Mandibular Florid Cemento-Osseous Dysplasia: A Case Report

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Case report

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Abstract

Background: Florid cemento-osseous dysplasia (FCOD) is a non-neoplastic lesion involving multiple quadrants of the jaw.

Case presentation: Patients are usually asymptomatic, and the diagnosis mainly depends on radiographic and clinical examinations. Here, we describe a 50-year-old female patient with clinical manifestations of bone defect, no pain or swelling, combined with imaging indicators of secondary infection. However, there are no accepted treatment guidelines for complex FCOD.

Conclusions: Therefore, based on a review of the literature, this article describes treatment approaches for symptomatic patients and details the successful treatment of FCOD with superimposed fungal osteomyelitis.

Background

The term florid cemento-osseous dysplasia (FCOD) refers to a group of fibro-osseous (cemental) exuberant lesions with multi-quadrant involvement¹. Melrose et al first described the disease in 1976; however, its pathogenesis remains unclear². These lesions are also known as sclerosing osteitis, multiple ossification, diffuse chronic osteomyelitis, and giant resistant bone cementoma. In 2005, the World Health Organization divided Cemento-Osseous Dysplasias (CODs) into three types: periapical, focal, and florid³. These three diseases are difficult to distinguish clinically, and accurate diagnosis mainly depends on histological and radiographic examinations. Although COD may be completely asymptomatic, some patients have pain, swelling, and purulent secretion⁴.

It is generally assumed that COD arises from the periodontal ligament, because of the physical proximity and histopathologic similarity⁵. The clinicopathologic features of COD are as follows: (1) the disease mainly occurs in black middle-aged women; (2) lesions are limited to the jaw and can involve multiple quadrants; (3) mandibular involvement is much more common than maxillary involvement; (4) pathological findings are similar to calcified foci of bone cement with fibroblast matrix⁵⁻⁷. However, the incidence of COD is unknown as a great number of patients are asymptomatic and the disease tends to be noted incidentally on routine radiographic examinations⁸. Symptoms such as dull pain, swelling, and purulent discharge are almost always associated with secondary infection^{6,7,9}.

Given the rarity of FCOD, there are currently no formal treatment guidelines. Retrospective studies over the last decade have indicated that, for asymptomatic patients, invasive surgery should be avoided. Invasive surgery may result in poor healing and increased infection in FCOD-affected lesion areas. For symptomatic patients, there is still considerable controversy as to the optimal treatment for this disease.

Case Presentation

The patient, 50 years old, was admitted to Department of Oral and Maxillofacial Oncology, Wuhan University. Two weeks ago, a "cyst" was found in her right mandible after a routine dental radiograph at the local hospital. The maxillofacial examination revealed facial symmetry; the range and track of mandibular movements were normal. I guarantee that the study is conducted in accordance with international, national and institutional rules, taking into account the right to know of patients in clinical studies. Approved by the Ethics Committee of Huazhong University of Science and Technology. Oral examination showed dentition defects and a removable partial denture. The remaining teeth had caries to varying degrees. An intraoral examination revealed a lack of fullness in the lingual alveolar bone of the right mandible, with normal mucosa and no pain on palpation. Curved tomography showed radial lesions of the mandible involving 44 ~ 46 and 36 ~ 38 spines, as well as in the maxillary region. To better evaluate the lesion, we performed jaw cone-beam computed tomography which showed multiple perforations of the buccal cortex plate (Fig. 1).

The operation was performed under general anesthesia and the intraoral path was selected. Cut along the gingival margin of the mandibular region 0.5 cm, separate and turn the gingival mucoperiosteal flap to the lesion jaw area. Scrape off the wall tissue and close the wound. After operation, anti-infection treatment was given. Seven days following treatment, the patient had recovered well with no allergies or signs of infection. Pathological sections showed typical fungal infection (Fig. 2). The patient was followed up after 1, 3, 5, 7, 12, and 24 months and surface tomography at each follow-up showed good bone regeneration in the original lesion area.

Discussion And Conclusion

Florid cemento-osseous dysplasia is a rare benign lesion that can involve periapical and alveolar areas; it is characterized by variable radiologic features in multiple quadrants of the jaw. To date, there is little research on this disease and the available studies are primarily small case report. In particular, for symptomatic patients, there are currently no accepted treatment guidelines. Further, the available studies are mostly retrospective; there is a lack of control and prospective research. Nonetheless, we have summarized the available treatment strategies that have been shown to be feasible in the small number of available case reports.

We reviewed 43 confirmed cases from 2009 to 2020 and found that 36 of them were female with a median age of 44 years (range 18–72 years) (Table 1); this is consistent with the characteristics of FCOD reported by MacDonald-Jankowski et al. Half of the patients identified in our review had no clinical features, and most of the X lines showed opaque masses (90%). The proportions of patients with pain, swelling, or purulent secretion, were 44%, 33% and 9%, respectively. Other symptoms such as sensory abnormalities and dilation were also been reported in some cases. Imaging examination combined with clinical characterization plays a vital role in FCOD diagnosis. Based on imaging features, FCOD can be divided into three stages: (1) the lesion is dominated by radiolucent changes, showing destructive osteolytic changes; (2) the lesion is characterized by mixed radiologic/intransparent changes; (3) the lesion is characterized by an opaque mass, a "sclerotic or inactive" stage. With regard to the above case study, we found multiple radiated opaque masses around the apex and the radiated opaque halo was limited to the dental region. However, cortical perforation is often associated with bone tissue infection. Therefore, we

can reasonably assume that this patient's unusual bone breakdown was due to secondary infection. The presence of new bone formation or a periosteal reaction is key to distinguishing between fungal and suppurative osteomyelitis; thus, we excluded the possibility of suppurative infection.

According to current treatment guidelines, FCOD patients should avoid any invasive surgery. However, for asymptomatic patients who need to recover masticatory function, routine treatment is not enough. The retrospective review of the literature revealed that resection or curettage of lesions for symptomatic FCOD is the most common surgical treatment. However, the resection range should be carefully selected to obtain a balance between functional recovery and prevention of recurrence. Although the disease is histologically benign, multiple relapses are reported in some cases. When there are no invasive imaging findings, only lesions that cause clinical symptoms should be removed. Active surgical intervention is required when associated with fungal osteomyelitis. For patients with uncomplicated fungal infections, systemic antifungal agents such as amphotericin B, voriconazole, itraconazole, and caspofungin can be used exclusively. However, when these infections are secondary to FCOD, the response to systemic antifungal antibiotics is weak due to altered vascular properties in the lesion area. Hence, complex FCOD requires surgical debridement and removal. For some invasive cases, in situ treatment is performed with aggressive benign tumors or carcinoma; however, this will leave a severe dentition defect.

Florid cemento-osseous dysplasia is a benign, nonneoplastic, self-limiting disease. Few cases have been reported in the current literature. FCOD often presents with no symptoms and is found incidentally on radiographs. In asymptomatic individuals, conservative approaches, such as broad-spectrum antibiotics in the case of infections along with surgical debridement and enucleation, are required. In asymptomatic individuals, only periodic follow-up and good oral care are advised.

Abbreviations

florid cemento-osseous dysplasia (FCOD)

Cemento-Osseous Dysplasias (CODs)

Declarations

Ethics approval and consent to participate

The case report was reported with the informed consent of the patient.

Consent for publication

The images in the manuscript had been given the informed consent of the patient.

Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

Author [Zirong Tang](#): Contributed to conception, design, acquisition, drafted manuscript, critically revised manuscript, gave final approval and agree to be accountable for all aspects of work ensuring integrity and accuracy.

Author [Lin Wang](#): Contributed to conception, design, acquisition, drafted manuscript, critically revised manuscript, gave final approval and agree to be accountable for all aspects of work ensuring integrity and accuracy.

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Author [Yulin Jia](#): Contributed to design, acquisition, analysis, and interpretation ,drafted manuscript, critically revised manuscript, gave final approval and agree to be accountable for all aspects of work ensuring integrity and accuracy. All authors gave their final approval and agree to be accountable for all aspects of the work.

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Tables

Table 1
Review of 43 cases documented as florid cemento-osseous dysplasia during 2009–2020

Patient (year)	Reference	Location	Sex, age (years)	Chief Complaint	Clinical Presentation	Infection	Familial History	Radiologic Findings	Management	Follow up (month)
1 (2018)	Esfahanizadeh et al ¹	Bilaterally in posterior mandible	F/62	Mandibular partial edentulous and demanding implant restoration	N	N	N	Radio-opaque masses with radiolucent halo	Conservative step-by-step 2-stage implant surgery	18
2,3 (2019)	Brody et al ²	Anterior and posterior mandible	F/50	Uncertain pain in the right mandibular region.	Pain and paresthesia	NA	Y	Radio-opaque masses with radiolucent halo	Removed the apical lesion and performed endodontic treatment	18
		Anterior of right mandible	F/19	NA	N	NA	Y	Radiolucent lesion	Follow-up	12
4 (2019)	Yang et al ³	Periapical distribution throughout the mandible and maxillae	F/73	A sharp bony area arising in the upper right maxillary region	Expansion	N	N	Radio-opaque masses	Follow-up	NA
5 (2018)	Min et al ⁴	Posterior mandible and maxilla	F/40	Pain in the tooth on chewing after repaired with a metal crown	Pain	N	NA	Radio-opaque masses with radiolucent halo	Mandibular lesions were removed	42
6,7 (2018)	Toledano-Serrabona et al ⁵	Bilateral maxilla and right of mandible	F/42	Acute and pulsating 1-month-long pain on the fourth quadrant	Pain	Y	NA	Radio-opaque masses with radiolucent halo	Extraction of tooth 47 and the elimination plus curettage of the residual bone cavity	6
		The left of mandible	F/37	Assessment and management of numerous mandible fibro-osseous lesions	N	N	NA	Radio-opaque masses	Endodontic treatment and periapical surgery plus enucleation of the lesion	< 1
8 (2018)	Aiuto et al ⁶	Bilateral mandible and maxilla	F/49	Recurrent episodes of abscesses and exhibiting a significant degree of mobility inferiorly to the second right molar	Purulent discharge and looseness of teeth	Y	NA	Radio-opaque masses with radiolucent halo	Extracted the second right inferior molar and enucleated the apical lesion	24
9,10,11 (2017)	Daviet-Noual et al ⁷	Bilateral mandible and maxilla	M/64	Mobile and painful due to a periodontal abscess.	N	N	NA	Radiolucent lesion	Extraction of the infection teeth	< 1 (last image date)
		Bilateral mandible	F/50	Chronic pain and tenderness on the lower left second molar	N	N	NA	Radio-opaque masses with radiolucent halo	Target specific symptoms only(endodontic treatment)	6
		Bilateral of posterior mandible	F/72	Multiple tooth pain (apical periodontitis)	N	N	NA	Radio-opaque masses with radiolucent halo	Endodontic treatment and incisional biopsy	NA

Patient (year)	Reference	Location	Sex, age (years)	Chief Complaint	Clinical Presentation	Infection	Familial History	Radiologic Findings	Management	Follow up (month)
12 (2017)	Sarmiento et al ⁸	Bilateral mandible	F/35	Facial asymmetry	N	NA	NA	Radio-opaque masses with radiolucent halo	Follow-up	NA
13 (2016)	Cavalcante et al ⁹	Bilateral of posterior mandible	F/49	Edema and extra oral fistula with pus drainage in hemiface submandibular of the right side	Pain and purulent discharge	Y	NA	Radio-opaque masses	Extracted the tooth 42 and curette the right lesion	12
14 (2016)	Yildirim et al ¹⁰	Bilateral of posterior mandible	F/29	To treat her cracked and missing teeth	N	N	NA	Radio-opaque masses with radiolucent halo	Follow-up	NA
15 (2015)	Delai et al ¹¹	Bilateral of posterior mandible	F/38	Pain on the right side of her face, particularly in the morning	N	NA	NA	Radio-opaque masses with radiolucent halo	Follow-up	lost to follow
16,17 (2015)	Thorawat et al ¹²	Mandible and maxilla	F/24	Pain and swelling in the right maxillary	Pain and swelling	Y	Y	Radio-opaque masses	Broad-spectrum antibiotics along with periodic recall	NA
		Bilateral mandible	F/45	Decayed teeth in the left mandibular posterior region of the jaw	N	N	Y	Radio-opaque masses	Follow-up	NA
18 (2014)	Ali et al ¹³	Bilateral mandible and maxilla	F/46	Acute episodes of chronic jaw discomfort	Pain	Y	NA	Radio-opaque masses with radiolucent halo	NA	NA
19 (2014)	Grewal et al ¹⁴	Bilateral mandible	M/26	Tenderness associated with a swelling in the left mandibular premolar region.	Pain	N	NA	Radio-opaque masses with radiolucent halo	Regular follow-ups	NA
20 (2013)	Senturk et al ¹⁵	Bilateral mandible and maxilla	M/35	Severe swelling, notably in the maxilla	Swelling	N	N	Radio-opaque masses with radiolucent halo	Extracted the involved teeth and removed the expanding bone	16
21 (2013)	Sarmiento et al ¹⁶	Bilateral mandible and maxilla	F/54	A swelling in the bone	Swelling	Y	N	Radio-opaque masses with radiolucent halo	Follow-up	NA
22 (2013)	Gerlach et al ¹⁷	Bilateral mandible	F/39	A spontaneous pain	N	Y	NA	Radio-opaque masses with radiolucent halo	Removed the mobile implant and curetted the expanding body	12
23 (2013)	Shah et al ¹⁸	Bilateral of posterior mandible	F/65	A request for the replacement of missing teeth	N	N	NA	Radio-opaque masses with radiolucent halo	Follow-up	NA

Patient (year)	Reference	Location	Sex, age (years)	Chief Complaint	Clinical Presentation	Infection	Familial History	Radiologic Findings	Management	Follow up (month)
24 (2013)	Rekabi et al ¹⁹	Posterior mandible and the right of maxilla	F/39	To restore the right mandibular first molar tooth, which had extensive caries	N	N	NA	Radio-opaque masses with radiolucent halo	Only removed the periapical lesion	12
25 (2013)	Huh et al ²⁰	Bilateral mandible	F/30	To evaluation and further treatment of the persistent lesions after root canal treatment	N	N	NA	Radio-opaque masses with radiolucent halo	Only removed the discomfortable area during palpation	21
26 (2012)	Kutluay Koklu et al ²¹	Bilateral mandible and the left of maxilla	M/45	For further investigation and treatment of his existing lesions.	N	N	NA	Radio-opaque masses with radiolucent halo	Follow-up	30
27,28 (2012)	Srivastava et al ²²	Bilateral mandible and maxilla	M/19	To correcte the spacing and malpositioned teeth	N	N	Y	Radio-opaque masses with radiolucent halo	Follow-up	NA
		Bilateral mandible and maxilla	F/NA	To correcte malpositioned teeth and pain	Pain and expansion	Y	Y	Radio-opaque masses	Curetted the painful area	< 1
29 (2011)	Smith et al ²³	Bilateral mandible and maxilla	F/53	Swelling, pain, and purulent in the left mandible area	Swelling, pain, and purulent discharge	Y	Y	Radiolucent lesion	Debridement and oral antibiotics after operation	NA
30 (2011)	Rao et al ²⁴	Bilateral mandible and maxilla	F/41	Pain in the right posterior region of the mandible	Pain	Y	NA	Radio-opaque masses with radiolucent halo	Curetted the multilocular lesion	6 (lack image date)
31,32 (2011)	Lawal et al ²⁵	Bilateral of mandible	F/70	Pain and discharge from a sinus located on the left jaw	Pain and infection	Y	N	Radio-opaque masses with radiolucent halo	Oral clindamycin tablets 300mg bid	Lost to follow
		Bilateral mandible and maxilla	F/60	Tooth ache and jaw swelling	Pain and swelling	Y	N	Radio-opaque masses	NA	NA
33 (2011)	Kim et al ²⁶	Bilateral mandible and maxilla	F/52	Acute pain and gingival swelling on the right mandibular molar region	Pain, swelling and inflammation	Y	Y	Radio-opaque masses	Surgical curettage was performed on the right mandibular posterior area	12(lack image date)
34,35 (2010)	Zillo Martini et al ²⁷	Bilateral mandible and maxilla	F/66	Mandibular lesions found in a panoramic X-ray taken for regular dental treatment	N	N	NA	Radiolucent lesion	Curettage	18
		Bilateral mandible	F/42	Facial asymptomatic swelling in the left mandible	Swelling, painless	N	NA	Radio-opaque masses with radiolucent halo	Curettage	Lost to follow

Patient (year)	Reference	Location	Sex, age (years)	Chief Complaint	Clinical Presentation	Infection	Familial History	Radiologic Findings	Management	Follow up (month)
36 (2010)	Sanjai et al ²⁸	Bilateral maxilla and the left of mandible	F/20	A swelling on the right zygomatic region	Swelling, painless	N	NA	Radio-opaque masses with radiolucent halo	Surgery was performed for all three quadrants	NA
37 (2009)	Gunduz et al ²⁹	Bilateral mandible and maxilla	F/43	A cute pain in the left molar region of the maxilla	Pain and swelling	N	NA	Radio-opaque masses	Only removed the loose teeth	11
38 (2019)	Choudhary et al ³⁰	Bilateral mandible	F/27	A swelling in the lower right front teeth region	Swelling	N	N	Radio-opaque masses with radiolucent halo	Facial recontouring under local anesthesia	NA
39 (2020)	Saikia et al ³¹	Bilateral mandible and maxilla	F/44	Draining lesion from her chin region	Pain, swelling and purulent discharge	Y	NA	Radio-opaque masses	extraction of the involved teeth was done along with Incisional biopsy	< 1
40 (2020)	Kungoane et al ³²	Bilateral mandible and maxilla	M/43	Painless bony protrusion	Swelling	N	NA	Mixed radiopaque-radiolucent lesions	Biopsies	NA
41,42,43 (2020)	Nel et al ³³	The right of maxilla	F/58	Pain in the right maxilla	Pain and swelling	Y	Y	Mixed radiopaque-radiolucent lesions	Surgical debridement of the necrotic bone	NA
		Bilateral mandible and maxilla	F/18	Mandible appeared expansive and protrusive	Mandible appeared expansive and protrusive	N	Y	Mixed radiopaque-radiolucent lesions	Follow-up	NA
		Bilateral mandible and maxilla	M/21	Pain in the lower-left side of the mandible	Pain and swelling	N	Y	Mixed radiopaque-radiolucent lesions	N	Lost to follow

Figures

Figure 1

Imaging examination of the patient.

Figure 2

Pathological sections of the patient.