

Case Reports

FLORID CEMENTO-OSSEOUS DYSPLASIA: A CASE REPORT STRESSING THE IMPORTANCE OF RADIOGRAPHIC INTERPRETATION AND DISCUSSION

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مقدمة :-

لا يزال الاختلاف في الآراء حول الأصل النسيجي والطبيعة البيولوجية لسوء التكوين العظمي الملاطي المعمم قائماً؛ حيث أن البعض يعتبر العملية سوء تكوين عظمي، والبعض يعتبرها سوء تكوين ملاطي، والبعض الآخر يعتبرها مزيجاً من الاثنين. هناك أيضاً من يعتبرها حالة التهابية. هذه الاختلافات في الآراء أسفرت عن عدة تسميات تصف هذه الآفة الفككية. لقد وصفت هذه الآفة بأنها في معظم الأحيان تصيب إنثاءً من أصول أفريقية في منتصف العمر. لكن هنالك عدة تقارير عن حالات حدثت في ذكور، وفي سلالات عرقية أخرى، وحالة في طفلة بالإضافة إلى حالات متعددة في بعض العائلات في هذا المقال. نصف حالة امرأة من أصل أمريكي لاتيني.

الحالة السريرية :-

مريضتنا امرأة مكسيكية تبلغ من العمر ٣٣ سنة، حوت لقلع بعض الاسنان بسبب التهاب الرباط حول السني. لدى الفحص الشعاعي، تم اكتشاف مناطق شفافية شعاعية مختلطة بمناطق ظلالية شعاعية في منطقة الناب والرباعية العلوية اليمنى (شكل ١)، وفي المنطقة الامامية من الفك السفلي (شكل ٢)، وحول ذروي جذري الثنيتين العلويتين. كانت الأسنان المعنية حية لدى الفحص. تم قلع بعض الأسنان وأخذت خزعة من المنطقة الفككية الأمامية السفلية. شخصت العينة في قسم علم أمراض وأشعة وطب الفم في جامعة أيوا على أنها ورم ليفي متعظم مركزي (شكل ٣) لاحقاً تم أخذ خزعة من نفس المنطقة ومن المنطقة الأخرى في الفك العلوي (شكل ٤).

وقد شخصت على أنها ورم ليفي متعظم مركزي أو سوء تكوين ملاطي حول ذروي. أيضاً تمت الإشارة الى منطقة إضافية مشابهة ولكن أصغر حجماً في الاتجاه الوحشي للرحى الثانية السفلية اليمنى، والى عدة مناطق عند ذروة جذور الرحي الثانية السفلية اليسرى والثانية العلوية اليمنى، تقرير الأشعة شخص الحالة على أنها اما سوء تكوين ملاطي حول ذروي أو سوء تكوين عظمي - ملاطي معمم وتقرير الأشعة والفحص المجهرى معا يبينان لنا أن الحالة تمثل سوء تكوين عظمي - ملاطي معمم. تمت متابعة المريضة ثلاثة وخمسة أشهر بعد قلع بعض الأسنان وأخذت الخزعات المذكورة، وكانت بحالة جيدة.

في عام ١٩٧١م في كتاب منظمة الصحة العالمية (مرجع ٩) استعمل بنديورغ ورفاقه الاسم الملاطي العملاق لوصف الحالة، بالإضافة لمرادف آخر هو الأورام الملاطية المتعددة العائلية. في عام ١٩٧٥م وصف والدرون ورفاقه العملية باسم الكتل الملاطية الفككية المتصلبة. واعتبروها جزءاً من طيف الآفات الليفية العظمية من أصل رباطي حول سني، واعتبروا أن المادة المتمعدنة ملاط. في عام ١٩٧٦م تقدم ملروز ورفاقه بالاسم سوء التكوين العظمي

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المعمم، واعتبروا العملية جزءاً من طيف سوء التكوين العظمي الذي يشمل آفات أخرى. معظم الحالات المدروسة سابقاً كانت في إناث من أصل إفريقي أسود، ومعدل اعمارهن هي ٤٢ سنة. تصف هذه الحالة المرضية تغيرات في عظام الفكين، تظهر شعاعياً على شكل بؤر شفافية شعاعية مترافقة بظلاية شعاعية، وقد تصاحبها أولاً تصاحبها أعراض سريرية، من ضمنها تمدد في الصفائح العظمية للفك، أو التهاب مزمن في العظم والنقي العظم، مع تكون أقنية قيحية. وقد لوحظ أن معظم الحالات التي تصاحبها هذه الأعراض تحدث بعد قلع الأسنان أو استعمال الأطقم السنوية المتحركة، ويعتقد أن السبب هو عدوى ناتجة عن مثل هذه العمليات، لا يستطيع النسيج المتصلب ذو التروية الدموية القليلة، والقدرة الضعيفة على ان يقاومها بجهرياً، هذه الآفة عبارة عن نسيج ليفي يحتوي على كتل من نسيج متمعدن، وهو الذي يدور النقاش حول كونه عظماً أو ملاطاً، أو خليطاً من الاثنين.

هنا لا نستطيع إلا أن نؤكد أهمية الدراسة لصور الأشعة، بما أن التشخيص المجهرى لا يعتمد عليه كلياً في هذه الحالة، حيث ١، الصورة النسيجية تشابه حالات أخرى من الآفات الليفية العظمية. ورغم أن والدرون ورفاقه يرون ان الملاط والعظم يمكن التفريق بينهما عن طريق الاستقطاب الضوئي المجهرى، وان النسيج في هذه الحالة هو ملاط، فإننا نرى ان نعتبره أحد النسيجين أو خليطاً منهما، بما أننا لا نعتمد فقط على التشخيص المجهرى، وإنما نرى أن الأشعة مهمة لعملية التشخيص النهائي، وهو ما يهمننا أن نتوصل اليه، لأن هذا الوضع يتطلب ان يكون العلاج تحفظياً قدر الامكان، مع محاولة تقادي القلع والاستعاضة السنوية المتحركة، بسبب المضاعفات التي ذكرناها آنفاً. وينصح معظم الأطباء بذلك ويمتابة المريض عبر زيارته المتعددة، دوننا حاجة إلى الافراط في العلاج جراحياً.

Florid cemento-osseous dysplasia is a controversial fibro-osseous lesion of the jaws. Literature regarding the subject has been briefly reviewed illustrating the wide range of opinions as to the tissue of origin, biological nature and nomenclature. This is the first reported case in a Hispanic individual. The report is presented to review the histopathologic similarity of this lesion to other fibro-osseous lesions, the importance of radiological interpretation in terms of differential diagnosis and its significance in avoiding overtreatment and the complication of osteomyelitis.

Introduction

Controversy concerning the tissue of origin and the biological nature of florid cemento-osseous dysplasia still exists. Opinions range from it being a dysplastic osseous lesion, illustrated by the names florid osseous dysplasia¹, multiple osteomas², multiple enostosis³, Paget's disease of the mandible⁴, and multiple periapical osteofibromatosis⁵ to a dysplastic cemental lesion, illustrated by the names sclerotic cemental masses⁶, multiple cementomas⁷, gigantiform cementoma^{3,9}, monstrous cementoma⁸, familial multiple cementomas⁹, periapical cementoblastoma¹⁰, periapical cemental dysplasia with multiple lesions¹¹, and sclerosing cementomas¹²; through a combined dysplastic cemento-osseous lesion, illustrated by the name multiple cemento-ossifying fibroma¹³; to it being an inflammatory lesion, as seen in the names sclerosing osteitis¹⁴, sclerosing osteomyelitis¹⁵, chronic sclerosing osteomyelitis, and chronic diffuse sclerosing osteomyelitis¹⁶. However, it is believed that these latter names have been errone-

ously used to describe florid cemento-osseous dysplasia, or florid cemento-osseous dysplasia with superimposed inflammation.^{17,18}

The entity designated as florid cemento-osseous dysplasia refers to changes in the jaws characterized radiographically by dense radiopaque foci, mixed with radiolucencies^{1,6,18,20} which, depending on the clinical condition, may or may not be symptomatic. There may be displacement of the cortical bony plates as well as inflammation due to infection.¹ Familial forms have been described.^{2,12,24} Although the vast majority of cases are stated to occur in middle-aged black females, several cases have been reported in other ethnic groups,^{1,18,19,21,26} and a few in males^{23,24}. A case in a Hispanic female is being reported.

Case Report

A 33-year-old Mexican female was referred to the fourth author for extraction of multiple posterior teeth with severe periodontitis. Her medical history was not significant except for intake of birth control "pills". Clinical examination revealed severe periodontitis with mobility of the maxillary

right first molar, right lateral incisor, and left first molar, and mandibular left second and first molars, left second premolar, right second premolar and right second molar.

Radiographically, radiolucent-radiopaque areas were noted in the maxillary right canine-lateral incisor region [Fig. 1], and in the mandibular anterior region from the left canine to the lateral incisor [Fig. 2]. A radiopaque mass was noted at the apices of the maxillary central incisors. The pulps of all involved teeth tested were vital. During subsequent appointments, extraction of the mobile teeth mentioned, except the maxillary right lateral incisor, and incisional biopsy of the mandibular lesion were carried out. Extraction of mandibular incisors, maxillary right lateral incisor, and left second molar, with removal of the mandibular and maxillary lesions, and endodontic treatment of the maxillary right canine and mandibular left canine were carried out later.

The first biopsy specimen of the mandibular lesion [Fig. 3] was diagnosed in the Department of Oral Pathology, Radiology & Medicine, The University of Iowa, as central ossifying fibroma. The rebiopsy and the maxillary biopsy [Fig. 4], done more than a month later, were diagnosed as central ossifying fibroma versus periapical cemental dysplasia. Further interpretation of the submitted radiographs referred additionally to another similar, but smaller, mandibular mixed radiolucent-radiopaque mass distal to the mandibular right second molar, and to multiple, poorly to moderately well-defined, radiolucent areas superimposed over the roots of the mandibular left first molar, and maxillary left second molar. The appearance was interpreted as suggestive of periapical cemental dysplasia versus florid cemento-osseous dysplasia. The radiolucent areas associated with the mandibular second molars were interpreted to represent either manifestations of the same disease process, or rarefying osteitis due to periodontal disease. At appointments three and five months post-operatively, the patient was asymptomatic and recovering well.

Integrating the histopathology and radiology reports, we came to the conclusion that this lesion represents a case of florid cemento-osseous dysplasia.

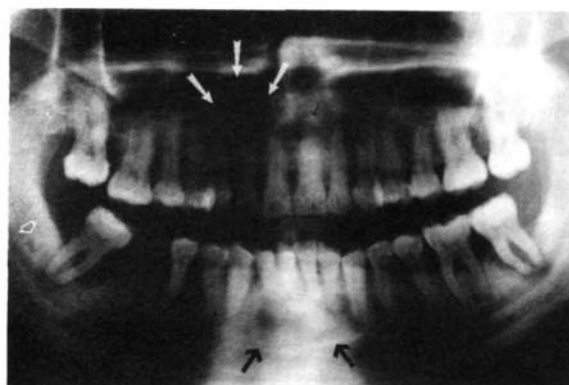


Figure 1. A pantomograph of the jaws showing the right maxillary lesion at the lateral incisor-canine area (solid white arrows), the maxillary lesions at the apices of the central incisors (small black arrow), the anterior mandibular lesion (large black arrows), and the small mandibular lesion distal to the second right molar (open white arrow).



Figure 2. Periapical view of the lower incisor region showing more clearly the anterior mandibular lesion which has a radiopaque center and radiolucent peripheries with corticated margins.

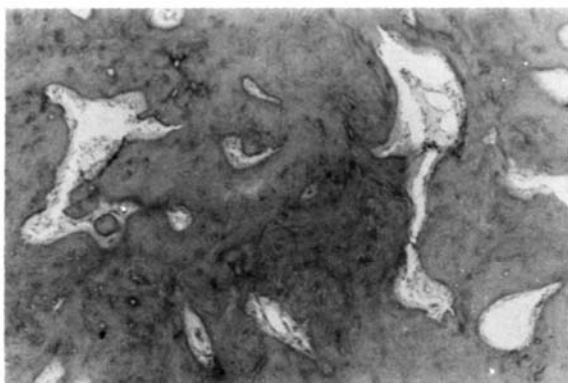


Figure 3. Histopathologic picture of the anterior mandibular lesions (H&E 25X) showing a poorly cellular fibrocollagenous connective tissue matrix, with large confluent mineralized masses.

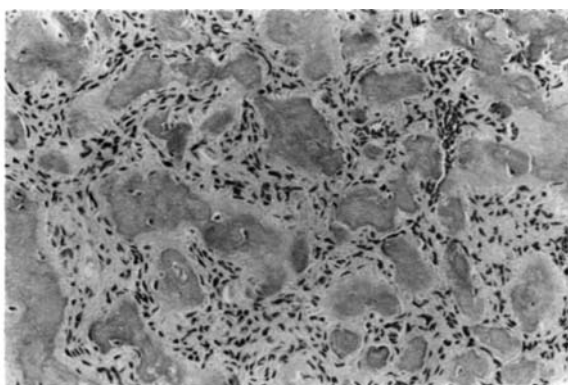


Figure 4. Histopathologic picture of the right maxillary lesion (H&E 63X) showing a highly cellular fibrocollagenous connective tissue matrix, with small foci or mineralization.

Discussion

In the 1971 World Health Organization publication *"Histological typing of odontogenic tumors, jaw cysts and allied lesions"*, Pindborg *et al* used the term gigantiform cementoma to describe a lesion which predominantly affected middle-aged black females, in which multiple sclerotic masses occupied the jaws. The term familial multiple cementomas was used as a synonym.⁹ The literature before and after that publication contains a large number of case reports of similarly described disease processes under several names mentioned in the introduction.

In 1975, Waldron *et al* described the condition as sclerotic cemental masses of the jaws.⁶ They considered these lesions as part of the spectrum of benign fibro-osseous lesions of periodontal ligament origin, representing an exuberant proliferation of cementum. They maintained that the dense mineralized tissue is cementum, and elaborated on its differentiating characteristics under polarized light microscopy.

In 1976, Melrose *et al* introduced the term florid osseous dysplasia, describing the process as an exuberant, multiquadrant involvement that represents the end of the spectrum of osseous dysplasia which includes, according to their classification, cementoma, periapical cemental dysplasia, periapical fibrous dysplasia and periapical osteofibrosis.¹ The subjects in their study group were 34 females, out of whom 32 were black, with a mean age of 42.

Although in almost all references the lesion is thought to affect mainly middle-aged black females, several cases have been reported in other groups including Caucasoid^{21,25}, Oriental^{11,19,26} Indian¹⁹, and West Indian²⁶. Here, we report a case in a Hispanic (Mexican) female. Likewise, a few cases have been reported in males and in a child.^{23,24} Cases with a familial pattern were notably reported in Caucasoids.^{21,24} Melrose *et al* found that 14 of their 34 cases were associated with simple bone cysts.¹ There is also one report in a white male with hereditary hemorrhagic telangiectasia²⁵ but this was probably coincidental.

Clinically, the condition may be asymptomatic, and discovered as a coincidental radiographic finding, or may cause facial deformity resulting from displacement of cortical bony plates.¹ When symptomatic, there may be pain alone or in association with sinus tracks with minimal purulent drainage.⁶ Interestingly, the majority of symptomatic cases occur in edentulous patients, either after extractions or wearing of dentures.¹ M 7,19,20,27 I, J, reasoned that the symptoms are probably related to secondary infection following tooth extraction or denture trauma, where the poorly vascularized hard tissue, with relatively little or no capacity for physiologic resorption, is incapable of combating,¹¹⁹ thus, resulting in chronic osteomyelitis. This is by far the most common complication,^{1,6,19} and probably the source of the confusion of this

lesion with primary osteomyelitis and the erroneous names which describe an inflammatory process^{17,18}.

Radiographically, there is often multiquadrant involvement²⁰ by mixed radiolucent-radiopaque masses, referred to by some as a "pagetoid appearance". These masses are sometimes surrounded by cystic radiolucencies,^{1,6,18,19} which may also be seen as isolated entities, forming the previously mentioned simple bone cysts seen in some cases. If the lesional area communicates with the oral cavity, the margins at the exposure site(s) may appear irregular.⁶ Sequestra may form as well.²⁰

Histopathologically, the lesions are usually found to consist of a bland fibrous matrix with globules, trabeculae or large masses of mineralized tissue which are argued to be bone, cementum or both [Figs. 3 and 4].^{1,6}

The importance of radiological interpretation as an adjunct to diagnosis cannot be over emphasized.^{21,26} We are of the opinion that histopathological distinction between this condition and other fibro-osseous lesions is not always reliable, nor is the distinction between cementum and bone.²¹ Waldron *et al* are of the opinion that bone and cementum could be differentiated on the basis of their polarized light microscopy features.⁶ We agree to considering the sclerotic masses as representative of bone, cementum, or a combination of both, which depends on the way the responsible mesenchymal progenitor cells differentiate.²⁰ Hence, we prefer the term florid cemento-osseous dysplasia, which was used by Waldron in 1985, although he insisted that the lesional tissue was more compatible with cementum than bone.²⁷ Even if it were cementum only, radiographs would still be necessary to differentiate this florid condition from localized ones, such as periapical cemental dysplasia. Furthermore, identical mineralizations may be found in fibro-osseous lesions affecting bones other than the jaws.¹

It is important that the correct diagnosis of florid cemento-osseous dysplasia be established in order that conservative treatment be initiated, thereby avoiding the cumbersome complication of chronic osteomyelitis. The condition can be monitored through routine patient recall visits, with surgical intervention only to avoid secondary infection through apical lesions or decubitus ulcers, and there is no need for overtreatment of the condition,

such as by attempting to excise such diffusely distributed lesions.^{1,6,18,20,27} The process is benign, and there is only one report in the literature of osteosarcoma of the mandible, which developed in a patient after irradiation of the area.²³

In our case, the patient did not develop complications after surgical intervention. Had her periodontitis been controlled at an earlier stage and the nature of the lesions recognized earlier, surgery would not have been the treatment of choice. In our experience with some of the cases referred to our surgical oral pathology service, this condition is unfortunately only recognized after surgical intervention and after osteomyelitis has developed.

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