

# Pigmented epidermal cyst with dense collection of melanin: A rare entity – Report of a case with review of the literature

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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.

**Key words:** Foreign body giant cells, keratin granuloma, melanin, pigmented epidermal cyst, ruptured cyst

## INTRODUCTION

Epidermal cyst is the most common cystic lesion of the skin. There is no diagnostic difficulty in these cases. Sometimes, the cyst lining may ulcerate focally or completely without any trace of the lining squamous epithelium, eliciting 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 producing the “Keratin granuloma.”<sup>[1]</sup> Massive melanin pigment deposition in an epidermal cyst is a very rare finding. We report such a case with review of the literature.

Microscopy showed a cyst wall with complete ulceration of the lining epithelium. Instead of the usual squamous lining, the inner wall was bordered by histiocytes and foreign body giant cells. Corresponding to the thickened area of the wall, there was a circumscribed nodule of histiocytes, foreign body giant cells and cholesterol clefts. Entrapped keratin material was seen as pale eosinophilic strands. The epidermal cyst had ruptured into the dermis forming the typical keratin granuloma [Figure 1]. An interesting finding was the presence of a dense collection of golden-brown pigment extra and intracellularly in the histiocytes [Figure 2] and multinucleate giant cells [Figure 3].

Suspecting a fungal infection and assuming the pigment to be hemosiderin, special stains were requested – Gomori Methenamine silver (GMS) to look for fungus and Perl’s prussian blue stain for hemosiderin. The stain for hemosiderin was negative [Figure 4]. No fungus was seen in the GMS stain, but the brown pigment had taken up a black color, indicating that the pigment is melanin [Figure 5].

Further confirmation of the melanin pigment was performed by a Masson Fontana stain, in which the pigment took black color [Figure 6] and a bleaching reaction in which the pigment completely disappeared [Figure 7].

## CASE REPORT

A 56-year-old male patient presented with a swelling in the right gluteal region of 5 years 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 3 cm diameter filled with whitish-creamy material with specks of brown to black color in the wall. The wall was thickened in one area.

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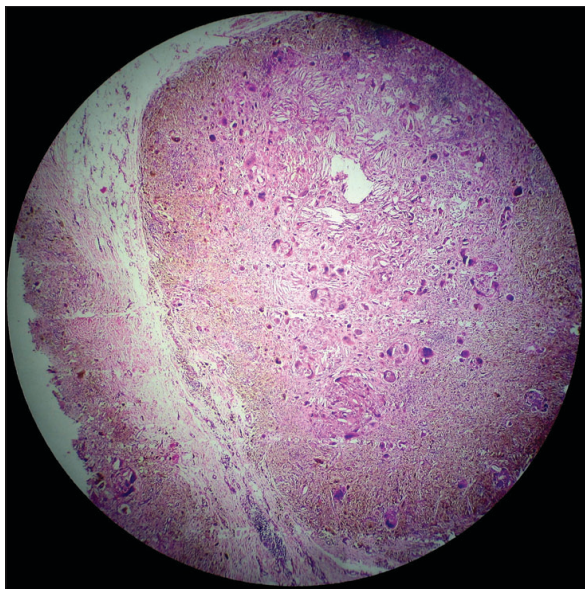
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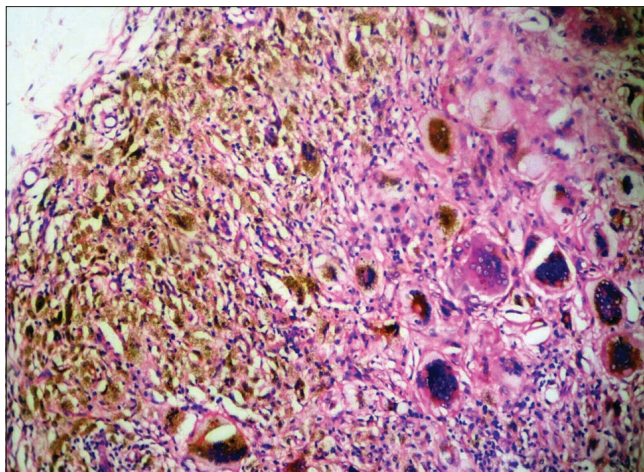
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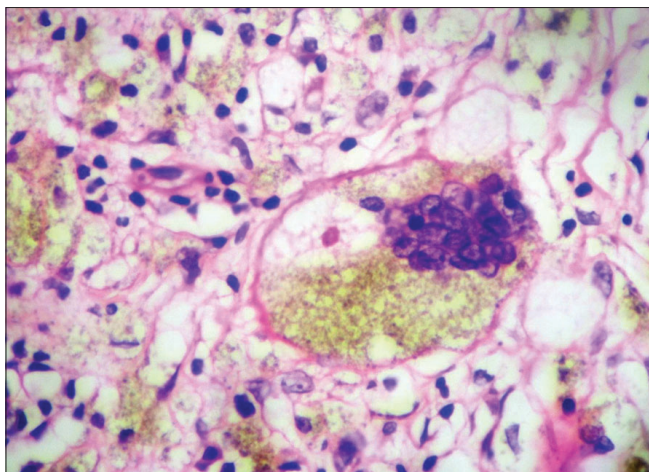
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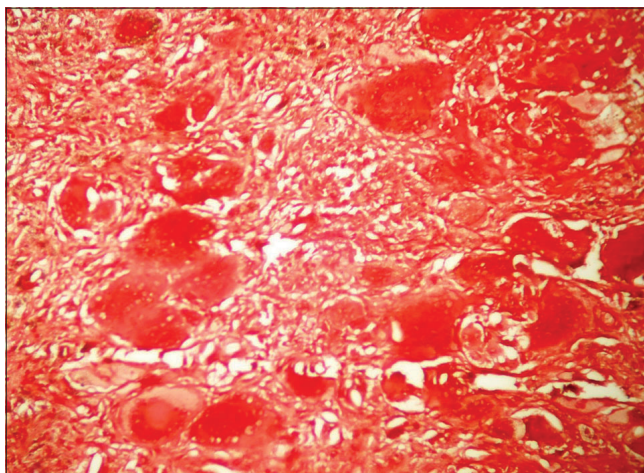
**Figure 1:** Ulcerated epidermal cyst wall with keratin granuloma in the deep dermis (H and E, scanner view x40)



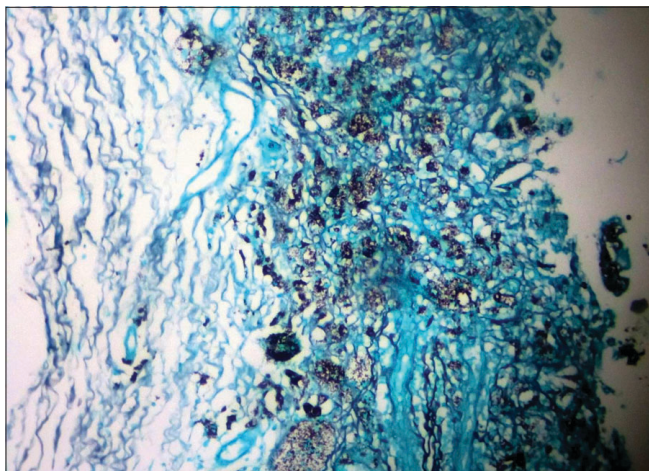
**Figure 2:** Brown pigment seen intra/extracellularly (H and E, x100)



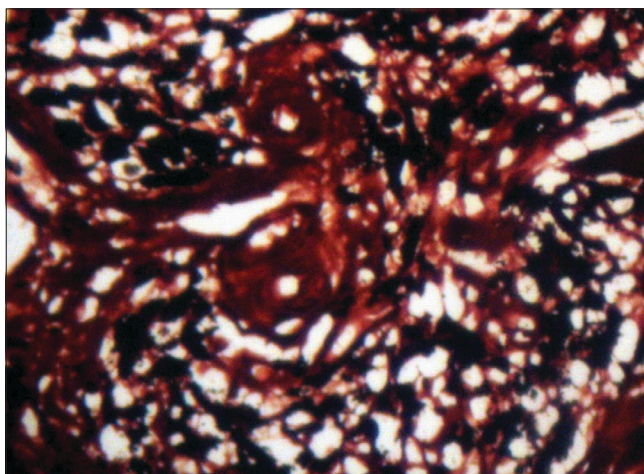
**Figure 3:** Higher power view of the pigment in multinucleate giant cells (H and E, x400)



**Figure 4:** Perls prussian blue for hemosiderin is negative (x100)



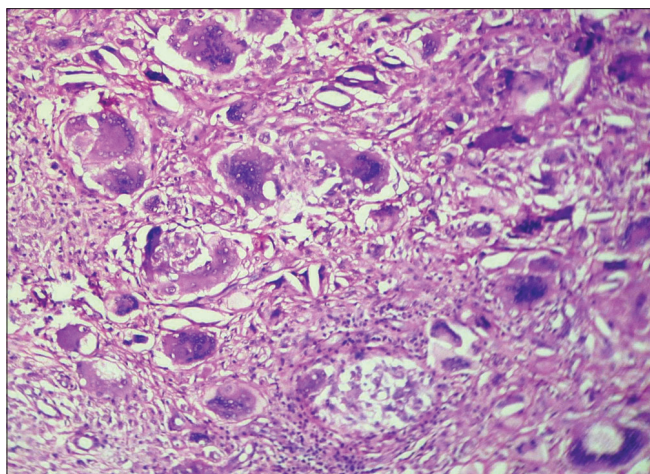
**Figure 5:** The pigment stained black in Gomori Methenamine silver stain (x100)



**Figure 6:** Masson Fontana stain showing positivity for melanin-stained black (x400)

## DISCUSSION

Epidermal cyst, also known as infundibular cyst, a common benign cystic lesion of the skin is seen in the mid/lower dermis



**Figure 7:** Bleaching reaction – The brown pigment has completely disappeared, confirming the pigment as melanin (x400)

or subcutaneous tissue. It occurs most commonly on the face, scalp, neck and trunk.<sup>[1]</sup> Usually, these cysts are solitary. Rarely, multiple infundibular cysts are seen as a manifestation of Gardner's syndrome or the basal cell nevus syndrome.<sup>[2]</sup>

Epidermal cysts are lined by stratified squamous epithelium showing epidermal type of keratinization.<sup>[3]</sup> These cysts may become infected and rupture into the dermis, resulting in a heavy inflammatory cell infiltrate in the adjacent dermis.<sup>[3]</sup> In the absence of infection, when an epidermal cyst ruptures and the contents of the cyst are released into the dermis, a 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.<sup>[1]</sup> 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.

Shet *et al.* observed that a large amount of pigment accumulation within epidermal cysts occurs after cyst rupture,<sup>[4]</sup> which was seen in our case also. Vaideeswar *et al.*<sup>[5]</sup> have also reported a case of ruptured epidermal cyst with exuberant melanophage proliferation and melanin pigment deposition.

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.<sup>[1]</sup> 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,<sup>[6]</sup> which was effectively treated by laser therapy.

Shet *et al.* studied 125 epidermal cysts from Indian patients to look for the presence of melanin, and found that 63% of the epidermal cysts (79 of 125 biopsies) showed presence of melanin pigment or melanocytes to a variable extent.<sup>[4]</sup> Extensive accumulation of melanin pigment is seen in only 10 of these 79 biopsies (12.6%). In our institution, we had 210 epidermal cysts in the year 2009 and 155 cases in 2010. None of these cases showed extensive collection of melanin pigment or proliferation of melanocytes in hematoxylin and eosin (H and E) sections. This is the first case of pigmented epidermal cyst with massive melanin deposition that was seen in our institution.

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.<sup>[2]</sup> In our case, pigmented hair shafts were not seen.

Akasaka *et al.* reported a case with increased number of melanocytes in the basal layer, large globular cells of melanocytic origin in the spinous layer and hair-germ-like aggregates of basaloid cells and melanocytes.<sup>[7]</sup> Because there was complete ulceration of the lining epithelium in our case, melanocytic proliferation in the basal layer or spinous layer could not be appreciated.

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. 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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