RESEARCH PAPER

Pigmented Epidermal Inclusion Cysts: An Uncommon Histopathological Variant — Case Series and Literature Review

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ABSTRACT

An epidermal inclusion cyst (EIC) is a common, benign cystic lesion of the skin. Ulceration of lining epithelium, rupture of cyst wall with chronic inflammation and foreign body type giant cell reaction is commonly seen. A pigmented epidermal inclusion cyst (EIC) is a rare (Citation) variant of the classic epidermal inclusion cyst that features increased melanin pigmentation within the cyst wall or keratinous debris. This pigmentation is due to the presence of melanocytes within the cyst wall or melanin transfer from adjacent melanocytes.

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INTRODUCTION

An epidermal inclusion cyst arises from cystic dilation of the infundibular portion of hair follicles. It can also arise due to traumatic inclusion of the epidermis. The epidermal inclusion cyst can present with a ruptured cyst wall eliciting local inflammation and eliciting a foreign body giant cell reaction. Massive melanin pigment deposition in an epidermal cyst is a very rare finding. Hence we report such a case with review of the literature.

CASE 1

- A 24 year old male presented with a swelling in the right knee in the popliteal fossa for a duration of 5 years. He presented to Surgical department with swelling measuring 3x2cms with presence of punctum. Incision and drainage was performed under local anesthesia. The specimen was sent for histopathological examination. The swelling was diagnosed as a sebaceous cyst so gross image is not available.
- Macroscopic examination of the swelling was a skin covered soft tissue mass measuring 2x1.6x1.6cm. External surface is grey brown and cut surface was grey brown.
- Microscopic examination of the sections studied from a cyst with surrounding fibrocollagenous tissue. The cyst is ruptured and is lined by stratified squamous epithelium with intact granular layer beneath which abundant keratin flakes noted. The surrounding fibrocollagenous tissue shows inflammatory cell infiltrates such as neutrophils, lymphocytes, plasma cells along with foreign body giant cells, abundant collection of melanocytes intermixed with keratin flakes showing reactive changes and also granulation tissue .
- To demonstrate the presence of melanin, Melanin bleach and Masson fontana – Positive for melanocytes. Perl's stain – Negative for hemosiderin laden macrophages

CASE 2

- A 77 year old female presented with a swelling in the scalp for 2 years. On macroscopic examination of the swelling was a skin covered soft tissue mass measuring 2x2x1.5cm. External surface is grey brown. Cut surface shows a cystic cavity filled with grey brown material measuring 1.6x1.5cm.
- Microscopic examination of the sections studied shows skin lined by keratinized stratified squamous epithelium. Underlying dermis shows cyst lined by stratified squamous epithelium with intact granular layer beneath which abundant keratin flakes noted. The surrounding fibrocollagenous tissue shows inflammatory cell infiltrates such as neutrophils, lymphocytes, plasma cells along with foreign body giant cells intermixed pigmented cells with keratin flakes showing reactive changes and also granulation tissue

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CASE 3

- A 41 year old female presented with a swelling in the left axilla for 3 years. On macroscopic examination a grey brown soft tissue bit with attached fibrofatty tissue measuring 2x1.5x0.5cm. Cut surface shows grey yellow area with focal grey brown area.
- Microscopic examination of the section studied shows fibrocollagenous tissue and fibroadipose tissue with focal areas showing free lamellated keratin flakes surrounded by inflammatory cell infiltrate composed of lymphocytes, plasma cells, histiocytes and few neutrophils admixed with foreign body type multinucleated giant cells. Also seen are areas of hemorrhage, pigment laden macrophages, adnexal structures and skeletal muscle bundles. No lining epithelium seen in the section studied.

DISCUSSION

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.[1] Usually, these cysts are solitary. Rarely, multiple infundibular cysts are seen as a manifestation of Gardner's syndrome or the basal cell nevus syndrome.[2] Epidermal cysts are lined by stratified squamous epithelium showing epidermal type of keratinization.[3] These cysts may become infected and rupture into the dermis, resulting in a heavy inflammatory cell infiltrate in the adjacent dermis.[3]

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.[1] 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,[4] which was seen in our case also.

Vaideeswar et al.[5] 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.[1] 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,[6] 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.[4] 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.[2] 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.[7] 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 [9][10].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[13][14]. 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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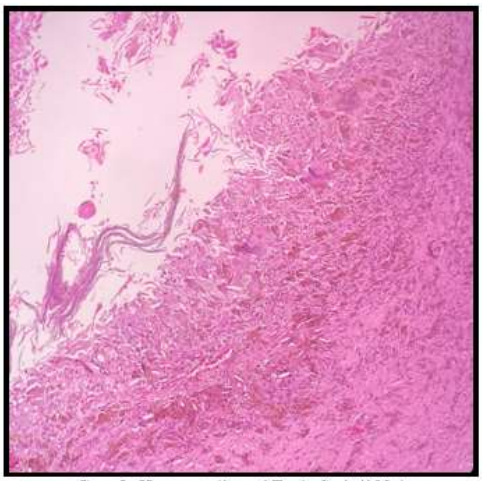
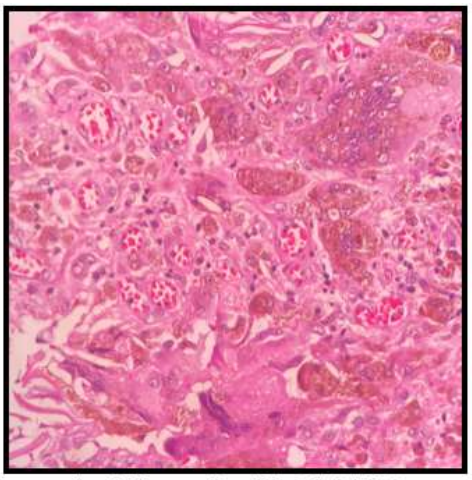
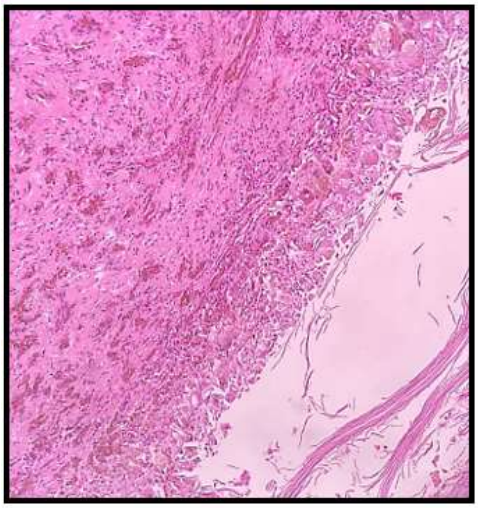


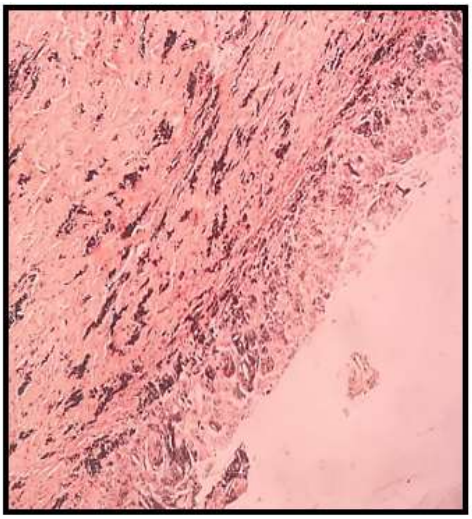
Case 1: Hematoxylin and Eosin Stain(100x)



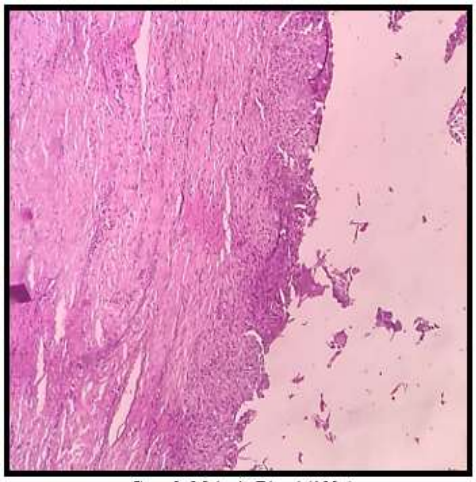
Case 2: Hematoxylin and Eosin Stain(400x)



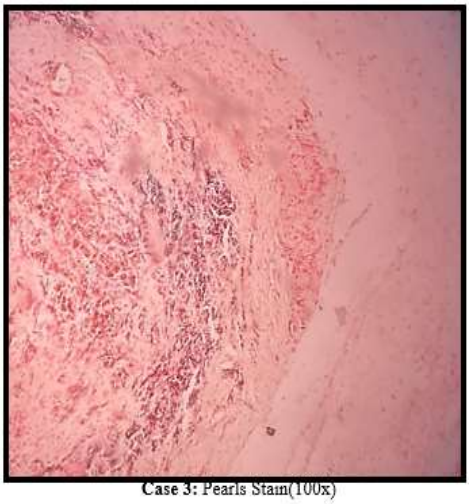
Case 3: Hematoxylin and Eosin Stain(100x)



Case 3: Masson Fontana(100x)



Case 3: Melanin Bleach(100x)



Case 3: Pearls Stain(100x)