Management of Pigmented Botryoid Odontogenic Cyst - A Case Report

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Abstract

Rationale: Botryoid odontogenic cyst (BOC) is a rare multilocular odontogenic cyst, considered a subtype of the lateral periodontal cyst (LPC). BOC shows a slight male predominance and most often affects the mandibular body. After enucleation, recurrence rates for BOC are higher (22%) compared to LPC (2%). Pigmented odontogenic cysts and tumours are rare, with only 71 reported cases, and one of them was a pigmented BOC. **Patient Concerns and Diagnosis:** This report describes a 62-year-old Afro-Brazilian female patient with a radiolucent lesion in the apical region of mandibular incisors, which, after surgical enucleation, was diagnosed as pigmented BOC. Literature data show that pigmented odontogenic cysts predominantly affect the mandible, without sex predilection. Most cases are found in Asian and Black patients. **Outcome and Take-away Lessons:** The presence of melanocytes within epithelial tissue may explain the deposition of melanin, suggesting a possible racial/ethnic predisposition for these lesions.

Keywords: Botryoid odontogenic cyst, melanin pigment, melanocytes, odontogenic cysts, odontogenic tumours

Introduction

Botryoid odontogenic cyst (BOC) is a rare odontogenic lesion with a multilocular, grape-like appearance, as described by Weathers and Waldron in 1973.[1] BOC is considered a multilocular subtype of lateral periodontal cyst (LPC), typically asymptomatic but prone to causing large swelling due to its growth potential.[2-4] Radiographically, BOCs show a multilocular appearance, and histologically, they are characterised by cystic cavities lined by a thin, non-keratinised epithelium.[2] The pathogenesis remains unclear, although epithelial rests are likely involved. Melanin pigmentation in odontogenic cysts is rare, with only one BOC case reported.[3-5]

Case Report

A 62-year-old Afro-Brazilian female patient was referred for evaluation of a mandibular lesion, which showed a radiolucent aspect during a radiographic examination one year ago. During anamnesis, the patient denied any medical condition or allergy. The extraoral examination was not remarkable. Intraorally, the patient was wearing an upper total prosthesis and a lower partial prosthesis. The panoramic radiograph showed a well-defined, multilocular radiolucency, located in the apical region of the lower incisors, and extending towards the mental foramen on the right side [Figure 1]. The incisors overlying the lesion were vital. The main clinical differential diagnoses were odontogenic keratocyst and residual cyst. The patient underwent a surgical procedure under local anaesthesia. A mucoperiosteal incision followed by tissue detachment allowed complete removal of the lesion [Figure 2]. Histopathological examination showed multiple cystic spaces delimited by thin epithelium and fibrous connective tissue without inflammation. The epithelial lining was composed of flat and cuboidal cells, which were arranged

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2–5 layers thick in most areas, as well as focal nodular areas with whirlpool-like thickening and clear cells [Figure 3a-d]. Notably, the epithelium showed a uniform presence of brown pigment. Due to this, the Fontana-Masson stain was performed, which showed extensive melanin deposits throughout the cystic epithelial lining [Figure 3e and f]. The definitive diagnosis was pigmented BOC. The postoperative period was uneventful. The patient was referred for restorative treatment of the lower incisors. Currently, after 12 years of follow-up, there is no evidence of recurrence or alteration.

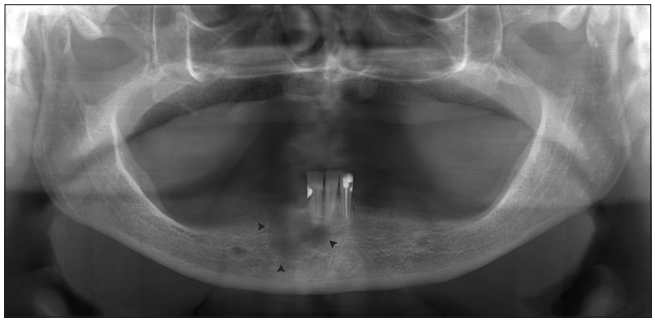


Figure 1: Panoramic radiograph showing a well-defined, multilocular radiolucency in the anterior region of the mandible. Arrowheads delimiting its edges

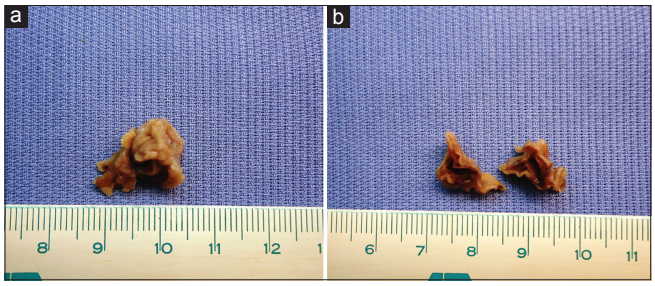


Figure 2: (a) Completely enucleated lesion, with a smooth surface and continuous contours. (b) Longitudinal section reveals internal compartmentalized aspect by fine fibrous trabeculae

Discussion

Melanocytes and/or melanin pigment are rare components of the odontogenic cysts and tumours.[6] Melanocytes are distributed in the skin, mucosal membranes, inner ear, eyes and nervous system, but are not normally present in bone tissue.[7] Lawson *et al*., (1976) performed the first study on intraoral melanocytes, which assessing human fetuses related to the presence of melanocytes in the dental lamina, in some ducts of minor salivary glands, and in oral mucosal epithelium.[8] Thus, we believe that the presence of melanocytes in the dental lamina may explain the histogenesis of melanin in pigmented odontogenic lesions.[5,6,8]

Melanin pigmentation is a rare component in odontogenic cysts, firstly described by Grand and Marwah (1964) who reported a pigmented gingival cyst.[6] Here, we performed a detailed literature review of pigmented odontogenic cysts and tumours, identifying 71 cases [Table 1]. Of them (excluding the current case), 41 cases were pigmented odontogenic cysts, equally involving both genders (19 males, 21 females, 1 unspecified), with a mean age of 22 years (range, 11–68 years). The site most affected was the mandible (n = 23), and for 35 cases with available race information, 31 were Asians and Blacks, whereas four were whites. According to the histopathological diagnosis, calcifying odontogenic cyst (n = 21) and odontogenic keratocyst (n = 11) were frequent, followed by dentigerous cyst (n = 4), LPC (n = 2), BOC (n = 1), gingival cyst (n = 1) and odontogenic cyst with verrucous proliferation (n = 1).[6,7,8] It is interesting that inflammatory odontogenic cysts (e.g., periapical cyst, the most common odontogenic lesion) are practically spared. Considering the

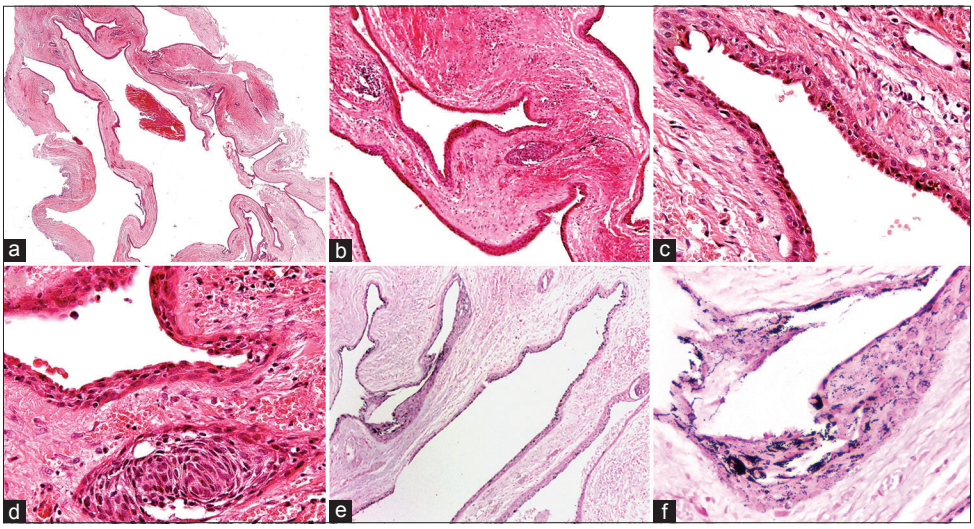


Figure 3: (a) Histological examination on Hematoxylin and Eosin stain showing multiple cystic spaces delimited by thin epithelium and fibrous connective tissue without inflammation (×4). (b) The epithelial lining was composed of flat/cuboidal cells, in 2–5 layers thick in most areas, as well as focal nodular areas with swirling architecture and clear cells (×10). (c and d) The epithelium showed a uniform presence of brown pigment (×40). (e and f) The Fontana-Masson stain revealed extensive melanin deposits throughout the cystic epithelial lining (x10 and x40)

Table 1: List of pigmented odontogenic cysts and tumours reported to date, including the current case

	Pigmented odontogenic cysts*	Pigmented odontogenic tumours	Pigmented odontogenic botryoid cysts*
Number of cases	40	30	2
Age (years), mean (range)	22.4 (11–68)	19.1 (6–53)	42 and 62
Gender, *n* (%)			
Female	21 (52.5)	10 (33.3)	1 (50.0)
Male	18 (45)	8 (26.6)	1 (50.0)
NA	1 (2.5)	12 (40)#	-
Location, *n* (%)			
Maxilla	15 (37.5)	7 (23.3)	0
Mandible	23 (57.5)	9 (30)	2 (100)
Maxillary gingiva	1 (2.5)	-	-
Mandibular gingiva	-	1 (3.3)	
NA	1 (2.5)	13 (43.3)#	-
Race, *n* (%)			
Black	7 (17.5)	6 (20)	2 (100)
Asian	23 (57.5)	11 (36.6)	0
White	4 (10)	1 (3.3)	0
NA	6 (15)	12 (40)	0

*Total cases of pigmented odontogenic cysts (*n*=42), #The references 12, 13, and 14 presented a large series of cases where sex data were not specified or detailed. NA: Not available

pigmented odontogenic tumours, there are 30 cases published, with a slight female predilection (10 females, 8 males, 12 unspecified), and a mean age of 19 years (range, 6–53 years). Both maxilla and mandible were similarly involved and except for 1 case (white), all others with available race information ($n = 17$) were Asians and Blacks. Regarding the microscopical diagnosis, the vast majority were adenomatoid odontogenic tumours ($n = 13$) and odontoma ($n = 8$, including ameloblastic fibro-odontoma/fibrodentinoma cases), followed by 2 cases each of ameloblastoma, calcifying epithelial odontogenic tumour and dentinogenic ghost cell tumour and 1 case each of ameloblastic fibroma, odontogenic fibroma and intraosseous odontogenic carcinoma.[7-9]

About 101 cases of BOC have been reported so far, with a slight male predilection, between the 5th and 7th decades of life, usually affecting the mandibular canine and premolar region.[5,9,10] Of them, only one case was pigmented, the clinicopathological features of which were not specified. Thus, we believe that in the current study, we present the first case of pigmented BOC documented in detail. The BOCs share clinical, radiographic, and histological similarities with LPCs and glandular odontogenic cysts. By microscopy, the multilocular appearance of BOCs allows its differentiation from LPCs.[5,9] To date, approximately 213 cases of LPC have been reported.[5,6,9] Of them, there is a single case of pigmented LPC affecting the mandible of a 38-year-old Black man.[5]

It is known that melanocytes respond to biological, physical and chemical factors from the surrounding microenvironment, with relevant functions including skin homeostasis, neutralizing free radicals/reactive oxygen species, hearing, neuroendocrine function, as well as inhibition of bacteria and fungi.[6] We think that some of these functions could be addressed in the pathogenesis of pigmented odontogenic cysts and tumours. Moreover, it is possible that the quantity of pigment melanin deposition and predisposing factors activating its production may be associated with racial pigmentation.[6]

Finally, several studies suggest that the pigmentation of odontogenic cysts and tumours may originate from the inductive effect of hard dental tissues or prominent calcifications.[6] Furthermore, the migration of melanocytes through the mesenchyme after complete neural crest morphogenesis is another possibility that should be considered.[7,8] Taken together, the presence of melanocytes within the dental lamina may explain the pigment melanin histogenesis in these odontogenic lesions, which expands their histopathological spectrum, strongly suggesting racial/ethnic predisposition.[5,6,8]

Conclusion

Pigmented odontogenic cysts and tumours are uncommon, with about 71 cases reported to date. Including the current case, 2 pigmented BOCs have been reported to date. Relevantly, almost all these cases corresponded to Asian and Black patients. The presence of melanocytes within epithelial tissue may explain the melanin pigment deposition in these odontogenic lesions, which expands their histopathological spectrum.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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