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CASE REPORT
Reconstructive

Squamous Cell Carcinoma following Epidermoid Cyst in the Buttock

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Summary: In the field of plastic surgery, subcutaneous masses in the buttocks are frequently observed. However, squamous cell carcinoma (SCC) after epidermoid cyst, which appears in the presacral space, is extremely rare. This report described a case of a 71-year-old woman, who previously received a skin incision by a doctor for treating a cystic lesion in the buttock; she was diagnosed with SCC by preoperative biopsy at the authors' department. In addition, computed tomography suspected that the tumor originated in the presacral space. Under general anesthesia, an extended resection of the malignant tumor with gastrointestinal surgery was performed. After resection, the defect of buttocks region was reconstructed with a V-Y advancement gluteus maximus myocutaneous flap. After pathological examination the tumor was diagnosed as SCC after epidermoid cyst; peplomycin sulfate at 50 mg/d was administered intramuscularly for 2 weeks as chemotherapy. No wound complications were observed after surgery, and no recurrence was noted for 5 years. For managing tumor in the gluteal region, a possibility of malignancy must be considered, and thorough radiographic studies must be pursued before surgery.
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In the field of plastic surgery, subcutaneous masses in the buttocks including epidermoid cysts are frequently observed. Many of these lesions are treated by surgical resection. Surgical skin incision is often performed when inflammation or infection is noticed. Few reports describing squamous cell carcinoma (SCC) after epidermoid cysts are found in this field.

Since the presacral space contains all 3 germ layers, various types of tumors can appear. However, retrorectal tumor recognized as a subcutaneous mass in the buttock are rare.[1] This report showed a rare case of SCC after an epidermoid cyst in the buttocks, which originated in the presacral space.

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CASE PRESENTATION

A 71-year-old woman had a chief complain of buttock and back pain. Medical history included hypertension, diabetes mellitus, and total hysterectomy for uterine cancer. There was no long-standing pyoderma and chronic pilonidal sinus/cyst in the buttock in the patient. The patient noticed a mass in the buttocks at 1 year before being referred to the authors' hospital and found the mass to become gradually larger and painful. Therefore, she visited a doctor, who performed only skin incision for treating the cystic lesion. After 6 months, the swelling recurred, and computed tomography (CT) revealed tumor invasion into the deeper tissue.

At the authors' department, a 10×7 × 6 cm-hyperpigmented, elastic, and soft-to-hard mass was observed. Blood tests revealed an SCC-related antigen level of 14.2 ng/dl, which far exceeded the upper limit of normal range at 1.5 ng/dl, and the mass was diagnosed as well-differentiated SCC (T4N0M0 type 3) by preoperative biopsy. CT findings revealed that the tumor spread from the presacral space to the gluteal region, possibly invaded the posterior rectum and destroyed the sacrococcygeal bone (Fig. 1), suggesting a possibility that the tumor originated in the

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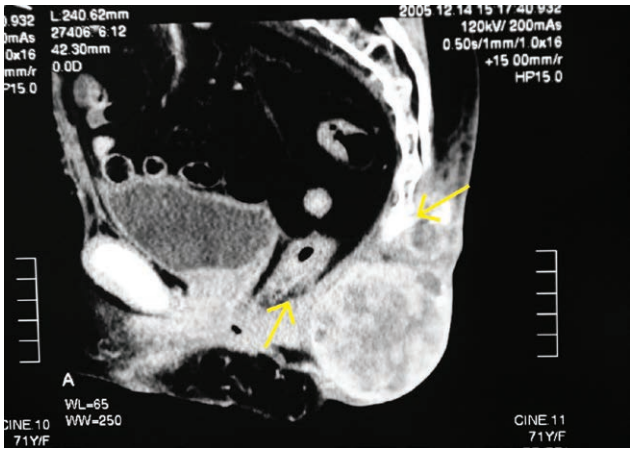

Fig. 1. Preoperative CT image. CT showed that the tumor spread from the presacral space to the gluteal region, possibly invaded the posterior rectum (yellow arrow), and destroyed the sacrococcygeal bone (yellow arrow).

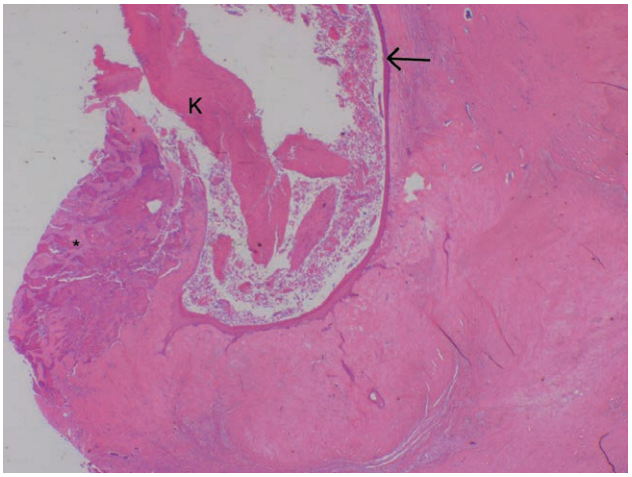

Fig. 3. Microscopic observations of the resected specimen. Histopathologic examination found tumor cells arising from the stratified squamous epithelium (black arrow) and keratin (K) in the epithelium at a magnification of 40. Asterisk indicates the resected tumor.

presacral space. An extended resection of the malignant tumor with gastrointestinal surgery was performed. Under general anesthesia, a skin incision was made with a 3-cm tumor margin (Fig. 2). The base of the tumor was resected at the attachment of the right gluteus maximus and the middle layer of the left gluteus maximus. Thereafter, the halves of fourth and fifth sacral bones were resected by a bone saw. The size of the defect after resection was 15×13 cm, and the bladder was exposed at the base of the defect, and after the patient was placed in the supine position, gastrointestinal surgery was performed. After performing colostomy, followed by abdominoperineal resection, the tumor and rectum were removed together. Intraoperative histopathology reconfirmed the pathological diagnosis of SCC, and negative margin after the resection of the tumor was observed. After the patient was placed in prone position again, a 15×20-cm gluteus maximus myocutaneous flap was made and moved into the tissue defect by V-Y advancement technique (Fig. 2). For preserving the superior and inferior gluteal arteries, only the lower portion of the gluteus maximus was resected at its insertion, and only half of the layer of the upper portion of the muscle was dissected. The origin of right gluteus maximus was partially resected for separating it from the skin and bone and moved to the midline. A continuous suction drain was placed under the flap (**see figure, Supplemental Digital Content 1**; after surgery, the flap color was favorable. (a) Donor site was able to be closed without any tension. (b) At 1 year after surgery, no recurrence was observed. The morphology of the surgical site was favorable, ***http://links.lww.com/PRSGO/A937***).

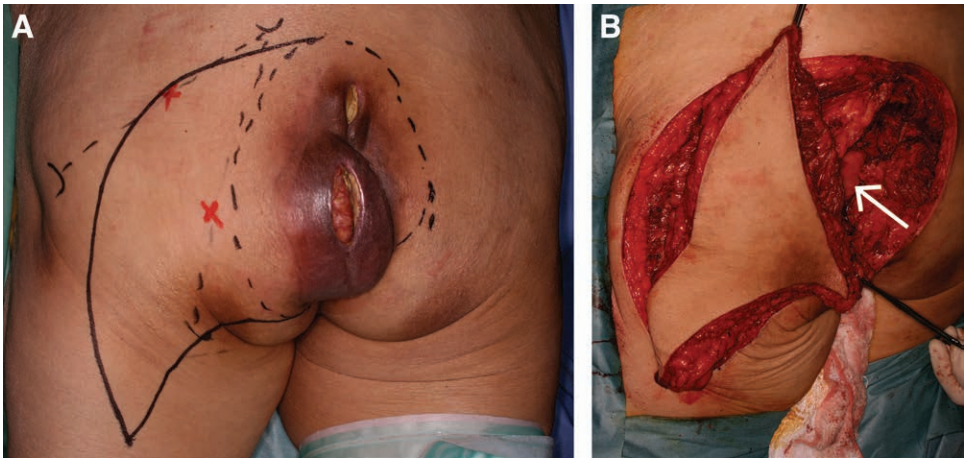

Fig. 2. Preoperative and intraoperative findings in the buttock of a 71-year-old female patient. A, Preoperative finding and surgical design. The red X marks showed the superior and inferior gluteal arteries. B, During surgery, the skin defect size after resecting tumor was 15×13 cm, and the bladder (white arrow) was exposed at the base of the defect. A 15×20-cm gluteus maximus myocutaneous flap was made and moved into the tissue defect by V-Y advancement technique.

Table 1. The Summary of Surgical Treatments for Squamous Cell Carcinoma Arising from Epidermoid Cysts in the Buttocks

Author (Country, Publication Year)	Age, Sex	Size (cm)	Duration (y)	Approach (Reconstruction)	Treatment	Outcome
Shah (India, 1989)	55, F	9×6	0.5	Posterior (not stated)	Excision (no detail was found)	Not stated
Wong (Singapore, 2000)	57, M	6	20	Posterior (free LD, rotation, and posterior thigh flaps)	Wide excision × 2	No recurrence
Debaize (Belgium, 2002)	38, F	20×15×12	20	Posterior (direct closure)	Marginal excision	No recurrence
Jehle (England, 2007)	48, M	5×5×3	28	Posterior (not stated)	Wide excision + inguinal lymph node dissection + RT	Lymph node + lung metastasis
Kshirsagar (India, 2011)	72, M	10×7	10	Posterior (STSG)	Wide excision	Not stated

F, female; LD, latissimus dorsi; M, male; RT, radiation therapy; STSG, split-thickness skin graft.

At 1 year after surgery, no recurrence was observed. The morphology of the surgical site was favorable (**Supplemental Digital Content 1**). Histopathologic examination of the tumor found atypical cells arising from the stratified squamous epithelium and tumor cells forming horn pearls, and the tumor was diagnosed as well-differentiated SCC (Fig. 3). No lymph node metastasis was found. Postoperatively, peplomycin sulfate at 50 mg/d was administered intramuscularly for 2 weeks as chemotherapy, and no wound complications were observed. At 5 years after surgery, no recurrence was observed on CT and physical findings, and the patient was able to walk with a cane.

DISCUSSION

Although epidermoid cysts are common skin lesions, they rarely become SCC.[2] Malignant tumors arising from epidermoid cysts are reported to appear at a rate of 0.011–2.2%.[3,4] PubMed search on epidermoid cysts becoming SCC in the buttocks in English literature yielded only 5 cases (Table 1).

In this study, the tumor was suspected to originate in the presacral space, because (1) tumor invasion was centered around the presacral space and (2) there was no history of a long-standing pyoderma. Tumors in the presacral space are frequently found in women. The presacral space has a caudal end containing many embryonic tissues and is known to be a site where the tumorigenesis of various cancers is frequently found. Jackman et al.[5] reported the concept of a "retrorectal tumor," which refers to tumors appearing in the presacral space. Reports of the malignant transformation of epidermoid cysts in the presacral space are extremely rare.[6,7]

Regarding surgery, the authors chose the procedure described above with considering the location of the tumor in the presacral space. Various surgical approaches including anterior, posterior, and combined approaches, and open abdominal and laparoscopic approaches are reported. Approach is selected by considering tumor size, malignant or benign, and invasion into other organs.[8] Because the present case had a malignant tumor with suspected invasion into the adjacent organs, combination surgery was selected.

As reconstructive surgery after tumor resection, free skin graft, free latissimus dorsi flap, gluteal rotation flap, and posterior thigh rotation flap are reported.[9] In this study, a single-side V-Y advancement gluteus maximus myocutaneous flap was selected for the following reasons: (1) the bladder was exposed at the base of the defect; (2) since the site was a weight-bearing region, reconstruction with adequate subcutaneous tissue was necessary; (3) the procedure was able to be performed quickly; (4) the flap was able to be moved over a wide range; (5) the flap had stable blood flow; and (6) postoperative gait disturbance was able to be minimized.

In this study, chemotherapy as an adjuvant therapy was administered with an analogue of bleomycin, which is effective against skin cancer, specifically SCC. Although adjuvant chemotherapy trials for cutaneous SCC are sorely insufficient, 5-fluorouracil (5-FU)/cisplatin, 5-FU/carboplatin, or paclitaxel/carboplatin combinations is used.[10] Further studies are needed to investigate the necessity of postoperative treatment.

CONCLUSIONS

A case of successful surgical resection and reconstruction of SCC after epidermoid cyst in the buttock was reported. In managing a gluteal subcutaneous tumor, a possibility of malignancy must be first considered, and thorough medical examination such as radiographic studies must be pursued before surgery.

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