

Melanin and odontogenic cysts: An ambiguous yet imperative relation

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Abstract

Pigmentation in the head-and-neck lesions is an uncommon feature. The presence of it is fascinating, but its relevance is unknown. Various odontogenic and nonodontogenic lesions show the presence of melanin pigment. Although the presence of melanin is normal in the epithelium and connective tissue, the role of melanin pigmentation in odontogenic cysts is unclear. Apart from maintaining the skin balance, varying other functions of melanin is being researched. However, not much study has been done regarding it in odontogenic lesions. As the literature regarding such cases are scanty, here, we present a case of pigmented dentigerous cyst and an attempt to understand their relationship.

Keywords: Case report, dentigerous cyst, melanin pigmentation, melanogenesis, odontogenic lesions

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INTRODUCTION

Lesions presenting in the jaws are of varying nature with odontogenic lesions being the most common type. Literature search reveals that the presence of melanin pigment is noted in odontogenic lesions such as odontogenic keratocyst (OKC), adenomatoid odontogenic tumor (AOT), lateral periodontal cyst, calcifying odontogenic cyst and ameloblastic fibroma to name a few.^[1] Although both cysts and tumors of odontogenic origin contain melanocytes and melanin-laden epithelial cells, its presence is variable. Here, we present a case of dentigerous cyst showing unusually extensive melanin pigmentation both in epithelial lining and connective tissue. Whether the presence of melanin has any impact on the treatment, prognosis or recurrence is yet to be ascertained.

CASE REPORT

A 28-year-old patient reported to our college with a complaint of decayed front teeth. The patient claimed to be systemically healthy with no relevant medical/dental history. On examination, endodontically treated 11 and 12 were noted. The teeth were nontender on percussion and there was no evidence of any swelling. Occlusal radiograph revealed the presence of a supernumerary tooth along with a well-defined radiolucent lesion in the region of 11 and 21. Clinically, a provisional diagnosis of the impacted supernumerary tooth with dentigerous cyst was given.

The supernumerary tooth was removed, and the specimen was sent for histopathological examination. H&E-stained sections revealed a cystic cavity lined by epithelium and

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connective tissue capsule. Epithelium was odontogenic in nature with 2–4 layers thick in most areas. Few foci showed hyperplasia of the epithelium. Melanin pigments were seen both in the epithelium and connective tissue as black deposits. Although the pigments were seen scattered in all the layers of the epithelium it was concentrated more in the basal layer. The presence of melanin was confirmed by staining with Masson Fontana [Figure 1]. In the connective tissue capsule, the pigments were concentrated in the subepithelial region. The capsule showed collagen bundles, scattered inflammatory cells, melanophages and blood vessels. Correlating with clinical, radiographical features, a diagnosis of the dentigerous cyst was given. Follow-up of 2 years is uneventful.

DISCUSSION

Dentigerous cyst is the second most common odontogenic cyst. The cyst surrounds the crown of an impacted tooth. It is caused by fluid accumulation between reduced enamel epithelium and the enamel surface. This is usually seen in mandibular and maxillary third molar and maxillary cuspid regions, commonly in second to third decade of life, with slight male predilection. It is seen either unilaterally, bilaterally or associated with various syndromes. Displacement of teeth, root resorption of adjacent teeth, pain, cortical plate expansion, and facial asymmetry are the possible sequelae to cyst enlargement. Radiographically, three varieties are seen: central, lateral and circumferential.^[2]

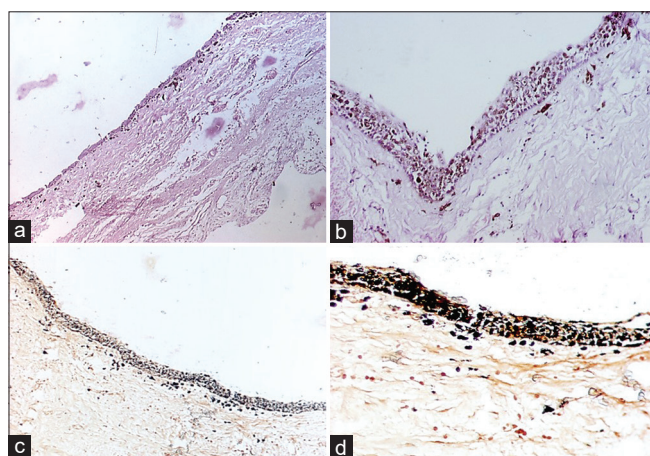


Figure 1: (a) H&E-stained section revealing cystic cavity lined by thin odontogenic epithelium and connective tissue capsule ($\times 4$). (b) H&E-stained section revealing melanin pigments in all the layers of odontogenic epithelium and few scattered melanophages in connective tissue capsule ($\times 10$). (c) Masson Fontana-stained section revealing cystic cavity lined by thin odontogenic epithelium with melanin pigments and melanophages in the connective tissue capsule ($\times 4$). (d) Masson Fontana-stained section revealing melanin pigments in all the layers of odontogenic epithelium and few scattered melanophages in connective tissue capsule ($\times 10$)

Histopathologically, the cyst shows a thin connective tissue wall with a thin layer of stratified squamous epithelium lining the lumen. Islands of odontogenic epithelium may also be seen within the capsule. In the presence of inflammation, rete ridge formation is seen. Rushton bodies are noted within the lining epithelium are noted. The content of the lumen is usually thin watery yellow fluid, occasionally tinged with blood.

The pluripotentialities of the epithelium in dentigerous cyst may be responsible for the varying presentation of the features seen in this cyst such as mucous secreting cells, respiratory epithelium or stratified squamous epithelium.^[2]

Previously, Melanin pigments in odontogenic lesions were said to be uncommon by some researchers.^[1] However, recently, melanin has been reported in jaw lesions with odontogenic origins such as OKC, AOT, calcifying epithelial odontogenic tumor, calcifying cystic odontogenic tumor, ameloblastoma, calcifying odontogenic cyst, dentigerous cyst, gingival cyst, botryoid odontogenic cyst, lateral periodontal cyst, ameloblastic fibroma, odontoma, ameloblastic fibroodontoma, ameloblastic fibrodentinoma, odontoameloblastoma and odontogenic fibroma.^[1-6] However, the pattern and distribution of melanin pigmentation is not clear.

Melanin deposition may vary in different odontogenic lesions, both in the epithelium and connective tissue. It can range from scattered deposit to involving a large area. In our case, it was most concentrated in the basal layer. In focal areas, the whole thickness of the epithelium showed the presence of melanin. Very few cases have reported extensive melanin deposition involving the whole thickness of the epithelium. In the connective tissue capsule, the melanophages were scattered in aggregates. The reason for this irregular deposition of melanin is not known.

Melanocytes are known to arise from the neural crest.^[7] Most researchers are of the opinion that melanocytes form part of the oral epithelium.^[6] While few others propose that their presence is a result of the migration of melanocytes.^[8] The enamel organ is derived from ectoderm where melanocytes have been found. Mucosal melanocytes have a tendency to concentrate around the attachment zone of the dental lamina.^[4,9] Melanocytes, which are normally present in the oral mucosa, are also found in the dental lamina or tooth bud of the fetuses.^[10]

Melanocytes respond to physical, chemical and biological stimuli from the surrounding microenvironment.^[11] Melanin and byproducts of melanogenesis have varying

functions in different parts of the body. Its functions include maintaining skin homeostasis, neutralizing free radicals and reactive oxygen species, body balancing, hearing, neuroendocrine function, functioning of heart valves, physical barrier against microorganisms, inhibition of bacterial and fungal microorganisms and neutralizing effects of bacterial toxins.^[12]

The mechanisms by which melanocytes and/or melanin pigment appear in odontogenic tumors remain unclear. The neural crest influence on the development of odontogenic tissues is well established, and the occurrence of melanocytes in these tissues is not surprising.^[10,13] A biochemical study of the cysts of jaws reported that the level of free radical concentration was found to be increased in case of inflamed cysts of jaws.^[14] Melanin is known to be one of the scavengers of free radicals.^[15] This could explain the reason for the presence of melanin in our case. The presence of inflammatory cells triggers melanogenesis whose intermediate product causes downregulation of inflammatory response.^[11] This could be the reason for the presence of melanin pigments and sparse inflammatory cells in our case [Flow Chart 1].^[11,12,16-19] Hence, the presence of melanin pigments in epithelium lining of the cyst may indicate the decreased function of inflammatory cells. Further studies are necessary to prove or refute these possibilities. Few odontogenic tissues may have the potential for neuroectodermal differentiation under certain settings. It is rational to speculate that the quantity of melanin produced by melanocytes and the

conditions or predisposing factors activating its production may be associated with racial pigmentation. A racial predisposition may be present.^[10,11,20,21]

CONCLUSION

Upon regular follow-up of the case, there was no evidence of recurrence. The importance of melanin pigments in odontogenic lesions is yet to be determined since such cases are seen in isolates. Its role as a scavenger of free radicals and downregulating inflammation needs to be investigated further. As of now, it is not known to have any impact on the treatment or prognosis. Further research needs to be undertaken extensively to know in details, the role of melanin and free radicals in odontogenic lesions.

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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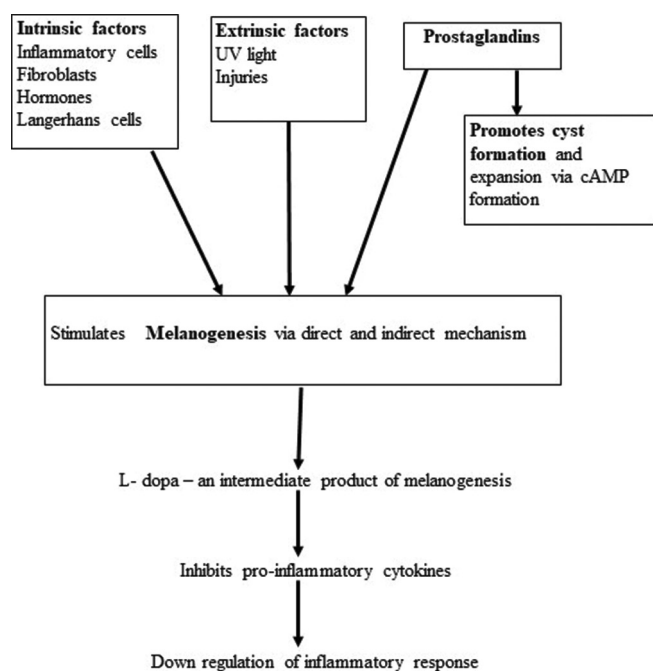
Nil.

Conflicts of interest

There are no conflicts of interest.

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Flow Chart: Role of prostaglandins in melanogenesis and cyst formation

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