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Research Letter

Adenoid cystic carcinoma of Bartholin's gland

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Adenoid cystic carcinoma (ACC) originating from Bartholin's gland is a rare carcinoma of the female reproductive tract, accounting for <1% of all female genital malignancies and only 0.1–7% of all vulvar carcinomas [1]. Histologically, ACC comprises only 15% of the many diverse Bartholin's gland tumors. Ever since the first documentation of ACC by Klob in 1864, only approximately 350 cases have been reported [2]. Due to the low incidence, knowledge regarding optimal management and clinical outcome is limited. This report describes the diagnosis, treatment and outcome of a patient with ACC of Bartholin's gland.

A 53-year-old multiparous, postmenopausal woman presented to the outpatient department with a painful, palpable, nodular mass in the left vulvar area which had been growing for 2 years. Vulvar examination revealed an elevated, firm, nodular mass measuring approximately 2.5 cm × 2.5 cm, localized in Bartholin's gland. Pelvic and rectal examination revealed no abnormalities. Tumor biopsy revealed a cribriform pattern of tubules and gland-like elements filled with homogeneous basophilic mucin (Fig. 1A and 1B). These characteristic features formed the basis for the diagnosis of ACC of Bartholin's gland. Subsequent cystoscopy, sigmoidoscopy, and thoracic radiographs revealed no abnormalities. Abdominal and pelvic magnetic resonance imaging and whole body bone scintigraphy were used to evaluate local and distant metastases. There were no abnormal inguinal lymph nodes, except for some minor local infiltration in the left vulvar area measuring 3 cm × 3 cm (Fig. 2). The patient underwent radical hemivulvectomy and bilateral inguinal–femoral lymphadenectomy. A minimum 2-cm margin of the normal-appearing skin or mucosa around the tumor was excised. Pathology of the excised tumor was consistent with the previous biopsy result. All resection margins were free of metastasis. The patient was discharged 1 month postsurgically without any complications. She remained tumor-free for 3 years after the operation.

The original diagnostic criteria for Bartholin's gland tumors were published in 1887 by Honan. The strict criteria did not correlate well with findings of advanced tumors [3]. Thus, in 1972, Chamlian and Taylor from the Armed Forces Institute of Pathology re-established the diagnostic criteria for Bartholin's gland carcinoma [4]. Histologically, areas of transition from normal to neoplasia must be found; the tumor must be compatible with Bartholin gland's origin; and no evidence of other primary tumor should be identified.

ACC of Bartholin's gland is a rare vulvar malignancy that is often disregarded as either a cyst or inflammation. There is a tendency for local perineural invasion, which is perhaps the cause for the initial infection-like itch and pain. The ambiguous symptoms of ACC lead to the delay in both diagnosis and treatment. The average age of ACC patients is 49 years old (range: 25–80 years). Bartholin's gland ACC should be considered in patients of this age with lesions near the gland [1].

Due to the lack of large case series, no consensus regarding the optimal treatment of ACC has been established. In most cases, one of two types of surgical procedure are generally performed: simple excision and radical vulvectomy, either with or without lymph node dissection. A review of the literature suggests that there is a higher recurrence rate in patients undergoing simple excisions compared with patients undergoing radical vulvectomy, (69% vs. 43%, respectively) [5]. It is a common belief that the most important aspect of treatment is to obtain tumor-free surgical margins to prevent recurrence; however, Yang et al have found a similar recurrence rate in patients with positive (52.9%) and negative (52.1%) margins [5]. This finding implies that the status of the margins might not be as important as previously thought. Postoperative adjuvant radiotherapy has been shown to be effective in controlling the disease in patients with positive margins and local recurrences [6].

The benefits of performing either unilateral or bilateral inguinal–femoral lymphadenectomy remain controversial in

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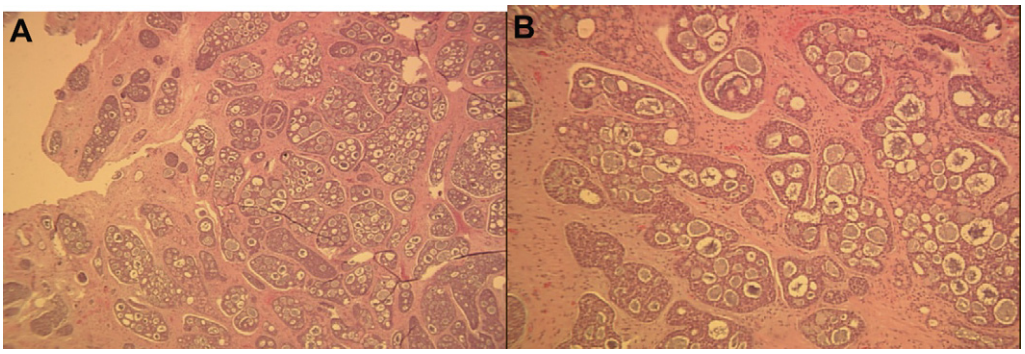

Fig. 1. Microscopic features of perineal biopsy showing the tumor has a cribriform pattern with the lumens containing basophilic mucin (hematoxylin and eosin, A: 100×, B: 400×).

the treatment of ACC; however, the main determinant of survival of ACC patients is the status of the inguinal–femoral lymph nodes [7]. Leuchter et al have reported a 5-year survival of 52%, 36% and 18% with zero, one or multiple positive lymph nodes, respectively [8]. With these results in mind, and the fact that contralateral inguinal node involvement increases with lesion size, some surgeons prefer bilateral lymph node resection as was performed in the case described here [9].

Bone and lung are the most common sites of distant metastasis for ACC of Bartholin's gland. Liver, kidney, and brain metastasis also occur but less frequently [10]. Information concerning the use and effectiveness of chemotherapy in the treatment of ACC is limited. Several chemotherapeutic agents have been developed for this disease including various combinations of adriamycin, dactinomycin, cyclophosphamide and methotrexate [5]. Although some reports of chemotherapy are encouraging, the number of cases is too few for meaningful conclusions to be drawn. As such, this option was not explored for the patient described in this report.

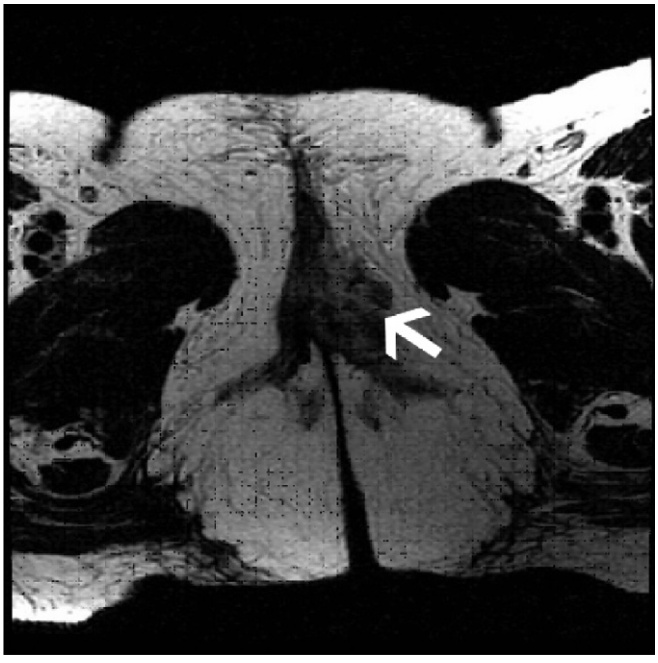

Fig. 2. Abdominopelvic magnetic resonance imaging shows a soft tissue mass infiltrated into the left vulvar area (arrow).

ACC of Bartholin's gland is a rare, vulvar malignancy with an aggressive and unpredictable biologic behavior. Due to the small number of reported cases, there is no consensus regarding standard treatment. Similar to the treatment of patients with other vulvar cancers, the present report and literature review suggest that an early diagnosis combined with radical vulvectomy and bilateral inguinal–femoral lymph node dissection will optimize the patient's chances of survival [7,8].

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