

## Cystic Basal Cell Carcinoma with a Giant Vulvar Cyst

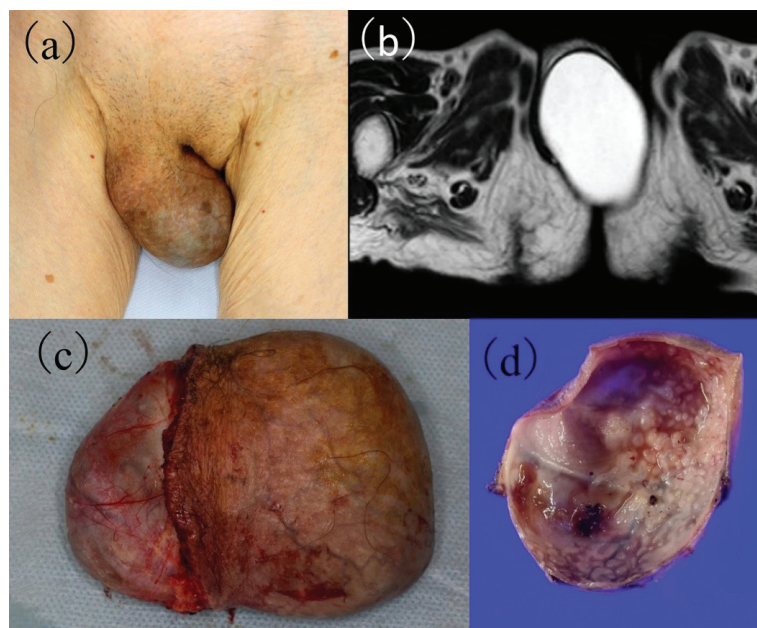
### Dear Editor,

Cystic basal cell carcinoma (BCC) is a rare subtype of BCC (1). Histologically, it is usually characterized by multiple small cysts without a clinical cystic appearance (2). Herein, we report an unusual case of cystic BCC with a large vulvar cyst.

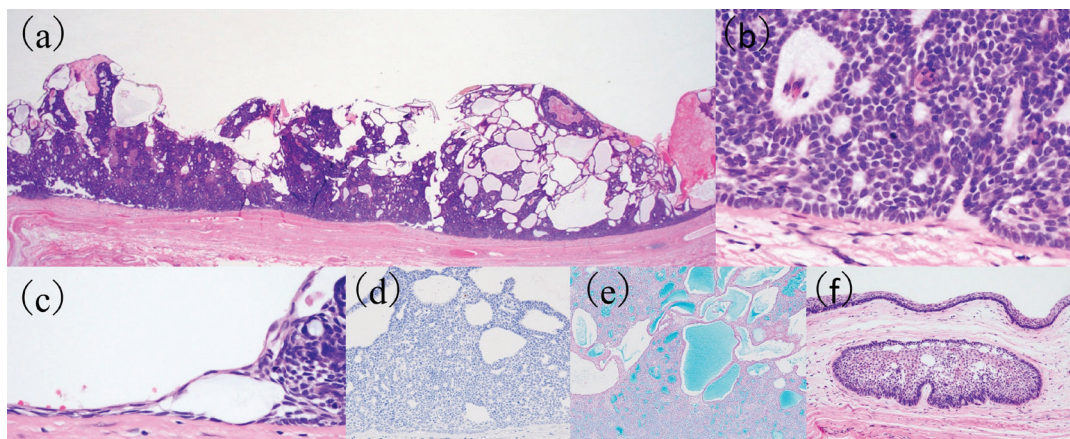
A 90-year-old Japanese woman visited our hospital with a pedunculated subcutaneous nodule on her right labia majora that had persisted for 10 years and had grown rapidly in the past 4 years. The initial examination revealed a cystic tumor (size 90×70×60 mm) (Figure 1, A). Magnetic resonance imaging revealed a cystic mass surrounded by a focally thickened wall on the vulva (Figure 1, B), with T2 high and T1 low intensities in the center. Therefore, an epidermoid cyst or other type of cystic tumor was considered for diagnosis.

The tumor was successfully excised with the overlying epidermis (Figure 1, C), the skin defect was primarily closed, and the deformity was corrected. During sample processing for pathological evaluation, brown serous fluid with no keratin leaked from the cyst, leading to cyst shrinkage (Figure 1, D). Histopathological evaluation revealed a thickened cyst wall and basaloid cells with peripheral palisading cell arrangements and slight atypia. Squamous epithelium with a granular layer and keratinization were absent (Figure 2, A), while mucin deposition was apparent in the tumor nests (Figure 2, B). Moreover, the walls of the cyst showed partial thinning (Figure 2, C).

On immunohistochemistry, the tumor nests were negative for epithelial membrane antigen (EMA) (Figure 2, D), carcinoembryonic antigen, and gross cystic



**Figure 1.** (A) A bluish cystic tumor (size 90×70×60 mm) on the right labia majora. (B) Magnetic resonance imaging showed a cystic mass surrounded by a focally thickened wall on the vulva (T2-weighted image). (C) Excised tumor with the overlying epidermis. (D) Brown serous fluid with no keratin leaked out of the cystic tumor during pathological specimen processing.



**Figure 2.** (A) Basaloid cells with atypical peripheral palisading cell arrangements were present in the wall of the cyst. Squamous epithelium with a granular layer and keratinization was absent (hematoxylin and eosin (H&E), original magnification  $\times 20$ ). (B) Mucin deposition in the tumor nests (H&E  $\times 40$ ). (C) The wall of the cyst showed partial thinning and few erythrocytes centrally, but no keratin (H&E  $\times 40$ ). (D) The tumor nests were negative for epithelial membrane antigen (EMA) (anti-EMA antibody  $\times 100$ ). (E) The mucin of the stroma was positive for Alcian blue stain ( $\times 100$ ). (F) The region of the BCC beneath the cyst. H&E  $\times 100$ .

disease fluid protein 15. The mucin of the stroma was positive for Alcian blue stain (Figure 2, E). Thus, the tumor was diagnosed as a cystic BCC. No evidence of recurrence has been observed as of 20 months after surgery.

BCCs that form single cysts, especially those completely composed of BCC cells or those that develop from epidermoid cysts, are very rare; however, in a few cases, the cyst walls comprised squamous cells with keratohyalin granules, and BCC cells were present in some parts of the tumor (3,4). In these cases, the tumor sizes were  $<50$  mm (3,4). Our patient had a cystic BCC with a single cyst that contained serous fluid without keratin, and the cyst wall was completely composed of BCC cells. The tumor cells were negative for EMA (Figure 2, D).

Whether the BCC in the present case originated from the overlying epidermis or from epidermoid cysts was unclear. Based on the complete lack of keratin and squamous epithelium in the wall of the cyst, the epidermoid cyst origin was less likely, despite a possible link with the overlying epidermis. However, no continuation between the cyst wall and the overlying epithelium was observed, which may reflect the large size of the tumor. Nevertheless, one section of the BCC was beneath the cyst (Figure 2, F), with a possible connection between the wall of the cyst and the overlying epidermis. Alternatively, the BCC may not have connected to the epidermis if it arose in the hair follicles.

Other histological differential diagnoses of cystic tumors include cystic trichoblastoma (5) and BCC

with ductal and glandular differentiation (6). In the present case, cystic trichoblastoma was excluded based on the lack of fibrous interstitium and structures such as follicular germinative cells. Moreover, ductal and glandular differentiation of the apocrine glands were absent.

The only previous reports of BCCs with a cystic appearance were a facial cyst with a 6 mm diameter in the English literature (2) and a genital BCC with a small size (3 $\times$ 2 cm) in the Japanese literature (7). To the best of our knowledge, no previous studies have reported a case of a cystic BCC with a single large genital cyst. Thus, BCCs and other malignant tumors should be carefully considered as differential diagnoses in cases of cystic tumors.

### References:

1. Patterson JW. Tumors of the epidermis. In: Weedon's Skin Pathology. 5th ed. London, UK: Churchill Livingstone; 2021. pp. 848-59.
2. Korekawa A, Nakajima K, Nakano H, Sawamura D. Translucent basal cell carcinoma with a single cyst of the upper eyelid. *J Dermatol*. 2017;44:e154-5.
3. Harada T, Fukumoto T, Shimizu H, Nishigori C. Basal cell carcinoma developed from an epidermal cyst: a case report and review of the literature. *Dermatol Reports*. 2021;13:9273.
4. Yamanaka K, Sato K, Masaki T, Nakano E, Nishigori C. Basal cell carcinoma derived from epidermal cyst of the knee of a patient with xeroderma pigmentosum group C. *J Dermatol*. 2022;49:e246-7.

5. Lopez-Takegami JCH, Wolter M, Löser C, Maiweg C, Jones M, Metze D, *et al.* Classification of cysts with follicular germinative differentiation. *J Cutan Pathol.* 2016;43:191-9.
6. Misago N, Satoh T, Narisawa Y. Basal cell carcinoma with ductal and glandular differentiation: a clinicopathological and immunohistochemical study of 10 cases. *Eur J Dermatol.* 2004;14:383-7.
7. Tsuboko T, Watanabe S, Sawada M, Dekio I, Ishizaki S, Kobayashi M, *et al.* A case of cystic basal cell carcinoma of the vulva. *Skin Cancer.* 2017;32:207-10.

**Takayuki Suyama<sup>1</sup>, Megumi Yokoyama<sup>1</sup>,  
Jun Matsushima<sup>2</sup>, Kazumoto Katagiri<sup>1</sup>**

<sup>1</sup>*Department of Dermatology, Dokkyo Medical University Saitama Medical Center, Saitama, Japan*

<sup>2</sup>*Department of Pathology, Dokkyo Medical University Saitama Medical Center, Saitama, Japan*

**Corresponding author:**

Takayuki Suyama, MD, PhD

Department of Dermatology, Dokkyo Medical University Saitama Medical Center

2-1-50 Minami-koshigaya, Koshigaya, Saitama, 343-8555, Japan

*suyama11@dokkyomed.ac.jp*

ORCID ID: 0000-0002-6986-411X

Received: March 20, 2024

Accepted: October 8, 2024

