DOI: 10.7860/JCDR/2024/68935.19746

Case Report

Dentistry Section

An Unusual Presentation of Cemento-ossifying Fibroma in the Anterior Maxilla: A Case Report

ANKITA CHAVHAN[1], SWATI PATIL[2], ALKA HANDE[3], ARCHANA SONONE[4], AAYUSHI PAKHALE[5]

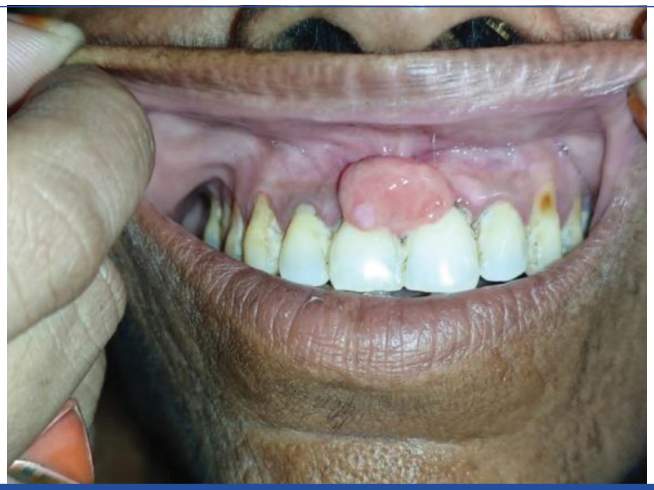
ABSTRACT

Cementifying Fibroma (CF) is a benign fibro-osseous lesion with sharply defined and well-confined edges. It is slow-growing and has a radiolucent peripheral component. It is mostly seen in the mandible in the 20-39 years age group with a female predilection. Hereby, the authors present a case report of a 60-year-old woman who had a painless growth in the upper front region of her jaw for two months and visited Outpatient Department (OPD). Extraoral examination revealed facial asymmetry due to swelling in the upper front region of the jaw. Intraoral examination showed round to oval swelling in the 11 and 21 regions. On radiographic examination, a round radiolucency in the 11 region was appreciated. After excision, the tissue was sent to the laboratory, and histopathologically the diagnosis was confirmed as peripheral CF. The patient was kept in a 12-month follow-up postoperatively, which reported no recurrence.

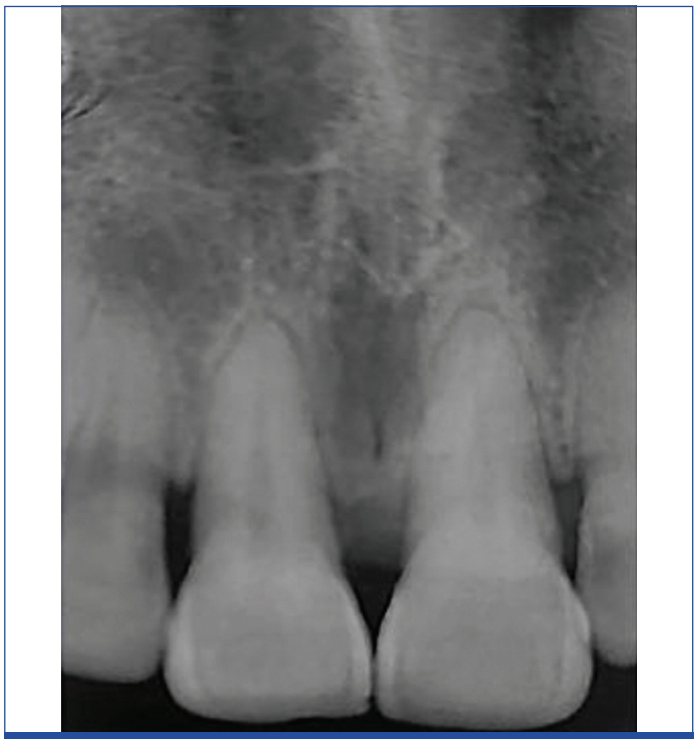
Keywords: Central cemento-ossifying fibroma, Fibro-osseous lesions, Ossifying fibroma

CASE REPORT

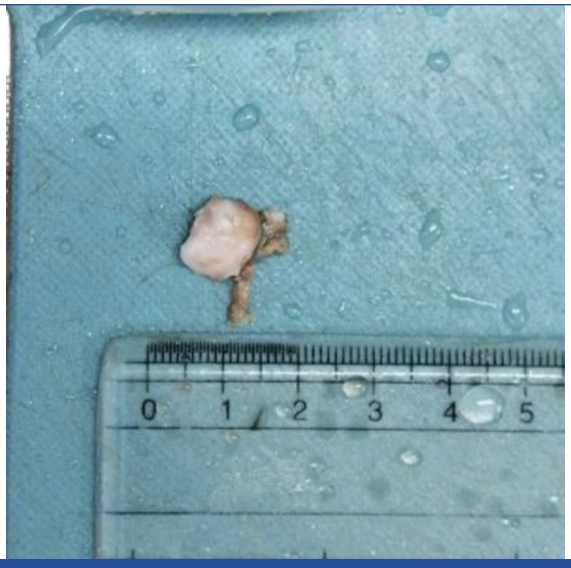
A 60-year-old female visited the Outpatient Department (OPD) of a dental hospital with a chief complaint of a painless swelling in her upper front region of the jaw for eight months. There was no history provided regarding any trauma or previous occurrence of the same condition. Following a complete evaluation of her history, nothing noteworthy was found in her dental, medical, or family history. On extraoral examination, slight facial asymmetry was observed in the upper anterior region due to a proliferative growth in the anterior maxilla. Upon palpation, the swelling was non tender and felt soft to firm in consistency. Temporomandibular joint movements were normal, with adequate mouth opening and incompetent lips. Intraoral examination revealed a small, well-defined, oval swelling over the upper anterior gingiva that extended from the maxillary central incisors region, measuring approximately 1.8×2.5 cm in size. On inspection, the lesion was smooth, pink in colour, the same as the surrounding normal mucosa, and covered by intact mucosa [Table/Fig-1]. Upon palpating the lesion, it was not tender and was soft to firm in consistency. Radiographic examination of the area of concern showed an ill-defined radiolucency which is round to oval in shape and approximately 1.3×1 cm in size. The lesion was well-demarcated radiographically and showed minute foci of calcifications, suggesting a Calcifying Odontogenic Fibroma (COF) in association with the right maxillary first incisor [Table/Fig-2]. After a complete blood examination, the lesion was excised under local anaesthesia, and the excised tissue was sent for histopathological examination {Haematoxylin and Eosin (H&E)}. In the laboratory, a single irregular whitish tissue piece measuring 1.4×0.9×0.6 cm was received and sent for further processing [Table/Fig-3]. Under microscopy, the scanner view showed hyperplastic parakeratinised stratified squamous epithelium and fibrous connective tissue stroma [Table/Fig-4]. At 10X magnification, long and broad rete ridges and bundles of collagen were appreciated, with closely packed growing fibroblasts in cellular fibroblastic tissue [Table/Fig-5]. In numerous areas of the connective tissue stroma, small foci of calcifications



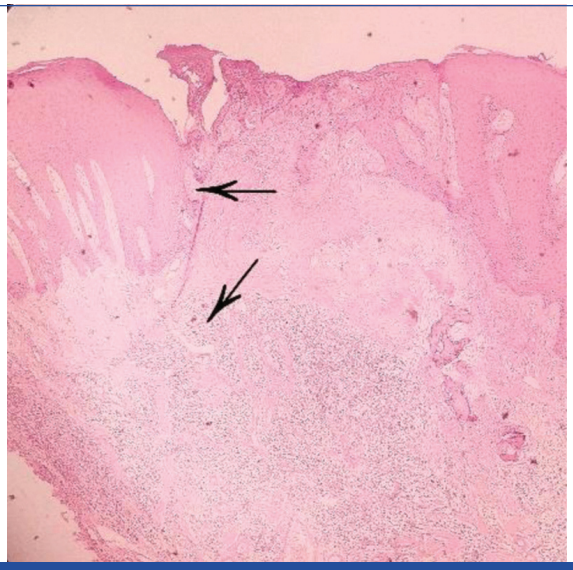
[Table/Fig-1]: Intraoral image showing a lesion on the upper lip.



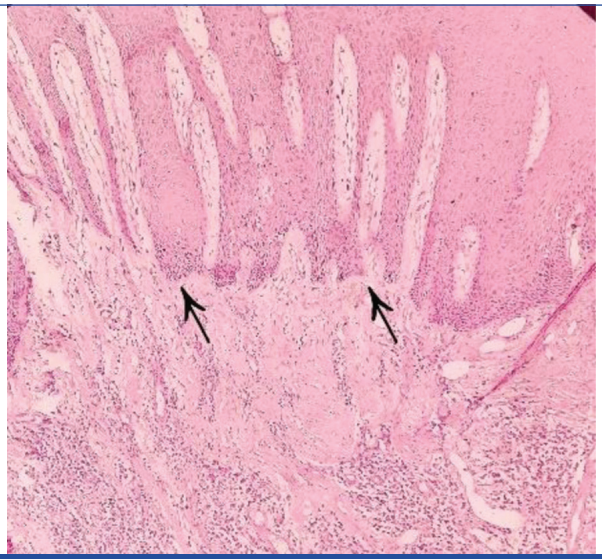
[Table/Fig-2]: Intraoral radiograph showing radiolucency in relation to maxillary central incisor.



[Table/Fig-3]: Excised tissue specimen measuring 1.4×0.9×0.6 cm.

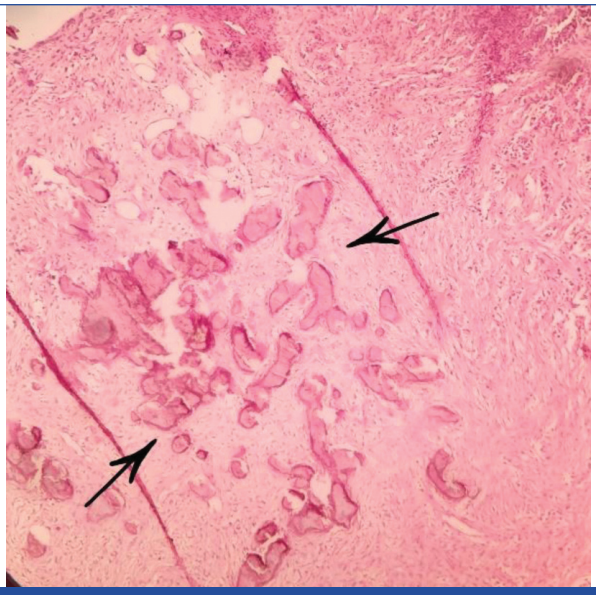


[Table/Fig-4]: Hyperplastic parakeratinised stratified squamous epithelium with connective tissue stroma (H&E, 4X).



[Table/Fig-5]: Epithelium shows long and broad rete ridges (H&E, 4X).

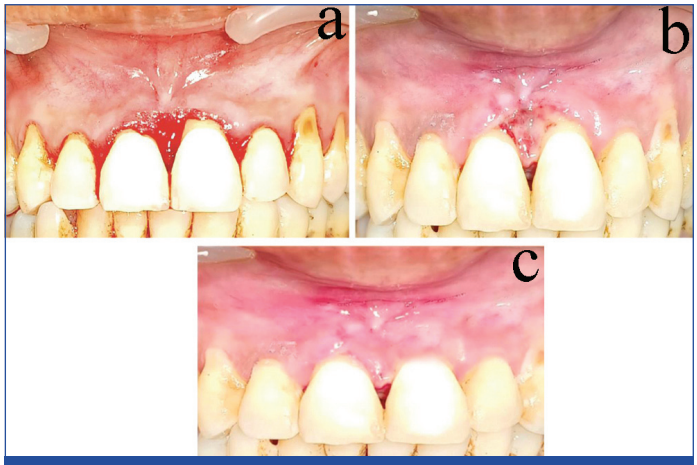
comprising bony trabeculae and cementum-like tissue with a few osteoid tissues were seen [Table/Fig-6]. These calcifications were in the form of single or multiple interconnecting trabeculae of bone or osteoid, suggestive of 'cemento-ossifying fibroma'. The lesion was initially considered to be a fibroma or traumatic fibroma due to the high concentration of collagen fibres in the



[Table/Fig-6]: Connective tissue stroma showing small foci of calcifications comprising bony trabeculae and cementum-like tissue with few osteoid tissues (H&E,10X).

connective tissue. However, upon histopathological examination, the presence of calcific foci confirmed the diagnosis as a COF.

After the final histopathological diagnosis, the patient was recalled after 15 days and one month for evaluation [Table/Fig-7a-c]. The healing after one month was good. The patient was then recalled at intervals of six months and 12 months, and she did not report any history of the recurrence of the same condition.



[Table/Fig-7a-c]: a) Postoperative view on day 1; b) Postoperative view after seven days showing satisfactory healing of the lesion; c) Postoperative view after 15 days showing complete satisfactory healing and improved aesthetics.

DISCUSSION

Fibro-osseous lesions of the skull are an uncommon category of diverse disorders that can develop either as a localised solitary process or as a symptom of a systemic skeletal disorder. This group includes benign mesenchymal tumours like CFs. CF, which develops from the mesodermal germ layer, is an uncommon, non odontogenic disease similar to the ossifying fibroma of the periodontal ligament [1]. The present locally damaging, non cancerous tumour affects the petromastoid region less frequently than the jaw bones, zygoma, paranasal air sinuses, and orbits. Since CF is a benign, slow-growing tumour, it is relatively less seen in younger children, and so this subtype is known as benign of a juvenile type [1]. Four different forms of cementum-containing lesions were identified by the World Health Organisation (WHO) in 1971: fibrous dysplasia, ossifying fibroma, CF, and cemento-ossifying fibroma [2]. The most widely accepted theory for its aetiopathogenesis holds that it is a tumour of periodontal origin. However, other theories have also been proposed. It is believed that it originates from multipotent mesenchymal blast cells,

Author/Country	Year	Age/gender	Site of lesion	Clinical features	Treatment modality
Qureshi MB et al., Karachi, Pakistan [12]	2021	37 years/female	Mandible	Bilateral swelling in right and left mandible	Complete surgical excision with piecemeal resection of both lesions
Divyadharshini V et al., Chennai [13]	2023	61 years/female	Anterior maxilla	Painless, progressive, slow-growing swelling on anterior maxilla	Excisional biopsy under local anesthesia. Extraction of teeth with poor prognosis
Deshapande A et al., Tamil Nadu [14]	2021	60 years/male	Anterior mandible	Single, non tender swelling in the anterior mandible	Complete excision under local anaesthesia followed by antibiotic therapy
Aburas S et al., Austria [15]	2020	19-year-old Caucasian woman	Left maxilla	Swelling from 23 to 26 region	Complete surgical excision under general anaesthesia
Christian INWS et al., Indonesia [16]	2023	39-year-old female	Left mandible	Mild tender, firm swelling in left mandible	Elective surgical procedure
Verma E et al., India [17]	2013	4 cases	Maxilla- 3 Mandible- 1	Painless swelling	Complete excision
Present study	2023	60-year-old woman	Anterior maxilla	Painless swelling in her upper front region of the jaw for 8 months	Complete excision

[Table/Fig-8]: Cases of COF reported worldwide [12-18].

which can produce cementum, alveolar bone, and fibrous tissue and are found in the periodontal membrane [3].

In the aetiopathological aspects, chromosome translocations have been observed in a small number of cemento-ossifying fibroma cases. Originating from mesenchymal blast cells found in the periodontal ligament, this entity has the capability to develop into fibrous tissue, cement, bone, or a blend of these components [4]. Due to microscopic commonalities with fibrous dysplasia and cemento-osseous dysplasia, researchers suggest that the lesion is an example of a localised dysplastic process in which bone metabolism has been altered [5]. Its origin may also be triggered by an irritant stimulus such as tooth extraction or, in some cases, trauma [6].

Early diagnosis is necessary for appropriate management to achieve local control and prevent spread; it can also be quickly obtained using minimally invasive Fine Needle Aspiration Cytology (FNAC) [7]. Histopathologically, the lesion is mostly made up of hyperplastic epithelium with numerous finely interlacing collagen fibres, along with significant areas of active fibroblasts interspersed in the connective tissue stroma. This connective tissue typically contains several tiny foci of atypical bone trabeculae [8]. This finding raises two areas of research that deserve more attention: first, whether instances of extremely aggressive CF affirm a more aggressive surgical approach, specifically when they recur; and second, whether the histologic manifestation may aid in predicting the nature of this tumour [9]. COF presents with different radiographic prototypes depending on the degree of mineralisation. Initially, it appears radiolucent; as it matures, calcific specks become more prominent until it eventually becomes a fully radiopaque mass [10]. Depending on the size and location of the specific lesion, conservative enucleation, curettage, or aggressive surgery has historically been used to treat CFs. They are distinguished by their effortless ability to absorb the environment. Mandibular central cemento-ossifying fibromas are easier to remove completely than maxillary central CFs. This difference might be due to the maxillary sinus's capacity for expansion and the differential in bone density between the mandible and maxilla. The prognosis is good, although recurrence is possible if, not treated with complete excision [11]. In the literature, various reported cases of CF are listed in [Table/Fig-8] [12-17].

CONCLUSION(S)

In summary, the case of cemento-ossifying fibroma in the maxillofacial region discussed in present report highlights its rarity within a timeframe, as well as the considerable challenges exposed in terms of differential diagnosis. Benign fibro-osseous lesions present a diagnostic conundrum due to their overlapping clinical, radiological, and histopathological features, which can perplex surgeons, pathologists, and radiologists. The divergence in the treatment plan and prognosis further complicates matters. Therefore, a comprehensive approach involving meticulous correlation of clinical, radiological, and histopathological findings is imperative for reaching an accurate diagnosis. Subsequently, a tailored treatment protocol can be devised to address the specific characteristics of each case effectively.

REFERENCES

[1] Venkataramana NK, Rao SAV, Kirshna Chaitanya N. Cementifying fibroma of the sphenoid wing in a child: A case report. J Pediatr Neurosci. 2021;16(1):49-54. Doi: 10.4103/jpn.JPN_162_11.

[2] Kaur T, Dhawan A, Bhullar RS, Gupta S. Cemento-ossifying fibroma in maxillofacial region: A series of 16 cases. J Maxillofac Oral Surg. 2021;20(2):240-45. Doi: 10.1007/s12663-019-01304-y.

[3] Mohan RPS, Verma S, Singh U, Agarwal N. Cementifying fibroma. BMJ Case Rep. 2013;2013:01-04. Accessed: March 18, 2024. Available from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3736223/.

[4] Ram R, Singhal A, Singhal P. Cemento-ossifying fibroma. Contemp Clin Dent. 2012;3(1):83-85. Doi: 10.4103/0976-237X.94553.

[5] Goyal S, Godhi S, Goyal S, Giraddi G. Cementifying Fibroma of the Mandible – A Case Report. J Oral Health Comm Dent. 2008;2(2):42-45. Accessed: March 18, 2024. Available from: https://www.johcd.net/abstractArticleContentBrowse/JOHCD/53/2/2/9948/abstractArticle/Article.

[6] Grewal M, Gautam S, Tanwar R, Saini N. Cemento-ossifying fibroma of maxilla: An unusual case report. J Dent Res Rev. 2020;7(4):206-09. Doi: 10.4103/jdrr.jdrr_12_20.

[7] Sivasubramaniam V, Faduyile F, Zarka MA, Deery A, Field A. Maxillary cementifying fibroma: A case report with fine-needle aspiration cytology highlighting the expanding role of e-medicine in cytopathologist collaborations. Diagn Cytopathol. 2012;40(9):810-14. Doi: 10.1002/dc.21636.

[8] Akkitap MP, Gümrü B, İdman E, Erdem NF, Gümüşer Z, Aksakallı F. Cemento-ossifying fibroma: Clinical, radiological and histopathological findings. Clin Exp Health Sci. 2020;10(4):468-72. Doi: 10.33808/clinexphealthsci.669796.

[9] Wu PC, Leung PKY, Ma KM. Recurrent cementifying fibroma. Journal of Oral and Maxillofacial Surgery. 1986;44(3):229-34. Accessed: March 18, 2024. Available from: https://www.sciencedirect.com/science/article/abs/pii/027823918690114X.

[10] Kahvec K, Ayrancı F, Ömezli MM, Ay M. Cemento-ossifying fibroma of the mandible: A case report. Eurasia J Oral Maxillofac Surg. 2023;2(1):09-10.

[11] Abdelsayed R, Buchanan A. Central ossifying fibroma of mandible arising in association with cemento-osseous dysplasia. An unusual case presentation. Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology. 2022;133(5):e137. Doi: 10.1016/j.oooo.2021.12.046.

[12] Qureshi MB, Tariq MU, Abdul-Ghafar J, Raza M, Din NU. Concomitant bilateral mandibular cemento-ossifying fibroma and cementoblastoma: Case report of an extremely rare occurrence. BMC Oral Health. 2021;21(1):437. Doi: 10.1186/s12903-021-01794-8.

[13] Divyadharshini V, Vadivel JK, Ganapathy D. A painless bump: A case report of cemento-ossifying fibroma of the anterior maxilla. Cureus. 2023;15(7):e41799. Doi: 10.7759/cureus.41799.

[14] Deshpande A, Hemavathy, Krishnan S. Peripheral cemento - ossifying fibroma of anterior mandible – a rare case report. Int J Dentistry Oral Sci. 2021;8(9):4226-29. Doi: 10.19070/2377-8075-21000861.

[15] Aburas S, Bandura P, Al-Ibraheem A, Berger S, Meier M, Turhani D. A large maxillary cemento-ossifying fibroma superimposed with solitary bone cyst documented over 18 years: A case report. Int J Surg Case Rep. 2020;68:257-62. Doi: 10.1016/j.ijscr.2020.03.011.

[16] Christian INWS, Michael, Setiawan K. Cemento-ossifying fibroma of the left mandible: A case report. World J Surg Proced. 2023;13(1):01-06. Doi: 10.5412/wjsp.v13.i1.1.

[17] Verma E, Chakki AB, Nagaral SC, Ganji KK. Peripheral cemento-ossifying fibroma: Case series literature review. Case Rep Dent. 2013;2013:e930870. Doi: 10.1155/2013/930870.

PARTICULARS OF CONTRIBUTORS:
1. Postgraduate Student, Department of Oral Pathology and Microbiology, Sharad Pawar Dental College (DMIHER), Sawangi (M), Wardha, Maharashtra, India.
2. Associate Professor, Department of Oral Pathology and Microbiology, Sharad Pawar Dental College (DMIHER), Sawangi (M), Wardha, Maharashtra, India.
3. Professor, Department of Oral Pathology and Microbiology, Sharad Pawar Dental College (DMIHER), Sawangi (M), Wardha, Maharashtra, India.
4. Assistant Professor, Department of Oral Pathology and Microbiology, Sharad Pawar Dental College (DMIHER), Sawangi (M), Wardha, Maharashtra, India.
5. Assistant Professor, Department of Oral Pathology and Microbiology Sharad Pawar Dental College (DMIHER), Sawangi (M), Wardha, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Ankita Chavhan,
Postgraduate Student, Department of Oral Pathology and Microbiology, Sharad Pawar Dental College, Sawangi (M), Wardha-442001, Maharashtra, India.
E-mail: ankichavhan21@gmail.com

PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Dec 07, 2023
- Manual Googling: Feb 15, 2024
- iThenticate Software: May 09, 2024 (12%)

ETYMOLOGY: Author Origin

EMENDATIONS: 8

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Dec 06, 2023**
Date of Peer Review: **Feb 17, 2024**
Date of Acceptance: **May 10, 2024**
Date of Publishing: **Aug 01, 2024**