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.

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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 #

Table: Summary of cases of cemento-osseous dysplasia in Jamaica

Cases	Gender	Age (years)	Site	Radiographic findings	Histological findings
1	F	56	Posterior mandible	Large area of radiopacity with surrounding radiolucency	Gigantiform cemento
2	F	49	Bilateral mandible in 876|678 region	Bilateral multiple radiopaque masses with surrounding zone of radiolucency 876|678 region.	Gigantiform cemento
3	F	47	Bilateral 54|678 region	Bilateral large radiopaque mass with surrounding rim of radiolucency in the 54|45 region	Gigantiform cemento
4	F	68	Right mandible	Mixed radiopaque and radiolucent area surrounding the periapical region of 21|123	Periapical cemental dysplasia
5	F	70	Posterior mandible	Radiopacity surrounded by a rim of radiolucency	Cementoblastoma
6	F		All four quadrants of jaw	See case report	No histology done

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).

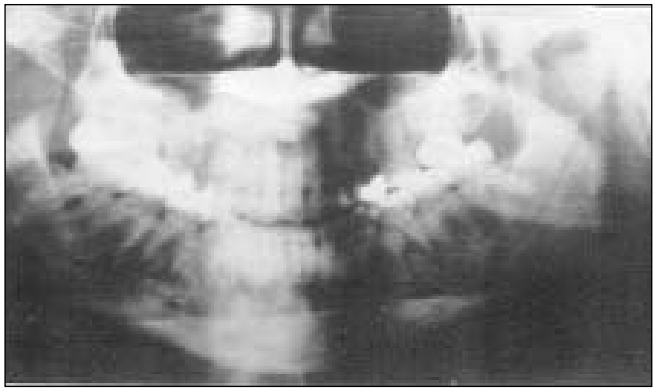

Fig. 1: Dental panoramic tomogram of a patient with FLCOD. Note the bilateral mixed lesion of the mandible and the increased radiopacification of the posterior maxilla

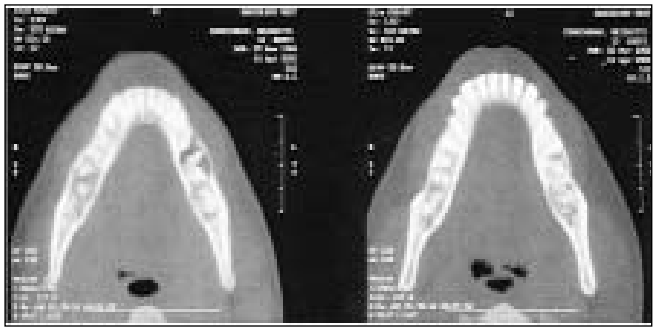

Fig. 2: CT-scan of the mandible confirming the extent of the mixed lesions of the mandible

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

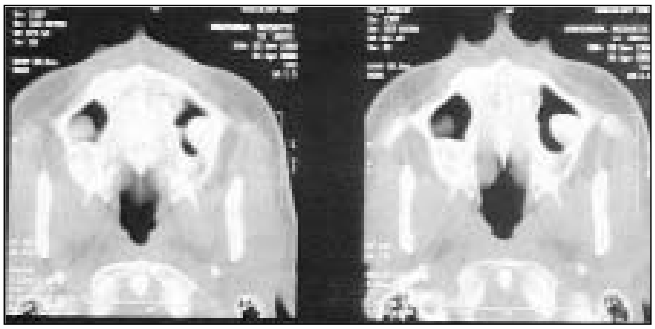

Fig. 3: CT-scan of the maxilla showing the radiopaque mass attached to the walls of the maxillary sinus bilaterally

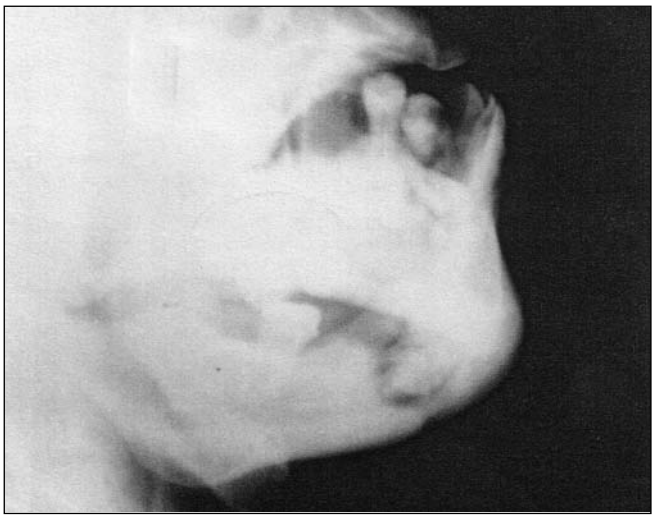

Fig. 4: Oblique lateral view of the mandible showing a radiopaque mass with a rim of radiolucency around it. The histological diagnosis was cementoblastoma

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 cememtoma 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 osteitis, osseous dysplasia and multiple exostosis. Similar masses have also been noted to occur in some cases of osteitis 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 enostosis. 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 radiographs. 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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