A retrospective clinico-pathologic analysis of cemento-osseous dysplasia in a South African patient population

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Abstract

Background: Cemento-osseous dysplasia (COD) is a fibro-osseous jaw bone lesion. The affected bone in COD progressively becomes sclerotic, poorly vascularized and susceptible to secondary osteomyelitis.

Objective: To provide a clinico-pathologic appraisal of COD in a South African patient population.

Methods: Archived records of 133 patients diagnosed with COD were reviewed for patient demographics, COD location, COD type, osteomyelitis or simple bone cyst secondary to COD.

Results: The mean age was 53.4 ± 13.5 years with a 94.7% female predilection. COD mainly affected the mandible (57.1%), followed by involvement of both jaws (38.3%) and maxilla (4.5%). Florid COD was the most prevalent (69.9%), followed by focal COD (18%) and periapical COD (12%). Florid COD showed a clear trend of increasing with age, peaking in the sixth decade and decreasing thereafter. Osteomyelitis and simple bone cyst presented as complications of COD in 74.4% and 5.3% of cases respectively, while 21.8% of all cases of jaw osteomyelitis during the study period were secondary to COD.

Conclusion: A higher frequency of jaw osteomyelitis secondary to COD was found compared to previous studies. No significant association was shown between any of the COD types and secondary osteomyelitis.

Keywords: Cemento-osseous dysplasia, South Africa, patient population.

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Introduction

Cemento-osseous dysplasia (COD) is a non-neoplastic fibro-osseous lesion which typically occurs in the tooth-bearing regions of the jaw bones. The classification of COD has evolved significantly since its initial categorization as a "cementoma" along with benign cementoblastoma and cementifying fibroma[1]. In subsequent classifications COD was placed in the group of non-neoplastic bone lesions[2,3]. The 2017 World Health Organization (WHO) classification of COD includes a focal, periapical and florid form of the disease[4]. Familial gigantiform cementoma is a rare form of fibro-osseous lesion characterized by early onset of multi-quadrant, progressively expansile jaw lesions, with some cases demonstrating an autosomal dominant inheritance pattern while other cases are sporadic[4]. The term "expansive osseous dysplasia" has been proposed by some authors for sporadic cases of osseous dysplasia that manifest as a solitary lesion but with progressive jaw expansion[5,6]. The expansive type, however, does not yet have parameters well known in the literature and is still not considered a type of COD by the WHO[4].

COD manifests histopathologically as a benign fibro-osseous lesion comprising a mixture of woven bone and cementum-like particles in a connective tissue stroma[1-4]. The bony trabeculae appear as thick, curvilinear structures with a characteristic "ginger-root" pattern. The in-

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dividual trabeculae gradually merge to form a sclerotic mass of cemento-osseous tissue[4]. Radiographically, COD may range from completely radiolucent to mixed to radiopaque with a radiolucent rim[7]. Focal COD presents as a single lesion in any tooth bearing area of the jaw, whether edentulous or not[4]. Periapical COD appears as a single or multiple lesions in the anterior mandible in the periapical region of vital teeth[4]. Florid COD usually presents bilaterally and symmetrically in the mandible but may involve all four quadrants of the jaws[4]. Due to overlapping histopathologic features between COD and the other fibro-osseous lesions of the jaw, often an accurate diagnosis of COD on a biopsy specimen requires correlation with the clinical and radiographic features[11].

There are relatively few studies that exclusively comprise a series of COD cases[8-12]. Some earlier studies incorporated COD into the spectrum of lesions previously termed "cementomas"[13,14], while in other studies this jaw disease was studied in the context of other fibro-osseous lesions in particular with ossifying fibroma and fibrous dysplasia[15–17]. Although COD mainly affects people of African descent[4], not much has been reported about them as an entity from Africa. Further, African data on the prevalence of these lesions is equivocal. In a hospital-based study conducted in Nigeria over 22 years, only one (0.8%) case of florid COD was found among 121 benign fibro-osseous lesions of the jaws[18]. Muwazi and Kamulegeya[19], reported a 10.9% prevalence of COD in Uganda with an age range from 6 to 69 years, while two (3.3%) cases of COD out of 60 benign fibro-osseous lesions were documented during a 5-year study in Northern Nigeria[20]. In many of these studies COD typing was not performed. This study aimed to identify the relative frequencies of the COD types, according to the 2017 WHO classification of COD, in a South African population sample. We further sought to determine whether specific trends exist between the COD types and their respective clinico-pathologic features.

Materials and methods

Study sample

The study comprised a retrospective record review of archived documentation of COD. The histopathology records of patients diagnosed with COD over the period spanning 1996 to 2015 were extracted from the files of the Department of Oral Pathology, School of Oral Health Sciences, University of the Witwatersrand, Johannesburg. Only cases with adequate clinical and radiographic data that allowed for a confirmed clinico-pathologic diagnosis of COD were included. Ethical clearance was obtained from the Human Research Ethics Committee of the University of the Witwatersrand (Clearance certificate number: M170814).

Data collection and analysis

Information on the patient's age and gender, location of the COD lesion/s, the COD type and the presence of osteomyelitis, simple bone cysts or other related complication either at the time of patient presentation or at follow-up were recorded. The COD type was determined by the radiographic characteristics that were described in the histopathology report. The panoramic radiographs were reviewed in those cases where a hard copy of the radiograph was filed with the histopathology report. The frequencies of the various COD types and osteomyelitis secondary to COD were statistically analysed. A p-value < 0.05 was regarded as significant.

Results

Age, gender and location of COD

There were 23,288 submissions of tissue specimens during the study period out of which a final diagnosis of COD was documented in 133 (0.6%) cases. The mean age of these patients was 53.4 ± 13.5 years. There were 126 (94.7%) females, and their ages ranged between 9 and 87 years (mean ± standard deviation 53.4 ± 13.4 years). The 7 (5.3%) male patients had an age range of 30 years to 70 years (mean ± standard deviation 52.4 ± 16 years). The mandible was far more commonly affected than the maxilla with 76 (57.1%) of the COD cases presenting in the lower jaw while only 6 (4.5%) of the COD cases presented in the maxilla. In 51 (38.3%) cases both jaws were affected.

Prevalence of COD types, radiographic findings and complications

The COD cases were classified as focal, periapical or florid type. Florid COD was the most prevalent (93 cases, 69.9%), followed by focal COD (24 cases, 18%) and periapical COD (16 cases, 12%). A diagnosis of periapical COD (57.1 ± 16.6 years), florid COD (53.9 ± 12.1 years) and focal COD (48.2 ± 15.3 years) was usually made in the fifth and sixth decades of life. The number of COD

cases collectively increased with age to about the sixth decade of life and thereafter showed a progressive decline (Figure 1). Florid COD cases showed a clear trend of increasing with age, peaking in the 51-60 year age group and then decreasing. The other COD types, however, showed no clear trend of decreasing with age. A female predominance was shown for all three COD types. Of the 7 cases of COD in males, there were three cases of florid COD, three cases of focal COD and one case of periapical COD.

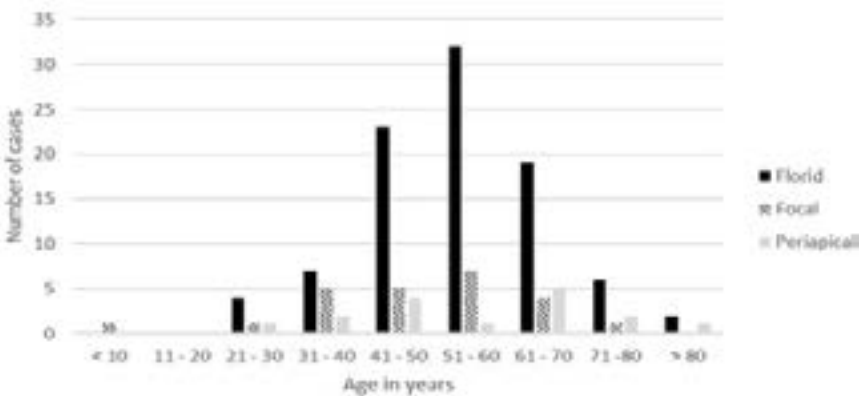

Figure 1. Distribution of cemento-osseous dysplasia types across the age groups.

Of the 76 (57.1%) cases that presented in the mandible slightly more than half the cases were of the florid type (40/76; 52.6%), followed by focal COD (20/76; 26.3%) and periapical COD (16/76; 21.1%). Florid COD affected both jaws in 51 (38.3%) cases, while the fewest number of COD cases were seen in the maxilla only (4.5%), which then mainly comprised focal COD (4/6 cases).

The panoramic radiographs were reviewed in 27 cases of florid COD, four cases of focal COD and three periapical COD cases. All three COD types typically presented as mixed radiodense, radiolucent lesions, affecting the tooth-bearing areas of the jaw and differing mainly in their extent of jaw involvement. Of the 133 COD cases, 99 (74.4%) cases showed osteomyelitis secondary to COD on biopsy, referred to as infected COD in this study. The COD types were statistically analysed to determine if any particular COD type was associated with a greater likelihood of secondary infection. Although most (74/99; 74.7%) cases of infected COD were of the florid type (Figure 2), florid COD was not significantly more likely to be associated with infection than the other COD types (Table 1). During the period of this study there were 355 cases with a histologically confirmed diagnosis of osteomyelitis of the jaw without associated COD, thereby indicating that 21.8% (99/454) of all cases of osteomyelitis of the jaw in this study were secondary to COD. Seven (5.3%) cases of COD were associated with simple bone cysts (Figure 3), which comprised four cases of florid COD and three cases of periapical COD.

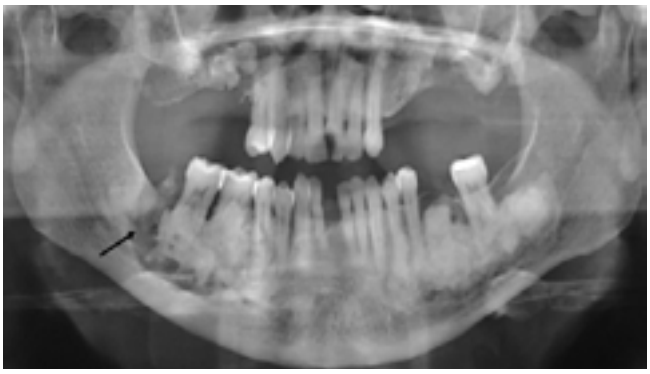

Figure 2. Florid cemento-osseous dysplasia showing multiple radiopaque lesions in the tooth-bearing areas of the mandible and maxilla. The area of bone destruction (arrow) represents osteomyelitis showing sequestration.

Table 1. Comparison between the cemento-osseous dysplasia (COD) types and secondary osteomyelitis (infected COD)

	Florid COD	Focal COD	Periapical	Fisher's exact test (p value)
Infected	74	15	10	
Non-infected	14	9	3	0.07

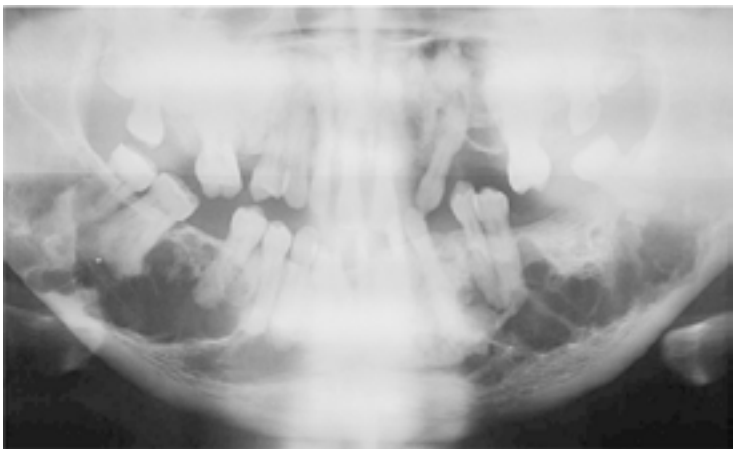

Figure 3. Simple bone cysts presenting as a complication of cemento-osseous dysplasia and manifesting as multilocular radiolucencies with scalloped margins bilaterally in the mandible.

Patient follow-up

Multiple surgical specimens were received from 10 patients who re-presented for treatment during the study period. The average period from the initial patient presentation to when they re-reported was 24.2 months (range=1-120 months). Of these, six patients required additional surgical debridement of COD lesions that were already infected at their initial presentation. Three of the six patients with recurrent sepsis underwent three surgical debridement procedures while one patient underwent a total of eight surgical debridement procedures during the follow-up period. In one of the patients who developed simple bone cysts, this manifested as a complication 3-years and 9 months after a diagnosis of florid COD was rendered.

Discussion

COD represented <1% of the total number of histologically diagnosed cases over a 20-year period in the Department Oral Pathology at the University of the Witwatersrand, constituting 133 cases of COD. Owosho et al.[21] reported 35 cases of COD over a six-year period in western Pennsylvania patients based on histopathologic reports, clinical records and radiographs. African studies on the prevalence of COD are infrequent. Butt et al.[22] noted 18 cases of COD out of 180 bone related jaw lesions in Kenya over 19 years. In a study conducted over a 5-year period in Uganda, the authors reported 17 cases of COD out of 155 fibro-osseous lesions[19], while a study in Northern Nigeria reported only two cases of COD out of 60 fibro-osseous lesions of the jaws over a 5-year period[20]. In the South African population, Thompson and Altini[14] reported 28 cases of florid COD over a 20-year period under the term "gigantiform cementoma" while Ackermann and Altini[13] described 127 cases of COD in their clinico-pathologic appraisal of "the cementomas".

In the present study the mean age at diagnosis of COD was 53.4 years, which is comparable with most other studies[21,23]. The majority of the COD subjects in this study were females (94.7%) with a peak incidence in the sixth decade, a finding that corresponds with most published work on COD[7,9]. The reason for the distinctive female predilection in COD is not known, but may allude to a complex interplay between genetic, hormonal and environmental factors in the development of this dis-

ease[7]. Males were affected in only 7 cases (5.3%). Analysis of the literature reveals fewer than 70 cases of COD reported in males[7,8,10,11,13,16,20-22]. The mean age reported in the literature at diagnosis of COD in males is 50.5 years[7,13,21,23], which is similar to the present study.

The most frequent COD type in the present study was florid COD (69.9%), demonstrating a higher frequency compared to studies from Korea, Brazil and western Pennsylvania where florid COD constituted 48.5%, 45.4% and 48.6% of their COD cases respectively[9,15,21]. This also contrasts with the rates in series involving Oriental, Ugandan, Nigerian and Thai populations, where florid COD accounted for 11%[24], 10.3%[19], 0.8%[18] and 0.8%[16] of all COD cases respectively. Osteomyelitis is a complication of COD that may have contributed to the increased number of COD diagnosis in the present study, since biopsies are mandatory in infected cases. The mean age of patients with florid COD in this study was 53.9 years, which is similar to the findings in a Jamaican (52.5 years)[25] and a Ugandan (50.3 years)[19] study.

Osteomyelitis and simple bone cyst are well known complications reported in COD.[4] In the current study 74.4% of the COD cases were symptomatic at diagnosis and histologic examination of the surgically debrided tissue confirmed osteomyelitis secondary to COD. The frequency of osteomyelitis presenting as a complication of COD varies in different studies. Kawai et al.[23], Alsufyani and Lam[7], Melrose et al.[12], Owosho et al.[21] and Netto et al.[15] reported significantly lower prevalence rates of osteomyelitis secondary to COD in the order of 14.8%, 11.3%, 5.9%, 5.7% and 4% respectively. Since the selection of cases in this study were based on histologic diagnoses there is an inherent bias that includes cases with pre-existing infection warranting surgical debridement and biopsy as part of patient management. Interestingly, however, those studies that report low prevalence rates of osteomyelitis secondary to COD are from non-African countries[7,12,15,21,23], while an African study reported a substantially higher frequency (78.7%) of osteomyelitis secondary to COD[8]. The former study data included all symptomatic cases of florid COD as well as asymptomatic cases detected during routine radiographic examination[8]. In another African study where, similar to this study, data was derived from a histopathology register in Uganda, the authors reported infected COD in 47% of cases in their COD study population[19]. Since the onset of symptoms is associated with oral exposure of the sclerotic bone,[7,23] early radiographic detection of this disease is prudent so that patients are encouraged to retain their teeth through reinforcement of good oral hygiene practice to minimize the risk of pulpal and periodontal infection. The higher percentage of symptomatic cases in studies from Africa is likely to be related to limited preventive and conservative dental services in these countries as well as limited use of routine panoramic radiography in public dental clinics[8]. An increased level of awareness for diagnosing asymptomatic cases of COD among dentists and other oral health care workers may contribute to a decrease in the frequency of osteomyelitis secondary to COD in these patients.

Limitation of this study

In many cases of periapical and florid COD, their distinctive clinico-radiographic patterns allow for a strong presumptive clinical diagnosis and a biopsy is avoided to minimize the risk of onset of symptomatic COD. Hence the exact incidence of COD in the South African population cannot be assessed from this study.

Role of each author

Mouna M Benaessa: Conception, data collection, data analysis and interpretation, drafting, critical review, final approval for submission.

Farzana Mahomed: Conception, data analysis and interpretation, drafting, critical review, final approval for submission.

Sizakele P Ngwenya: Conception, drafting, critical review, final approval for submission.

Declaration of conflict of interest

None.

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