

Squamous cell carcinoma arising from an epidermal cyst

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There are few case reports in the English literature concerning malignant transformation of an epidermal cyst into squamous cell carcinoma. We report a case of squamous cell carcinoma arising from a 40-year epidermal cyst in the proximal left thigh of a 74-year-old man. The epidermal cyst had increased in size in the previous 3 years. The patient presented with a discharge from the lesion. Ultrasonography and magnetic resonance imaging of the lesion suggested malignancy. An intra-operative frozen section of an incisional biopsy of the lesion found it to be an epidermal cyst with suspected malignant change, so a marginal excision of the lesion was performed. Squamous cell carcinoma arising from the epidermal cyst was confirmed histologically. A wide excision was done in order to obtain an adequate resection margin. A computed tomographic scan of the thorax and abdomen found no evidence of metastases. No evidence of recurrence was noted in the latest follow-up, 2 years postoperatively.

Introduction

Epidermal cysts and squamous cell carcinoma are both commonly encountered in practice. Malignant transformation of the benign lesion is rare. There are few reports of such malignant transformation in the literature.

Case report

A 74-year-old man presented to our department in February 2005 with a recent onset of haemoserous discharge from a mass on the posteromedial aspect of the proximal left thigh. The mass, that had been present for 40 years, began to enlarge progressively during the 3 years prior to his presentation. Clinically, the mass was cystic and measured 15 cm in diameter. There were no palpable groin lymph nodes.

Ultrasonography and magnetic resonance imaging (MRI) revealed a complex cystic mass on the posteromedial aspect of the left thigh with irregular walls and septations (Fig 1). From the MRI, the mass measured 13 cm x 9.5 cm x 12 cm and the superior part of the mass was closely related to the gracilis muscle. The differential diagnosis included a neoplastic lesion with cystic degeneration or infection.

Because the size of the lesion and MRI findings suggested malignancy, an incisional biopsy was performed to obtain a pathological diagnosis and to plan treatment. On exploration, the mass was thin-walled and multi-lobed. After incising the mass, a large amount of pus-like material leaked from the cavities. An intra-operative frozen section reported that it was an epidermal cyst with nuclear atypia, suggestive of malignant change

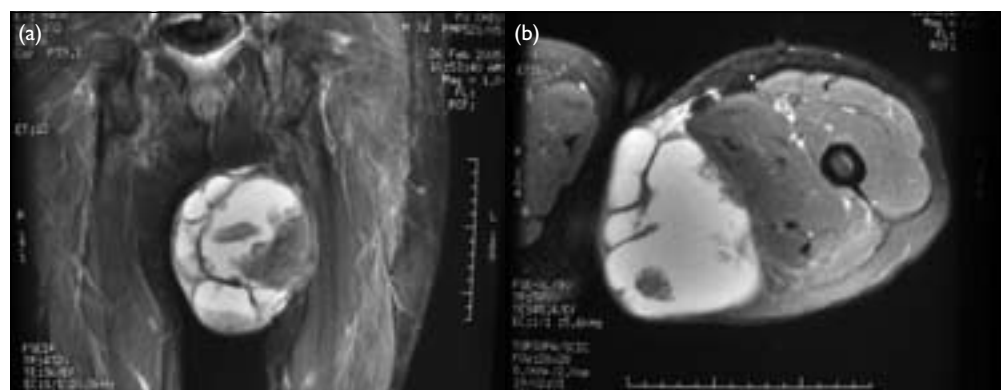


FIG 1. Magnetic resonance image of the left thigh mass (T2 image)

The mass is markedly hyperintense with obvious septations and hypointense materials in some of the compartments

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Carcinoma, squamous cell; Epidermal cyst; Skin neoplasms

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without definite stromal invasion but the exact nature of the tumour and its histological grading was undetermined. A marginal excision of the mass along with the overlying skin was performed because of the continuous leaking of pus-like material from the mass and the difficulty of sealing the multiple cavities within the mass. The pathological examination of the excised specimen found a well-differentiated squamous cell carcinoma arising from an epidermal cyst. The excision margin was 2 to 3 mm in most areas except one area with less than 0.5-mm margin. Re-excision with a 10-mm margin around the previous scar was performed. A histological examination of the excised tissue found no residual tumour (Fig 2).

A computed tomographic scan of the pelvis, abdomen, and thorax (with contrast) showed no evidence of distant organ or lymph node metastases. The wound healed. There was no clinical evidence of recurrence in the latest follow-up done 2 years postoperatively.

Discussion

The incidence of non-melanoma skin cancer is 0.34 per 100 000 Chinese people in Hong Kong.¹ Thus, cutaneous squamous cell carcinoma is not rare. It is well-recognised that chronic skin disease is a predisposing factor for squamous cell carcinoma.² Squamous cell carcinoma arising from an epidermal cyst is rarely reported. In 1999, López-Ríos et al³ found 27 reported cases in a search of the English literature covering the past 55 years. After critically reviewing those case reports, including his own case, he found that only eight were well-documented with a microscopic description and photomicrographs corresponding to a squamous cell carcinoma arising in a cutaneous epidermal cyst. In 2003, Cameron and Hilsinger⁴ identified two more such well-documented cases, one from a case report in the English literature and one from his centre. Besides these 10 cases, a series consisting of five cases retrieved from the pathology files of three centres has been published,⁵

making, with the addition of our patient, a total of 16 well-documented case reports in the English literature.

Reported rates of malignant transformation of an epidermal cyst into cutaneous squamous cell carcinoma range from 0.011 to 0.045%.³⁻⁵ Among the 16 well-documented cases, the mean age was 58.6 (range, 21-92) years.^{4,5} These cases included 10 male and 5 female patients.^{4,5} Nine of the 16 lesions were on the head and neck while the other seven lesions were on the trunk or limb.^{4,5} Of the 10 cases identified by López-Ríos et al³ and Cameron and Hilsinger,⁴ the size of the lesions ranged from 1.5 to 10 cm (mean, 5 cm) and the duration of lesions ranged from 2 to 132 months (mean, 33.5 months).^{3,4} In two cases the lesion duration was much longer, being 10 and 11 years respectively, while the lesion duration of the other cases was within 6 months.^{3,4} In our case, the size and duration of the lesion were 13 cm and 40 years respectively, representing a lesion with the largest size and longest duration of all well-documented case reports. The lesions in all reported patients were excised. An unusual gross appearance was reported in two cases, with either a thickened cyst wall or granular tissue within the cyst.⁴ All excised epidermal cysts had both a routine pathological examination⁴ and a close examination of the thick-walled areas in the cutaneous cysts to exclude malignancy.³ A metastasis was found in only one patient who had a primary lesion on the thigh with an inguinal lymph

表皮囊腫引致的鱗狀細胞癌

醫學文獻只有很少關於表皮囊腫轉化成鱗狀細胞癌的病例報告。本文描述一名74歲男性病人，左大腿處的表皮囊腫達40年之久，後轉化成鱗狀細胞癌。最近3年內，表皮囊腫不斷增大，並有溢液。超聲波及磁共振檢查均顯示有惡性組織。在患處的活檢中，術中冰凍切片發現表皮囊腫似乎有惡性病變，後進行胞膜切除。組織學證實是由表皮囊腫轉化成的鱗狀細胞癌。為手術切緣於是進行廣泛切除。術後電腦斷層掃描顯示胸腹沒有癌轉移，兩年內亦沒有復發跡象。

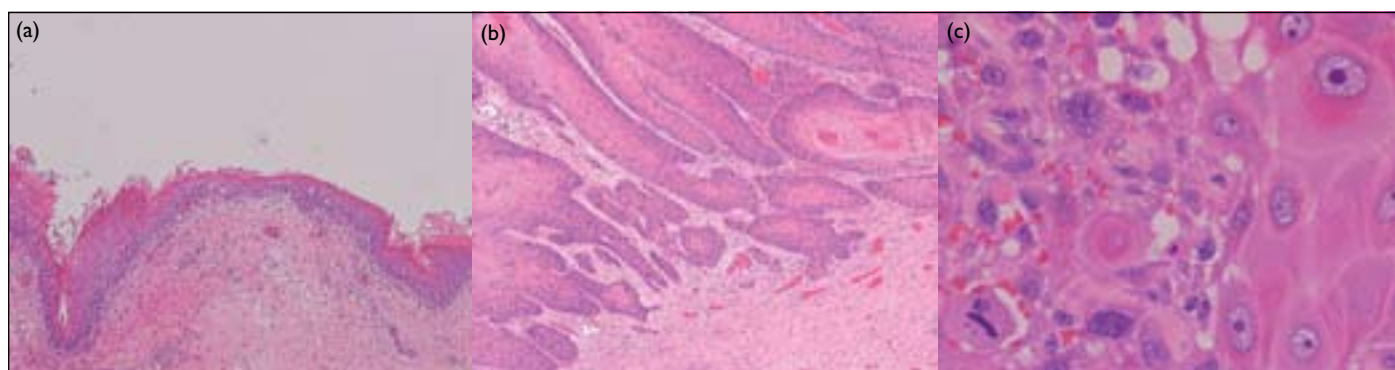


FIG 2. Histological specimen of the tumour

(a) The remaining benign epidermal cyst wall of the left thigh mass; (b) malignant squamous cell carcinoma arising from the epidermal cyst; and (c) high-power view of cavity wall showed stratified polygonal tumour cells and desmoplastic stroma

node metastasis. Inguinal lymph node dissection was performed in this case. A second excision procedure was needed in two other cases because the wound margins tested positive for carcinoma after the first excision procedure.⁴

Cutaneous squamous cell carcinoma has a multitude of clinical, histological, and aetiological subtypes with widely varying clinical behaviours and metastatic potential.⁶ The optimal treatment and the surgical margins for excision should be guided by the risk of metastasis, tumour size, differentiation, depth of invasion and perineural invasion.⁷ It is proposed that excision margins should be at least 4 mm for all but high-risk tumours (those that are 2 cm or larger, histologically grade 2 or above, show invasion of subcutaneous tissue and located in high-risk areas), in which case a margin of at least 6 mm is recommended.⁸ Intra-operative frozen section is an accurate means of evaluating the margins of squamous cell carcinoma,⁹ and is commonly used in cutaneous surgery. Few of the case reports discuss the subtypes of squamous cell carcinomas arising from epidermal cysts. Thus, the clinical course, prognosis, approach, and optimal management of this disease entity are not well-established.

In our patient, the frozen section of the incisional biopsy showed suspected malignancy arising from the epidermal cyst, but the exact pathology and the desirable resection margin could not be determined. Because it would be too tedious and time consuming to perform an intra-operative frozen section to assess the surgical resection margin for a lesion measuring 13 cm, further excision was not attempted during the first operation. Taking into consideration the

undetermined risk of metastasis and the large size of the tumour, it was decided to re-excise the tumour with a 10-mm margin even though the squamous cell carcinoma was well-differentiated.

The incidence of metastatic squamous cell carcinoma secondary to chronic inflammatory lesions ranges from 10 to 40%.^{10,11} Computed tomographic scans (with contrast) of the thorax, abdomen, and pelvis were performed in our patient to detect any metastases to the lymph nodes or other organs. Although a sentinel lymph node biopsy can accurately diagnose subclinical lymph node metastases in squamous cell carcinoma, whether such early detection and treatment of subclinical nodal metastases will lead to improved rates of remaining disease-free or overall survival is not known.¹² Recently, a retrospective analysis of 12 cases has shown that positron emission tomography (PET) is useful for the detection of squamous cell carcinoma metastases,¹³ but many questions, like diagnostic sensitivity, remain unanswered. Therefore, the role of sentinel lymph node biopsies or PET in the management of cutaneous squamous cell carcinoma is not yet defined.

Conclusions

This is one of very few well-documented case reports of squamous cell carcinoma arising from an epidermal cyst. The clinical features and management of this disease entity need further study for thorough understanding. This case also emphasises the importance of suspecting malignancy when faced with a mass that is large or changing size rapidly.

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