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Case Report

Melanophagic proliferation in a epidermal inclusion cyst with multinucleated giant cell reaction-A rare case report

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Abstract: Epidermal cyst is a very common benign cystic lesion of the skin. It is usual to find ulceration of the lining epithelium, rupture of the cyst wall with chronic inflammation and foreign body giant cell reaction. But, it is very rare to see an epidermal cyst with marked accumulation of melanin pigment. Only a few cases of pigmented epidermal cyst with dense collection of melanin pigment have been published in the literature. Here, we are reporting a case of ruptured epidermal cyst with keratin granuloma formation and showing dense collection of melanin pigment.
Keywords: Foreign body giant cells, keratin granuloma, melanin, pigmented epidermal cyst.

INTRODUCTION

Epidermal cyst is a very common benign cystic lesion of the skin. Such cysts can occur anywhere in the body, although they are more common in head and neck region, trunk, and extremities Epidermal inclusion cyst (EIC) refers to the cyst that results from proliferation and implantation of epidermal elements within a circumscribed space in the dermis. But, it is very rare to see an epidermal cyst with marked accumulation of melanin pigment. Sometimes this cyst may elicit a foreign body giant cell reaction. In such cases, the nature of the lesion is evident only by the presence of extruded keratin flakes surrounded by inflammatory cells and foreign body giant cells. Massive melanin pigment deposition in an epidermal cyst is a very rare finding. We report such a case with review of the literature [1].

CASE REPORT

We present a case of an pigmented epidermal inclusion cyst with giant cell reaction originating in the post aural area. We review the clinical, histopathological findings along with confirmation by special stains. A 35 year old female patient presented with a swelling in the post aural area since 4 months duration with recent increase in size. The specimen was excised and sent for histopathological examination. Clinical photograph of the lesion was not available because the clinical diagnosis was the common sebaceous cyst, and no special entity was expected by the clinicians. Grossly, the specimen was a cystic swelling of 1.5 cm diameter filled with cheesy material with specks of brown to black color in the wall. The wall was thickened in one area.

Fig-1: Specimen consists of grey white cyst measuring 1.5cm across with hairs, cut section shows cheesy and brownish material.

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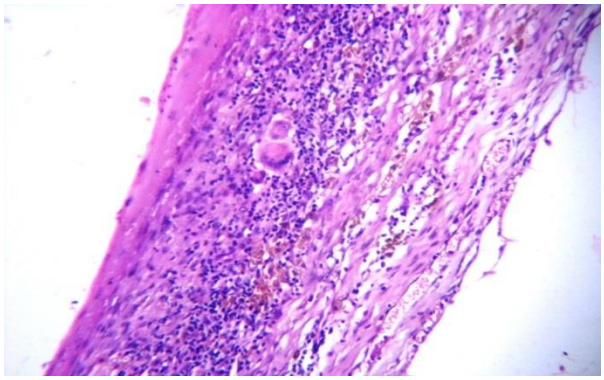

Fig-2: Hematoxylin &Eosin stain shows cystic lesion with a ruptured stratified squamous epithelial lining, keratinous debris, multinucleated giant cells, prominent inflammatory reactions, and pigment incontinence

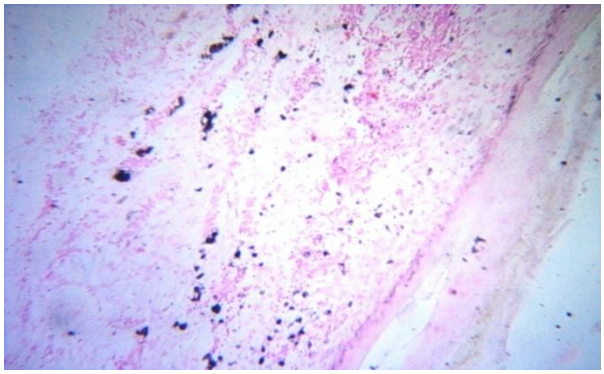

Fig-3: Special stain: Fontana Masson showing prominent melanophagic proliferation in the pigmented epithelial cyst.

Microscopy showed a cyst wall with complete ulceration of the lining epithelium. Instead of the usual squamous lining, the inner wall was bordered by foreign body giant cells. Entrapped keratin material was seen as pale eosinophilic strands. An interesting finding was the presence of a dense collection of golden-brown pigment extra and intracellular in the histiocytes and multinucleate giant cells

DISCUSSION

Epidermal inclusion cyst are common but pigmented epithelial inclusion cysts are rare**.** They may be congenital due to entrapment of ectoderm at the time of fusion of neural tube and secondary may be due to post-trauma and iatrogenic. Epidermal cysts arise from inclusion of epidermal structures in the dermis and other deeper tissues after trauma[2].

Epidermal cyst, also known as infundibular cyst, a common benign cystic lesion of the skin is seen in the mid/lower dermis or subcutaneous tissue. It occurs most commonly on the face, scalp, neck and trunk. Usually these cysts are solitary. Rarely, multiple infundibular cysts are seen as a manifestation of Gardner's syndrome or the basal cell nevus syndrome[3].

We presume that chronic irritation of this subcutaneous epidermal cyst was responsible for the rupture and increased production of melanin by melanocytes, which provoked a granulomatous reaction and melanophagic proliferation. Epidermal cysts are lined by stratified squamous epithelium showing epidermal type of keratinization. These cysts may become infected and rupture into the dermis, resulting in a heavy inflammatory cell infiltrate in the adjacent dermis. In the absence of infection the dermis shows considerable foreign body reaction with numerous multinucleated giant cells results, forming a “keratin granuloma.” The foreign-body reaction usually causes disintegration of the cyst wall. The presence of keratin material surrounded by the reactive inflammatory cells is the only sign of a previously existed epidermal cyst. In our case, there was no evidence of infection. Only ulceration, rupture and foreign body giant cell reaction with typical keratin granuloma in the deep dermis were seen with the unusual finding of dense collection of melanin pigment in the macrophages and giant cells. Melanocytes were not visible[5]. FNAC plays a crucial role in its diagnosis and management. Symptomatic cases should be readily excised and need histological correlation to rule out any potential complications that can arise in these cysts.

Shet *et al*. observed that a large amount of pigment accumulation within epidermal cysts occurs after cyst rupture, which was seen in our case also[1]. Vaideeswar *et al*. have also reported a case of ruptured epidermal cyst with exuberant melanophage proliferation and melanin pigment deposition[4]. In sections stained with hematoxylin–eosin, melanocytes and melanin pigmentation of keratinocytes can be seen only rarely in epidermal cysts of whites, but frequently in epidermal cysts of blacks. The present case was also from a dark-skinned patient and with massive collection of melanin in the dermis. The literature review showed a report of multiple pigmented epidermal cysts in the face of a white patient, which was effectively treated by laser therapy.

Pigmented epidermal cyst should be differentiated from the entity “pigmented follicular cyst” in which prominent rete ridge pattern of epidermal lining and several terminal-sized pigmented hair shafts containing abundant melanin pigment are seen within the cyst cavity. In our case, pigmented hair shafts were not seen [6]. Extensive search of the literature showed only a few articles published about pigmented epidermal cyst with massive melanin deposition in the dermis. The rarity may be because the pigment visible in H and E sections may be either ignored or interpreted as hemosiderin and no further methods might have been employed to demonstrate the nature of the pigment. Cameron and Hilsinger reported that malignant transformation of the cyst wall epithelium occurs very rarely

CONCLUSION

Most epithelial inclusion cysts owe their origin of trauma, we present a case of melanophagic proliferation in a epidermal inclusion with multinucleated giant cell reaction. The rarity may be because the pigment visible in Hematoxylin and Eosin sections may be either ignored or interpreted as hemosiderin and no further methods might have been employed to demonstrate the nature of the pigment. Hence, this case is being reported to remind the pathologists about the academic importance of diagnosing this entity as such and not simply as "epidermal cyst" ignoring the pigment.

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