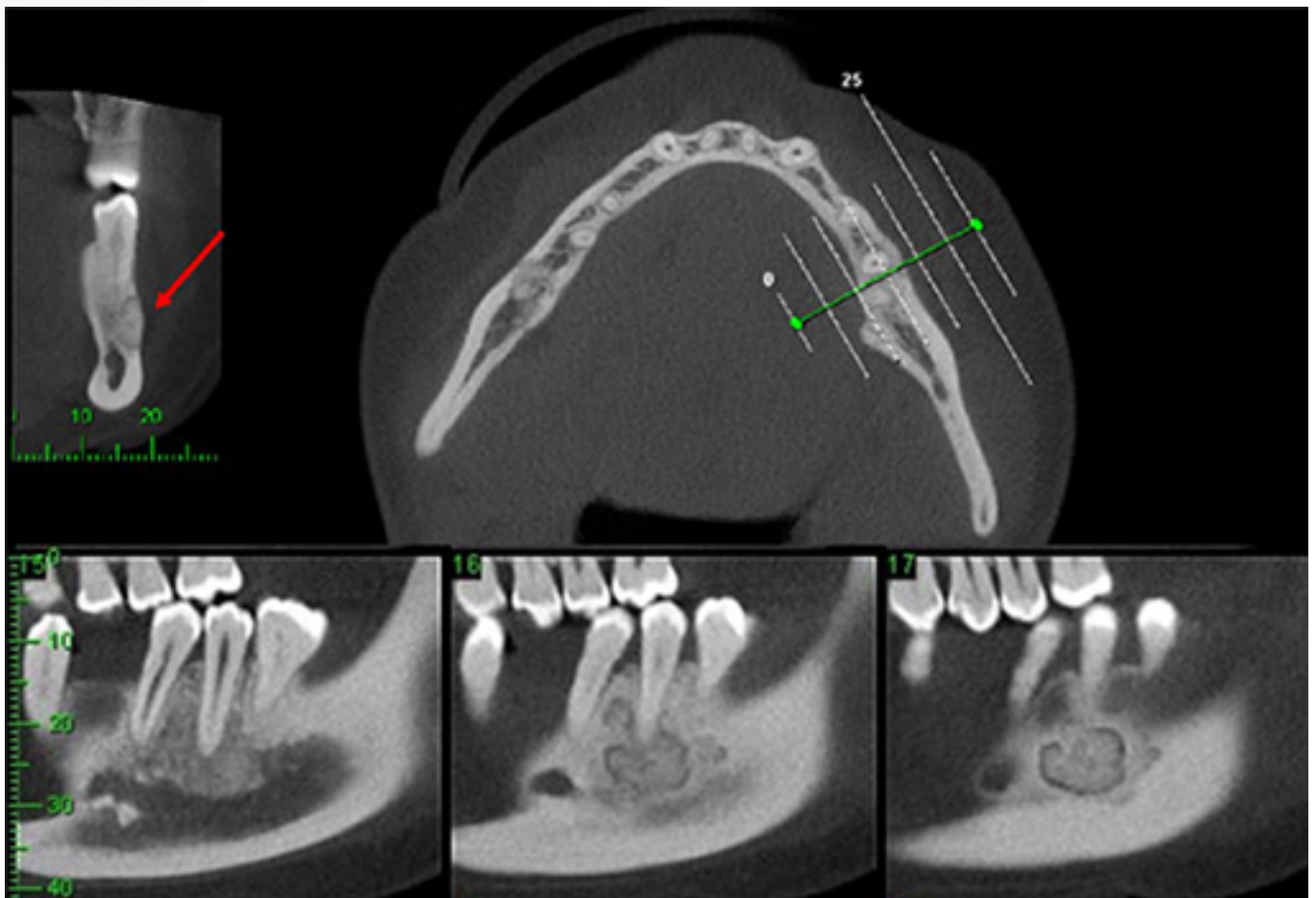


Figure 4. Heterogenous focal cemento-osseous dysplasia (ground-glass pattern) at the 35 periapical region. Extreme thinning of the adjacent buccal cortical plate is noted (red arrow).

Cortical thinning (88.7%) was a prominent feature of all types of COD, as was cortical expansion (51.6%). There were no cases of tooth displacement or root resorption in relation to COD in this study.

Cemento-osseous dysplasia in its intermediate stage (82.3%) was more commonly observed than the late stage COD (17.7%). Within the intermediate stage, two different internal calcification patterns were noted with near equal frequency: heterogenous (37.1%) and homogenous (45.2%)

Figure 5. Florid cemento-osseous dysplasia in the mandible. The cross-sectional images demonstrate the 44 periapical region with multiple internal calcifications and a ground-glass pattern. Significant expansion of the buccal and lingual cortices with thinning is also noted.

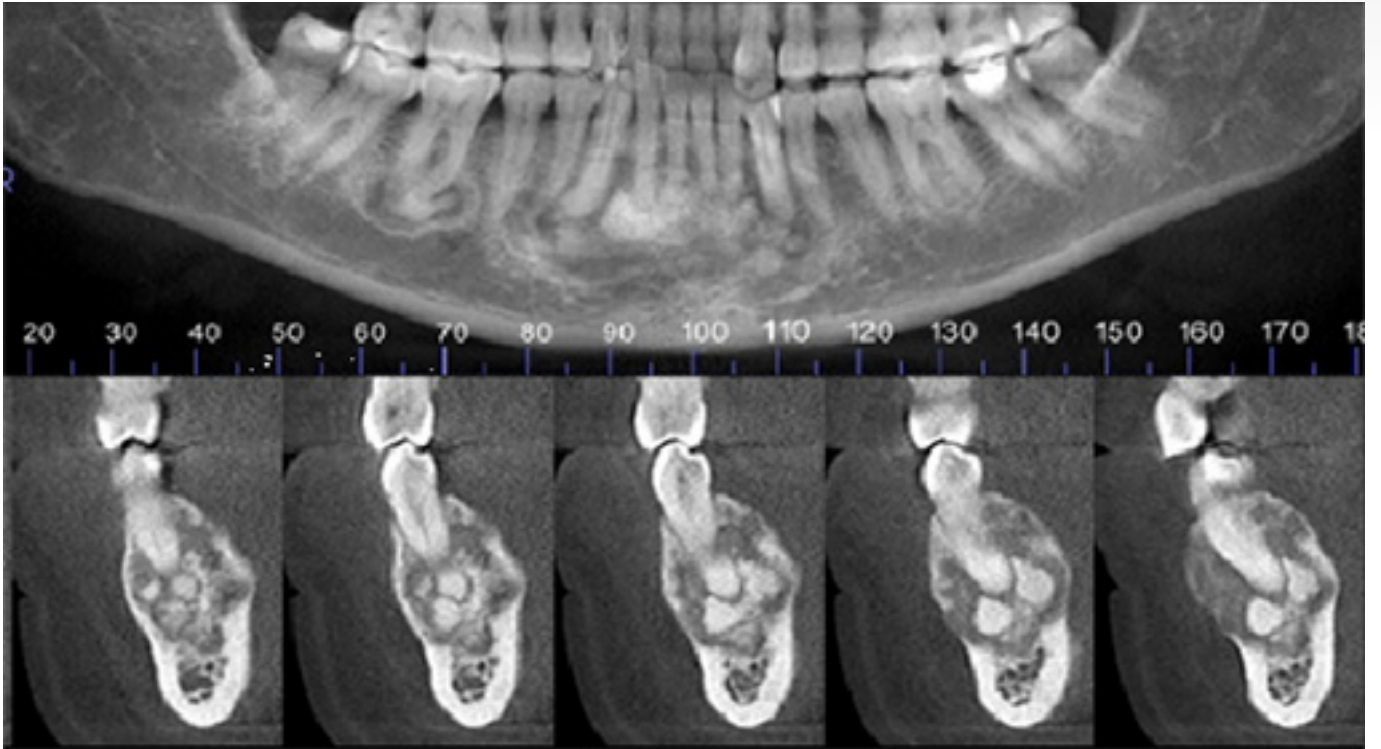


Figure 6. Homogeneous focal cemento-osseous dysplasia at the 45 periapical region. Cortical thinning and expansion adjacent to the lesion is noted (red arrow).

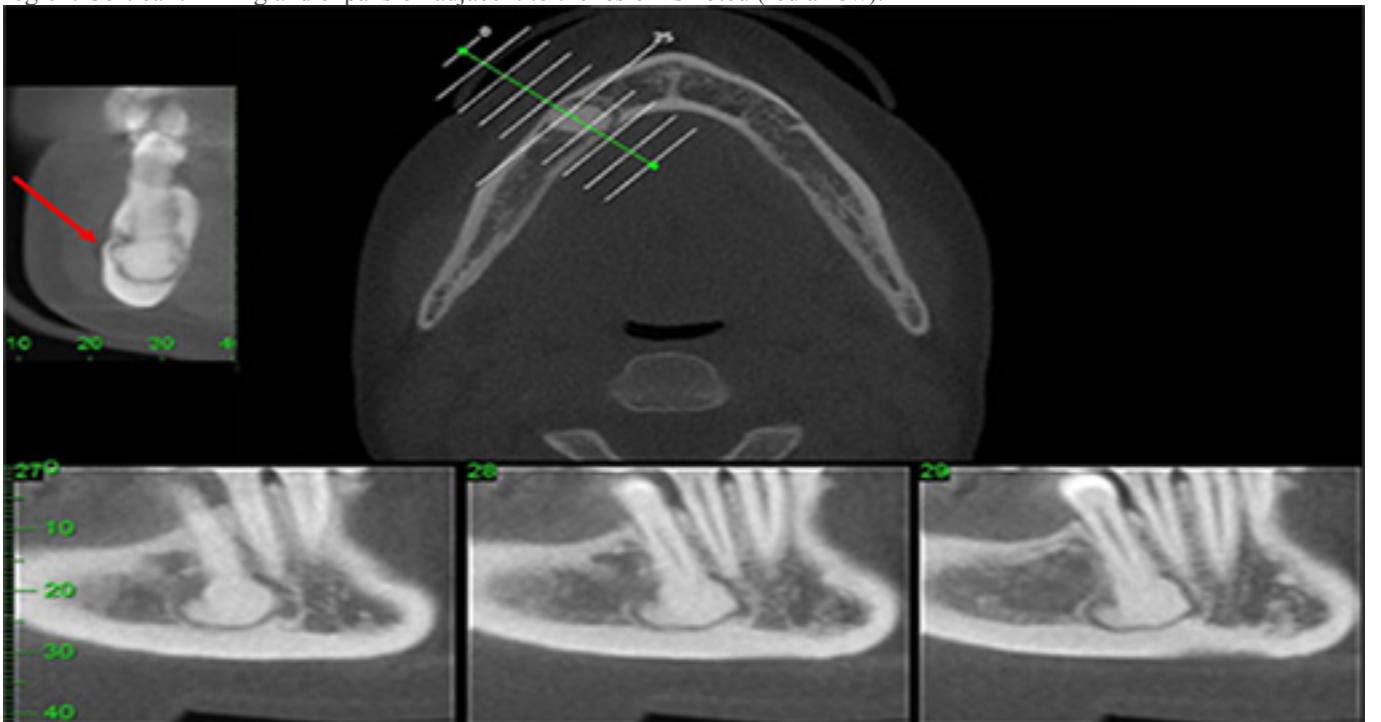
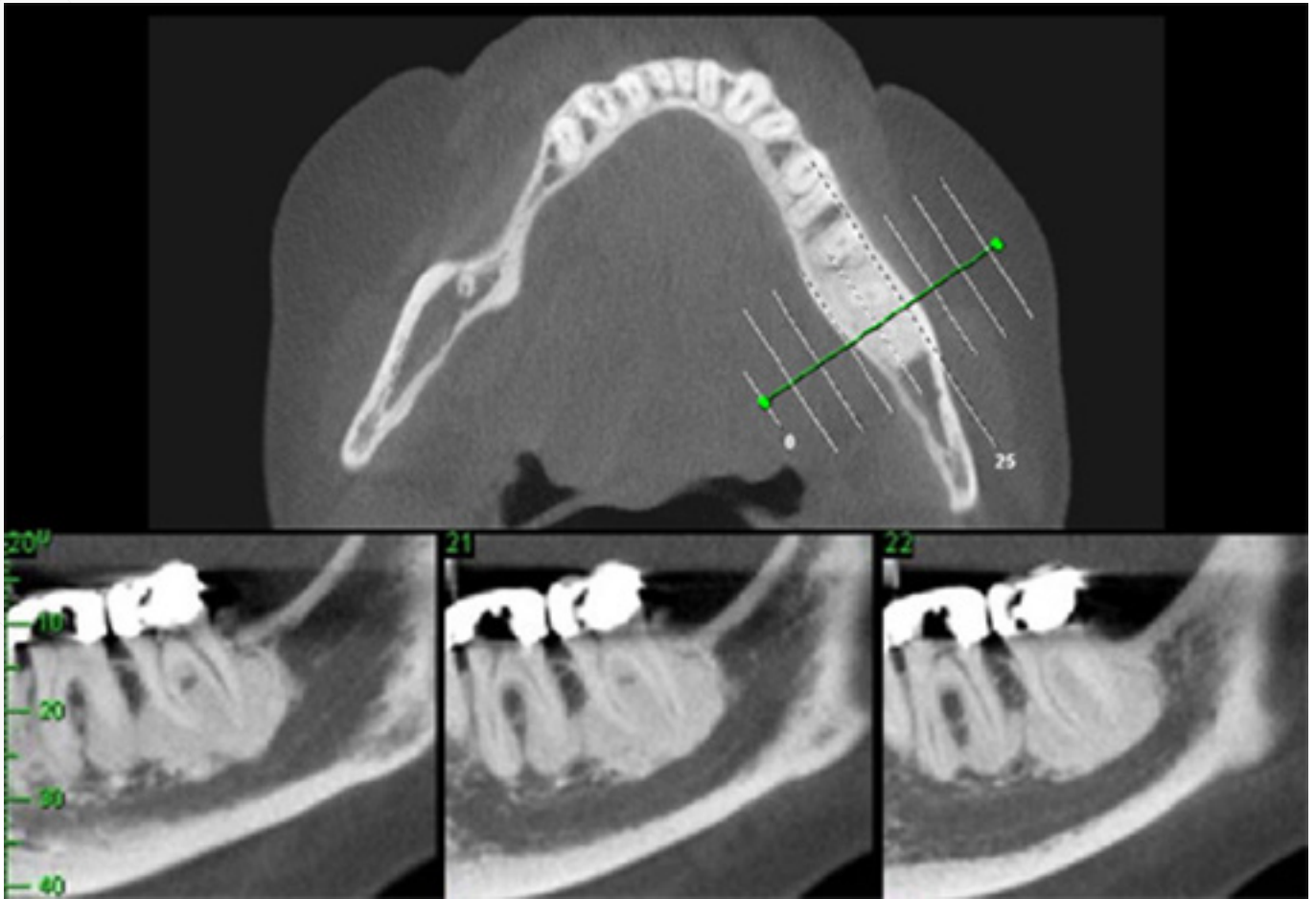


Figure 7. Late-stage cemento-osseous dysplasia at the 36 and 37 periapical regions. Diffuse homogenous increased radiodensity with ill-defined borders. There is fusion between the root apices and the lesion.



Discussion

Classification

In the literature, inconsistent nomenclature and descriptions of COD and its subtypes have led to much confusion. To avoid complexity, this paper uses the simplified classification proposed by Eversole et al in 2008: focal COD (FCOD) and florid COD (FL.COD) [3]. According to this classification, FCOD is described as discrete, localised lesions rarely exceeding 2 cm in size; and FL.COD is defined by the presence of multiple non-expansile COD lesions involving two or more quadrants in the jaws.

Based on this classification, if two small and discrete COD lesions were to be found over two quadrants of the jaws, these should be labelled as FL.COD. However, it would be more appropriate to describe these lesions as being multifocal FCOD, based on fundamental radiology. Furthermore, most FL.COD cases observed in this study presented with marked cortical expansion. To clearly differentiate FL.COD from multifocal FCOD, the authors propose a refined definition of FL.COD: multiple confluent COD lesions involving any quadrants of the jaws, with or without cortical expansion.

Radiographic Features

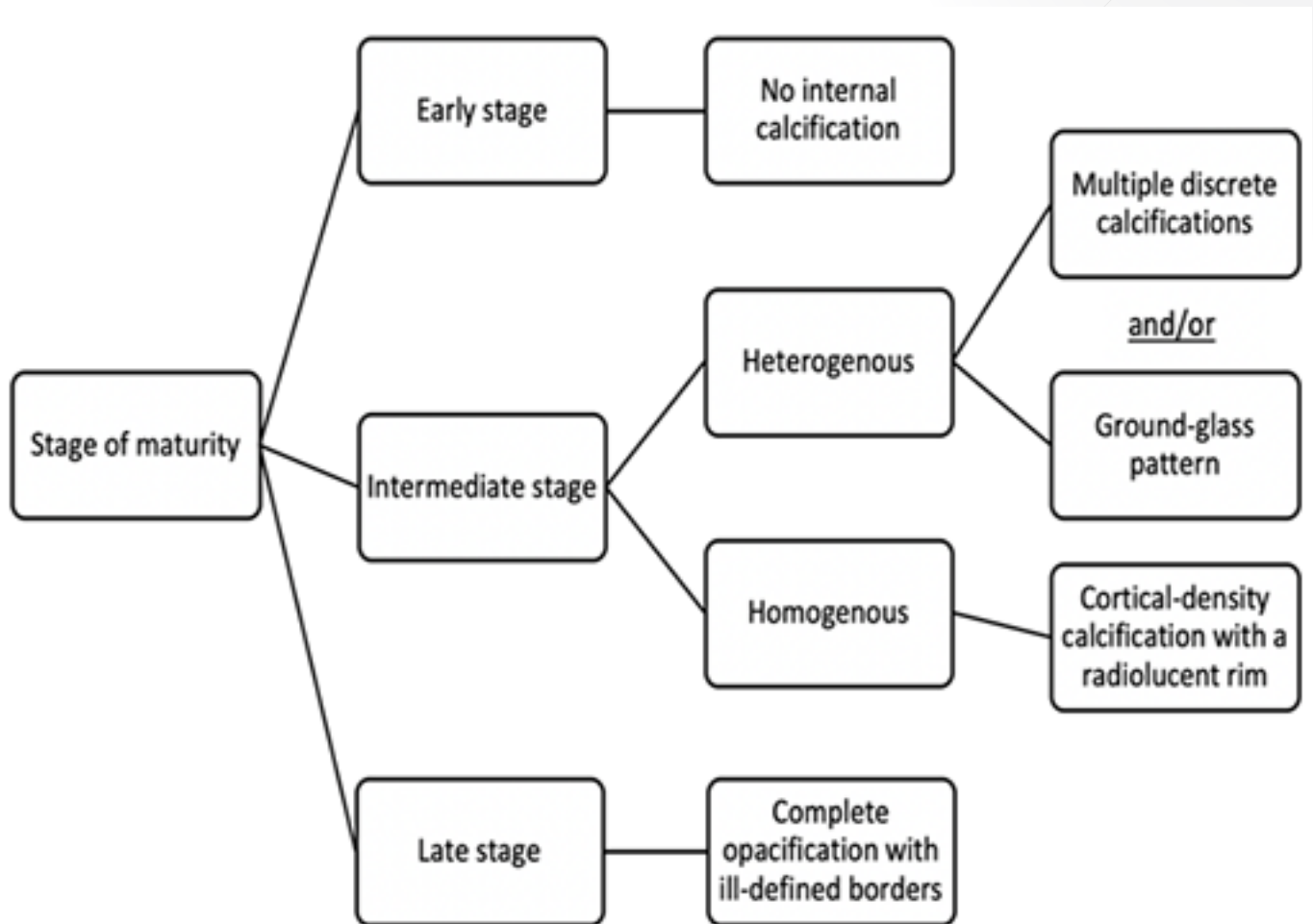
The posterior mandible was the most favoured site for FCOD, which is consistent with Eversole et al's classification. The current study includes cases of COD in edentulous regions, which has not been specified in previous literature. Given the retrospective nature of the study, it cannot be determined if these CODs developed before or after the loss of corresponding teeth. The authors propose further investigation into whether or not this requires its own classification.

In the 57 FCOD cases, the authors noted a difference in peripheral morphology; almost half of the cases demonstrated unilocular periphery and the remainder presented with multilocular periphery. The multilocular FCODs were generally larger than the unilocular FCODs. This finding infers that change in the size of FCOD is likely associated with alteration in peripheral morphology.

In this study, the intermediate stage of COD, defined as "mixed radiolucent-opaque pattern with a well-defined radiolucent rim around the radiopacity" by Eversole et al, was further divided into two subtypes based on the pattern of internal calcification: heterogenous and homogenous [3]. The heterogenous type of intermediate-stage COD had multiple discrete calcific bodies and/

or a ground-glass pattern of internal calcification as demonstrated in Figs 3-5. The homogenous type had a single smooth, dense (similar to cortical bone density) internal calcification surrounded by a well-defined radiolucent rim (Fig 6). The late stage (Fig 7) demonstrates a “diffuse radiopacity with ill-defined borders” as described by Eversole et al. A flowchart demonstrating different stages of COD maturity is shown in Figure 8.

Figure 8. Flowchart demonstrating different stages of cemento-osseous dysplasia according to the pattern of internal calcification.



Cortical thinning (88.7%) and cortical expansion (51.6%) were prominent features in all types of COD in this study. These features were not emphasised in previous studies using three-dimensional imaging such as CBCT. However, in the recent paper published by Cavalcanti et al, where CBCT imaging was used, of their 82 COD cases, 36.6% showed cortical thinning and 14.6% showed cortical expansion⁷. These figures are significantly lower than the results of the current study.

Diagnostic Dilemma for Dentists

The radiographic appearance of COD may appear similar to other lesions depending on location and maturity. The radiolucent appearance of early stage COD may mimic inflammatory apical pathology associated with non-vital teeth [8-11]. Furthermore, large early-stage unilocular FCOD demonstrating expansion and thinning of the adjacent cortices may resemble inflammatory periapical cysts, which generally require surgical intervention. Thorough clinical examination and history taking are critical to avoid misdiagnosis. Cemento-osseous dysplasia should always be considered as a differential diagnosis when periapical radiolucencies are associated with vital teeth.

Cemento-osseous dysplasia should also be differentiated from true neoplasms and ossifying fibroma [3]. Ossifying fibroma is larger in size than COD, is typically not associated with the roots of teeth and may cause significant expansion of the affected jaw.³ Florid COD can appear as an extensive diffuse lesion with mixed radiopacities; it should be differentiated from Paget's bone disease, chronic diffuse osteomyelitis and osteoma and metastatic diseases [12, 13]. It is important to note that secondary infection and osteomyelitis may develop concurrently in the setting of mature COD due to its poor vascularity.

Identification of COD may be further complicated by the presence of other pathology. A case report by Prodromidis et al identified a COD-like lesion co-existing with a compound odontoma [14]. It was unclear if the presence of the two lesions was a coincidental association, common development or if there were COD-like features within the odontoma. Gerlach et al presented a case of a patient that had radiographically definitive FL.COD, but then later developed a concomitant cemento-ossifying fibroma [15]. There has also been a reported case of osteosarcoma diagnosed in a patient with FL.COD, and this may represent coincidence or osteosarcoma arising from FL.COD [16]. As such, it would be prudent to monitor patients with FL.COD closely [16].

The diagnosis of COD can prove difficult and familiarity with the key radiographic features is important to establish a correct diagnosis. Alsufyani and Lam showed dento-maxillofacial radiologists were significantly superior in identifying cases of COD, compared with general dental practitioners [17]. The inability to diagnose COD may lead to unnecessary and invasive dental treatment being performed on patients. Biopsy is usually not indicated for diagnosis of COD and may predispose the area to infection due to decreased vascularity and abundant amorphous bone material; however, it may be required in atypical cases where a diagnosis cannot be clearly established [18].

Management

Cemento-osseous dysplasia is typically self-limiting and does not require intervention, and in most cases, careful monitoring with follow-up radiographs is sufficient [3, 6, 19-21]. Use of low-dose conventional radiographs such as orthopantomograms may be sufficient for review with minimal radiation exposure. Confirming the vitality of teeth associated with the lesion may assist in excluding the presence of inflammatory pathology and assist with diagnosis. In FL.COD, there is an increased risk of secondary infection, sequestrum formation and osteomyelitis due to poor vascularity of the lesions [20]. To minimise this risk, preventative strategies should be implemented to reduce the risk of caries and periodontal disease [21, 22]. In situations of severe facial deformity and symptoms affecting the quality of life, surgical intervention may be indicated [23-30].

Conclusion

Cortical thinning and cortical expansion are not commonly reported features of COD, and our study is the first to present that these features are prevalent despite the type of COD. It is therefore critical for all dental practitioners to recognise COD and to avoid confusion with other expansile lesions in the head and neck region, such as odontogenic tumours and cysts. As most cases of COD are incidental findings, comparison with previous radiographs and clinical correlation are essential.

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