

# Benign Fibro-Osseous Lesions of The Jaw: A Retrospective Analysis

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## ABSTRACT

**Objective:** The main goal of this retrospective study was to characterise FOLs in terms of their demographic distribution, prevalence, and clinical and radiological features, and to discuss the treatments for this condition.**Methods:** This study included patients with FOLs found in the archives of the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, University of Harran, Türkiye. The panoramic radiographs and histopathological results of all patients referred to our clinic between 2017 and 2020 were reviewed retrospectively. In total, 18,835 patient records were evaluated. Two oral and maxillofacial surgeons sequentially examined the panoramic radiographs of all patients who presented to our clinic for examination or treatment. In total, 10 patients showed radiological and histopathology results compatible with FOLs.**Results:** In total, 18,835 radiographs were evaluated, and 10 (0.00074%) FOLs were seen in 10 patients (8 females and 2 males) ranging in age from 18–64 years. Three of the cases were of FCOD, three were of FaCOD (father and two daughters), one was of FoCOD, one was of OF, and two were of FD.**Conclusion:** FOLs, and in particular FaCFOD, are rarely seen in the clinic. Accurate diagnosis of these diseases is important to avoid inappropriate treatment. In this study, we reported 10 FOLs in 10 patients seen at our institution, and presented a review of the literature.**Keywords:** fibroosseous lesions, familial osseous dysplasia, ossifying fibroma, florid cemental osseous dysplasia

## INTRODUCTION

Fibro-osseous lesions (FOLs) are a group of lesions of the jaw and facial bones arising from fibroblast cells. These lesions can be developmental, reactive-dysplastic, or neoplastic. In all FOLs, bone is replaced with fibrous and cement-like tissue, which shows gradual mineralization [1].

Although the classification system for FOLs has undergone several revisions, they are typically classified as fibro-osseous neoplasms (FOs), osseous dysplasias (ODs), or fibrous dysplasia (FD), based on the system of Waldron,[2] or Ossifying fibroma (OF), FD, or cemento-osseous dysplasias (CODs), as proposed

by the World Health Organization (WHO, 2017) [3]. The WHO (2017) supports the terminology of ‘cemento-osseous dysplasia’ because this term more accurately describes the relationship of the lesions to the teeth and their origin from the periodontal tissues. [3].

Radiographic modalities, such as cone-beam computed tomography (CBCT) and orthopantomography, are used to study FOLs in detail, including in terms of their relationship with adjacent structures. Radiographically, FOLs may be radiolucent (early stage), mixed radiolucent-radiopaque (mixed stage) or radiopaque (mature stage) in appearance. It may be well-circumscribed, or it may have an appearance that cannot be clearly separated from the surrounding bone tissue. Resorption and displacement of teeth may occur, in addition to bone expansion [4].

COD is a FOLs subtype classified as periapical cemento-osseous dysplasia (PCOD), focal cemento-osseous dysplasia (FoCOD), florid cemento-osseous dysplasia (FCOD), or familial form of florid cemento-osseous dysplasia (FFCOD) according to the clinical and radiographic findings. The term ‘florid’ is in reference to the possibility of FCOD being present in multiple quadrants of the jaw, although appearance in all four quadrants is rare [5].

FFCOD is an uncommon hereditary autosomal dominant disorder; only a few cases have been reported in the literature. FaFCOD is characterised by irregular, lobular, intense and opaque masses that spread along the jaws and alveolar processes, and affect multiple members of the same family [6].

#### Main Points:

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Usually, no treatment is required for FFCOD, such that only regular follow-up examinations are recommended. Accurate diagnosis is crucial because misdiagnosis can lead to the requirement for biopsy, endodontic treatment, or surgical treatment [7].

The main goal of this retrospective study was to characterise FOLs in terms of their demographic distribution, prevalence, and clinical and radiological features, and to discuss the treatments for this condition.

#### MATERIALS AND METHODS

This study included patients with FOLs found in the archives of the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, University of Harran, Türkiye. The Ethics Committee of Harran University Faculty of Medicine approved this study, which was conducted in accordance with the Declaration of Helsinki. The panoramic radiographs and histopathological results of all patients referred to our clinic between 2017 and 2020 were reviewed retrospectively. In total, 18,835 patient records were evaluated.

Two oral and maxillofacial surgeons sequentially examined the panoramic radiographs of all patients who presented to our clinic for examination or treatment. In total, 10 patients showed radiological and histopathology results compatible with FOLs, and were thus included in the study (Table 1). They were classified based on the system of WHO [3]. The data analysed included gender, age, symptoms, other medical problems, location of lesions, radiographic technique, histopathological findings, treatment and postoperative complications.

#### RESULTS

In total, 18,835 radiographs were evaluated, and 10 (0.00074%) FOLs were seen in 10 patients (8 females and 2 males) ranging in age from 18–64 years. Three of the cases were of FCOD, three were of FFCOD (father and two daughters), one was of of FoCOD, one was of COF, and two were of FD (Table 1).

Pathological growth of the jaw bones (resulting from displacement of cortical plates) caused pain and aesthetic problems in three cases, and a decrease in chewing function in five others (three FFCOD, two FD). Thus, contour plasty was performed under general anaesthesia. Postoperatively, there were no complications and patients attended the routine follow-up.

**Table 1.** Distribution of type of lesions, age/ gender, symptoms, other medical problems, location of lesions, type of radiograph, treatment and complication of patients with fibroosseous lesion.

Type of lesion	Gender/age	Symptoms	Other Medical Problems	Location of Lesions	Radiograph	Treatment	Post-operative Complications
FOD	F/50	-	-	Right and left mandible	OPG	No treatment	-
FOD	F/37	pain	-	Right and left mandible	OPG	No treatment	-
FOD	F/34	-	-	Right maxillary anterior region, right and left mandible	OPG	No treatment	-
FaOD	M/37	swelling and dull pain	-	involving four quadrants of the maxilla and mandible, left subcondyle region and right mandibular angulus region	OPG	contour plasty	-
FaOD	F/18	swelling and dull pain	-	involving four quadrants of the maxilla and mandible, left mandibular incisura	OPG	contour plasty	-
FaOD	F/15	and dull pain swelling	-	involving four quadrants of the maxilla and mandible, left mandibular angulus	OPG	contour plasty	-
FoOD	F/51	-	-	Anterior mandibular region	OPG	No treatment	-
OF	F/64	Dull pain	-	Right mandibular premolar-molar region	OPG	No treatment	-
FD	M/42	asymmetry	-	Left mandibular molar region	OPG	contour plasty	-
FD	F/22	asymmetry	-	Right maksillar anterior region	OPG	contour plasty	-

## DISCUSSION

The aetiology of FOLs is not yet fully understood, but the proximity of the lesions to the periodontal ligament suggests that they originate from the same tissue. FOLs are classified as developmental, reactive-dysplastic or neoplastic. FDs are developmental lesions, fibromas are neoplasms, and CODs are reactive lesions [8].

In the most recent WHO classification, COF was classified as an odontogenic tumor having a mesenchymal origin, distinguishing it from the juvenile types [3]. However, it is still a fibro-osseous lesion and was discussed in detail with the other OFs in the fibro-osseous lesions section of the most recent WHO edition.

OF, which is classified as a benign odontogenic tumour of mesenchymal origin that can occur in any facial bone, is usually seen in the third and fourth decades of life. Among BFOLs, OF is the only group that is neoplastic in origin, and divided into two subgroups as cemento-ossifying fibroma and juvenile ossifying

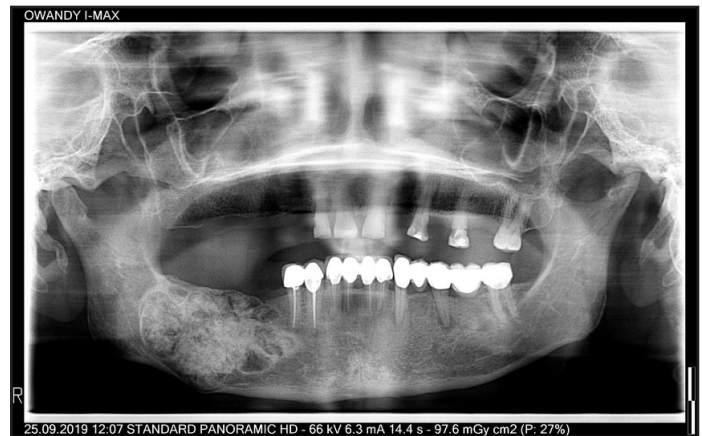
fibroma [9]. Opinions vary regarding gender differences in the prevalence of OF. MacDonald et al. suggested that OF affects both genders equally overall, but women are affected more frequently in the fourth decade of life [10]. OF is frequently seen in the mandibular premolar-molar region, and the maxilla, zygomatic arch and canine fossa are also often affected [9]. OF may present as a well-defined, completely radiolucent mass in the early stages, or with areas of opacity varying according to the degree of calcification. In advanced stages, OF can appear as a mass surrounded by radiopaque and radiolucent bands. Migration and root resorption may be observed in adjacent teeth, and the bone cortex may show expansion [11]. In this study, we found one case of OF. Patient (women aged 64) had chief complaints of expansion of the bone cortex and pain (Figure 1). The lesion was located in the right mandibular premolar-molar region. Radiographically, the lesion appeared as well-defined, dense radiopaque areas surrounded by radiolucent area. The demographic, clinical and radiographic findings of the patient was consistent with previous studies.

The aetiology of FD, a hamartomatous developmental bone disease that arises due to a failure of immature bone to mature, is not fully understood [12]. FD lesions may occur in one bone (monostotic), or in multiple bones (polyostotic). One form of polyostotic FD, namely McCune-Albright syndrome, is accompanied by skin and endocrine anomalies. The most common form of FD is the monostotic type, which accounts for approximately 80–85% of all cases [10]. FDs are frequently seen in the jaw and facial bones. They primarily affect the maxilla, but involvement of the mandible, zygoma, sphenoid, frontal and occipital bones may also be seen [10]. Eversole et al. [11] reported that teeth remain in place when there is bone involvement, but displacement may occasionally occur. Tooth root resorption is rare. Monostotic-type FD is a slow-growing, painless mass generally observed in young adults (second and third decades of life), which affects both sexes equally [13]. The term craniofacial fibrous dysplasia is used to describe monostotic-type FD [11]. On radiographic images, FD may show a radiolucent, sclerotic or mixed radiopaque-radiolucent appearance. Extragnathic FD lesions have better-defined margins compared to gnathic lesions, and the lack of a diffuse structure and borders is an important radiographic feature of FDs of the jaw and facial bones [10]. In this study, we found two cases of FD (female and male aged 22 and 42 years, respectively). In the first patient, the lesion was located in the right maxillary incisor and premolar region, whereas it was located in the left mandibular molar region in the second patient (Figure 2). In both cases, the lesions caused the bone to expand; the expansion was much greater in the second case, enlarging the lingual and buccal cortex and alveolar bone and being in contact with the opposite teeth. Contour plasty was performed in both cases due to the patients' complaints. Radiography revealed sclerosis in the first case, with resorption seen in the roots of the right maxillary first and second incisors. In the second case, the lesion had displaced the teeth and inferior alveolar canal. Expansion of the alveolar bone was also clearly visible. There were no skin or endocrine anomalies in either case. Both cases showed single bone involvement, compatible with monostatic-type FD.

COD is a group of dysplastic bone lesions most commonly seen in the mandible. COD is divided into four subgroups: PCOD, FoCOD, FCOD, and FFCOD [5]. FFCOD is a hereditary form of COD that is much rarer than the other forms. All forms of COD affect the area around the tooth and emerge from the periodontal ligament or cement-like tissue. Although the histopathological features of the different types of CODs are the same, their clinical

and radiographic features are different [14].

PCOD is more common in women between the third and fifth decades of life [15]. Radiographically and histopathologically, the lesions show three distinct stages, where progression from the first to the third stage may take months or even years. Early (lytic) stage lesions appear as radiolucent areas in the periapical region, and can be confused with chronic inflammatory lesions [11]. Clinical examination is important for diagnosis. In the second stage, as the lesion matures, radiopaque foci appear within the radiolucent area. In the third (mature) stage, the lesion shows a solid, radiopaque appearance with a surrounding radiolucent band. According to Koenig et al. [16], PCOD lesions are round, typically multifocal, well-defined and smaller than 1 cm. Cortical bone and mucosa are normal. Multifocal lesions can be in different stages within the same patient. In this study, we didn't find any PCOD case.



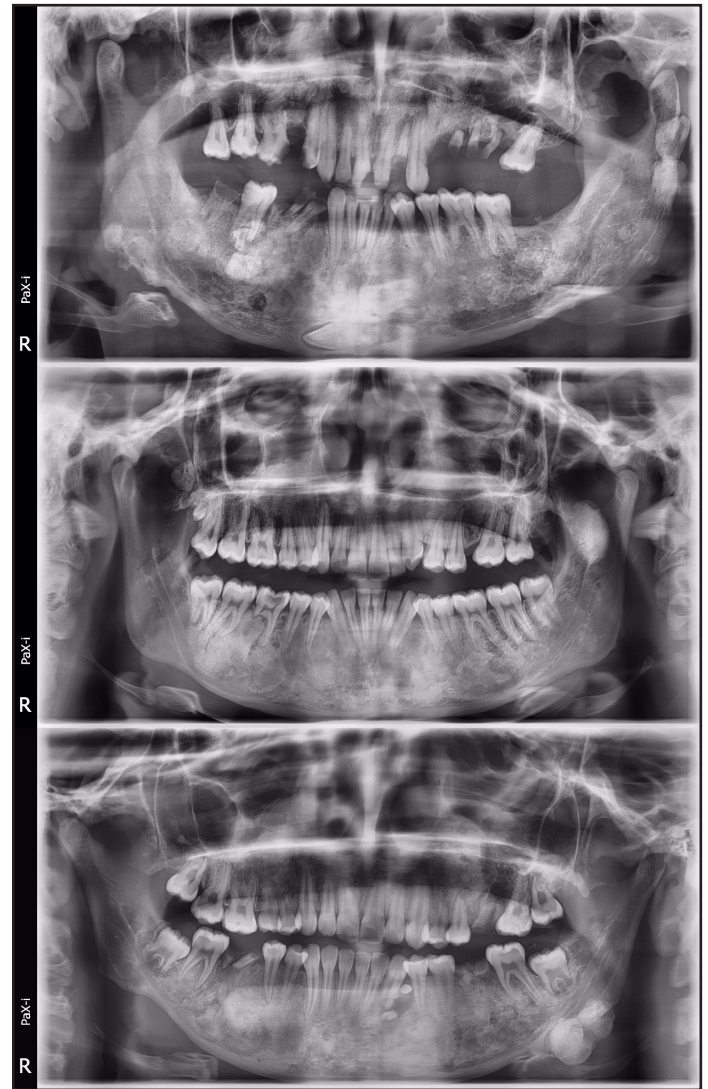
**Figure 1.** Panoramic radiograph showing large cemento-ossifying fibroma lesion which involve right mandibular premolar- molar region.



**Figure 2.** Panoramic radiographic appearance of fibrous dysplasia

FoCOD is the most common type of FOL, and is seen most frequently in African women in the third and fourth decades of life. Although the posterior mandibular region is the most affected area, FoCOD can develop anywhere in the jaw bone. FoCOD usually affects the apical region of the tooth [10]. Radiographically, FoCOD may be well-defined, fully radiolucent, or radiopaque with a partially sclerotic surrounding band. However, these two patterns typically cooccur. FoCOD can also occur in edentulous patients and tooth extraction areas; the lesions are usually 1–2 cm in size and do not tend to show a multifocal distribution. Although the lesion is usually asymptomatic, it can cause pain and swelling if there is secondary infection. FoCOD is generally detected incidentally on routine dental radiographs; the presence of vital teeth in close proximity to the periapical or tooth extraction region is an important diagnostic finding [9]. In this study, we found one FoCOD case (women aged 51 years). The lesions were located in the anterior mandibular region. Radiographically, the lesions were in the mature stage, and appeared as well-defined, dense radiopaque areas.

FCOD is a common form of PCOD [16] White et al. [15] showed that FCOD is most prevalent in women in the fourth and fifth decades of life, typically presenting symmetrically and bilaterally in the mandible or mandibular canal. FCOD can be asymptomatic and is often detected incidentally. Koenig et al. [16] stated that FCOD differs from FoCOD in that it affects more than one quadrant of the jaw and has a larger size. If the lesion becomes very large, it can expand the bone. Radiographic images of FCOD show multiple sclerotic radiopacities surrounded by a peripheral radiolucent rim, accompanied by mixed lesions with ill-defined borders [17]. Extensive lesions can cause cortical expansion and displacement of the mandibular canal [15]. In this study, we found three FCOD cases, (females aged 50, 37, and 34 years). The lesions were well-defined, located bilaterally in the mandible, and showed a multifocal distribution. In the first and second patients (Figure 3), the lesions were well-defined, located bilaterally in the mandible, and showed a multifocal distribution. In the third patient the lesions were well-defined, located bilaterally in the mandible and right maxillary anterior region, and showed a multifocal distribution. Lesions in all three patients show early, mixed and mature stages. The demographic, clinical and radiographic findings of the patients were consistent with previous studies.



**Figure 4.** Panoramic radiographs of family members



**Figure 3.** In the radiographic image, multiple lesions are visible in both quadrants of the mandible. Similar pathologic lesions involvement of mandible and the surrounded the teeth.

**Table 2.** Review of the literature on familial florid osseous dysplasia (FaOD)

Author	Year	Ethnicity	Involved family members	Reported as
1. Agazzi and Belloni <sup>24</sup>	1953	Italian family	Unspecified	Hard odontomas of the jaws
2. Cannon et al <sup>23</sup>	1980	Unspecified	Mother and son	Familial gigantiform cementoma
3. Sedano et al <sup>21</sup>	1982	White family	10 members	Autosomal dominant cemental dysplasia
4. Young et al <sup>20</sup>	1989	White family	55 members	Familial gigantiform cementoma
5. Musella and Slater <sup>25</sup>	1989	Italian family	Mother and daughter	Familial florid osseous dysplasia
6. Oikarinen et al <sup>22</sup>	1991	Caucasian family	Father and 2 children	Familial gigantiform cementoma
7. Thakkar et al <sup>30</sup>	1993	Caribbean family	Mother, 2 daughters and 1 son	Familial periapical cemental dysplasia
8. Coleman et al <sup>19</sup>	1996	African family	Mother and 2 children	Familial florid osseous dysplasia
9. Toffanin et al <sup>18</sup>	2000	Italian family	Grandmother, son, daughter, grandson, granddaughter	Familial florid osseous dysplasia
10. Hatori et al <sup>26</sup>	2003	Japanese family	Father and daughter	Familial florid osseous dysplasia
11. Srivastava et al <sup>29</sup>	2012	Indian family	Mother and son	Familial florid osseous dysplasia
12. Sim et al <sup>27</sup>	2014	Asian family	Mother and her identical twin daughters	Familial florid osseous dysplasia
13. Thorawat et al <sup>28</sup>	2015	Black family	Mother and daughter	Familial florid osseous dysplasia
14. Kucukkurt et al <sup>17</sup>	2016	Unspecified	Mother, son and mother's brother	Familial florid osseous dysplasia
15. Mingming et al <sup>6</sup>	2019	Chinese family	The family comprises three generations	autosomal dominant Familial florid osseous dysplasia

FFCOD is rarely seen; in our review, only 15 cases were found (Table 2) [6, 17-30]. Küçük Kurt et al. [17] noted that although FFCOD is usually asymptomatic, it can cause pain, swelling and facial deformity due to infection. FFCOD tends to occur in younger individuals [6]. Radiographically, FFCOD shows similar features to the other CODs; moreover, maturation and the quantity of mineralised tissue in the lesion affect the radiographic appearance. A completely radiolucent appearance is common in early lesions, while radiopaque foci appear as the lesions mature. In mature lesions, a radiolucent border around the lesion separates it from the adjacent normal bone [15]. We found a case of FFCOD where in a father and two daughters showed similar clinical and histopathological findings. The father was 37 years old and his daughters were aged 15 and 18 years. Orthopantomography revealed multiple radiopaque masses with radiolucent borders in all four quadrants of the jaw, in all three family members (Figure 4). The thin lesions appeared as irregular and lobular radiopacities surrounded by a radiolucent border. All patients exhibited regional swelling. The father had right mandibular angle and left ramus swellings; his older daughter had a left

mandibular incisura swelling and the younger daughter showed a left mandibular angle swelling. All three patients complained of extreme swelling and dull pain. Root resorption was present in some teeth in all three cases. In all patients, contour plasty was performed via an extraoral submandibular and intraoral approach. No postoperative complications developed in any of the patients and they were routinely followed-up. Regarding FFCOD cases previously reported, they were more common in white families; only two families were black. Moreover, the mother and children were affected more frequently; in only two cases were the father and children affected, as in our case (Tables 1 and 2).

**Limitations:** The major limitation of our study is the small number of cases and it was studied single center. The research needs to be supported with more cases and it is thought that multi-center studies will contribute more to the literature.

## CONCLUSION

FOLs, and in particular FFCOD, are rarely seen in the clinic. Accurate diagnosis of these diseases is important to avoid

inappropriate treatment. In this study, we reported 10 FOLs in 10 patients seen at our institution, and presented a review of the literature.

**Conflict of interest:** The authors declare that they have no conflict interest.

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**Ethical Approval:** The study was approved by Harran University local ethics committee (Approval Number: 2020/02/012, Date: 2024-02-19).

**Author Contributions:** Concept – S.Y., M.E.P.; Design – S.Y., M.E.P.; Supervision – M.E.P, S.Y.; Resources – S.Y., M.E.P.; Materials S.Y., M.E.P.; Data Collection and/or Processing– M.E.P, S.Y.; Analysis and/or Interpretation – M.E.P., S.Y.; Literature Search – S.Y., M.E.P.; Writing Manuscript – S.Y., M.E.P.; Critical Review – M.E.P, S.Y.

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