

Case Report

Chemoradiotherapy with irinotecan (CPT-11) for adenoid cystic carcinoma of Bartholin's gland: A case report and review of the literature



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Introduction

Malignant tumors arising in the vulva reportedly account for 3% to 5% of gynecological malignancies, and Bartholin's gland carcinoma was reported to account for 0.1% to 7% of vulvar malignancies (Nasu et al., 2005). In particular, adenoid cystic carcinoma (ACC) is extremely rare, accounting for approximately 10% of Bartholin's gland malignancies. The tissue types of ACC vary. While its frequency is high in the salivary gland, paranasal sinus, breast, and skin, this carcinoma is also occasionally observed in the cervical glands and ovaries in the gynecological field. However, ACC of Bartholin's gland is extremely rare, with only about 80 cases having been reported in the international literature (Alsan et al., 2011). Moreover, there is as yet no established postoperative treatment. We experienced a case in which chemoradiotherapy with irinotecan (CPT-11) was performed for ACC of Bartholin's gland. This case is presented with a review of the relevant literature.

Case report

Our patient was a 68-year-old woman, gravida 3, para 2, with menopause at age 58 years. Family and past histories were unremarkable. She had not undergone gynecological cancer screening. She visited a neighborhood hospital with a chief complaint of a painful mass that had been present in the left vulva for one year, for which antibiotics had been administered based on the diagnosis of an infected Bartholin cyst. Because relief was not obtained with this treatment, she was referred to our hospital.

During the examination at the first visit, a 2×2 cm, highly mobile, hard and painful mass was palpated around the left Bartholin's gland. Macroscopic observation revealed no abnormalities of either the mucosa or the vulva around the vaginal ostium. No macroscopic abnormalities were observed in either the uterine cervix or the vaginal wall. The uterus was 7×4 cm in size and highly mobile. Neither adnexa was palpable. There was no inguinal lymphadenopathy.

Clinical examinations

Transvaginal ultrasonography revealed a 3.2×2.0 cm mass with a clear border and an irregular internal echo pattern at the left side of the vaginal ostium (Fig. 1A). There were no abnormalities in either ovary. Magnetic resonance imaging (MRI) revealed a 2.0×1.9 cm mass with an irregular border and contrast enhancement around the left Bartholin's gland (Fig. 1B). Routine hematologic and biochemical tests revealed no abnormalities. The following tumor marker results were obtained: CEA of 2.9 ng/mL, CA19-9 of 6.8 U/mL, and CA 125 of 19.3 U/mL, and SCC was below 0.5 ng/mL. Cervical cytology showed no evidence of either an intraepithelial lesion or malignancy, and endometrial cytology results were also negative.

Treatment course

Tumor resection was performed for treatment and diagnosis. The tumor was located below the mucosa of the vaginal opening and had not infiltrated the surrounding tissue, based on macroscopic observation. The border was unclear, and the tumor had infiltrated and proliferated within the tissue around the pubis corresponding to the left Bartholin's gland. The resected tumor was 1.5×2.0 cm in size, white, and hard. A honeycomb structure containing mucus was observed inside the tumor mass (Fig. 2). The postoperative histopathological diagnosis was ACC. Although continuity with the normal Bartholin's gland was not confirmed, the carcinoma was considered to have originated in Bartholin's gland, based on the site of occurrence (Fig. 3). After surgery, the patient refused to undergo a detailed lower gastrointestinal tract examination. But the patient was examined by the positron emission tomography-computed tomography (PET-CT) and routine CT, and there was no area of abnormally increased uptake of FDG in the whole body. Because neither PET-CT nor routine CT revealed any potential primary lesions, the patient was diagnosed as having ACC of Bartholin's gland. The resection margin was positive. After obtaining her consent for postoperative treatment, chemoradiotherapy (whole pelvis irradiation at a total

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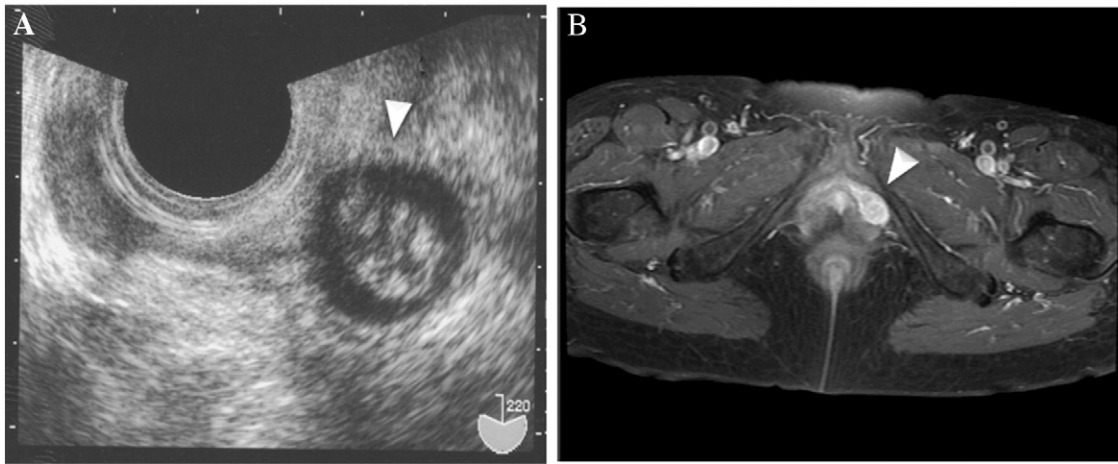


Fig. 1. A: Transvaginal ultrasonography. 3.2×2.0 cm low echo area with an irregular internal pattern was observed at the left side of the vaginal ostium. B: MRI of the pelvic cavity. 2.0×1.9 cm mass imaged with contrast enhancement corresponded to the left Bartholin's gland. The border with adjacent bone was clear.

dose of 59.4 Gy in 33 fractions combined with CPT-11 at a dose of 50 mg/m²×5) was administered. Although Grade 2 neutropenia (1024/mm³) and Grade 2 anemia (9.4 g/dL) were observed as hematologic toxicities, both were manageable. Moreover, non-hematologic toxicities were Grade 1 nausea and Grade 2 radiation dermatitis, whereas diarrhea did not occur. For the 24 months since treatment completion, to date, there have been no signs of recurrence.

Discussion

Bartholin's gland carcinomas account for approximately 0.1% to 7% of vulvar malignancies (Nasu et al., 2005). However, as noted above, ACC of Bartholin's gland has been reported in only about 80 cases in the international literature. According to a review of 49 articles conducted by Alsan et al., the median age of 79 cases was 48 years (range: 25–80 years) (Alsan et al., 2011). Although no characteristic test finding was revealed, there were many patients with a chief complaint of a vulvar mass associated with pain and burning sensation

of the vulvae. Thus, it is preferable to actively perform biopsy or resection for early diagnosis in patients age 40 and older with a painful indurated mass involving Bartholin's gland. While cases preoperatively diagnosed by aspiration biopsy cytology have been reported, there are also many in which this disease was definitively diagnosed after tumor resection performed for both diagnosis and treatment. Although no therapy has yet been established, the recommended surgical strategies are simple vulvectomy, radical vulvectomy, and lymph node dissection to the affected inguinal lymph node. Because ACC also occurs in relatively young women, lymph node dissection is recommended for such patients. However, it appears that vulvectomy should be recommended for elderly patients while lymph node dissection is not performed given the risks of postoperative complications such as deep venous thrombosis and lymphedema. Also, very little data are available regarding lymph node dissection at any age for this disease. According to the review by Alsan et al., simple and radical vulvectomy had been performed in 54% and 46% of cases, respectively. However, the recurrence rate was not low. Recurrence was observed in 35% of cases with

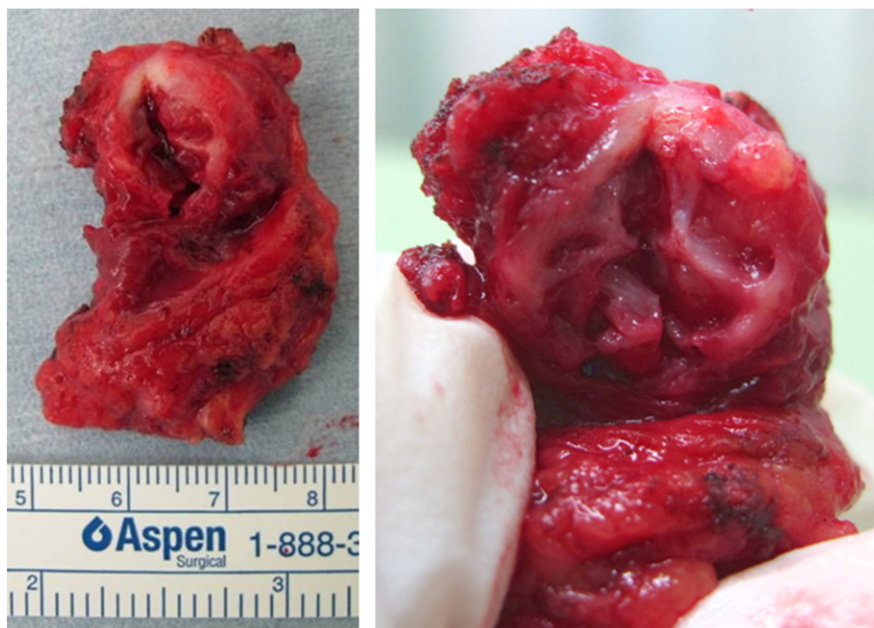


Fig. 2. Macroscopic findings of the left Bartholin's gland tumor. The resected tumor was 1.5×2.0 cm in size, white, and hard. A honeycomb structure containing mucus was observed within the tumor mass.

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