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# SQUAMOUS CELL CARCINOMA ARISING ON A BACKGROUND OF EPIDERMAL INCLUSION CYST OVER THE GLUTEAL REGION – A CASE REPORT

General Surgery

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## ABSTRACT

**Background:** Epidermal inclusion cysts are the most common cutaneous cysts, but malignant transformation in them is extremely rare. **Methods:** We present a case of a 63-year-old female with a swelling in the right gluteal region with associated discharge that was resected based on the clinical impression and cytology of an epidermal inclusion cyst. Histopathologic exam showed atypical cells with enlarged nuclei and prominent nucleoli with brisk mitotic activity, and on IHC, these cells were positive for PanCK and P40 and negative for Sox10, confirming the diagnosis of Poorly Differentiated Squamous Cell Carcinoma. **Conclusion:** There are 56 reported cases of SCC arising on a background of epidermal inclusion cysts in literature. Although epidermal inclusion cysts are usually benign, excision is recommended for atypical cysts with high suspicion for malignancy. They should be excised intact and should undergo histopathological examination to rule out incipient malignancy.

## KEYWORDS

Epidermal Inclusion Cysts; Squamous Cell Carcinoma; Malignant transformation of cutaneous cysts; Aggressive SCC

## INTRODUCTION

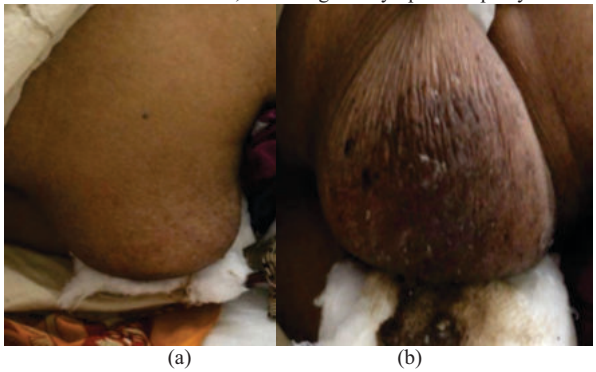
Epidermal inclusion cysts are the most common cutaneous cysts, arising from the infundibulum of the hair follicle. They often occur in areas where hair follicles have been inflamed or repeatedly irritated. They are benign, can be hereditary in rare syndromes such as Gardner syndrome, nodular elastosis with cysts and comedones (Favre-Racouchot syndrome), and basal cell nevus syndrome (Gorlin syndrome).

Malignant transformation in epidermal inclusion cysts is extremely rare, but has been reported – they include Squamous Cell Carcinoma, Basal Cell Carcinoma, Paget's Disease, Bowen's Disease, Mycosis Fungoides, Merkel Cell Carcinoma and Melanoma [1]. Incidence of SCC arising from epidermal cysts, ranges from 0.011 to 0.045% [2]. Here we report a rare case of SCC arising from within an epidermal inclusion cyst.

## CASE REPORT

A 63 year old female came to the OPD with complaints of a painless slow growing swelling in the right gluteal region since 8 months, which showed rapid growth in the last 2 months. The patient complained of discomfort while sitting but it did not bother her at night. She denied any history of trauma to the region. The rest of her history was unremarkable.

Physical examination was unremarkable, with stable vital signs, and an average body build. Examination of the swelling revealed a 12x6 cm soft, cystic, fluctuant, mobile swelling with a smooth surface over the right gluteal region, around 2 cm from the anal verge, with discharge of purulent material from one end of the swelling, with ulceration of the skin over it, and no regional lymphadenopathy.

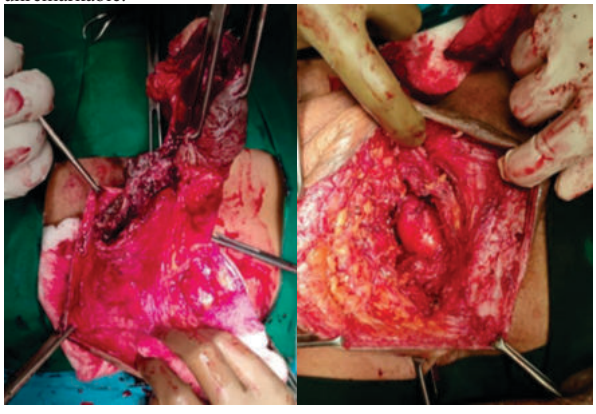


(a) (b)

**Figure 1.** (a) Cystic swelling over the right gluteal region. (b) Purulent discharge from the swelling

Routine blood investigations were within normal limits. Ultrasound of the swelling revealed fat and muscle echoes within the swelling, with focal hyperechoic areas and mild vascularity. The underlying gluteus muscle was normal, with normal fat planes. Cytopathology was suggestive of an epidermal inclusion cyst.

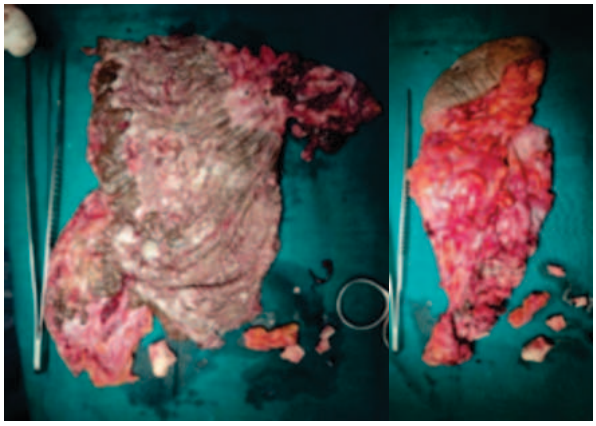
Patient underwent excision biopsy of the lesion under spinal anaesthesia, via an elliptical incision including the discharging sinus. Intra-operatively, the cyst was noted to be adherent to the lateral rectal wall. The cyst was dissected and excised intact and intoto and sent for histopathological examination. The wound was closed primarily with a negative suction drain insitu, with minimal blood loss. Patient withstood the procedure well and the post-operative period was unremarkable.



(a) (b)

**Figure 2.** (a) Wide local excision done. (b) Deep extension of the cyst, upto the right lateral rectal wall.

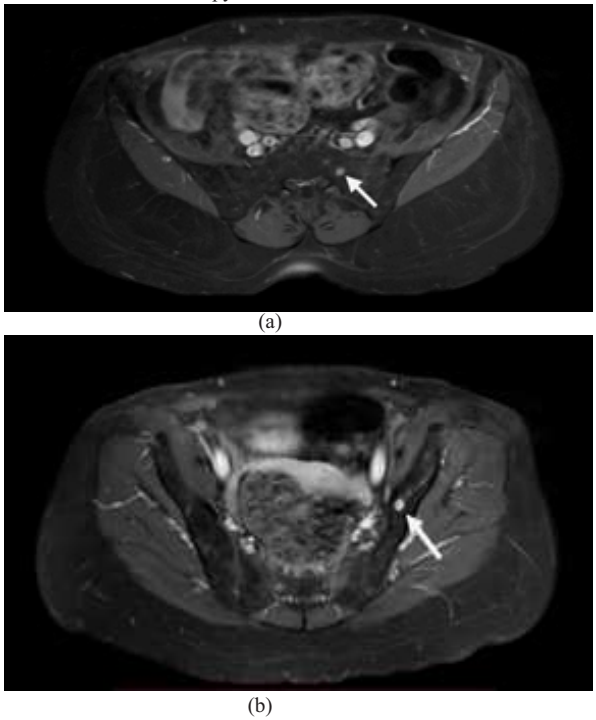
The external surface of the cyst showed fat attached grey brown areas, with the cut sections showing the presence of a sinus tract and variable grey-brown areas with epidermidization. Histopathological examination described the presence of marked hyperkeratosis, acanthosis and atypical melanocytes. These atypical melanocytes had enlarged nuclei, prominent nucleoli with brisk mitotic activity, with the cytoplasm staining positive with Masson Fontana stain – suggestive of Melanoma. Margin status of the tumor was unavailable. On Immunohistochemistry, the tumor cells were positive for PanCK and P40 and negative for Sox 10 with occasional cells staining with S100 and HMB45 – confirming the diagnosis of Poorly differentiated Squamous Cell Carcinoma, arising on a background of epidermal inclusion cyst.



(a) (b)

**Figure 3.** (a) External surface (b) Cut section of the excised cyst.

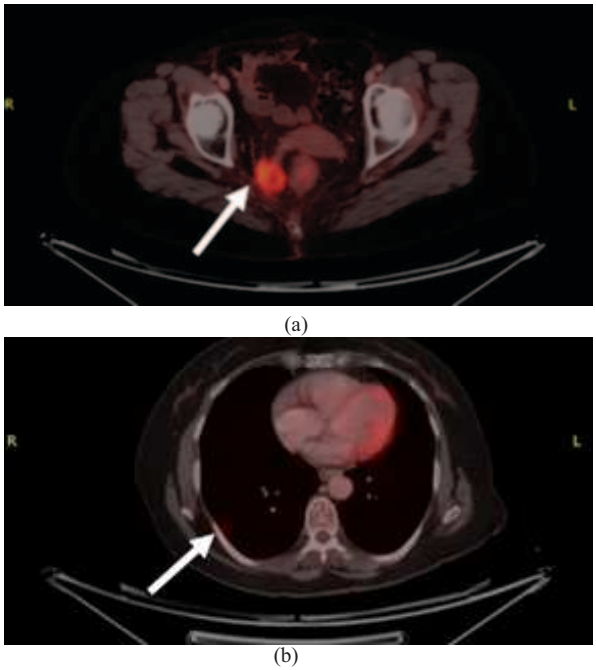
PET-CT revealed a 7x3.1x2.3 cm lobulated heterogeneously enhancing soft tissue lesion in the right mesorectal / pararectal region, extending across the right pelvic diaphragm and few discrete adjacent right lateral mesorectal metastatic lymph nodes. Patient was advised re-surgery for the same, but patient refused, hence advised Palliative Chemotherapy – Inj. Paclitaxel and Inj. Carboplatin. After 2 cycles of chemotherapy, patient underwent colonoscopy, showing no mucosal involvement of the rectum. Patient developed hip pain, radiating to bilateral legs. MRI Abdomen and Pelvis revealed near total resolution of previous lesion but showed mildly enhancing rectal wall thickening with maximum thickness of 6mm with adjacent mesorectal fat stranding, with bony metastatic lesions in the left iliac bone measuring 8.2x6mm and in the left ala of sacrum measuring 6x6mm. Patient underwent concurrent Chemotherapy – 4 cycles of Paclitaxel and Carboplatin and Radiotherapy to the primary and the sacral metastatic lesions. Patient's radiating hip pain worsened, PET-CT revealed 3.3x2.1 cm mass extending from the right pararectal region extending across the pelvic diaphragm and ischial fossa, abutting on the sacral nerve plexus and a 9mm metastatic lesion in the lower lobe of right lung. On follow up, patient was found to have a hard fixed growth on the right lateral wall of the anal canal involving more than half the circumference, around 1 cm from the anal verge, with upper limit just palpable. Patient refused further imaging and evaluation, and has been on Palliative chemotherapy since.



(a)

(b)

**Figure 4.** MRI Pelvis – axial sections showing T1 post-contrast hyperintense enhancing lesions (a) In the left ala of sacrum. (b) In the left iliac bone.



(a)

(b)

**Figure 5.** PET-CT showing lesions in (a) The right pararectal region. (b) The lower lobe of right lung.

## DISCUSSION

Epidermal inclusion cysts are usually benign and asymptomatic, but may get secondarily infected and inflamed, and rarely, undergo a malignant transformation. Primary cutaneous squamous cell carcinoma (SCC) is a malignant tumor that may arise from the keratinizing cells of the epidermis or its appendages. It is locally invasive and has the potential to metastasize to other organs of the body. Squamous cell carcinoma (SCC) arising from a cutaneous epidermal cyst is an unusual phenomenon. Clinically, a malignant transformation in an otherwise benign cyst should be suspected when there is persistent disproportionate pain, rapid growth, size > 2cm, heterogenous content within the cyst, ulceration, persistent drainage [3,4]. Confirmation of SCC in the background of an epidermal inclusion cyst requires histopathological examination and immunohistochemistry. It needs to be differentiated from SCC with cystic changes. In the former, there is absence of contact between the tumor and the epidermis [5,6].

One of the major risk factors for cutaneous SCC include chronically diseased or injured skin [7], this might be the pathogenesis behind development of SCC from longstanding epidermal inclusion cysts. Malignant transformation may occur in periodically inflamed lesions or even in asymptomatic lesions subjected to low levels of recurrent irritation (e.g. friction and pressure). In the current case, chronic pressure and friction over the cyst in the gluteal region, could be the reason behind the development of SCC in the lesion. Other etiological factors reported to be involved in malignant transformation of cysts include immunosuppression, actinic damage, and HPV infection, however, confirmation is yet to be reported [8]. The current case did not have any of the above factors contributing to the malignant transformation.

Our diagnosis of SCC arising from an epidermal inclusion cyst was made on the basis of the long-standing nature of the cyst, with recent rapid growth; the presence of both malignant and non-malignant parts in the cyst; and the non-malignant part of the tumor was contiguous with the region of atypical cell proliferation.

In 2021, Alkul S et al. reviewed 382 case reports in the literature from 1976 to 2021, and found that only 56 cases had characteristic histopathologic features consistent with SCC arising from within a epidermal inclusion cyst [9]. Of the literature till date, incidence of SCC arising from epidermal cysts, ranges from 0.011 to 0.045% [2].

## CONCLUSIONS

Although epidermal inclusion cysts are usually benign, excision is recommended for large or long-standing cysts with recent rapid

growth, or recent ulceration, cysts which do not respond to medical therapy, or those that recur. They should be excised intact and intoto, should undergo histopathological examination to rule out incipient malignancy, and should undergo re-excision if necessary. Our case presented with a symptomatic lesion (recent rapid growth and discharge), but previous studies have demonstrated that clinically asymptomatic lesions may also contain malignancy [1,9,10,11,12].

Although the majority of cases reported in literature were successfully diagnosed and treated without complications and recurrence, three cases had advanced disease with metastases and succumbed within 5–10 months of initial presentation [13,14,15], while one patient had recurrence with metastases and survived following chemotherapy. Our patient had residual disease and bone and lung metastases, not responding to chemo-radiotherapy, hence long-term prognosis appears grim. Further studies are warranted to determine long-term prognosis of these patients.

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