Florid cemento-osseous dysplasia with a concurrent glandular odontogenic cyst: a rare combination of an odontogenic cyst with a benign fibroosseous lesion.



Faculty of Health Sciences Fakulteit Gesondheidswetenskappe Lefapha Ia Disaense tša Maphelo

Tsholofelo Kungoane, Liam Robinson

Background: Cemento-osseous dysplasias (CODs) are a group of nonneoplastic lesions, associated with a strong female predominance, with three clinical and radiological presentations. Focal COD is commonly seen in the mandibular first molar region in female patients in their mid-thirties. Periapical COD is noted in the anterior mandibular region associated with the apices of vital teeth in female patients of African descent. Florid COD involves teeth-bearing regions of both the mandible. The most common cysts associated with benign fibro-osseous lesions are non-odontogenic and non-epithelial lined cysts such as simple bone cyst and aneurysmal bone cysts. Literature is scant when documenting benign fibro-osseous lesions with odontogenic cysts, with only a single case documenting this phenomenon. This is the second documented case of a benign fibro-osseous lesion with an odontogenic cyst.

Case study: A 43-year-old female patient presented with a painless bony protrusion in the left mandibular first molar region and an expansile bony lesion involving the left posterior maxilla, associated with a painful left maxillary first molar. A panoramic radiograph showed multiple mixed radiopaque-radiolucent lesions involving the apices of teeth in both the mandible and maxilla, and an incidental unilocular cystic lesion in the left posterior maxillary region* (Fig. 1)

Discussion: The current case shows two clear distinct radiological and pathological entities, whilst those reported in the literature showed a fibro-osseous proliferation in the wall of an odontogenic tumour or cyst. The only case in literature similar to the current case is that of Sanjai et al. whereby florid COD was seen in conjunction with a dentigerous cyst. An interesting observation was that majority of these lesions occurred in females, as summarised in Table 1.

Author (s)	Year published	Number of cases	Age/ gender	Location of both lesions	BFOL	Odontogenic Lesion
lida et al.	2006	1	37/ F	Max L	OD-like	Odontoma
Sanjai et al.	2010	1	20/ F	Man L	FCOD	Dentigerous cyst
Prodromidis et al.	2011	1	36/ F	Man L	COD-like	Odontoma
Li et al.	2013	1	22/F	Max R	OF-like	AOT
Naidu et al.	2016	2	34 F 23 F	Max R Max R	OF-like	AOT
Reddy et al.	2019	1	12/M	Man L	BFOL-like	Odontoma
Kungoane and Robinson	2021	1	43/F	Man, Max	FOD	GOC

This study was approved by the University of Pretoria Faculty of Health Sciences Research Ethics Committee (Reference No.: 712/2019). The case study was published in a journal: Kungoane T, Robinson L. Florid cemento-osseous dysplasia with a concurrent glandular odontogenic cyst. Head and Neck Path. 2021;15,262–266 DOI: 10.1007/s12105-020-01146-y

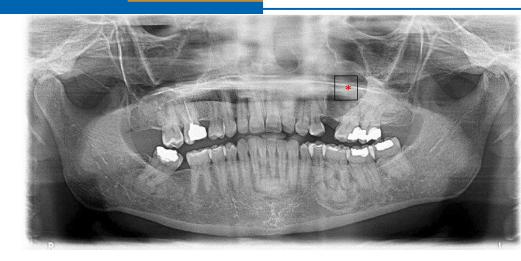


Figure 1

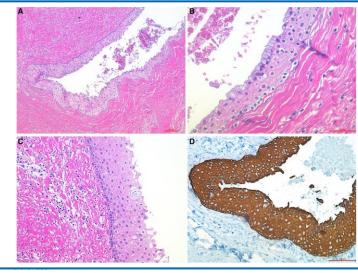


Fig. 2 Haematoxylin and eosin (H&E)-stained section showing the maxillary cyst lined by epithelium of varying thickness showing prominent squamous metaplasia (a). The epithelial lining shows prominent superficial hobnailing with areas of ciliated cells and scattered mucous cells (b). Intra-epithelial cyst is noted (c). Cytokeratin 19 (CK19) immunohistochemical staining of the epithelium confirming odontogenic origin (d)

1. Eversole R, Su L, ElMofty S. Benign fibro-osseous lesions of the craniofacial complex a review. Head Neck Pathol. 2008;2(3):177–202. 2. El-Mofty SK. Fibro-osseous lesions of the craniofacial skeleton: an update. Head Neck Pathol. 2014;8(4):432–44. https://doi.org/10.1007/s1210 5-014-0590-0.

 Mahomed F, Altini M, Meer S, Coleman H. Cemento-osseous dysplasia with associated simple bone cysts. J Oral Maxillofac Surg. 2005;63(10):1549–54. https://doi.org/10.1007/s1210 5-016-0753-2.

4. Bishop JA, Yonescu R, Batista D, Warnock GR, Westra WH. Glandular odontogenic cysts (GOCs) lack MAML2 rearrangements: a finding to discredit the putative nature of GOC as a precursor to central mucoepidermoid carcinoma. Head Neck Pathol. 2014;8(3):287–90. https://doi.org/10.1007/s1210 5-014-0534-8.

5. Sanjai K, Kumarswamy J, Kumar VK, Patil A. Florid cemento osseous dysplasia in association with dentigerous cyst. J Oral Maxillofac Pathol. 2010;14(2):63-8. https://doi.