



Bowen's Disease and Eccrine Porocarcinoma Developed after Herniorrhaphy: A Case Report and Literatures Review

Chea K^{1,2}, Huang Q¹, Chena B^{1*} and Hua X^{1*}

¹Department of Dermatology, Shenzhen Hospital, Peking University, China

²Shantou University Medical College, China

Abstract

Eccrine Porocarcinoma is a rare cutaneous tumor with high invasiveness and risk of metastasis. It usually presents clinically as red nodule, plaque, ulcerated lesion or neoplasm. It is hard to diagnose without histopathology. Surgery is the mainstream treatment. We are reporting a case of eccrine porocarcinoma arising in right mons pubis, where had Bowen's disease previous after herniorrhaphy, as well as reviewing literatures about eccrine porocarcinoma.

Keywords: Eccrine porocarcinoma; Bowen's disease; Scar; Herniorrhaphy; Trauma

Introduction

Eccrine Porocarcinoma (EPC) is a rare cutaneous tumor which was first described by Pinkus and Mehregan in 1963 [1] and the term of eccrine porocarcinoma was coined by Mishima and Morioka in 1969 [2]. It has low morbidity, high invasiveness and high risk of metastasis or recurrence [3]. The clinical presentation is red nodule, plaque or neoplasm accompanied by ulcerated lesion, erosion or scab [4,5]. Diagnose is established on histopathology. There is no consensus about treatment at present while it is reported that Mohs Micrographic Surgery (MMS) can reduce the local recurrence [6].

Case Presentation

A 73-year-old male patient presented with dark red plaque in right mons pubis for 2 years in April 2021. He accepted an abdominal surgery for right oblique inguinal hernia in other hospital 6 years ago (2015) and the mesh was placed (the material was unspecified). A 7 cm long white linear scar was formed after herniorrhaphy. The patient presented with dark red plaque in right mons pubis 2 years ago (2019) with ulceration, scab and no pain. No treatment was accepted. Physical examination revealed dark red and erythematous plaques with well-circumscribed and rough surface in right mons pubis near groin. The lesion was 4 cm × 3 cm in size with ulceration and scab. There was a 7 cm white linear scar observed on the medial side of the plaques (Figure 1A). Histopathological examination after surgery showed feature characteristic of Bowen's Disease (BD): Atypical squamous cells with infiltrative growth into interstitial, polygonal oncocytes with hyperchromatic, enlarged nuclei and atypical mitoses (Figure 2A). The incision recovered well during follow-up visit after therapy. However, 1 year later (September 2022) patient visited again with dark red plaques in the same area of previous BD for a month. It presented as grain of rice in size and grew gradually with spontaneously ulceration in surface and no pruritus. It grew a little better treated with topical medication. Physical examination revealed two dark red plaques, 1 cm × 1 cm in size, with rough surface, ulceration and scab in right mons pubis near groin (Figure 1B). Histopathology examination revealed the characteristic of EPC: Papillary hyperplasia in tumor cells, with polar disorder, Bowenoid change and mitoses. Ductal differentiation was observed (Figure 2B). Patient accepted MMS in other hospital and the follow-up by telephone after half a year showed that the oral recovery was good and no new tumor was found. Patient denied of other systemic disease and was free of the history of personal and family.

Discussion

Eccrine Porocarcinoma (EPC) is a rare cutaneous tumor with morbidity of 0.005% to 0.01% [3,4,7]. EPC has high risk of metastasis. About 20% cases had lymph node metastases at the first

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*Correspondence:

Xiaoping Hu, Department of Dermatology, Shenzhen Hospital, Peking University, Lianhua Street, 1120, Futian District, Shenzhen, 518036, China

Bancheng Chen, Department of Dermatology, Peking University Shenzhen Hospital, Lianhua Street, 1120, Futian District, Shenzhen, 518036, China

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Figure 1: **a:** The lesion was 4 cm x 3 cm in size with ulceration and scab. There was a 7 cm white linear scar observed on the medial side of the plaques. **b:** Two dark red plaques, 1 cm x 1 cm in size, with rough surface, ulceration and scab in right mons pubis near groin.

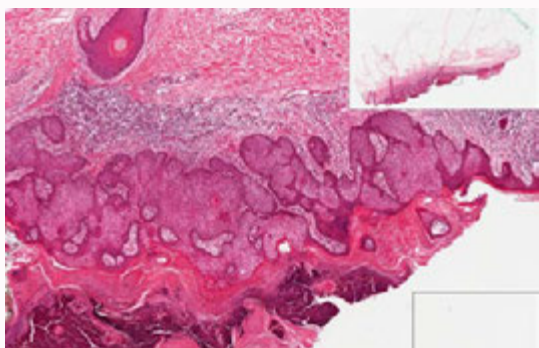


Figure 2a: (H&E, x300).

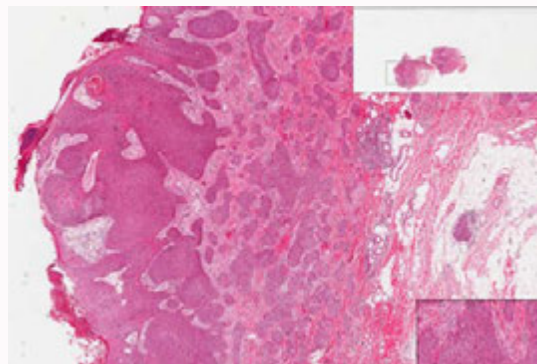


Figure 2b: (H&E, x200).

visit. The most common organ to metastasis is the nearby lymph node followed by lung. EPC is highly invasive with 67% mortality of the patients who have lymph node metastasis [4]. It may malignantly transform of benign poroma or arise as malignancy in first presentation [7]. The patient in our case had a short course of disease with 1 month from the discovery of the mass to diagnosis. Therefore, it was unlikely that the malignant was transformed from benign eccrine pore tumor. Sometimes it develops base seborrheic keratosis or Bowen's disease [8-11]. The mean age of diagnose is 65-year-old and mean course of disease is 5.7 years. There is approximately equal incidence among males and females [3,12]. Tumors are found at higher rates in head/neck region and lower limbs followed by trunk [3,12].

Clinical characteristic of EPC is various. It may present as nodular lesion, plaque or neoplasm with swell, ulceration, seepage, scab, spontaneous bleeding, itching or pain [4,5]. For its multiformity of clinical presentation, EPC is often misdiagnosed as basal cell carcinoma, squamous cell carcinoma, sebaceous carcinoma or pyogenic granuloma [5]. Therefore, biopsy is necessary for diagnose.

Histopathology shows the feature of poroma-epidermis is thickened with hyperkeratosis or parakeratosis, the tumor cells are smaller than acanthocyte showing basophilic, ductal differentiation and the ducts with eosinophilic pericarp may be seen. The characteristic of malignant tumor also could be seen-pleomorphic cells with nuclear hyperchromasia and increased mitotic activity [5,13]. Immunohistochemical analysis is favorable for diagnose. It is reported that positive Epithelial Membrane Antigen (EMA) and negative Carcinoembryonic Antigen (CEA) indicate the diagnosis

of EPC [5]. However, a few cases with positive EMA were reported [7,14]. Meanwhile, some researchers pointed out that EMA was not a useful marker to make a differential diagnosis with Bowen's disease in EPC with Bowenoid change [15]. CK19 is a helpful marker to distinguish EPC and Bowen's disease [5,8].

For its rarity and seldom reports, there is no guideline of treatment yet. The therapy for EPC includes fulguration, chemotherapy, radiotherapy, Wide Local Excision (WLE) and Mohs Micrographic Surgery (MMS) [5,6]. Given its high invasiveness and rate of metastasis, complete resection is extremely important for prognosis. Most of cases accepted WLE (average >2 cm to 3 cm surgical margins) with 20% for local recurrence. Studies have shown that patients who accepted MMS have a lower local recurrence rate than WLE [6]. Amitotic index of more than 14 mitotic cells per high-power field, lymphatic vessel invasion, and a tumor depth exceeding 7 mm often predict a worse prognosis [6]. Due to its high rate of metastasis to lymph nodes, regional lymph node dissection may necessitate. However, there is insufficient data to support to routine lymph node dissection currently because of its low morbidity [12].

The cases of EPC developed by postoperative of Bowen's disease are rare. We searched in PubMed by using the key words "(eccrine porocarcinoma) and (Bowen's disease)" and found only 12 cases (Table 1) [8-10,15-24]. KAO [17] described "Bowen's disease with invasive adnexal carcinoma (BD-CA)", suggesting that BD-CA has the potential for adnexal differentiation [18], but in our case, postoperative pathology of Bowen's disease showed clean incisal edge, which indicated that the possibility of secondary EPC after Bowen's disease could be excluded. A case of EPC rising in a scar was reported in China and the authors presented that scar might be associated with the development of EPC [14]. The patient in our case suffered from local excision for Bowen's disease 1 year ago and a white atrophic scar was formed. The development of EPC at this location might associate with the scar. Patrick A [19] reported a case of EPC rising at vulva, which had a history of cervical cancer for 10 years and received the treatment of radiotherapy and implants. At the same time, De Giorgi et al. [20] pointed out that inflammation caused by trauma and foreign bodies could promote the occurrence and metastasis of tumors. The patient in our case also accepted herniorrhaphy and the mesh was placed. After that Bowen's disease appeared near the scar of incision, and EPC occurred at the same site after the local excision of the Bowen's disease, which suggested that the occurrence of cutaneous tumors might related of surgery or foreign bodies. Searching the cutaneous cancer related to surgery or foreign bodies in PubMed, we found a handful of case reports on cutaneous

Table 1: The cases of EPC with other skin lesions reported.

Authors	Year	Journal	Gender	Age	Sit	Other lesions
Zheng et al. [8]	2015	Clin Exp Dermatol	F	63	right axilla, right groin, right waist and medial left thigh	Bowen's disease
Kottler et al. [9]	2014	Ann Pathol	F	88	Right upper limb	Bowenoid changes
Hoshina et al. [10]	2007	Clin Exp Dermatol	F	66	abdomen	Seborrheic keratosis; Bowen's disease
Jiuli et al. [14]	2011	The Chinese Journal of Dermatovenereology	F	72	low part of right face	scar
Mariko et al. [15]	2003	Acta Dermato-Venereologica	F	70	left thigh	Bowenoid changes
Chen et al. [16]	2005	Dermatol Surg	M	58	left side of the chest wall	multiple Bowen's disease
Kamiya et al. [18]	2006	J Dermatol	F	66	lateral side of her right lower leg	Bowen's disease
Kamiya et al. [18]	2006	J Dermatol	F	87	right temple	Bowen's disease
Kamiya et al. [18]	2006	J Dermatol	F	94	anterior aspect of her right tibia	Bowen's disease
Lowney et al. [21]	2011	Clinical dermatology	F	56	right shin	Bowen's disease
Rajesh et al. [22]	2015	Indian J Surg	F	69	back	Bowen's disease
Limin et al. [23]	2009	Diagn Cytopathol	M	86	left lateral ankle	Bowen's disease
Cribier et al. [24]	1999	Eur J Dermatol	F	75	leg	Bowen's disease and actinic keratoses

cancer such as squamous cell carcinoma and Marjolin's ulcer related to tattoos [25,26], synthetic hair grafts [27], grenade fragments [28], total knee arthroplasty [29]. However, in the cases above the foreign bodies came into direct contact with the skin, which was different to our case. In our case, the mesh was placed in the body without direct contact with the epidermis after herniorrhaphy. Therefore, the correlation between the mesh and EPC and Bowen's disease occurred at the surgical incision remained to be investigated. The case also suggested the in clinical work, the past medical history of patients, including trauma history and surgical history, should not be ignored to further more exploration for correlation between trauma, foreign bodies and cutaneous tumors.

Conclusion

In summary, EPC is rare with less than 300 cases reported worldwide and only 12 cases of Bowen's disease complicated or secondary to EPC. We reported a case of Bowen's disease and EPC after herniorrhaphy, which remained dermatologist pathological biopsy and tissue immunological examination should be performed when patient get new lesion with a history of Bowen's disease to avoid misdiagnosis of EPC. However, the link between these two diseases is unclear yet and more clinical data is needed to explore.

Authors' Contribution

All authors made the contributions to this case and review. Bancheng Chen provided the clinical case. Keying Che prepared the draft of this manuscript. Qiufeng Huang searched the literatures. Xiaoping Hu contributed to revise the manuscript. All authors revised and approved of the final manuscript.

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