Cemento-osseous Dysplasia in Jamaica
Review of Six Cases
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ABSTRACT
Six cases of cemento-osseous dysplasia (COD) of the jaw bone in Jamaicans are reviewed. Five were documented over a 15-year period (1980 – 1995). These include a case of florid cemento-osseous dysplasia (previously called gigantiform cementoma). Three of the initial cases were histologically diagnosed as gigantiform cementoma. There was no indication in the patient’s case file whether these were familial or non-familial. The other two cases were diagnosed histologically as periapical cemento-osseous dysplasia and cementoblastoma respectively. Based on the current understanding of the nature of florid-cemento-osseous dysplasia (FLCOD), a new case was diagnosed as such solely on radiological findings. This single case of FLCOD is reported and discussed against the background of other cemento-osseous lesions. Special emphasis is placed on the radiology of COD in this paper. The confirmative role of radiology without the need for histophathology and treatment for asymptomatic FLCOD is emphasized.

INTRODUCTION
The gigantiform cementoma (GC) is a very rare condition which is classified by the World Health Organization as a distinct histopathological entity. According to Agazzi and Belloni (1), the lesions have their onset at a young age, develop slowly and usually involve all four quadrants of the jaws. The lesion occurs in families and appears to be inherited as an autosomal dominant characteristic, although other reported cases are non-familial and do not support this claim. The benign cementoblastoma is a common lesion (2-6). Langdon (7) pointed out the exceptionally rapid growth and aggressive behaviour of cementoblastoma.

The periapical cemental dysplasia is classically described as a lesion of rather common occurrences; its nature is not fully understood (8). Some authors adhere strongly to a theory of it originating from odontogenic tissue (cementum) while others believe that it represents only an unusual reaction of the periapical bone. It is not considered a neoplasm (8). Florid cemento-osseous dysplasia (FLCOD) was initially reported as florid osseous dysplasia by Melrose et al (9) who described a condition that has come to be accepted as the most clinically extensive form of cemento-osseous dysplasia (COD), thus the use of the term florid. The diagnosis of FLCOD is a clinical and radiographic one, and biopsy is not necessary. A patient must manifest the typical changes in at least two quadrants for a clinical and radiologic diagnosis of FLCOD to be made. A four quadrant disease may be suggestive of a familial nature.

PATIENTS AND METHODS
All cases histologically diagnosed as COD in Jamaica over a 15-year period (1980 – 1995) were reviewed in terms of clinical and radiographic findings. An additional new case of FLCOD diagnosed in 2002 is also documented in this paper in the form of a case report.

RESULTS
A total of six cases are documented in this study from Jamaica. Five cases of COD were seen over a 15-year period (1980 – 1995); three cases of gigantiform cementoma (total of five lesions) and one case each of cementoblastoma and periapical cemental dysplasia. These cases are summarized in the Table together with a new case of FLCOD seen in 2002.

All were females with an age range of 23 – 70 years and an average age of 52.2 years. All cases were symptomatic prior to discovery, the first five cases presenting as jaw swelling and the sixth presented as persistent pain from the right mandible without any obvious dental cause.

Case Report
A 23-year-old Jamaican female of African descent presented to the Cornwall Dental Centre Montego Bay, Jamaica, with severe pain of the right mandible in the molar region. Localization of the pain to one of the molars was not possible. Clinical and initial radiographic examination with a periapical radiograph did not show any dental cause for the pain.

Dental Radiologic Findings
A dental panoramic view revealed radiopaque and radiolucent lesions of the mandible bilaterally with radiographic evidence of root resorption of teeth # 37 and #
47 (Fig.1). The radiograph also showed a well-circumscribed radiopaque mass of the maxillary antra. Computed tomography confirmed the presence of these large diffuse sclerotic mandibular masses and revealed involvement of the incisor region (Fig. 2). The bilateral sinus lesions were interpreted as osteomata; however, their appearance and close proximity to the maxillary molar and premolar apices suggest that these lesions were also extrinsic in origin and part of the same cemento-osseous condition (Fig. 3).

A diagnosis of FLOD was made based on the above clinical and radiographic findings.

**DISCUSSION**

The scanty literature on gigantiform cementomas has been reviewed by Punniamoorthy (9) who noted that the origin of the lesion is still a mystery. Kramer (10) and his colleagues...
in the WHO odontogenic group have suggested that
gigantiform cementoma is a form of dysplasia or even
harmartomatous in nature. Punniamoorthy (9) also pointed
out that numerous cases of lesions of the jaws have been
reported in the literature which are very similar to
gigantiform cementoma in terms of clinical, radiographic
and histologic features but yet have been described under
different terms such as chronic sclerosing osteomyelitis,
sclerosing/sclerotic cemental mass, chronic productive
ostitis, osseous dysplasia and multiple exostosis. Similar
masses have also been noted to occur in some cases of
ostitis deformans or pagets disease of bone.

It is significant to note the difference in the
characteristic radiographic appearance of both the
cementoblastoma and the gigantiform cementomas. The
cementoblastomas do have a rim of radiolucency around
them (Fig. 4). Radiologically, however, the three cases of
gigantiform cementoma in the series of jaw bone tumours in
Jamaica closely resembles that which is detailed of the
cementoblastomas (11, 12) as all three cases do have a
radiolucent rim around the periphery of the radiographic
mass. As such, it would seem that both cementoblastomas
and gigantiform cementomas constitute an important
differential diagnosis for each other at radiologic level.

From the only case documented so far in Jamaica of
periapical cemental dysplasia, it is very difficult to state
conclusively that the clinical and radiological behaviour of
this lesion differs from what has been previously
documented. It is of interest that only one case of periapical
cemental dysplasia was found, despite its prevalence
amongst the West Indian population in the United Kingdom.
It is very likely that this condition is presently under-reported
in Jamaica because it is symptomless and is only a chance
finding on routine radiography which is not a common
practice in Jamaica. Hence, the real incidence of periapical
cemental dysplasia in Jamaica is more than that recorded in
this series of COD.

In this review, we report a multi-quadrant fibro-osseous
lesion which has been designated gigantiform cementomas or
familial multiple cementomas in the first edition of the WHO
histological typing of odontogenic tumours, jaw cysts and
allied lesion. In retrospect, Case 1 in this study may actually
be a case of focal cemento-osseous dysplasia. Summerlin
and Tomich (13) pointed out the unique features of this
condition that place it in the spectrum of COD. They
presented data on 175 examples of what they termed focal
cemento-osseous dysplasia (FCOD) for the first time. They
also contrasted these cases with 45 cases of cemento-
ossifying fibroma, a benign neoplasm. They pointed out that
FCOD was probably more common than is appreciated and
that it is likely to be misdiagnosed as cemento-ossifying
fibroma. Focal cemento-osseous dysplasia lesions are
solitary and patients are asymptomatic in most instances;
there is no cortical expansion. Almost all cases are
discovered on routine radiography. Radiographically, a
solitary lesion of FCOD may present as a radiopacity with a
narrow rim of decreased radiodensity in the mandible. It is
for this reason that we can diagnose case 1 as FCOD
retrospectively. Melrose et al (14) described a condition that
has come to be accepted as the most clinically extensive form
of COD, hence the use of the term florid. Prior to the
publication of their articles, cases of FLCOD had been
published under at least 12 different names, such as
gigantiform cementoma, chronic sclerosing osteomyelitis,
sclerotic cemental masses and multiple exostosis. This
terminologic jungle has fortunately come to an end. FLCOD
is more common in middle-aged black women. In their
series of 34 patients, Melrose et al (14) reported on 33
females and a male. It is now established that the condition
occurs in all ethnic groups (14-16).

Clinically FLCOD may present in patients as cortical
expansion, particularly of the mandible. The expansion may
be pronounced enough to cause the practitioner to suspect a
neoplasm of pagets diseases of the bone. Infection may be
absent, but dull aching sensation of intermittent nature may
be the presenting feature in the mandibular molar region.
The teeth are vital and there is no evidence of other pathology
that may be responsible for the symptoms and the disease is
discovered by examination of routine radiographs.

Florid-cemento-osseous dysplasia was a term proposed
in the 2nd edition (10) of the World Health Organization
(WHO) “international histological classification of
odontogenic tumours” to replace the 1st edition gigantiform
cementoma (17). FLCOD lesions are lobulated masses of
dense, lightly mineralized almost acellular cemento-osseous
tissue typically occurring in several parts of the jaw (10). It
is important to note that whilst the 2nd edition WHO
classification maintained the definition of FLCOD, it
modified the definition of periapical cemental dysplasia (17),
another cemento-osseous dysplasia which mostly affected
the mandibular incisor region. Unfortunately, this
modification confused the boundary between FLCOD and
PCD, if they are actually two distinct pathological entity (we
have been very careful not to say distinct histopathological
entity). The main distinction between FLCOD and PCD is
that for PCD each periapical lesion is self-limiting and rarely
exceeds 1cm in diameter. The two lesions are similar
histologically and also similar histologically to cemento-
ossifying fibroma and fibrous dysplasia. Waldron
commented that the majority of cases called chronic
sclerosing osteomyelitis were actually FLCOD (18).

Radiographically a wide spectrum is seen (16). The
reported case presented as a four-quadrant lesion on radi-
ographs. Radiographically FLCOD usually presents as diffuse
distribution of lobular irregularly shaped radio-pacities
throughout the alveolar process. They may also have a
ground glass appearance.

These variable features of FLCOD mitigate against the
diagnosis of fibrous dysplasia, cemento-ossifying fibroma
and to a lesser extent, pagets disease of the bone.
Concomitant simple bone cysts are occasional features of
FLCOD. In a patient without simple bone cyst formation, the
diagnosis of FLCOD is a clinical or radiographic one. Biopsy is not necessary. A patient must manifest the typical changes in at least two quadrants for a clinical diagnosis of FLCOD to be made. Evaluation of serum alkaline phosphatase level and skeletal survey would suffice to rule out any suspicion of pagets disease of the bone.

FLCOD may have a familial nature, hence familial FLCOD. In 1953, Agazzi and Belloni (1) described an Italian family in which several members manifested four-quadrant disease that had begun at an early age and resulted in facial disfiguration. The management of FLCOD consists of clinical and radiographic observation for the life of the patient as well as excellent periodontal and restorative care to the dentition. This was the management of choice for this reported case.

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REFERENCES