Craniofac Res. 2023; 2(1):22-26. Received: 2023-01-23 / Accepted: 2023-03-03

Synchronus central cemento-ossifying fibroma and supernumerary tooth in the maxilla. Case report and review of the literature

Presentación simultánea fibroma cemento-osificante central y diente supernumerario en el maxilar. Reporte de un caso y revisión de la literatura

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ABSTRACT: Cemento ossifying fibroma (COF) is a benign fibro-osseous lesion, which occurs mainly in women, between the 3rd and 4th decade, affecting the posterior mandibular region. In this article, an atypical presentation of COF is presented synchronously with a supernumerary tooth included in the maxilla, and a review of the literature is carried out. A 17-year-old male patient, with a radiographic finding of a supernumerary tooth included and an asymptomatic increase in vestibular volume in the maxilla, which causes dental malposition. Extraction of the supernumerary tooth and excisional biopsy of the vestibular lesion were performed under local anesthesia. The histopathological study reports a fibro cellular tissue with calcifications similar to bone trabeculae, compatible with Central Ossifying Cement Fibroma. The etiology of COF is uncertain and its mechanism is still unknown, but it could have a genetic or traumatic component as a triggering factor. The literature shows limited reports presented in the maxilla, its characteristics are summarized in this article. A presentation of OFC simultaneously with supernumerary teeth has not been reported, however, it has been presented in conjunction with other injuries or multiple COF.

KEY WORDS: Fibroma, ossifying; maxilla; tooth supernumerary.

INTRODUCCIÓN

The cemento-ossifying fibroma (COF) is a fibro osseus lesion with the normal bone architecture replaced by a variable amount of fibrous connective tissue, osseus tissue and/or cement. Its etiology is not clear, but there have been studies for possible COF triggering agents, such as trauma and genetic mutation (Nilesh *et al*., 2020).

It mainly affects the craniofacial region, being that 70% of the cases, showing itself mostly in the jaw, in the bicuspid and molar region. It's most frequently found in adults, between their third and fourth decade, predominantly within females (Nilesh *et al*., 2020; Ram *et al*., 2012; Titinchi *et al*., 2016).

The COF show a variable behavior, such can range from a slow growth to an aggressive local destruction. It usually presents itself as an asymptomatic osseus expansion, in the literature there have been reported cases of painful evolution, paresthesia, sinus obstruction, facial asymmetry, proptosis and intracranial complications (Titinchi *et al*., 2016; Nilesh *et al*., 2020; Ram *et al*., 2012).

X-ray features vary accord to the evolution time, showing a radiolucent image in early stages, a mixed one in intermediate stages, and a radiopaque one in late stages. In general, it's observed as a mixed density, well delimited unilocular lesion, with a sclerotic border that can cause cortical expansion. (Nilesh *et al*., 2020; Ram *et al*., 2012; Titinchi *et al*., 2016).

The definitive diagnosis must be drawn from a histopathological study, in correlation with the clinical and radiological features and characteristics that the case may present. The chosen treatment for a COF is a surgical excision (Nilesh *et al*., 2020; Titinchi *et al*., 2016).

In the other hand, supernumerary teeth are pieces that exceed the human's dental formula number. Those are usually asymptomatic and detected in routinary x-rays. In literature there have been reported signs of eruption failure, displacement, root resorption and mispositioning of adjacent pieces, cysts formation, cortical perforation, dilacerations and malocclusion of the teeth (Mossaz *et al*., 2014; Suljkanovic *et al*., 2021).

The present article displays a clinical case about a patient that presents an atypical central cemento-osifying fibroma in a synchronic form with a supernumerary tooth located in the maxilla. A reviewing of literature is carried out, that includes COF cases with atypical presences in the maxilla. Finally, the surgical procedure for its solving is descripted, and the signs and possible etiologies of the COF are discussed.

CASE REPORT

A 17-year-old male patient shows up at the Pontificia Universidad Católica de Chile's dental clinic. Without relevant morbid background. He's been referred from the orthodontic unit, due to an x-ray finding of a supernumerary tooth located in the maxilla.

At the extraoral clinical examination there were no findings of relevance, nevertheless, intraorally an increase of volume is observed in the buccal surface related to the upper left incisor, with a dimension of approximately 0.8 cm of diameter, single, hard consistency, not adhered to soft tissues, without coloration changes, of defined limits and painless. The upper left incisor was in vestibule version without mobility (Fig. 1a). Without relevant discoveries in the palate.

In the computed tomography, a hypodense area is observed, with calcifications in its interior, producing expansion and thinning of the buccal cortical, related to the upper left incisor. The supernumerary tooth was included in the palate between the upper right incisor and canine, without damage towards adjacent structures (Fig. 1b).

Under local anesthesia and the usage of a surgical operating field, the excisional biopsy of the lesion took place along with the extraction of the supernumerary tooth and proceeded to be sent to a histopathological study (Fig. 1c). The patient recovery proceeded without postoperative complications.

The histopathological study resulted in a well delimited tumor, formed by fibro cellular tissue with little amount of blood vessels of small caliber. The presence of round shaped calcifications stands out, among others that are like small trabeculae, made from osteoid tissue, which makes it compatible with the central cement-ossifying fibroma (Fig. 1d).

DISCUSSION

This lesion's etiology is uncertain, and its mechanism is still unknown. It is believed that the trauma could result in inflammation in the periodontalligament and act as a trigger for its development (Nilesh *et al*., 2020; Titinchi *et al*., 2016; Bala *et al*., 2017). There's also been claims of a genetic origin taking part, given that there have been observed molecular changes such as translocations and gene deletions (Nilesh *et al*., 2020, Alawi, 2002).

The COF is an uncommon tumor lesion, mostly found located in the jaw and the premolar and molar region (Nilesh *et al*., 2020, Ram *et al*., 2012, Titinchi *et al*., 2016). Titinchi and Chang *et al*. report a 74,6% and 93% of cases that display presence in the jaw, with a 55,5% and 89% located at the posterior region, respectively (Titinchi *et al*., 2016, Chang *et al*., 2008). In the present case, there's a report of infrequent COF presence, located in the anterior region of the maxilla, a summary table reporting the COF cases with presence in the anterior region of the maxilla and its characteristics is presented ahead (Table I).

According to the literature, the COF usually appears in women in their third and fourth decade (You Qiu et. Al 2021, Bilal et. Al 2021). Nevertheless, there have been few reports of COF presence in male patients in their 20's; Akcam et. al 2012 and Sudarshan et. al 2016 studies describe a

COF case located in the jaw of a 20 years-old patient. The age range for the cases of COF presence in the anterior region of the maxilla is set at 11 to 55 years of age, with an average of 27,3. In the case presented, the injury was observed in an uncommon population, regarding age, as well as gender, being the example, a 17 years-old male patient.

This benign tumor is characterized for being of slow development and expansive, presenting itself as an asymptomatic volume increase, that doesn't generate alterations in the adjacent tissues in early stages. As it sizes increases, alterations such as facial asymmetry, paresthesia, pain, displacement, and tooth mobility start to appear (Nilesh *et al*., 2020, Chang *et al*., 2008).

In this review, the reported cases in the upper jaw, provided a secondary alteration, being the most prevalent, the expansion of bone tables and tooth displacement and even producing facial asymmetry in the cases presenting a bigger size. Even though tooth displacing occurred, there wasn't any tooth resorption observed in the lesion-affected pieces (Pérez García *et al*., 2004, Sarwar *et al*., 2008, Cagri *et al*., 2009, Shailesh *et al*., 2011, Akcam *et al*., 2012, Prakash *et al*., 2013). In this case report, the patient presented a slight intraoral volume increase, which caused alteration to the adjacent tissues that resulted in vestibule version of the upper left incisor, without presence of tooth resorption to the adjacent pieces.

In X-ray matters, the COF has been described to appear with a variety of forms. Research describes the image of COF as a radiolucent and well-defined area (Hombal et. al 2007, Sarwar et. al 2008). As the lesion progresses, the amount of radiopaque material increases, giving it a mixed aspect at intermediate stages and a fully radiopaque look in

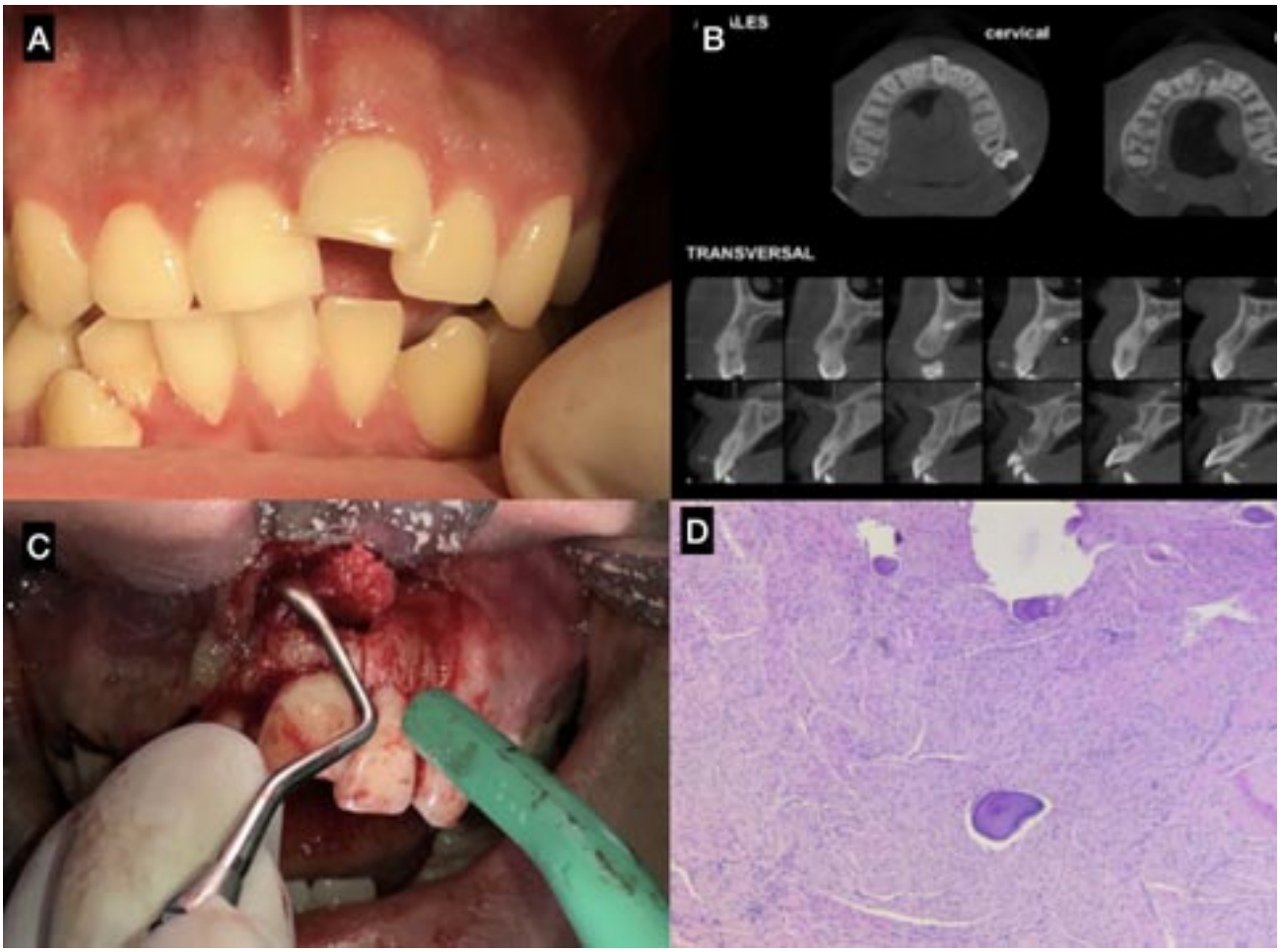

Fig. 1. A) Buccal volume increase related to tooth 2.1, B) Cone beam computed tomography showing the area related to central incisor with the tumor, C) Surgical approach to remove the tumor, D) Histopathological study (HE). Fibro cellular bone tissue and calcifications are observed.

Table I. Articles showing reports related to the disease.

Title/Author/Year	Age/ SEX	Location/Evolution/Size	XR imaging	Secondary alteration	Histopathology	Treatment	Relapse
Ossifying fibroma of the upper jaw: report of a case and review of the literature. Pérez García *et al*. (2004).	22, male.	Anterior maxilla, 3 months 2 x 2 cm.	Mixed density.	Bone table expansion, root divergence, DMA.	Fusiform cells, amorphous calcifications.	Surgical excision, extraction of associated teeth.	No relapse.
Cement-ossifying fibroma a rare case. Sarwar *et al*. (2008).	11, male.	Anterior maxilla, 6-8 months, 3 x 3 cm.	Well defined, mixed density.	Bone tables expansion, tooth displacement.	Pseudoepitheliomatous hyperplasia, cementum and osseus trabeculae rimmed by fibroblasts.	Surgical excision.	Not mentioned.
A Large Mass in the Maxilla: Clinical Features and Differential Diagnosis. Delilbasi *et al* .(2009).	21, male.	Posterior maxilla, 8 months, 7 x 5 x 4 cm.	Mixed density.	Facial asymmetry, perforation and bone table expansion, root divergence.	Fusiform fibroblast cells, cementum, and irregular bone structure.	Surgical excision, extraction of associated teeth.	No relapse after 2 years.
Ossifying fibroma of the jaws: Report of two cases and literature review - Gondivkar *et al*. (2011).	35, male.	Posterior palatal zone, maxilla, 5 years, 4 x 3 cm.	Well defined, radiopaque density.	Root divergence.	Fusiform fibroblast cells, bone spicules and osteoblasts.	Partial maxillectomy.	No relapse.
Synchronous ossifying fibromas of the jaws: a review. Akcam *et al*. (2012).	20, male.	Posterior maxilla, not mentioned, 7 x 4,5 cm.	Well defined, radiolucent density.	Facial asymmetry, bone table expansion, tooth displacement.	Stromal fibroblast and calcifications.	Surgical excision and curettage, extraction of associated teeth.	No relapse after 8 months.
Cementifying fibroma. Mohan *et al*. (2013).	55, male.	Posterior maxilla, 1 year, 5 x 6 cm.	Well defined, mixed density.	Bone tables expansion.	Disorganized collagen, calcification deposits in fibrous stroma, irregular osseus trabeculae.	Surgical excision.	No relapse after 6 months.

late stages. From this, a sort of differential diagnosis can come up from its initial, intermediate, and late stages, such as cement-osseous dysplasia, fibrous dysplasia and odontomas respectively (Nilesh *et al*., 2020, Chang *et al*., 2008). Based off the clinical and x-ray characteristics, the reported case is consistent with the most frequent COF description, presenting itself at an intermediate stage.

The most common surgical treatment, and the one performed in this case, is the enucleation and curetting of the lesion, which presents a recurrence rate between 0 – 28% (Ram *et al*., 2012, Titinchi *et al*., 2016, Chang *et al*., 2008). In this review, the treatment of the cases was through surgical excision, with variations regarding the treatment's aggressiveness. Considering 6 to 24 months of follow up there were no relapses (Pérez García *et al*., 2004, Sarwar *et al*., 2008, Cagri *et al*., 2009, Shailesh *et al*., 2011, Akcam *et al*., 2012, Prakash *et al*., 2013). A 0% recurrence rate indicates an achieved success through surgical management of this type of lesions, but it is necessary to consider the absence or short follow-up time of reported cases, so enlarging the clinical and x-ray follow-up time is necessary. The reported lesion is incapsulated, achieving a complete and total removal, which decreases the recurrence rate. Yet despite the above, maintaining a track of the patient and evaluate the postoperative development are highly recommended. 12 months follow up of the patient took place, in which a successful result was observed, without relapsing signs, considering clinical and x-rays aspects.

In the present literature review, there were not any reports of synchronous presentation of COF with supernumerary teeth, nevertheless, the simultaneous occurrence of COF with other lesions such as giant-cell fibroma and other COF has been described (Kim *et al*., 2012, Akcam *et al*., 2012). In this case report, the patient shows no traumatic injury background, with a synchronous presentation of COF and a supernumerary tooth in the upper jaw. Considering these kinds of affections, an associated genetic factor could be considered. But to be certain, it is required an exhaustive examination of the patients that present this kind of diseases and carry out future investigations where the etiology and its associated factors can be analyzed in detail.

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RESUMEN: El fibroma cemento osificante (COF) es una lesión fibro ósea benigna, que se presenta principalmente en mujeres, entre la 3ra y 4ta década, afectando la región posterior mandibular. En este artículo se expone una presentación atípica de COF de forma sincrónica con un diente supernumerario incluido en el maxilar junto a una revisión de la literatura. Paciente de sexo masculino de 17 años, con un hallazgo radiográfico de un diente supernumerario incluido y un aumento de volumen vestibular asintomático en el maxilar, que provoca malposición dentaria. Se realizó la exodoncia del diente supernumerario y la biopsia excisional de la lesión vestibular bajo anestesia local. El estudio histopatológico informa un tejido fibrocelular con calcificaciones semejantes a trabéculas óseas, compatible con Fibroma Cemento Osificante Central. La etiología de COF es incierta y su mecanismo es aún desconocido, pero podría tener un componente genético o traumático como factor desencadenante. La literatura muestra limitados reportes presentados en el maxilar, sus características se encuentran resumidas en este artículo. No se ha reportado una presentación de COF de forma simultánea con dientes supernumerarios, sin embargo, sí se ha presentado en conjunto con otras lesiones o COF múltiples.

PALABRAS CLAVE: Fibroma osificante, maxilar, diente supernumerario.

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