

Pigmented lateral periodontal cyst: A case report and review of pigmented odontogenic cysts

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Abstract

Pigmented odontogenic cysts are uncommon and to date, only 37 cases have been reported in the English literature. Here, we report a case of a pigmented lateral periodontal cyst (LPC) in the maxilla of a 48-year-old female. The patient presented with clinical swelling in the maxillary anterior region. Microscopic features of the biopsied specimen were consistent with a diagnosis of LPC. The epithelial cyst lining exhibited numerous coarse granules of melanin pigment, which was confirmed by S-100 immunohistochemistry and Fontana-Masson bleach histochemical method. Almost all documented cases of pigmented odontogenic cysts have occurred in Asians and African-Americans, with only three cases in white patients. Racial pigmentation may have a role in the pathogenesis of these lesions. Although the origin and pathologic significance of melanocytes in these pigmented intraosseous lesions cannot be explained, it may be something to consider for investigation in future.

Keywords: Fontana-Masson bleach, lateral periodontal cyst, melanin, pigmented odontogenic cysts

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INTRODUCTION

The lateral periodontal cyst (LPC) accounts for <1% of odontogenic cysts.^[1] The first well-documented case of LPC was presented in 1958.^[2] LPCs are considered noninflammatory developmental odontogenic cysts which may arise from the dental lamina, reduced enamel epithelium, or the rests of Malassez. They are typically seen on the lateral aspect or between the roots of erupted teeth, and most frequently arise in the mandibular incisor, canine and premolar region. Less than 20% may arise in the maxilla.^[1] A slight male predilection is noted with a peak incidence in the sixth and seventh decades of life. Radiographically, LPCs are well defined, often corticated

unilocular radiolucencies which are approximately <1 cm in diameter. A larger multilocular variant known as botryoid odontogenic cyst (BOC) was first reported in 1973.^[3] Melanocytes, which are derived from the neural crest cells and are widely distributed throughout the skin, nervous system and certain types of the mucosa, are not a common finding in intraosseous gnathic lesions. Intraosseous pigmented lesions have often been reported to occur as metastatic lesions of malignant melanoma. Few cases of melanin pigmentation within the jaw bone in association with many gnathic tumors and cysts such as melanotic neuroectodermal tumor of infancy (MNTI), adenomatoid odontogenic tumor, calcifying odontogenic

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cyst (COC), ameloblastic fibroma, odontoma, ameloblastic fibro-odontoma, odontogenic keratocyst (OKC), LPC and dentigerous cyst (DC) have been documented. With the exception of MNTI, racial pigmentation may have an important role in the occurrence of such lesions as majority of these were reported in Asian and African-American patients. In this article, we present a rare pigmented variant of LPC which, to the best of our knowledge, is the second case of pigmented LPC documented in the English literature, and the first to occur in the maxilla.

CASE REPORT

A 48-year-old African-American female presented with a clinically evident swelling in the anterior hard palate in the region of maxillary right central incisor. Past medical and dental histories revealed a congenitally missing tooth # 8 with no other significant contributory conditions. The patient was informed about the possibility of a cyst-like lesion by her previous dentist. Radiographic examination revealed a unilocular radiolucent lesion in the area of the maxillary right central incisor [Figure 1]. The borders of the radiolucency were well defined and corticated. An excisional biopsy was performed and revealed a gray cyst-like soft tissue measuring 1.5 cm × 1.3 cm × 0.8 cm. Histopathological evaluation revealed multiple sections of the specimen consisting of connective tissue and luminal odontogenic epithelium. A central lumen-like area was rimmed at the periphery by a band of epithelium that varied from one or three cells in thickness. Focal areas exhibiting nodular swirls of squamous cells were noted [Figure 2a]. The lining epithelium exhibited scattered melanocytes and fine granules of dark brown pigment which was interpreted as melanin [Figures 2b and 3a]. Histochemical evaluation with Fontana-Masson bleach method confirmed the pigment to be melanin [Figure 3b]. Numerous melanin-containing cells were immunohistochemically positive for S-100 protein,

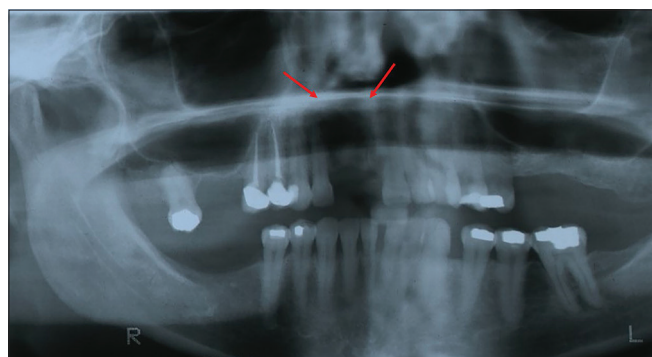


Figure 1: Panoramic radiographic presentation of the pigmented lateral periodontal cyst shows a radiolucent lesion in the region of congenitally missing maxillary right central incisor (shown with red arrows). The cystic lesion grossly measured 1.5 cm × 1.3 cm × 0.8 cm

and these were interpreted as melanocytes [Figure 4a and b]. These spindle- and dendritic-shaped cells containing abundant cytoplasmic brown–black pigment were also seen in the superficial connective tissue subjacent to the cystic lining epithelium [Figure 4b]. In addition, numerous cholesterol clefts associated with foreign body type of giant cells were also noted within this specimen. A diagnosis of pigmented LPC was rendered. No recurrence has been noted till date.

DISCUSSION

Pigmented odontogenic lesions of the jawbones are a rare phenomenon but not unusual. The existence of melanocytes in the oral mucosa, for example, gingiva and buccal mucosa, is not unusual and is often seen in patients of dark complexion. Majority of the physiological pigmentation in human tissues is a result of melanin production by the melanocytes. Melanocytes are derived from neural crest cells, and though the majority are present in the epidermis, nervous system, hair and iris, they are also seen in the inner ear, certain types of mucosa and other tissues.^[4] They produce melanin in intracellular granules called melanosomes which upon maturation release melanin into the surrounding keratinocytes or epithelial cells. However, the presence of melanocytes and melanin in intraosseous sites may suggest a pathological occurrence.

Reports of intraosseous odontogenic pigmentation as a primary lesion, other than metastases of malignant melanoma, are very few. Upon review of the English literature, 37 documented cases of pigmented odontogenic cysts were found [Table 1]. Melanin pigmentation has been reported most frequently in association with COC (21)^[5-21] followed by OKC (9),^[8,22-25] DC (4),^[26-29] LPC (1),^[30] gingival cyst (GC) (1)^[31] and BOC (1)^[32] [Table 1]. Buchner *et al.* were the first to report a pigmented LPC in the mandible of a 38-year-old African-American male.^[30] In a study of 20 cases of LPC, Altini and Shear found melanin pigment

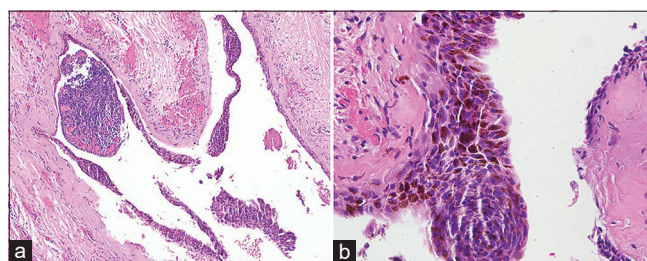


Figure 2: Histopathologic examination shows a thin epithelial lining and numerous localized thickened nodular plaques (a, H&E stain, ×100 for ×10 magnification). Brown–black granules interpreted as melanin noted within the lining epithelium and thickened nodular plaques (b, H&E stain, ×400 for ×40 magnification)

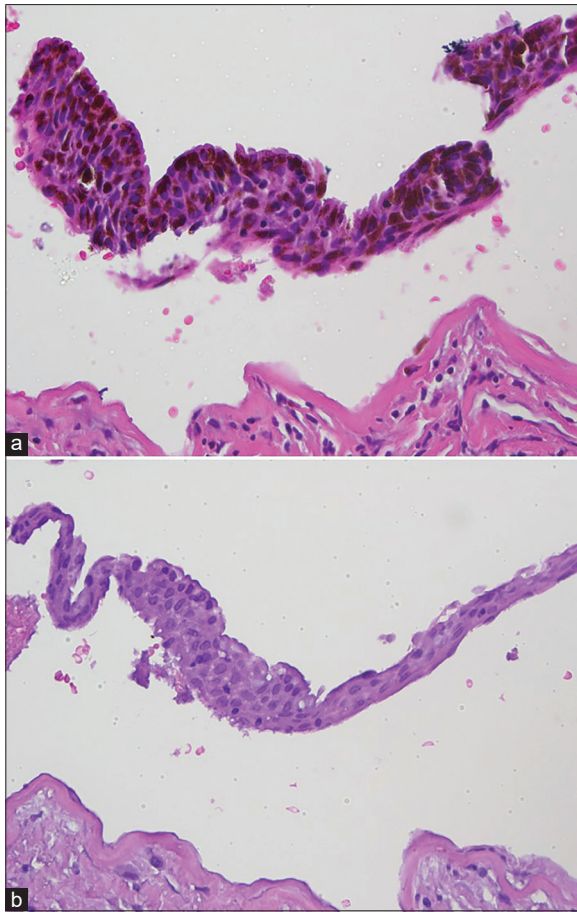


Figure 3: Histopathologic image shows a lining exhibiting numerous dark brown stained granules (a, H&E stain, $\times 400$ for $\times 40$ magnification). Fontana-Masson bleach shows the disappearance of brown granules in the tissue, confirming the pigment as melanin (b, H&E stain, $\times 400$ for $\times 40$ magnification)

in the epithelium of one lesion which was classified as a BOC.^[32] To our knowledge, the present case is the second one to document melanocytes and melanin pigment in an LPC, and the first to report a pigmented LPC in the maxilla. When data from all known cases of pigmented odontogenic cysts were combined, no gender predilection was found (18 males, 19 females and one unreported). These cysts had an increased predisposition to occur in the mandible than the maxilla. The age range varied from 8 years to 68 years, with an average age of 23.4 ± 15.8 years at the time of diagnosis. Most lesions occurred mainly in the Asian and African-American ethnicities [Table 1]. The incidence of such lesions in Asians, specifically the Japanese population, exceeded others by far. Fourteen of the 38 (36.8%) lesions occurred in patients of Japanese ethnicity, seven each in males and females.

The exact mechanism of the presence of melanin pigmentation in odontogenic cysts is still obscure. Most of the odontogenic cysts arise from the dental lamina

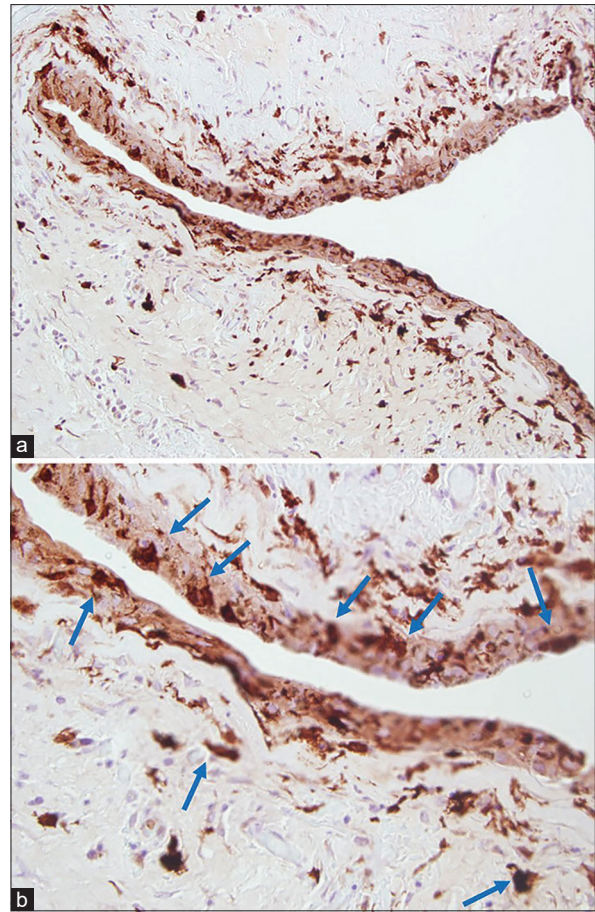


Figure 4: Immunohistochemistry with S-100 shows a few cells in the epithelial lining to be positive for S-100 protein (a, IHC stain, $\times 200$ for $\times 20$ magnification). Few S-100-positive cells are seen in the lining and in superficial connective tissue subjacent to the lining, marked with blue arrows (b, IHC stain, $\times 400$ for $\times 40$ magnification)

which originates from the primitive oral ectoderm. In their study of facial skeletons of human fetuses, Lawson *et al.* found melanocytes within the dental lamina and outer enamel epithelium of 6 Negro fetuses and in 3 of the 11 Caucasian ones.^[33] This may justify the occasional presence of melanocytes in odontogenic lesions. However, this hypothesis fails to explain the presence of melanocytes and melanin pigment in lesions that do not originate from the rest of the dental lamina. Alternatively, it has been hypothesized that the presence of pigmentation may be attributed to either a close relationship between the oral epithelium and the cranial neural crest-derived ectomesenchyme during odontogenesis,^[34] and/or neuroectodermal differentiation of the odontogenic cells.^[35] Neural crest cells play an important role in odontogenesis as seen by the reciprocal induction between inner enamel epithelium and dental papilla, which arises from ectomesenchyme, a derivative of neural crest cells. As dentin is laid down, the odontoblasts and melanocytes retract, but the melanin pigment remains in the

Table 1: Reported cases of pigmented odontogenic cysts

Case	Age/race (years)	Gender	Location	Diagnosis	References
1.	11/Japanese	Female	L mandible, molar region	COC	Takeda <i>et al.</i> ^[5]
2.	17/Japanese	Male	R mandible, canine	COC	
3.	16/Chinese	Female	Maxilla	COC	Siar and Ng ^[6]
4.	31/Indian	Male	Maxilla	COC	
5.	68/Malay	Female	Mandible	COC	
6.	15/NR	Female	R maxilla, canine	COC with odontoma	Kezler and Guglielmotti ^[7]
7.	21/Japanese	Male	Maxilla	COC	Takeda <i>et al.</i> ^[8]
8.	16/Japanese	Male	R mandible, molar area	OKC	Takeda <i>et al.</i> ^[9]
9.	15/Japanese	Female	L mandible, molar area	OKC	
10.	26/Japanese	Female	R mandible, molar area	OKC	
11.	15/Japanese	Male	L and R mandible, molar	OKC	
12.	11/Japanese	Female	Maxilla and mandible	OKC	
13.	13/Japanese	Female	R mandible	COC	Nagao <i>et al.</i> ^[10]
14.	13/Japanese	Male	R mandible	COC	Saito <i>et al.</i> ^[11]
15.	9/Japanese	Female	L mandible	COC	
16.	35/Japanese	Female	R maxilla	COC	
17.	15/West Indian	Female	R mandible, canine region	COC	Soames ^[12]
18.	13/Hispanic	Male	Mandible	COC	Schwimmer <i>et al.</i> ^[13]
19.	11/black	Female	Maxilla	COC	Petri and Stump ^[14]
20.	64/NR	Male	Mandible, midline	COC	Sauk ^[15]
21.	21/white	Female	L maxilla	COC	Abrams and Howell ^[16]
22.	27/Indian	Male	R mandible	COC	Chandi and Simon ^[17]
23.	24/black	Female	L maxilla, canine region	COC	Duckworth and Seward ^[18]
24.	16/NR	Male	R maxilla, incisor region	COC	Gorlin <i>et al.</i> ^[19]
25.	23/Bantu	Female	L maxilla, incisor region	COC	Lurie ^[20]
26.	15/Japanese	Male	L mandible, canine-premolar region	COC	Han <i>et al.</i> ^[21]
27.	20/white	Female	R mandible, 3rd molar area	OKC	MacLeod <i>et al.</i> ^[22]
28.	nr/black	Male	NR	OKC	Brannon ^[23]
29.	NR/West Indian	Female	NR	OKC	Browne ^[24]
30.	13/Indian	Male	L maxilla, molar region	OKC	Manaktala <i>et al.</i> ^[25]
31.	13/white	Female	L mandible, premolar	DC	Parrini <i>et al.</i> ^[26]
32.	37/Iranian	Male	R maxilla, canine	DC	Shahrabi <i>et al.</i> ^[27]
33.	29/Japanese	Male	L mandible, retromolar area	DC	Takeda and Yamamoto ^[28]
34.	8/black	Male	R mandible, cuspid and bicuspid area	DC with AOT	Warter <i>et al.</i> ^[29]
35.	38/black	Male	L mandible, between canine and premolar	LPC	Buchner <i>et al.</i> ^[30]
36.	62/black	NR	L maxilla, lateral incisor and canine region	GC	Grand and Marwah ^[31]
37.	42/black	Male	NR	BOC	Altini and Shear ^[32]
38.	48/black	Female	R maxilla, incisor region	LPC	Present case

AOT: Adenomatoid odontogenic tumor, BOC: Botryoid odontogenic cyst, COC: Calcifying odontogenic cyst, DC: Dentigerous cyst, GC: Gingival cyst, L: Left, LPC: Lateral periodontal cyst, M: Male, NR: Not reported, OKC: Odontogenic keratocyst, R: Right

odontoblastic processes trapped between the new dentin formed. Thus, it might be considered that the presence of melanocytes in odontogenic lesions is developmental in nature. This hypothesis also explains the presence of melanocytes and melanin pigment in lesions that do not originate from the rest of the dental lamina. Interestingly, the propensity of pigmentation to be seen predominantly in Asians and African-Americans, and being almost nonexistent in white populations, points to a correlation with racial pigmentation. Although genetic factors have not been implicated in the occurrence of odontogenic lesions *per se*, the possible effects of genetic background on racial pigmentation cannot be neglected. Except for three cases, all patients were either African-American or Asian [Table 1]. This leads us to an understanding that the pigmentation is physiologic rather than pathologic and hence does not contribute to the outcome of the lesion. An additional explanation might be that under certain circumstances, few lesional odontogenic tissue

cells have a potential for neuroectodermal differentiation. It is possible that odontogenic lesions contain inactive melanocytes which produce melanin after being activated by certain predisposing triggers that might be related to factors associated with racial pigmentation.^[30]

CONCLUSION

The exact etiopathogenesis of the pigmentation in intraosseous odontogenic lesions still remains unclear. More documented case studies and careful assessment may be necessary to derive an insight into the association between the cranial neural crest-derived ectomesenchyme and the odontogenic epithelium, as well as to understand the pathologic significance and prognostic implications if any.

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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