

Central Odontogenic Fibroma, its Treatment and its Recurrence: A Systematic Review

Review Article

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Abstract

Introduction: The central odontogenic fibroma (COF) is an extremely rare benign neoplasm of intraosseous or central presentation, being itself only 0.1% of all odontogenic tumors. The purpose of this systematic review is to provide an overview of the treatment of the central odontogenic fibroma and its recurrence.

Methods: Following PRISMA guideline, a systematic review was carried out.

Results: The initial search retrieved 51763 articles identified through database searching, and 41 articles identified through manual search. After examination of the title and the abstract, 83 articles were retrieved for further examination. Sixty-two articles were included, thus obtaining a total of 104 patients. The recurrence of the 84 patients was 4.76%.

Discussion: The recurrence reports vary in the literature. The most recent review by Garcia-Cano E, et al., showed no recurrence whatsoever; Ramer and colleagues reported only 5 cases of recurrence. However, only 39 cases out of 68 were followed up. In this systematic review out of 84 patients, 4 (4.7%) presented recurrence. From these, 3 cases were treated by enucleation and one by curettage.

Conclusion: The recurrence rate in these tumors is very low and mainly related to an incomplete resection. Therefore, conservative surgery must be done as a first-line treatment.

Keywords: Systematic Review; Central Odontogenic Fibroma; Treatment; Recurrence.

Introduction

The central odontogenic fibroma (COF) is an extremely rare benign neoplasm of intraosseous or central presentation, being itself only 0.1% of all odontogenic tumors [1, 2]. It is histologically considered among the odontogenic tumors of mesenchymal tissue, with or without formation of odontogenic epithelium [1]. The World Health Organization (WHO) defines it as a benign proliferation of ectomesenchyme fibroblastic tissue characterized by a variable number of apparently inactive odontogenic epithelium [3]. There are two distinct histological types:

1. Simple COF, characterized as a poorly cellular and myxoid fibroblastic neoplasia without odontogenic epithelial component.
2. WHO or complex COF, with abundant odontogenic epithelium cell with foci of calcification [4].

The treatment of COF in the literature mainly involves

conservative surgery [5]. However, this treatment is based only in case reports or literature reviews without a complete follow-up of all the patients.

The purpose of this systematic review is to provide an overview of the treatment of the central odontogenic fibroma and its recurrence, based on the following key questions:

- (1) What are the surgical treatments reported?
- (2) What is the recurrence with these treatments?

Methods

Search Strategy

A research through public domain databases was performed to identify articles focusing on COF and its treatment by two

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authors Lombardi-Azócar JJ and García-Cano E, including the following databases: PubMed, Embase, Cochrane Library, and Web of Science (all searched up to January 4th, 2016). In addition, we performed a manual search of secondary sources including references of the articles initially identified. The goal was to identify all studies addressing COF in relation to its treatment and its recurrence.

The following search terms were used: ((((((Central Odontogenic Fibroma) OR COF) AND Treatment) OR Enucleation) OR curettage) OR Resection) AND Recurrence.

Data Extraction and Analysis

We followed the PRISMA guideline for systematic reviews.

All the articles regarding the prevalence and treatment of patients with COF and its recurrence were included. The full texts of articles that met the inclusion criteria and of articles whose abstract was lacking information were obtained.

Data on the number of patients, patient characteristics such as gender, age, COF localization, correlation with unerupted tooth, treatment used, follow-up, and recurrence were tabulated.

Inclusion Criteria

Patients with histopathological diagnosis of COF, with follow-up and information regarding recurrence.

Exclusion Criteria

Patients with histopathological diagnosis of odontogenic fibroma, without diagnosis of being central.

Patients without follow-up.

Patients without information about recurrence. Patients with histopathological findings of COF and other types in the same tumor. Articles that did not have an English abstract with the above-mentioned characteristics.

Results

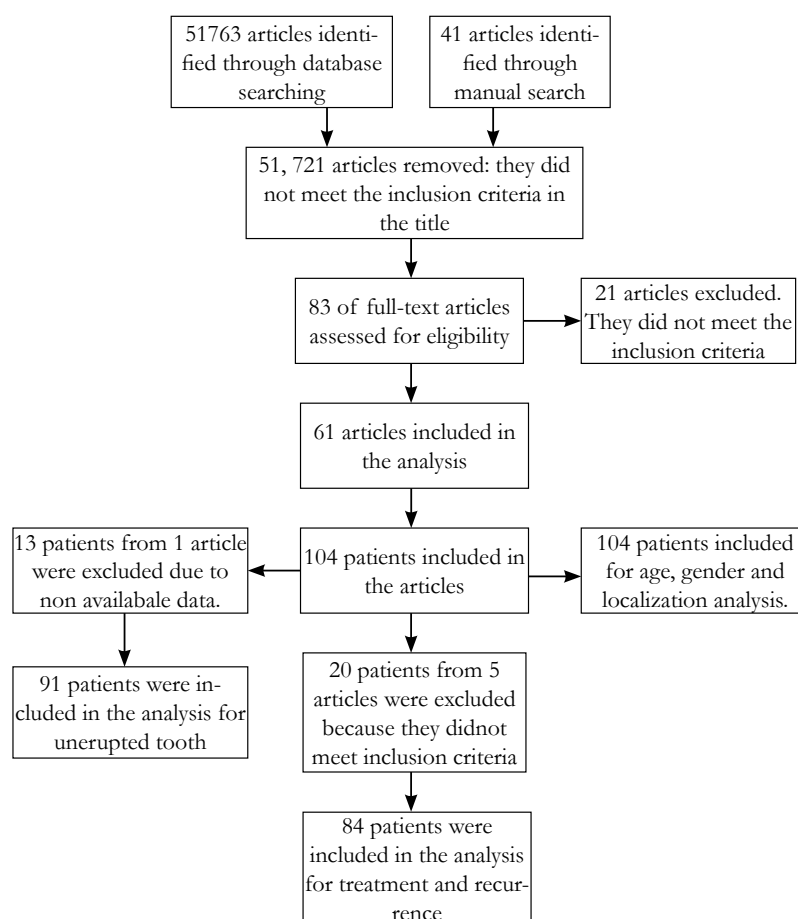
The initial search retrieved 51763 articles identified through database searching, and 41 articles identified through manual search. After examination of the title and the abstract, 83 articles were retrieved for further examination. Sixty-two articles were included, thus obtaining a total of 104 patients (Table 1).

For the analysis of the correlation with an unerupted tooth, 13 patients from one article were excluded because no information was available, resulting in a total of 91 patients.

For the analysis of the tumor localization, all of the 104 patients were included.

For the treatment and recurrence analysis, 20 patients from 5 articles were excluded due to a lack of information about follow-up, diagnosis of being central, or information about recurrence.

Figure 1. Data Search Flow Chart.



Eighty-Four patients were included in the analysis.

The age of the patients ranged from 3 to 74 years old, with a mean age of 31.11 years (n=104), with a gender relation of 0.79:1 (46 males – 58 females).

The unerupted tooth relation for the 91 patients was 28.57% (26 patients).

The most frequent localization for all patients was the mandible in 52% (54 patients), and the maxilla 48% (50 patients).

The surgery type of the 84 patients included were as shown in the Table 2.

The recurrence of the 84 patients was 4.76% (4 patients) Table 3.

Discussion

Unerupted Tooth Relation

Cicconetti and colleagues made a literature survey which included 64 patients, from which 25% of them were associated with the

Table 1. Summary of Patients and Articles Included in the Systematic Review.

Reference	Age	Gender	Unerupted tooth relation	Localiza-tion	Treatment	Follow-up	Recurrence
Wesley RK, et al., [6]	11 y	M	Yes	Mandible	Enucleation/Curettage	24 Mo	No
Heimdal A, et al., [7]	20 y	F	No	Mandible	Enucleation	108 Mo	Yes
Schofield ID [8]	11 Y	F	No	Mandible	Enucleation	N / A	N / A
Dahl EC, et al., [9]	27 Y 80 Y	M M	No Yes	Maxilla Mandible	Enucleation No treatment	60 Mo 48 Mo	No N / A
Dunlap CL, Barker BF [10]	33 Y 35 Y	F F	No No	Maxilla Maxilla	Curettage Curettage	120 Mo 108 Mo	No No
Janssen JH, Blijdorp PA [11]	44 Y	M	No	Maxilla	Enucleation	6 Mo	No
Sepheriadou- Mavropoulou TH, et al., [12]	16 Y	F	No	Maxilla	Enucleation	30 Mo	No
Watt-Smith SR, et al., [13]	63 Y	F	No	Mandible	Enucleation	9 Mo	No
Handlers JP, et al., [14]	14 Y 72 Y 28 Y 45 Y 41 Y 23 Y 26 Y 28 Y 30 Y 22 Y 42 Y 61 Y 14 Y 46 Y 24 Y 66 Y 39 Y 52 Y 34 Y	F M M F F F F F F F F F F F M F F F M	N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A	Maxilla Maxilla Maxilla Maxilla Maxilla Maxilla Maxilla Maxilla Maxilla Maxilla Maxilla Maxilla Maxilla Maxilla Maxilla Mandible Maxilla Mandible Mandible	Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation Enucleation	3 Mo 12 Mo 12 Mo 12 Mo 36 Mo N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A	No No No No No N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A N / A
Gunhan O, et al., [15]	55 Y	F	N / A	Maxilla	Enucleation	60 Mo	No
Thomopoulos G, et al., [16]	53 Y	M	No	Maxilla	Enucleation	6 Mo+	No+
Chen CH, Huang YL [17]	32 Y	M	N/A	Mandible	Enucleation	36 Mo	No
Allen CM, et al., [18]	66 Y 14 Y 30 Y	F F F	No N / A No	Maxilla Maxilla Maxilla	Curettage Curettage Curettage	6 Mo 48 Mo 14 Mo	No No Yes
Huey MW, et al., [19]	42 Y	M	No	Maxilla	Enucleation	4 Mo	No
Mosqueda-Taylor, et al., [20]	17 Y	F	No	Mandible	Curettage	72 Mo	No
Calvo N, et al., [21]	61 Y	M	No	Maxilla	Enucleation	36 Mo	No
Daniels JSM [22]	30 Y	F	Yes	Mandible	Enucleation	60 Mo	No
Covani U, et al., [23]	26 Y	F	No	Maxilla	Enucleation	72 Mo	No
Cicconetti A, et al., [24]	17 Y	F	Si	Maxilla	Enucleation	24 Mo	No
Cercadillo-Ibarguren I, et al., [25]	38 Y	F	Si	Mandible	Enucleation / Curettage	18 Mo	No

Silva CO, et al., [26]	45 Y	F	No	Mandible	Enucleation	12 Mo	No
Chuang GP, et al., [27]	20 Y	F	No	Mandible	Enucleation / Curettage	8 Mo	No
Araki M, et al., [28]	40 Y	M	Yes	Mandible	Enucleation	84 Mo	No
Daskala I, et al., [29]	71 Y	M	No	Mandible	Enucleation	1 Mo	No
Kishino M, et al., [30]	29 Y	M	No	Maxilla	Enucleation	14 Mo	No
Brazao-Silva Mt, et al., [31]	28 Y	M	No	Mandible	Curettage	156 Mo	No
Melo AR, et al., [32]	16 Y	M	No	Maxilla	Enucleation	60 Mo	Yes
de-Matos FR, et al., [33]	36 Y	F	No	Maxilla	Enucleation	11 Mo	No
Mosqueda-Taylor A, et al., [34]	39 Y	M	No	Maxilla	Enucleation	3 Mo	No
	48 Y	M	No	Maxilla	Enucleation	12 Mo	No
	42 Y	F	No	Mandible	Enucleation	48 Mo	No
	17 Y	F	No	Mandible	Enucleation	156 Mo	No
	25 Y	F	No	Maxilla	Enucleation	6 Mo	No
	14 Y	M	No	Mandible	Enucleation	16 Mo	No
	14 Y	M	No	Mandible	Enucleation	24 Mo	No
	49 Y	M	No	Maxilla	Enucleation	6 Mo	No
	31 Y	F	No	Maxilla	Enucleation	48 Mo	No
	28 Y	M	No	Mandible	Enucleation	6 Mo	No
	24 Y	M	No	Maxilla	Enucleation	N / A	N / A
	39 Y	F	No	Maxilla	Enucleation	N / A	N / A
	51 Y	F	No	Mandible	Enucleation	N / A	N / A
	25 Y	F	No	Maxilla	Enucleation	N / A	N / A
Nah KS [35]	17 Y	M	No	Mandible	Enucleation	24 Mo	No
Kiklander S, et al., [36]	9 Y	M	Yes	Mandible	Enucleation	12 Mo	No
Bologna-Molina R, et al., [37]	14 Y	M	No	Mandible	Enucleation / Curettage	24 M	No
Ahmadi SK, et al., [38]	16 Y	M	No	Mandible	Enucleation	36 Mo	No
Hrichi R, et al., [39]	17 Y	M	Yes	Mandible	Enucleation/Curettage	60 Mo	No
	30 Y	M	Yes	Mandible	Enucleation/Curettage	60 Mo	No
	18 Y	F	Yes	Maxilla	Enucleation / Curettage	60 Mo	No
	38 Y	F	Yes	Mandible	Enucleation / Curettage	60 Mo	No
	12 Y	M	Yes	Mandible	Enucleation / Curettage	60 Mo	No
	16 Y	M	Yes	Mandible	Enucleation / Curettage	60 Mo	No
	11 Y	F	Yes	Maxilla	Enucleation / Curettage	60 Mo	No
	17 Y	M	Yes	Mandible	Enucleation / Curettage	60 Mo	No
Hara M, et al., [40]	24 Y	F	No	Maxilla	Enucleation	12 Mo	No
	12 Y	F	Yes	Mandible	Enucleation	28 Mo	No
Chhabra V, Chhabra B. [41]	16 Y	F	No	Mandible	Enucleation	12 Mo	No
Kimura T, et al., [42]	74 Y	F	No	Mandible	Marginal Mandibulectomy	48 Mo	No
Takeota T, et al., [43]	29 Y	M	Yes	Mandible	Enucleation	24 Mo	No
Pushpanshu J, et al., [4]	18 Y	F	No	Mandible	Curettage	30 Mo	No
Iordanidis S, et al., [44]	39 Y	F	Yes	Mandible	Enucleation / Curettage	12 Mo	No
Salgado H, et al., [2]	24 Y	M	No	Maxilla	Enucleation	18 Mo	No
Sachdeva SK, et al., [45]	18 Y	F	Yes	Mandible	Enucleation	12 Mo	Yes
Gopinathan PA, et al., [46]	51 Y	M	No	Mandible	Enucleation	36 Mo	No
Batson JP, et al., [47]	18 Y	M	No	Mandible	Enucleation / Curettage	18 Mo	No
Schussel JL, et al., [48]	54 Y	F	No	Maxilla	Enucleation	6 Mo	No
Venugopal S, et al., [49]	49 Y	M	No	Mandible	Enucleation	12 Mo	No
Chrcanovic BR, et al., [50]	7 Y	M	Yes	Mandible	Enucleation	9 Mo	No
Liu X, et al., [51]	41 Y	M	No	Maxilla	Enucleation	13 Mo	No
Thankappan P, et al., [52]	10 Y	M	Yes	Mandible	Enucleation	12 Mo	No
Soolari A, Khan A [5]	53 Y	M	No	Mandible	Enucleation / Curettage	12 Mo+	No+
Hedge U, Rekha M [53]	35 Y	M	No	Maxilla	Enucleation	24 Mo	No
Salehinejad J, et al., [54]	10 Y	M	N / A	Mandible	Right Hemimandibulectomy	14 Mo	No
Shiraishi T, et al., [55]	3 Y	F	Yes	Mandible	Left Segmentary Mandibulectomy	24 Mo	No
Anbiaee N, et al., [56]	4 Y	M	Yes	Mandible	Right Segmentary Mandibular Resection	6 Mo	No
Monteiro LS, et al., [57]	74 Y	F	No	Mandible	Er:YAG (2940nm)/enucleation/Curettage	48 Mo	No

El-Harti K, et al., [58]	15 Y	F	Yes	Maxilla	Enucleation	12 Mo	No
Santoro A, et al., [59]	12 Y	M	Yes	Mandible	Enucleation	12 Mo	No
Prakash U, et al., [60]	45 Y	F	No	Mandible	Enucleation/Curettage	6 Mo	No
Nakamura Y, et al., [61]	6 Y	F	Yes	Maxilla	Enucleation	96 Mo	No
Pippi R, et al., [62]	16 Y	F	Yes	Mandible	Enucleation/Curettage	60 Mo	No
García-Cano E, et al., [64]	15 Y	F	Yes	Mandible	Enucleation/Curettage/Liquid Nitrogen	12 Mo	No

* Y: Years; M: Male; F: Female; N/A: Non Available; Mo: Months; +: Information asked through e-mail to author.

Table 2. Summary Data of the Patients Included in the Systematic Review.

Total number of patients	104
Average of patients	31.11 years
Gender relation (Male:Female)	0.79 : 1
Unrupted tooth relation (n=91)	26 (28.57%)
Localization (n=104):	
Mandible	54 (52%)
Maxilla	50(48%)
Surgery type (n= 84):	
Enucleation	53 (63.09%)
Curettage	8 (9.52%)
Enucleation and Curettage	17 (20.23%)
Resection procedure	4 (4.76%)
Er:YAG 2940nm and Enucleation and Curettage	1 (1.19%)
Enucleation and Curettage and liquid nitrogen	1 (1.19%)
Mean follow –up time (n=84)	34.17 Mo 2.84 years
Recurrence (n=84)	4 (4.76%)

* n = number of patients

Table 3. Procedures Perform in the Recurrence Cases.

Recurrence (n=84)	4 (4.76%)
Type of surgery (4):	
Enucleation	3 (66.66%)
Curettage	1 (33.33%)

n = number of patients

presence of an unerupted tooth [24]. However, in our systematic review, we included 91 patients, and only 26 of them correlated with an unerupted tooth (28.57%), while the rest did not (65 patients, 71.43%). Therefore, its presence is not necessary to make a differential radiological diagnosis.

Localization

The localization of these tumors varies in the literature. The larger the number of patients, the more the percentage changes. Handlers described 39 cases of COF, reporting 56% occurring in the maxilla and 44% in the mandible [14]. Ramer showed an incidence ratio in mandible and maxilla of 1:1 (34:34 patients) [64]. Meanwhile, in the most recent review by Cicconetti, they reported 52 patients, founding 61.53% in the mandible and 38.47% in the maxilla [24].

Nonetheless in our systematic review, out of 104 patients, 54 (52%) were located in the mandible, while the rest 50 (48%) were located in the maxilla.

Treatment

The first authors who reported curettage as a first-line treatment were Dunlap and Barker, who presented two cases of maxillary odontogenic fibroma treated by this method with a follow-up of 9 to 10 years without evidence of recurrence [10]. Since these lesions readily separate from their bony crypt and show no evidence of bony infiltration [5], generally the treatment described in the literature has been conservative.

However, there are special cases in which resections are necessary due to the extension of the tumor that compromises the bony structures, thus not allowing a conservative treatment; such

treatments include mandibulectomy, hemimandibulectomy, resection and reconstruction with bone grafts or free flaps [42, 54-56]. As the technology and new treatments are discovered, various methods have been implemented for treating this tumor and to lower its recurrence, such as laser treatment described by Monteiro LS [57], or the use of liquid nitrogen as described by Garcia-Cano et al., [63].

In this review 63% (53 patients) of the patients were treated only by enucleation, followed by enucleation and curettage with 20.2% (17 patients), and only curettage in 9.5% (8 patients). Only 7.14% were treated by other methods due to the tumor extension.

Recurrence

The recurrence reports vary in the literature. The most recent review by Garcia-Cano et al., of 27 cases (23 treated by enucleation and 4 by a resection procedure) with a mean follow up of 20.73 months, showed no recurrence whatsoever [63]. Ramer and colleagues reported only 5 cases of recurrence. However, only 39 cases out of 68 were followed up [64].

In this systematic review out of 84 patients, 4 (4.7%) presented recurrence. From these, 3 cases were treated by enucleation and one by curettage (Table 3).

Heimdal and colleagues explained that the recurrence presented with their patient may have resulted due to incomplete removal [7]. On the other hand, Allen did not describe a possible cause [18]. Melo described that mistakes made in the histological diagnosis of the tumor and an inadequate surgical technique are considered to be possible causes for recurrence; they also described that a probable explanation would be the maintenance of the impacted maxillary right canine and its periodontal ligament as a tumor reactivating factor [32]. Sachdeva SK, et al., described the most recent case for recurrence, mainly due to a probable incomplete removal of the tumor [45].

Conclusion

The COF is an extremely rare benign neoplasm found only in 0.1% of all odontogenic tumors.

The recurrence rate in these tumors is very low and mainly related to an incomplete resection. Therefore, conservative surgery must be done as a first-line treatment.

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Special Issue on

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