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Case Report

Perineal Squamous Cell Carcinoma Arising in an Epidermal Cyst

Fuminori Teraishi[a]*, Yuki Matsumi[a], Kota Tachibana[b], Toshiyuki Watanabe[c], and Toshiyoshi Fujiwara[a]

Departments of [a]Gastroenterological Surgery, [b]Dermatology, Okayama University Graduate School of Medicine, Dentistry and Pharmaceutical Sciences, [c]Department of Plastic and Reconstructive Surgery, Okayama University Hospital, Okayama 700-8558, Japan

A 90-year-old Japanese woman who had been aware of a subcutaneous mass on the right perineal region for 5 years was referred to our hospital for further examination and treatment because of the rapid growth of the mass and bleeding that began 3 months earlier. A biopsy of the mass revealed a diagnosis of well-differentiated squamous cell carcinoma. On preoperative examination, the tumor was 90×40 mm in size and was suspected to have partially invaded the levator ani muscle and external sphincter. Since a preoperative cardiac evaluation indicated severe aortic stenosis, we performed transcatheter aortic valve implantation. A radical resection was then performed with general anesthesia. The skin and subcutaneous tissue defects were reconstructed with a posterior gluteal-thigh propeller flap, and a sigmoid colostomy was created. The patient had a good postoperative course and was transferred to a rehabilitation facility 28 days after the surgery. Epidermal cysts are a common benign tumor, and clinicians should keep in mind that these cysts can become malignant.

Key words: squamous cell carcinoma, epidermoid cyst, gluteal thigh flap

Epidermal cysts are benign tumors that are commonly encountered in daily medical practice. They most frequently develop on the head and neck, but they have also been documented on the trunk, extremities, and buttocks, and malignant transformation has been reported on rare occasions [1]. We provide the details of our patient's perineal squamous cell carcinoma that arose from an epidermal cyst.

Case Presentation

A 90-year-old Japanese woman who had been aware of a subcutaneous mass on her right perineal region for ~5 years was referred to our department for an examination and treatment because of the rapid growth of the mass and bleeding that she had observed beginning 3 months earlier. A preoperative examination identified a 90×40-mm oval mass extending from the right buttock to the perineal lesion, and mucus production was seen from the cutaneous fistula (Fig. 1). No mass or induration was palpable on digital examination.

Other than a hemoglobin level at 8.6 g/dL and mild renal dysfunction, no other abnormalities were detected. Pelvic computed tomography showed a well-defined, 80-mm-sized mass with slightly heterogeneous internal density on the right perineal region. Pelvic magnetic resonance imaging (MRI) revealed the 75×45-mm mass as a cystic lesion with a high internal signal at T2 and an irregularly marginated elevated lesion in the lumen. The mass was in contact with the external sphincter and the levator ani muscle, with suspected invasion to both muscles (Fig. 2). Rectal endoscopy revealed no abnormal findings of note in the rectal to anal canal.

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*Corresponding author. Phone: +81-86-235-7257; Fax: +81-86-221-8775
E-mail: pkc1940h@okayama-u.ac.jp (F. Teraishi)

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A tumor biopsy with the patient under local anesthesia demonstrated carcinoma with atypical squamous cell proliferation in the deep dermis, and a diagnosis of squamous cell carcinoma was made. However, a preoperative cardiac function test indicated severe aortic stenosis, and thus the patient underwent transcatheter aortic valve implantation (TAVI) at our institution's department of cardiology, and then a tumor resection under general anesthesia. A skin incision was made with a margin of ≥5 mm from the tumor in the jackknife position, and the tumor was removed without rupturing the tumor capsule. The levator ani muscle and the external sphincter, which were in contact with the tumor, were partially resected combined with tumor (Fig. 3A). After the resection, the skin and subcutaneous tissue defects were reconstructed with a posterior gluteal-thigh propeller flap (Fig. 3B), and a sigmoid colostomy was created. The excisional specimen revealed a white-toned mass lesion measuring 8.6×5.5 cm (Fig. 4A). A mass lesion developing within the cyst was also observed (Fig. 4B).

Histologically, squamous cell carcinoma in a solid pattern was identified (Fig. 4C), and there was residual cyst wall with slight atypia within the tumor (Fig. 4D). There was no invasion into the partially resected levator ani muscle, but tumor cells were exposed at the deep

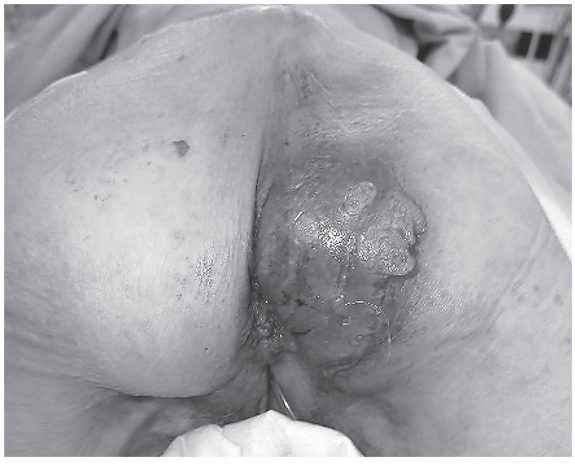

Fig. 1 Gross findings in the operative field. An approx. 8-cm oval mass is present in the right perineal area near the anus.

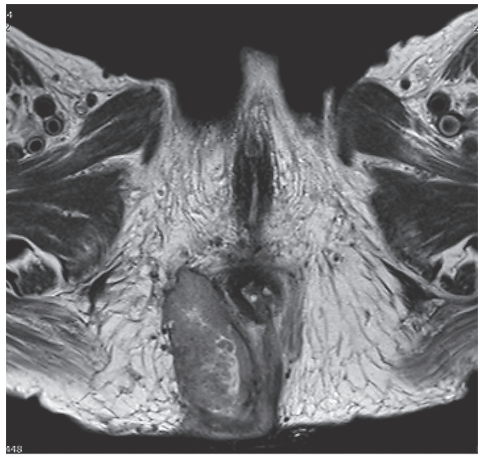

Fig. 2 MRI of the pelvis (transverse section) demonstrates a 75 × 45-mm lobulated tumor in the right perineal area.

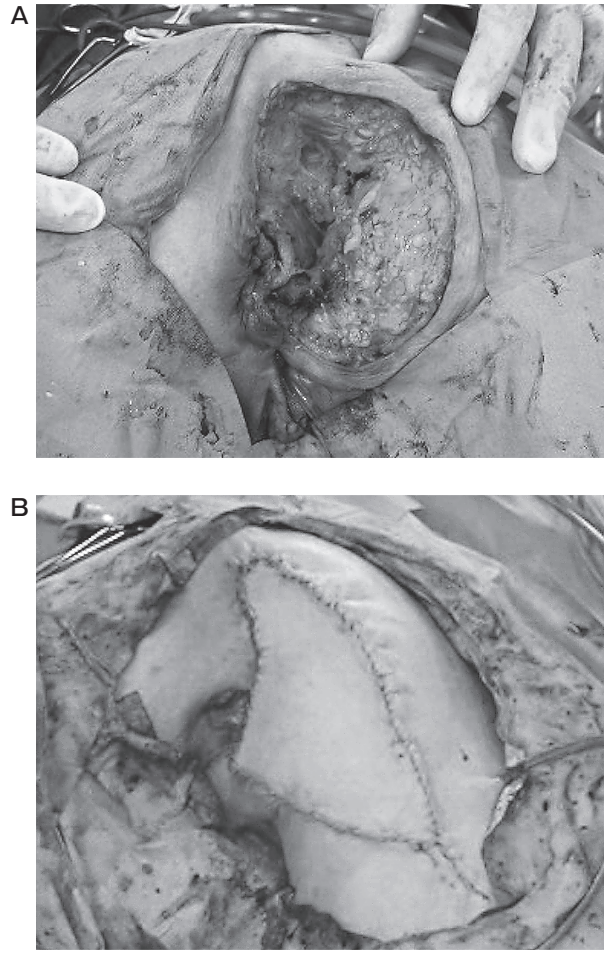

Fig. 3 **A**, With the patient in the jackknife position, the tumor was removed with partial resection of the levator ani muscle and the external sphincter muscle; **B**, The skin and subcutaneous tissue defects were reconstructed with a posterior gluteal-thigh propeller flap.

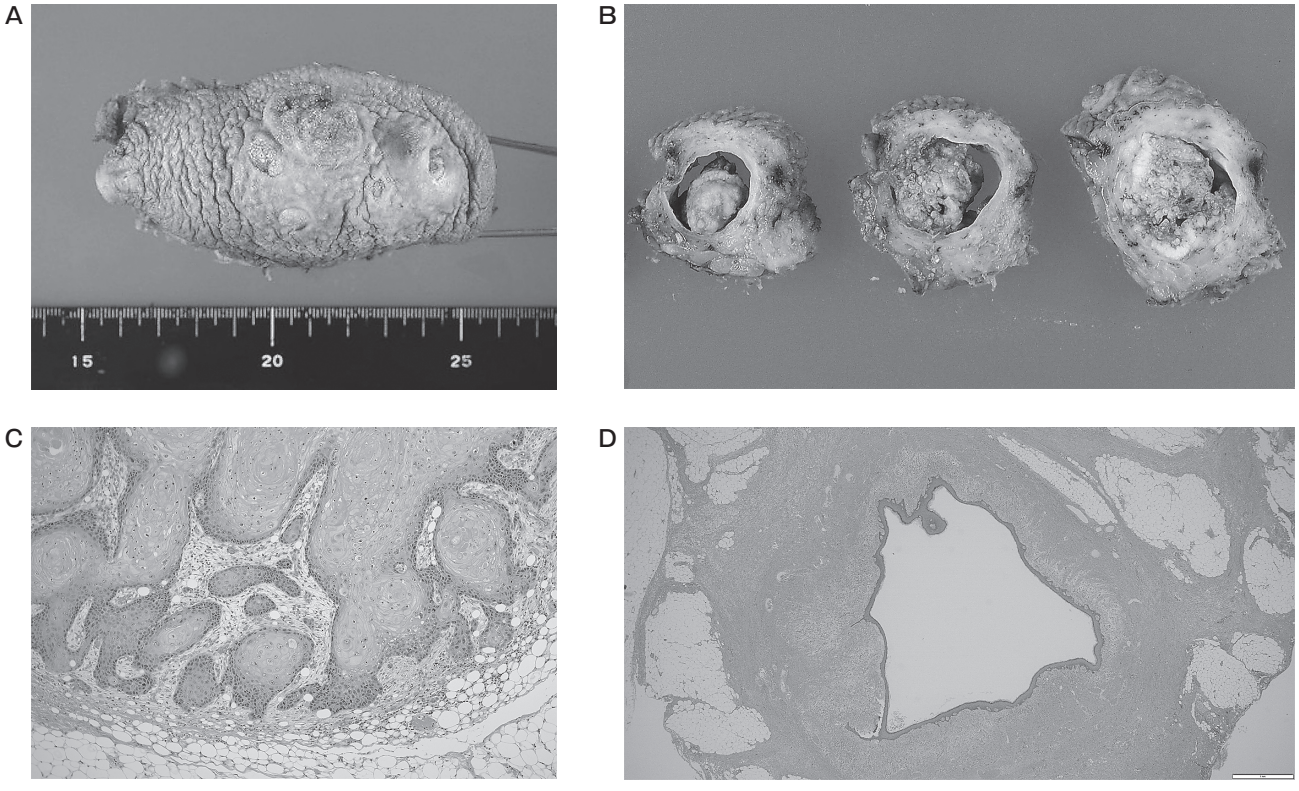

Fig. 4 **A**, The resected specimen revealed a white-toned mass lesion measuring 8.6 × 5.5 cm; **B**, A sectional image of the tumor showed a mass lesion developing within the cyst. The histopathological examination revealed squamous cell carcinoma in a solid pattern **C**, and a residual cyst wall with slight atypia within the tumor **D**.

margin on some of the dissected surfaces. The patient had a good postoperative course and was transferred to a rehabilitation facility 28 days after the surgery. No postoperative chemotherapy was administered, and the patient has been under observation with no signs of recurrence for > 1 year after her surgery.

Discussion

Epidermal cysts are benign diseases caused by the invasion of epidermal elements from hair follicles into the subcutaneous fat. Their skin lesions are frequently observed in clinical settings, but they rarely become malignant [1]; the incidence of malignant transformation of epidermal cysts into squamous cell carcinoma (SCC) is 0.011-2.2% [2, 3]. However, it is quite possible that many patients either do not undergo a histopathological examination after the excision of an epidermal cyst or are left untreated, and the actual frequency of malignant transformation of epidermal cysts into SCC may thus be even more rare than the range quoted above. The average age of patients with an epidermal cyst is the 50s, with no significant difference in the rate of occurrence between males and females [3] or among the sites of occurrence include the buttocks, head and neck, and lower extremities [3, 4].

The cause of the malignant transformation of epidermal cysts remains unclear, but it has been suggested that chronic inflammation triggered by infection leads to dysplasia and/or malignancy [5]. SCC associated with epidermal cysts in the head and neck, which is the most frequent form, has been reported to be triggered by exposure to UV light and chronic irritation [6].

Clinically, it is difficult to distinguish between benign and malignant disease in epidermal cysts, but malignant transformation should be considered when there is a rapid growth in the size of the cyst or symptoms such as pain, ulceration, or discharge of secretions [7]. In our patient's case, although a detailed history could not be obtained due to her advanced age, the possibility of

malignancy was strongly suggested by the rapid growth of the previously observed buttock mass and the thickening of the tumor wall and irregular margins on MRI images, which led to the diagnosis of SCC based on a biopsy. Although the possibility of collision carcinoma (in which an SCC arising from a different site collides with an epidermal cyst) cannot be ruled out, the rationale for diagnosing our patient as having an SCC arising in an epidermal cyst was based on two factors: (*i*) the development of a mass lesion within the cyst, and (*ii*) the residual non-malignant cyst wall in part of the tumor.

MRI findings of malignant epidermal cysts often show thickening of the cyst wall, irregular margins, and an enhancement of the contrast effect [8, 9]. The treatment of epidermal cysts suspected of malignant transformation is resection with appropriate margins, similar to other malignant cutaneous lesions. With regard to minimum margins, 6-mm margins have been recommended for high-risk tumors and extensive excision is performed with resection margins of ≥ 2 cm for advanced cancer [10]. Most patients have a good prognosis, but if the cyst wall ruptures, the cyst can easily invade surrounding tissues, resulting in lymph node metastasis or metastasis to other organs, with a poor prognosis [11]. In our patient's case, the pathology did not show invasion of the anorectal muscles as suspected on MRI, but a portion of the dissected surface was positive for deep margins. Because of her advanced age (90 yrs), no postoperative treatment was performed, but she has been under observation with no signs of recurrence for > 1 year after the surgery.

With regard to reconstruction after the extensive resection of a patient's perineum, reconstruction using a gluteal artery perforator flap is a good option for repairing extensive defects in the perineum as it provides a wide, thick flap containing the inferior descending femoral artery and the perforator of the profunda femoris artery [12]. In our patient's case, it was also possible to completely fill the entire perineal defect by harvesting a wide and thick flap that included both of the perforators.

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