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

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 García-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.

Abstract

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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.

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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 51,763 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.
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 was 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

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

| Reference                          | Age | Gender | Unerupted tooth relation | Localisation | 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| M      | No                       | Maxilla      | Enucleation                 | 60 Mo     | No         |
| Dunlap CL, Barker BF [10]          | 33 Y| F      | No                       | Maxilla      | Curettage                   | 120 Mo    | No         |
| Janssen JH, Blüdorp 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| F      | N / A                    | Maxilla      | Enucleation                 | 3 Mo      | No         |
| 72 Y                               | M    | N / A  | Maxilla                  | 12 Mo        | No                          |
| 28 Y                               | M    | N / A  | Maxilla                  | 12 Mo        | No                          |
| 45 Y                               | F    | N / A  | Maxilla                  | 12 Mo        | No                          |
| 41 Y                               | F    | N / A  | Maxilla                  | 36 Mo        | No                          |
| 23 Y                               | F    | N / A  | Maxilla                  | N / A        | N / A                       |
| 26 Y                               | F    | N / A  | Maxilla                  | N / A        | N / A                       |
| 28 Y                               | F    | N / A  | Maxilla                  | N / A        | N / A                       |
| 30 Y                               | F    | N / A  | Maxilla                  | N / A        | N / A                       |
| 22 Y                               | F    | N / A  | Maxilla                  | N / A        | N / A                       |
| 42 Y                               | F    | N / A  | Maxilla                  | N / A        | N / A                       |
| 61 Y                               | F    | N / A  | Maxilla                  | N / A        | N / A                       |
| 14 Y                               | F    | N / A  | Maxilla                  | N / A        | N / A                       |
| 46 Y                               | F    | N / A  | Maxilla                  | N / A        | N / A                       |
| 24 Y                               | M    | N / A  | Maxilla                  | N / A        | N / A                       |
| 66 Y                               | F    | N / A  | Mandible                 | N / A        | N / A                       |
| 39 Y                               | F    | N / A  | Maxilla                  | N / A        | N / A                       |
| 52 Y                               | F    | N / A  | Mandible                 | N / A        | N / A                       |
| 34 Y                               | M    | N / A  | Mandible                 | 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| F      | No                       | Maxilla      | Curettage                   | 6 Mo      | No         |
| 14 Y                               | F    | N / A  | Maxilla                  | 48 Mo        | No                          |
| 30 Y                               | F    | No     | Maxilla                  | 14 Mo        | 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]         | 38 Y| F      | Si                       | Mandible     | Enucleation / Curettage     | 18 Mo     | No         |
| Cercadillo-Ibarquen I, et al., [25] |      |        |                          |              |                             |           |            |
| Author(s) | Age | Sex | Site | Treatment | Follow-Up | Recurrence |
|-----------|-----|-----|------|-----------|-----------|------------|
| Lombardi-Azócar JJ, García-Cano E, Malagón-Hidalgo HO, González-Magaña F, Montiel-Janquín AJ, et al., (2017) | 45 Y | F | 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 |
| Ahmad SJ, et al., [38] | 16 Y | M | No | Mandible | Enucleation | 36 Mo | No |
| Hrchi 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 |
| | 29 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 |
| Takeo 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 |
| Jordanidis 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 |
| Gopinath 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 |
| Schueller 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 |
| Chreanovic 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 |
| Thananko 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 |
| Salehnejad J, et al., [54] | 10 Y | M | N / A | Mandible | Right Hemimandibulectomy | 14 Mo | No |
| Shirashi T, et al., [55] | 3 Y | F | Yes | Mandible | Left Segmentary Mandibulectomy | 24 Mo | No |
| Anbiace N, et al., [56] | 4 Y | M | Yes | Mandible | Right Segmental Mandibular Resection | 6 Mo | No |
| Monteiro LS, et al., [57] | 74 Y | F | No | Mandible | Er:YAG (2940mm)/enucleation / Curettage | 48 Mo | No |
Lombardi-Azócar JJ, García-Cano E, Malagón-Hidalgo HO, González-Magaña F, Montiel-Jarquín AJ, et al., (2017) Central Odontogenic Fibroma, its Treatment and its Recurrence: A Systematic Review. Int J Dentistry Oral Sci. S10:001, 1-7.

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. 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 cases are listed in Table 3.

**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 |
| Unerupted 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 2940mm 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

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