Heliyon 7 (2021) e07594

Contents lists available at ScienceDirect

Heliyon

journal homepage: www.cell.com/heliyon

Case report

Large cemento-ossifying fibroma of the mandible involving the infratemporal and parapharyngeal spaces

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ARTICLE INFO

Keywords:
Cemento-ossifying fibroma
Mandible
Infratemporal
Parapharyngeal space
Computed tomography

ABSTRACT

Cemento-ossifying fibroma is a benign fibro-osseous lesion of the jaws. Cemento-ossifying fibroma develops from the periodontal ligament and contains multipotent stem cells that can form cementum, lamellar bone, and/or fibrous tissue. These tumours occur in the third and fourth decades of life with higher predilection of occurrence in the female population and seldom attain a large size. We report a rare case of cemento-ossifying fibroma in a 45-year-old man involving the body of the mandible and extending into the para-pharyngeal and infratemporal region. This article describes the clinical, radiographic, and histological features of a large cemento-ossifying fibroma of the mandible.

1. Introduction

Cemento-ossifying fibroma (COF) is a well-encapsulated neoplasm that comprises a variable mixture of fibrous and calcified tissues. The calcified tissue may resemble bone, cementum, or both [1, 2]. COF is considered a central neoplasm of the bone and periodontal ligament [3]. There is a considerable disagreement regarding the terminology, origin, and criteria for the diagnosis of COF [3]. Menzel first reported ossifying fibroma in a 35-year-old female patient in 1872 and coined the term COF [4]. Several synonyms for COF such as osteo-fibroma, fibro-osteoma, and benign fibro-osseous lesion of periodontal ligament origin have been used [5].

COF shares similar clinicoradiographic and pathological features with cementifying fibroma. However, the term COF is used in the contemporary literature for all lesions with similar features [5]. Certain cardinal features, such as aggressive local growth and high recurrence rate, favour a more neoplastic categorisation of COF [5]. We report an unusual case of a patient who presented with a COF in the mandibular body extending into the infratemporal and parapharyngeal spaces. To the best of our knowledge, COFs with such anatomical extension have been infrequently reported in the literature.

2. Case report

2.1. Clinical features

Written informed consent was obtained from the patient for publishing the photographs and radiographs of this case. A 45-year-old man presented to a private dental clinic in Bengaluru, India with a complaint of swelling on the lower aspect of the left side of the face since one year and reported a history of difficulty in swallowing since 4 months. As narrated by the patient, the swelling was initially small and gradually increased in size. The medical history was unremarkable.

Extraoral examination revealed a diffuse solitary swelling on the left lower one-third of the face (Figure 1a) extending from the left corner of the mouth to the ramus of the mandible anteroposteriorly and from 4 cm below the ala-tragal line to the submandibular region superoinferiorly. The overlying skin was normal. On palpation, the swelling was firm, non-tender, and fixed to the underlying tissues. Intraoral examination revealed a solitary oval swelling measuring approximately 4 × 3 cm that obliterated the buccal vestibule near the mandibular left second premolar and molars (Figure 1b). The colour of the overlying mucosa was normal. A 2-cm ulcer covered by white pseudo-membranous slough was evident over the swelling. The mandibular left first molar was mobile. The buccal and lingual cortical plates in the mandibular left second premolar and molar region were expanded.

2.2. Radiographic features

A panoramic radiograph revealed a well-demarcated multilocular lesion measuring approximately 8 × 4 cm with radiopaque specks and

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https://doi.org/10.1016/j.heliyon.2021.e07594
Received 22 February 2021; Received in revised form 20 April 2021; Accepted 13 July 2021

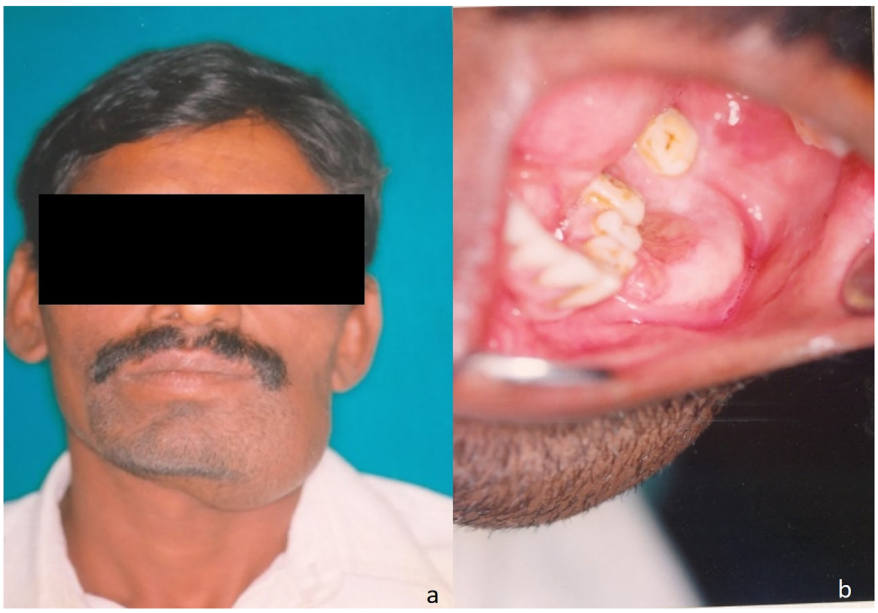

Figure 1. (a) Extraoral photograph of the patient showing the extension of the swelling. (b) Intraoral component of the swelling.

sclerotic borders. The lesion extended from the mandibular left canine region to the left condylar neck and coronoid process anteroposteriorly and from the alveolar ridge of the mandibular left premolar and molar region to the lower border of the mandible superoinferiorly (Figure 2).

Axial computed tomography (CT) showed a large, lobulated, mixed-density lesion causing buccolingual cortical expansion in the mandibular left body and ramus. Thinning of the buccal cortical plate and punctuate areas of calcification within the lesion were observed. The lingual expansion of the swelling had led to the displacement and compression of the oropharyngeal airway (Figure 3a).

Coronal CT showed an expansile, mixed-density, lobulated mass involving the mandibular left body and ramus extending into the left infratemporal region (Figure 3b and c). The results of blood investigations were within the reference range. Biopsy of the lesion was performed intraorally under local anaesthesia. Histopathological examination of the tissue showed a background of fibro-cellular stroma interspersed with formation of new bone in a trabecular pattern and spherules of cementum-like tissue (Figure 4a). Based on the clinicoradiographic and histopathological findings, the lesion was diagnosed as COF. Hemi-mandibulectomy was performed on the left side under general anaesthesia in a private hospital (Figure 4b). The surgical defect of the mandible was reconstructed using an acrylic plate at another centre because of logistical and financial reasons.

3. Discussion

OFs are rare and benign osseous neoplasms commonly occurring in the maxillofacial bones. The lesions are characterized by the presence of a rich fibrous connective tissue stroma with varying degrees of mineralization [6]. Based on the type of mineralized tissue, the lesions can be further histologically classified into ossifying and cementifying fibroma [7]. The term COF was coined for lesions containing both bone- and cementum-like tissues [6]. Further, based on the location, COF can be classified into central COF and peripheral COF [8]. Earlier classifications had categorized OF as an osteogenic lesion and COF as an odontogenic lesion. However, the World Health organization revisions of head and neck neoplasms included both OF and COF in osteogenic neoplasms [5, 9].

Considerable controversy exists regarding the origin of these tumours. Although they are listed as osseous lesions, the origin of these lesions in the jaws is attributed to the blast cells of the periodontal ligament [4]. Factors such as traumatic tooth extraction and localized infection followed by subsequent inflammation and fibrosis in the tooth apices can trigger the periodontal ligament cells [10, 11]. In this case, the tumour could have developed from the remnants of the periodontal ligament following the extraction of a second mandibular molar.

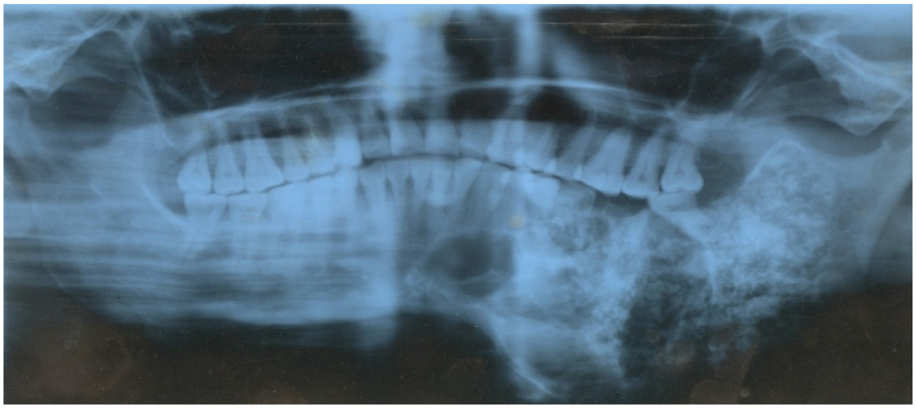

Figure 2. Panoramic radiograph showing extension of the lesion on the left side of the mandible. Mixed radiopaque and radiolucent areas extending from the mandibular symphysis region to the left side.

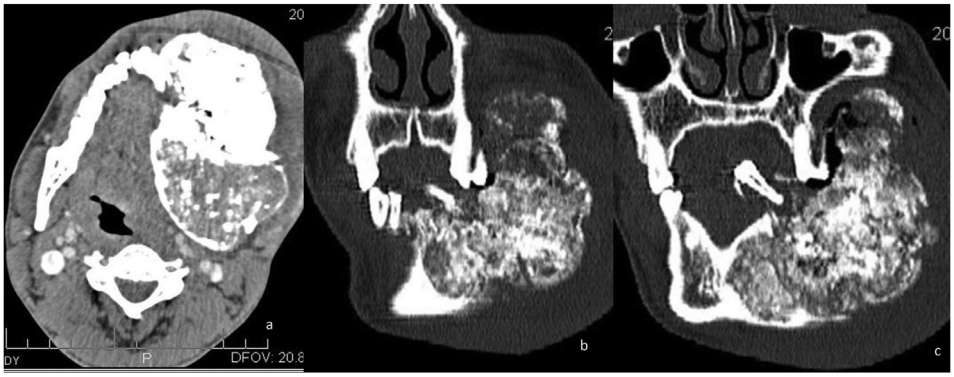

Figure 3. (a) Axial computed tomography showing an expansile lesion extending into the left pharyngeal and buccal space. (b) Coronal computed tomography showing the extension of the lesion in the mandibular anterior region. (c) Coronal computed tomography showing the extension of the lesion in the mandibular posterior region.

On appropriate stimulation, these cells can potentially differentiate into fibrous tissue, bone, and/or cementum, thereby justifying the presence of both types of calcified tissues in the lesions. Therefore, previous classifications that categorized these lesions as primary odontogenic neoplasms are more appropriate [2, 12]. Brademann et al. stated that the presence of ectopic periodontal tissue segregated from primitive mesenchymal cells could be attributed to the occurrence of COF elsewhere in the facial skeleton, other than in the jaws [13].

COF are usually slow-growing lesions that present during the third and fourth decades of life with a very high female predominance [5, 14, 15, 16]. However, the juvenile variant of the lesion is clinically aggressive and highly recurrent [8, 17]. The prevalence of COF is higher in the Caucasian population than in the African-American population [5, 7]. The lesion is more common in the mandible than in the maxilla, though the lesion may involve multiple quadrants [18]. Further, they are more common in the posterior part of the jaws than in the anterior region [6]. In our report, the patient was a 45-year-old man with COF in the posterior part of the mandible.

Clinically, COF is asymptomatic in the early stages. Swelling and jaw deformity or loosening of the teeth might be the early symptoms [19]. Root resorption is noted in approximately 12.7% cases [8, 14]. Mandibular lesions often lead to expansion of the bone cortices and downward displacement of the lower border. Involvement of the maxillary sinus is noted in 90% maxillary lesions [9]. In our report, the patient exhibited tooth displacement and significant cortical expansion.

Extension to the nasal septum, orbital floor, and infraorbital foramen might be evident in large lesions [8]. COF is rarely associated with destruction of extraosseous soft tissue components [20]. In our report, the patient presented with an unusually large COF extending to the infratemporal region and pharyngeal soft tissue space. The pharyngeal extension caused dysphagia in the patient, which is an uncommon finding of COF.

Radiographically, COF presents as a well-circumscribed, unilocular or multilocular, mixed-density lesion [8]. The radiographic findings vary according to the maturity of the lesion, with an increase in radiopacity associated with an increase in maturity [14]. Shokri et al. showed that cone-beam computerized tomography is effective in determining the size and extent of the lesion and can aid in distinguishing the lesion from other similar lesions [21]. In our report, CT was performed to determine the extent of the lesion.

Radiological differential diagnosis of COF includes osseous lesions such as fibrous dysplasia (FD), cemento-osseous dysplasia, and odontogenic lesions including keratocystic odontogenic tumour (KOT), calcifying odontogenic cysts, and calcifying epithelial odontogenic tumour (CEOT) [1, 8, 22].

A significant diagnostic feature of COF is that it is a well-circumscribed lesion and exhibits a centrifugal pattern of growth. Therefore, the lesion expands equally in all directions and tends to present as a round tumour mass [15]. FD displays a ground-glass appearance and blends with the surrounding bone, KOT shows more anteroposterior growth unlike COF, and CEOT is generally associated with an unerupted or impacted tooth [8, 22].

Histologically, the presence of fibrous connective tissue with irregular deposits of eosinophilic bone and basophilic cementum is highly suggestive of COF. Further, significant immunopositivity to keratan sulphate aids in differentiating COF from FD [8].

The treatment of COF must be directed towards the complete removal of the tumour mass using enucleation or curettage techniques. Surgical resection of the jaw is advised only for large neoplasms. The recurrence rate of these lesions is less than 5% [23, 24]. In our case, surgical resection of the tumour with hemi-mandibulectomy was performed. Some surgeons perform enucleation for smaller COFs [25]. However, it is important to note that the recurrence rate of COF is 10–28% after enucleation and 5% after resection [15]. In conclusion, COF are

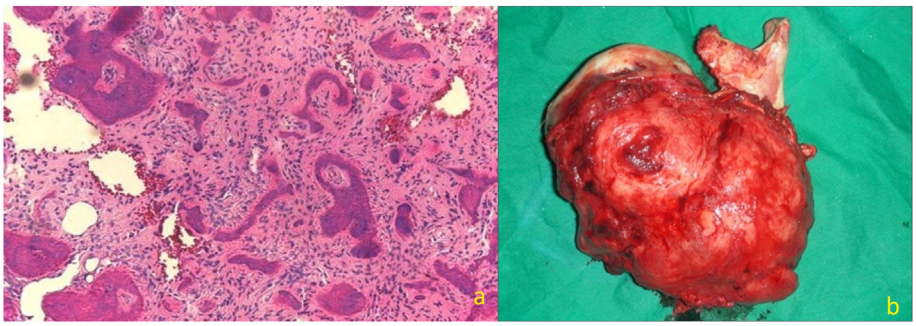

Figure 4. (a) Haematoxylin- and eosin-stained pictomicrograph showing areas of new bone formation in trabecular pattern intermingled with spherules of cementum-like tissue. (b) Gross specimen after hemi-mandibulectomy showing the extent of the lesion.

slow-growing benign lesions that resemble many other osseous neoplasms both clinically and radiographically. Though the lesions are well-circumscribed, they occasionally grow into large lesions extending into vital spaces. Thorough histological examination aids in distinguishing these lesions from OFs. In addition to conventional radiography, CT aids in determining the size and extension of larger lesions. Complete surgical excision and long follow-up are advised to prevent the recurrence of these lesions.

Declarations

Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

Funding statement

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Data availability statement

Data included in article/supplementary material/referenced in article.

Declaration of interests statement

The authors declare no conflict of interest.

Additional information

No additional information is available for this paper.

Acknowledgements

None.

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