Perineal squamous cell carcinoma arising from an epidermal cyst: a case report

Byung-Soo Park¹, Dong Hoon Shin², Soo-Hong Kim¹, Hyuk Jae Jung¹, Gyung Mo Son¹ and Hyun Sung Kim¹*

Abstract

Background: Epidermal cysts and squamous cell carcinomas (SCCs) are common skin lesions. However, a malignant change in an epidermal cyst is very rare. The incidence of a malignant change from an epidermal cyst to cutaneous SCC is 0.011–0.045%. In particular, malignant transformation of an epidermal cyst in the perineum is extremely rare. To date, three cases have been reported in the English literature.

Case presentation: We report a case of 51-year-old male with an approximately 15-cm perineal mass. This mass started to grow suddenly 4 months previously and caused great discomfort in the perineum due to the large size. The patient underwent excision of the mass with a negative margin. Histopathological analysis confirmed a microinvasive SCC arising from a proliferating epidermoid cyst.

Conclusions: Even if benign tumors are suspected, a change in size, pain, ulceration, or discharge should indicate the need for surgical resection due to the possibility of a malignant change.

Keywords: Squamous cell carcinoma, Epidermal cyst, Malignant transformation, Perineum, Excision

Background

Epidermal cysts and squamous cell carcinomas (SCCs) are common skin lesions [1]. However, cutaneous SCC arising from an epidermal cyst is quite rare [2]. The most common sites are the head and neck, and it has also been reported in the trunk, limb, and gluteal regions [3]. In particular, SCC arising from an epidermal cyst in the perineum is extremely rare. To the best of our knowledge, three cases have been reported in the English literature until now [1, 4]. Here, we report the case of a perineal cutaneous SCC arising from an epidermal cyst.

Case presentation

A 51-year-old male was referred to our center due to a large perineal mass. The mass was first discovered 30 years ago as a chestnut-sized small movable cystic nodule. Subsequently, he watched himself for a long time because of no unusual changes. The cyst started to grow suddenly 4 months previous to the visit and caused great discomfort in the perineum due to its large size.

On physical examination, an approximately 15-cm cystic mass was observed in the left perineum near the anus. There was no sign of inflammation such as tenderness or redness. Ulceration or discharge was not observed. On a digital rectal examination, there were no specific findings in the anus. A colonoscopy was performed and was unremarkable. Magnetic resonance imaging revealed a 6.7 × 16 cm lobulated mass in the medial aspect of the left perineum with an intermediate signal on T1WI, a high signal on T2WI, and peripheral wall and internal septal enhancement (Fig. 1). There was no significantly enlarged inguinal lymph node. Laboratory values were within normal ranges, except for TPLA (+), FTA-ABS IgG (+), and FTA-ABS IgM (−). Preoperative pathologic tests such as fine needle aspiration and core needle biopsy were not performed because the mass was considered as an epidermal cystic mass.

The patient underwent excision of the mass with a negative margin. On exploration, a cystic mass with sebum and keratin was identified in the left perineum (Fig. 2). It had a clear margin without invasion of anal sphincter and urologic tissues. Because the preoperative diagnosis was a cystic mass such as an epidermal inclusion cyst made by the MR pelvis, the surgery was performed with minimal gross margin. The skin was preserved as...
much as possible and closed easily without any recon-
struction. Histopathological analysis showed the cyst had
a thin wall composed of benign squamous epithelium.
Some sections of the wall were thick, indicating a microin-
vasive squamous cell carcinoma (pTisN0M0, pStage0)
(Fig. 3).
The patient was discharged 2 days after the surgery
without a significant postoperative complication. We
performed regular follow-up examinations with CT every
6 months, and he showed no evidence of recurrence at
3 years postoperatively.

Discussion and conclusions
An epidermal cyst is a benign disease caused by invagin-
ation of epidermal elements into subcutaneous fat from a
hair follicle [5]. Although it is a common skin lesion, malig-
nant transformation of an epidermal cyst is very rare [1, 2].
The incidence of a malignant change from an epidermal
cyst to cutaneous SCC is 0.011–0.045% [6]. A recent review
of the literature has found 41 well-documented cases of
SCC arising from cutaneous epidermal cysts [2]. Most cases
have occurred in the head and neck, and cases in trunk,
limb, and gluteal tissues have also been reported [2, 3]. In
particular, perineal SCC arising from an epidermal cyst is
extremely rare, and to date, three cases have been reported
in the English literature. In one case each, the lesion was
found in the right labia major of a 76-year-old woman [4],
in the scrotum of an 86-year-old man in 2013 [7], and in
the right vulvar region of a 65-year-old woman in 2016 [1].
The cause of a malignant change in an epidermal cyst is
not yet clear. It is known that chronic inflammation or
infection can trigger this event, which seems to create
dysplasia and/or malignant changes. The most frequent
reports are SSCs in the head and neck regions, indicating
that the change may be a side effect of ultraviolet radi-
ation. Exposure to ultraviolet radiation and chronic stimuli
can be triggered [4, 5]. Although a study was conducted to
determine whether human papilloma virus (HPV) can
cause malignant transformation of epidermal cyst, the
presence of HPV in the lesions was not demonstrated [8].
However, a recent report showed that p16 immunoreac-
tivity was detected in the epidermal cyst wall and accom-
panying invasive tumors, which is currently used as a
surrogate for HPV detection in SCC. Therefore, these
authors suggested that SCC arising from an epidermal cyst
might be associated with HPV infection [1]. Another pos-
sible explanation could be that epidermal cysts contribute
to induce local immune destabilization, as observed in
other pathological conditions [9]. Further research is still
needed on this issue.
Clinically, it is difficult to distinguish between benign
cystic lesions and malignant lesions. When an epidermal
cyst changes in size or is associated with atypical condi-
tions such as pain, ulceration, or discharge, cancerous
transformation should be considered [10]. In the present
case, a dramatic change in size was observed. SCC arising from an epidermal cyst should be distinguished from a cystic change of SCC; therefore, for diagnosis, it is essential to show no connection between the tumor and epidermis on a microscopic level [11].

Treatment is not different from that for other skin malignant lesions. Excision with a proper margin is the standard of care. It is recommended that minimal margin of excision be 4 mm, but for high-risk tumors, a 6-mm margin is suggested. Advanced cancer requires extensive surgery with a resection margin of 2 cm or more [12]. The postoperative course is mostly known to have low malignant potential, but there are some reports of an aggressive course, such as metastasis and even mortality [1].

Conclusion
In conclusion, malignant transformation can occur, even in benign skin lesions. In particular, when there is an atypical situation such as a change in size, pain, ulceration or discharge, surgical resection should be performed due to the possibility of a malignant change. After complete resection, a thorough pathologic examination should be conducted to confirm a malignant transformation.

Abbreviations
HPV: Human papilloma virus; SCC: Squamous cell carcinoma

Availability of data and materials
As a case report, all data generated or analyzed are included in this published article.

Authors’ contributions
HSK and BSP designed the report; SHK and HJJ collected the patient’s clinical information; GMS analyzed the patient’s data; DHS performed the pathologic analyses; and BSP wrote the paper. All authors read and approved the final manuscript.

Ethics approval and consent to participate
The case report was exempt from review by the Institutional Review Board of Pusan National University Yangsan Hospital.

Consent for publication
The patient provided written consent to publish this case report.

Competing interests
The authors declare that they have no competing interests.

Publisher’s Note
Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

Author details
1Department of Surgery, Pusan National University Yangsan Hospital, 20 Geumo-ro, Mulgeum-eup, Yangsan, Gyungsangnam-do 50612, Republic of Korea. 2Department of Pathology, Pusan National University Yangsan Hospital, Yangsan, Republic of Korea.

Received: 29 March 2018 Accepted: 5 July 2018
Published online: 28 July 2018

References
1. Sze S, Richmond I, Bickers A, Saha A. Squamous cell carcinoma arising from a vulval epidermal cyst. J Obstet Gynaecol Res. 2016;42:1623–6.
2. Frank E, Macias D, Hondorp B, Kerstetter J, Inman JC. Incidental squamous cell carcinoma in an epidermal inclusion cyst: a case report and review of the literature. Case Rep Dermatol. 2018;10:631–8.
3. Sridevi HB, Shariff MH, Pushpalatha Pai K. Squamous cell carcinoma arising in an epidermal cyst. Indian J Cancer. 2015;52:335–6.
4. Sumi Y, Yamamoto N, Kyosawa T. Squamous cell carcinoma arising in a giant epidermal cyst of the perineum: a case report and literature review. J Plast Reconstr Aesthet Surg. 2012;65:209–11.
5. Cameron D. Squamous cell carcinoma in an epidermal inclusion cyst: case report. Otolaryngol Head Neck Surg. 2003;129:141–3.
6. Lopez-Rios F, Rodriguez-Peralto JL, Castano E, Benito A. Squamous cell carcinoma arising in a cutaneous epidermal cyst: case report and literature review. Am J Dermatopathol. 1999;21:174–7.
7. Yeh L-P, Kuo-Sheng L. Squamous cell carcinoma arising from an epidermal cyst of the scrotum. Tzu Chi Med J. 2013;25:117–8.
8. Morritt AN, Tiffin N, Brotherston TM. Squamous cell carcinoma arising in epidermoid cysts: report of four cases and review of the literature. J Plast Reconstr Aesthet Surg. 2012;65:1267–9.
9. Fabbrocini G, Ruocco E, De Vita V, Monfrecola G. Squamous cell carcinoma arising in long-standing hidradenitis suppurativa: an overlooked facet of the immunocompromised district. Clin Dermatol. 2017;35:225–7.
10. Jehle KS, Shakir AJ, Sayegh ME. Squamous cell carcinoma arising in an epidermoid cyst. Br J Hosp Med (Lond). 2007;68:446.
11. Antón-Badiola I, San Miguel-Fraile P, Petrieo-Cancelo A, Ortiz-Rey JA. Squamous cell carcinoma arising on an epidermal inclusion cyst: a case presentation and review of the literature. Actas Dermosifiliogr. 2010;101:349–53.
12. Shabbir A, Loss L, Bogner P, Zeitouni NC. Squamous cell carcinoma developing from an epidermoid cyst of the ear. Dermatol Surg. 2011;37:700–3.

Fig. 3 Histopathological findings. a Gross findings: microinvasive squamous cell carcinoma (arrow), epidermal cyst wall (thin portion). b Microinvasive squamous cell carcinoma (H&E, × 400). c Benign cyst (H&E, × 1)