Adenoid cystic carcinoma of the vulva: A case report and literature review

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Abstract. Adenoid cystic carcinoma (ACC) of the vulva represents a rare malignancy, characterized by its low incidence and often non-specific clinical manifestations, which may lead to misdiagnosis and ultimately compromise patient prognosis. Therefore, early diagnosis and prompt intervention are crucial. The present study described the case of a middle-aged female patient who presented with a notable mass in the vulvar region. Following comprehensive imaging studies and pathological examination, the patient was conclusively diagnosed with vulvar ACC. Pathological examination following surgery revealed tumor invasion into adjacent nerves and skeletal muscle. However, the surgical margins were clear, and the patient showed no signs of recurrence during the 6 month follow-up period. Initial symptoms of vulvar ACC may resemble those of benign conditions such as Bartholin gland cysts or abscesses. In conclusion, the diagnosis of vulvar ACC requires comprehensive clinical evaluation, imaging and pathological examination. In the future, developing a comprehensive treatment plan and conducting long-term follow-up are necessary to improve the prognosis and management of patients with ACC of the vulva.

Introduction

Adenoid cystic carcinoma (ACC) of the vulva is a rare and aggressive malignant tumor that typically originates from the Bartholin gland located near the vaginal introitus. Bartholin gland carcinoma accounts for <1% of gynecological cancers and 3-7% of all vulvar malignancy (2).In the vulva, ACC is a rare malignancy that was represents ~30 % of Bartholin gland carcinoma (2) and was first described by Bernstein *et al* (3) as a rare histological subtype of adenocarcinoma, affecting mainly the glandular mucosa, salivary glands, mammary glands and the female genital tract, particularly the cervix. Due to its rare occurrence (4) and non-specific clinical manifestations, the disease is easily misdiagnosed as a Bartholin gland cyst and abscess. (1,5). Risk factors may be associated with chronic inflammation, abnormal hormone levels, etc. (6). In addition, treatment methods mainly include surgical excision, radiation therapy, and chemotherapy, but due to the lack of standardized treatment protocols, clinical effects are often inconsistent. The overall 10-year survival rate is 64% (7), underscoring the challenges in managing this rare and complex disease.

Case report

The patient was a 45-year-old Chinese female who underwent regular physical examinations and had no notable past medical history., with menarche at 13 years, having had five pregnancies and one delivery, and with no family history of disease, smoking or drinking. The patient presented with a subcutaneous mass in the right vulvar area that had slowly enlarged over 4 months without pain. The patient had sought medical attention at Central Hospital, Tianjin University in January 2025, due to continuous pain from the mass for 3 days. Upon examination, the skin showed no damage, redness or swelling. Gynecological examination revealed a subcutaneous mass on the right vulva, measuring ~2x1 cm in size. The mass was hard in texture, with mild tenderness and unclear boundaries. As a vestibular gland cyst could be excluded due to clearly defined boundaries, further examination of the patient was conducted. The patient was negative of human papillomavirus (HPV) and tested positively for syphilis. Alanine aminotransferase (ALT) was 30 U/l and aspartate aminotransferase (AST) was 25 U/l, with the normal reference range for both typically being 0-40 U/l. Total bilirubin (TBIL) was 10 μmol/l (normal reference range of <34 μmol/l. Serum creatinine (Cr) was 50 μmol/l, with a normal reference range for females of 44-97 μmol/l. Blood urea nitrogen (BUN) was 7 mmol/l, with a normal reference range typically being 2.9-8.2 mmol/l. Carbohydrate antigen 125 (CA125) was 10 U/ml (reference range typically <35 U/ml. Ultrasound examination showed a heterogeneous echogenic nodule measuring 2.4x1.4x0.9 cm, subcutaneously located on the right vulva, which had uneven internal echoes and blood flow signals visible on the inner edge (Fig. 1). Due to the presence of pain, hard texture, uneven ultrasound echoes and visible blood flow signals, malignancy could not be

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excluded. The patient underwent surgery on the following day after the initial consultation. The procedure performed was extended local excision. The specific operational steps were as follows: Under local anesthesia, the surgeon excised the lesion along with a surrounding margin of normal tissue using an electrosurgical knife to ensure negative surgical margins. The entire procedure lasted 1 h and 10 min. The resected tissue specimen was sent for pathological examination.

Pathological examination revealed a gray-brown nodular tissue measuring 3.0x1.5x1.4 cm. The section was gray-white with a solid hard consistency. Microscopic observation showed tumor cells composed of glandular epithelial cells and myoepithelial cells (arranged in a cribriform or tubular pattern), with some areas exhibiting a solid growth pattern. Various sizes of clear or basophilic mucinous deposits were observed in the glandular cavities and cribriform structures (Fig. 2A). Accompanying perineural invasion (Fig. 2B) and skeletal muscle invasion could be observed at the invasive front of the tumor (Fig. 2C), with residual Bartholin gland tissue in the surrounding local area (Fig. 2D), considered to be of Bartholin gland origin. No cancer was found at the margins of the tumor. The shortest distance was >1 cm. Tissue specimens were fixed in 10% neutral buffered formalin at room temperature for 8 h, followed by paraffin embedding. Sections of 5 μm thickness were prepared. For detection, sections were first deparaffinized in xylene and rehydrated through a descending alcohol series (100, 95, 80, 70%) to distilled water. Subsequently, heat-induced antigen retrieval was performed by immersing the sections in citrate buffer (pH 6.0) and heating at 95-100˚C for 15-20 min. After natural cooling to room temperature, the sections were washed with PBS). CK5/6 (Cat. No.: M7237, Supplier: DAKO), CK7 (Cat. No.: M7018, Supplier: DAKO), P63 (Cat. No.: CM163A, Supplier: Biocare Medical), and KI-67 (Cat. No.: M7240, Supplier: DAKO) were incubated at a dilution of 1:100 overnight at 4˚C; S100 (Cat. No.: Z0311, Supplier: DAKO) and GCDFP-15 (Cat. No.: MAB-0240, Supplier: Maixin Bio) were incubated at a dilution of 1:200 overnight at 4˚C. For the negative control, PBS was used to replace the primary antibodies. A ready-to-use horseradish peroxidase (HRP)-labeled anti-mouse/rabbit universal secondary antibody (Cat. No.: K5007, Supplier: DAKO) was applied at 1:200 and incubated for 30 min at room temperature. Color development was performed using DAB, followed by counterstaining with hematoxylin for 5 min at room temperature. Observation by light microscopy showed positive CK5/6, CK7, P63 and KI-67 (~20%) staining, and negative S100 and GCDFP-15 staining (Fig. 3A-D). A follow-up ultrasound examination conducted 2 weeks post-surgery showed no significant masses in the bilateral groin superficial lymph nodes (data not shown). Visible lymph nodes did not exhibit abnormal blood flow signals, suggesting reactive hyperplasia. Transvaginal Doppler ultrasound examination revealed no notable abnormalities in the uterus, cervix or bilateral adnexa. Computed tomography scans showed no markedly enlarged lymph nodes in the abdominal or pelvic cavities. Enhanced pelvic magnetic resonance imaging scans revealed multiple lymph nodes in both pelvic walls and bilateral groin areas, some of which were enlarged, with the largest measuring 1.4 cm, with a regular shape and without abnormal blood flow signals. No groin lymphadenectomy was performed, nor was adjuvant radiotherapy or chemotherapy administered (data not shown). The follow-up protocol was clinical examinations conducted every 3 months, with MRI every 6 months for 2 years and annually thereafter. No recurrence of tumors or lymphadenopathy was observed 6 months post-operation. The patient recovered well after surgery and was clinically healthy as of January 2026.

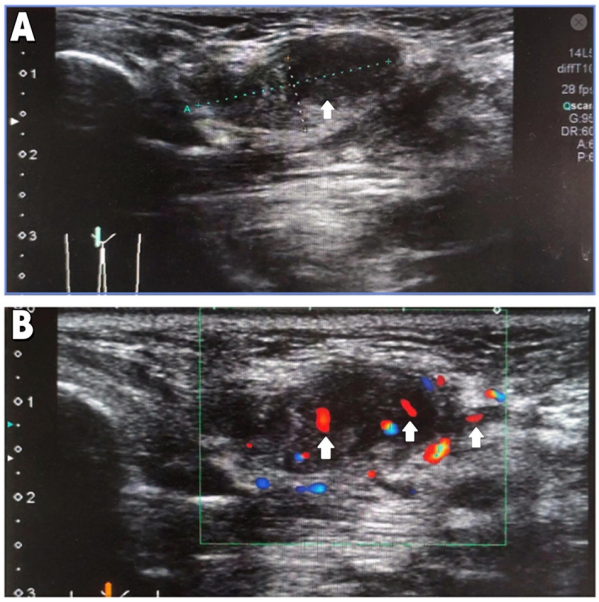

Figure 1. Ultrasound examination (A) Ultrasonography showed a 2.4x1.4x0.9 cm inhomogeneous echogenic nodule (arrow), subcutaneously located on the right vulva. (B) Internal echo was uneven and dot-and-strip blood flow signals (arrow) were visible at the inner edge.

Discussion

The present review searched PubMed (pubmed.ncbi.nlm.nih.gov/)for case reports on ACC of the vulva in the past 5 years and obtained the references listed in each article. The literature review is summarized in Table I. We identified 12 articles reporting on 36 patients.

Regarding clinical features, the median onset age for vulvar ACC is 48 years, with an age spectrum ranging from 25 to 80 years, and similar incidence rates in younger and older patients (7). The predominant clinical manifestations are similar to those of salivary gland ACC, marked by a gradually enlarging mass accompanied by discomfort, characterized by a firm texture and indistinct margins, often adherent to surrounding tissues (8). Tumor cells proliferate along the bundles of peripheral nerve fibers, which not only increase surgical complexity but frequently leads to postoperative pain and numbness due to nerve impairment (9). Vulvar ACC exhibits notable invasiveness, with advanced cases often accompanied by local recurrence and distant metastasis; the lungs emerge as the most common sites of distant metastasis, although the involvement of bones, liver and brain has also been reported (9).

The pathogenesis of vulvar ACC includes: i) Hormonal fluctuations during pregnancy, which may influence cell proliferation and differentiation within vulvar tissues, thereby increasing the risk of cancer; however, the precise mechanisms

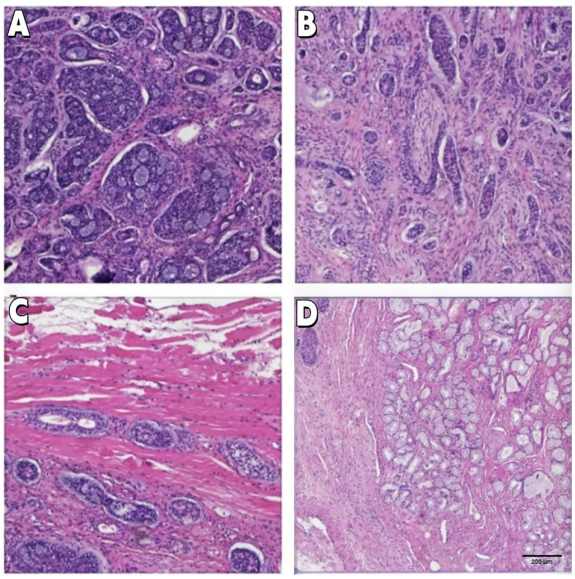

Figure 2. Representative histopathological images of the tumor. (A) Transparent or basophilic mucoid material was deposited within the lumens and varying sizes of sieve-like pores. (B) There was evidence of perineural invasion and (C) skeletal muscle invasion was detected at the leading edge of tumor infiltration. (D) Residual Bartholin's gland tissue was present in localized areas surrounding the tumor. Scale bar, 200 μm).

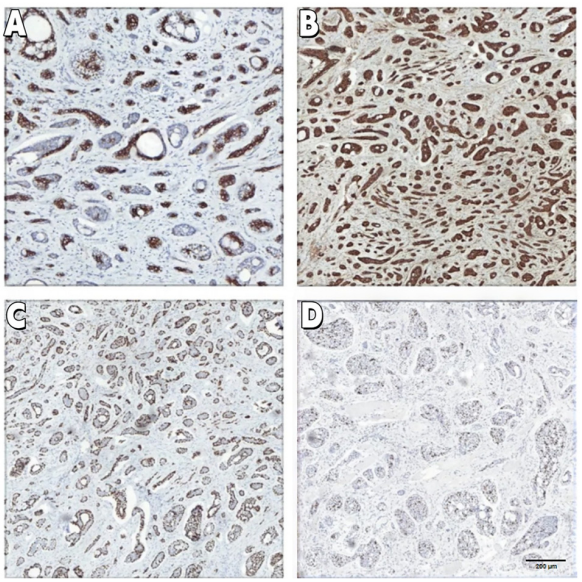

Figure 3. Representative immunohistochemical images of the tumor. (A) CK7(+), (B) CK5/6(+), (C) P63(+), (D) KI-67(~20%). +, positive. Scale bar, 200 μm).

demand further exploration (6); ii) viral infection, as HPV infection is predominantly associated with cervical squamous cell carcinoma and vulvar squamous cell carcinoma (10); however, HPV infection itself does not directly induce vulvar or cervical ACC (11) and the present case was negative for HPV; and iii) genetic mutations and chromosomal irregularities, including the presence of the MYB-NFIB fusion gene, which has been suggested in previous studies (11,12). Additional mutations such as KRAS and KDM6A have also been identified in vulvar adenocarcinoma, with cytogenetic assessments revealing complex karyotypes involving chromosomes 1, 4, 6, 11, 14 and 22 in Bartholin gland adenocarcinoma. These mutations may facilitate tumor development by impacting cell proliferation, differentiation and apoptosis (12,13).

Regarding the pathological diagnosis, histologically, ACC arising from the Bartholin gland is indistinguishable from its counterparts in salivary glands, upper respiratory tract or skin. In regard to its cellular characteristics, it is composed of glandular epithelial and myoepithelial cells. Concerning its histomorphological features, the tumors exhibit tubular, sieve-like or solid growth. The sieve-like arrangement of glandular structures is its typical feature, containing true and false glandular structures (14). True glandular cavities are formed by myoepithelial cells surrounding glandular epithelial cells, whereas false glandular structures are surrounded by myoepithelial cells, with sieve pores appearing round or oval (7). The tubular type is characterized by round or elongated tubular structures lined by epithelial and myoepithelial cells (14). Perineural infiltration is an important feature of vulvar ACC (14). Immunohistochemical markers support epithelial and myoepithelial differentiation, including epithelial markers such as CK and epithelial membrane antigen and myoepithelial markers such as CK5/6, P63, P40, S-100, smooth muscle actin, carcinoembryonic antigen and CD117 (7). ACC of the vestibular glands is extremely rare. To diagnose primary vestibular gland ACC of the vulva, it is essential to exclude ACC that has metastasized from the salivary glands, lacrimal glands, nasopharynx, mammary glands, skin or cervix. The tumor should be located in the Bartholin gland area, and there should be a transitional zone between benign Bartholin gland tissue and the tumor (15,16). Other tumors should be excluded, such as the more common squamous cell carcinoma and adenocarcinoma tumors. Basaloid squamous cell carcinoma primarily has solid structures without cystic cavities or basal membrane-like substances, with focal keratinization observed. *In situ* squamous cell carcinoma is calponin negative (14). Compared with ACC, adenocarcinoma lacks consistent acinar structures, and there is no basal membrane-like material within the lumen (5,11). Metastatic carcinoid tumors and small cell carcinoma are solid masses, with rare glandular cavities, containing argentaffin cells, and the majority are positive for neuron-specific enolase (14). Carcinoid tumors almost universally express CgA and other neuroendocrine markers (14). This often requires a comprehensive judgment based on clinical history, imaging studies and immunohistochemical markers (8). Genetic testing is essential in diagnosing vulvar ACC, detecting MYB-NFIB fusion genes, other relevant gene mutations and possible genetic susceptibility genes aids in establishing an accurate diagnosis and differentiation of tumor types, providing a basis for personalized treatment plans (11,12,17).

As a rare and highly invasive malignant tumor of the vulva, currently no consensus has been yet reached on the recommended treatment for ACC. Thus, the general consensus on vulvar cancer (18) applies, including radical surgery, which is the main treatment for vulvar ACC, and aimed at achieving complete cure through extensive excision of tumor tissue (3,6). Ensuring sufficient negative margins during surgery is essential in preventing postoperative recurrence; patients with negative margins have a survival time 15-30 years longer

Table I. Literature review of ACC of the vulva cases in the past 5 years.

First author/s, year	Age, years	Location	Size, cm	Lymph node Involvement	Resection margin	Perineural invasion	Treatment	Relapse and follow-up status	(Refs.)
Evin *et al*, 2023	31	RV	3.3	-	+	+	LE, RHV, ARTx	PD, 38 months lung; 36 months AWD	(20)
Nieuwenhuyzen-de Boer *et al*, 2020	61	RV	1	-	+	+	LE, RHV, IILND	Simultaneous lung metastasis	(26)
Güral *et al*, 2024	56	V	Nts	-	-	+	HV, BILND, ARTx	48 months lung and right humerus; 98 months DOD	(21)
Samba *et al*, 2024	42	V	Nts	-	-	-	RHV, BILND, ARTx	NED	(23)
	43	LV	Nts	-	+	+	LE, HBSRC, ARTx	60 months NED	
	38	V	3.5	Nts	+	Nts	ELE, ARTx	8 months NED	
	52	RV	2	-	-	Nts	ELE, BILND, ARTx	36 months NED	
Alhashemi *et al*, 2023	64	LV	7	Nts	Nts	Nts	LE, ARTx	12 months local	(5)
Wang *et al*, 2022	58	LV	3.2	-	-	-	HV, IILND	18 months NED	(11)
Santiago *et al*, 2021	77	RV	5	-	-	+	ELE, ARTx	24 months lung; 36 months DOD	(17)
Verta *et al*, 2022	42	VA	3.4	-	-	Nts	PC+RA, BILND	8 months NED	(6)
Nakamura *et al*, 2020	63	V	Nts	Nts	-	Nts	LE	4 years lung, 5 years local	(13)
Doutel *et al*, 2024	61	V	Nts	Nts	-	Nts	TPR	2 years lung and liver	(27)
	52	LMA	2.3	Nts	Nts	+	LE	3 years NED	
Zhu *et al*, 2020	41	BG	5	Nts	-	+	LE, ARTx	Local and lung; 82 months AWD	(28)
	41	BG	Nts	Nts	+	+	LE, ARTx	Multiple metastases (lung, bone, liver); 27 years AWD	
	70	VA	0.5	Nts	+	-	LE	129 months AED	
	58	LMA	Nts	Nts	+	Nts	LE, ARTx	13 years and 4 months AED	
	63	RV	1.5	-	-	+	RLE, IILND	76 months NED	
	50	RV	2.1	Nts	+	+	RLE, CT	Local; 74 months SD	
	59	LV	3	Nts	+	+	RLE, ARTx	73 months NED	
	40	LV	1.5	-	-	+	RHV, IILND	44 months NED	
	54	RV	1.2	-	-	+	RLE, IILND	34 months NED	
	71	LV	3	Nts	-	+	RLE	30 months NED	
	62	LV	2.2	Nts	-	+	RLE	29 months SD	
	37	LV	1.5	-	+	+	RLE, IILND, ARTx	NED	
	40	RV	3	Nts	+	+	RHV, ARTx	54 months NED	
	59	LV	2.5	Nts	-	+	RLE	46 months NED	
	57	RV	1.6	Nts	-	+	RLE	37 months NED	
	69	RV	1.8	Nts	-	+	RLE	24 months NED	

Table I. Continued.

First author/s, year	Age, years	Location	Size, cm	Lymph node Involvement	Resection margin	Perineural invasion	Treatment	Relapse and follow-up status	(Refs.)
	61	RV	3	-	+	+	RLE, IILND, ARTx	23 months NED	
	51	LV	2.5	-	+	+	RLE, IILND, ARTx	NED	
	26	LV	1.8	Nts	-	+	RLE	NED	
	56	LV	2.8	Nts	-	+	RLE	NED	
	50	RV	2	-	-	+	RLE, IILND	78 months NED	

RV, right vulva; LV, left vulva,; LMA, labia majora; VA, vagina; ELE, extended local excision; LE, local excision; RV, radical vulvectomy; RHV, radical hemivulvectomy; PC + RA, partial colpectomy + rectal amputation; IILND, ipsilateral inguinal lymph node dissection; BILND, bilateral femoral lymph node dissection; ARTx, adjuvant radiotherapy; CT, chemotherapy; SD, stable disease; NED, no evidence of disease; AWD, alive with disease; DOD, died of disease; Nts, not stated; N, negative; P, positive.

than those with positive margins (7). For patients with positive margins, postoperative adjuvant radiotherapy is often recommended to reduce the risk of local recurrence (19-21). The role of lymphadenectomy in the treatment of vulvar ACC remains controversial. Since this tumor rarely metastasizes to groin lymph nodes, not all patients require lymphadenectomy. However, for patients with clinically or radiologically suspected lymph node metastasis, systemic lymphadenectomy is still necessary. Radiotherapy serves an important role in the treatment of vulvar ACC, particularly in patients with positive margins or those who cannot undergo complete surgical excision. Radiotherapy can effectively control local lesions, reduce the risk of recurrence and may improve patients' quality of life. Chemotherapy is predominantly employed for patients exhibiting advanced or recurrent metastatic conditions, with the objective of inhibiting the proliferation and migration of tumor cells through systemic intervention. However, due to the infrequency of vulvar ACC, a standardized chemotherapy protocol remains elusive, necessitating a tailored approach to treatment that is contingent upon the individual circumstances of the patient (22,23). Regarding targeted therapy, a number of ACC cases exhibit expression of the c-Kit receptor (also known as CD117), with the tyrosine kinase inhibitor imatinib emerging as a targeted therapeutic agent aimed to block the c-Kit signaling cascade (24). In phase II clinical trials investigating salivary gland ACC, imatinib has demonstrated efficacy in preventing disease progression (25), while its implications for vulvar ACC warrant further exploration.

A previous study reported the overall recurrence rate of Bartholin's gland ACC as 30% (7), with the simple resection group having a higher recurrence rate compared with the R0 resection group (26 vs. 7%). The recurrence rate for patients with positive margins was 35%, and the metastasis rate was 50%. For the negative margin group, the recurrence rate was 10%, and the metastasis rate was 24%. The distant metastasis rate was 31%, with bone and lung being the main metastatic sites. The effects of chemotherapy varied, with a few patients showing positive responses. The overall 10-year survival rate was 64%, and the median survival time for patients with negative margins (31 years) is significantly longer compared with that of patients with positive margins.

This case underscores the importance of maintaining a high index of suspicion and performing timely biopsy for persistent or slowly enlarging painful vulvar masses in middle-aged and older women . From pathogenesis perspective, the patient in this case was positive for syphilis infection. While previous research has predominantly focused on the role of HPV, existing evidence suggests that Bartholin's gland ACC may be unrelated to HPV infection (26). Molecular pathological studies (11-13) have untangled the potential genetic basis of vulvar ACC. Although the classic MYB-NFIB fusion gene appears less common at this site compared to salivary glands, MYB gene rearrangements and mutations in genes such as KRAS and KDM6A have been reported, which may drive tumor initiation and progression (27). Despite the presence of perineural invasion and skeletal muscle involvement, extended local excision alone was chosen given the negative surgical margins and the absence of clinical or radiological evidence of lymph node metastasis. This approach aligns with the view that surgery alone may be sufficient for early-stage

cases with negative margins (28). Regarding inguinal lymph node dissection, its necessity remains controversial. Routine prophylactic dissection may not be mandatory (29). For metastatic or recurrent disease, precise radiotherapy techniques such as stereotactic ablative radiotherapy have shown potential value (30). In the future, with a deeper understanding of the molecular characteristics of the tumor, targeted therapies against specific targets (CD117) may provide new options for patients with advanced disease.

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Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

Authors' contributions

ZD conceived and designed the study, acquired patient imaging and pathological images, was responsible for data collection, organization, and contributing to writing the paper. ZD and GL interpreted the data. ZD, GL and CZ reviewed and edited the manuscirpt. CZ was responsible for designing the study, providing recommendations for patient treatment plans, and contributed to overseeing the overall work of the research team. All authors agree to take responsibility for all aspects of the research and ensure that appropriate investigations and resolutions are made regarding the accuracy or completeness of any part of the work. All authors have read and approved the final version of the paper. ZD, GL and CZ confirm the authenticity of all the raw data.

Ethics approval and consent to participate

Not applicable.

Patient consent for publication

Written informed consent was obtained from the patient for the publication of the present case report and relevant images.

Competing interests

The authors declare that they have no competing interests.

References

1. Chang Y, Wu W and Chen H: Adenoid cystic carcinoma of the Bartholin's gland: A case report and literature review. J Int Med Res 48: 300060519863540, 2020.
2. Kostov S, Kornovski Y, Ivanova V, Metodiev D, Yordanov A, Slavchev S, Ivanova Y, Seidel A, Juhasz-Böss I, Hasan I, *et al*: Bartholin gland carcinoma: A state-of-the-art review of epidemiology, histopathology, molecular testing, and clinical management. Cancers (Basel) 17: 3819, 2025.
3. Bernstein SG, Voet RL, Lifshitz S and Buchsbaum HJ: Adenoid cystic carcinoma of Bartholin's gland. Case report and review of the literature. Am J Obstet Gynecol 147: 385-390, 1983.
4. Woida FM and Ribeiro-Silva A: Adenoid cystic carcinoma of the Bartholin gland: An overview. Arch Pathol Lab Med 131: 796-798, 2007.
5. Alhashemi M, Oubari S, Mohamad AH, Alhashemi M, Kabel O, Alhelue A and Fattal F: Adenoid cystic carcinoma (ACC) of the Bartholin's gland misdiagnosed for three times: A case report. BMC Womens Health 23: 156, 2023.
6. Verta S, Christmann C and Brambs CE: Adenoid cystic carcinoma of Bartholin's gland: A case report with emphasis on surgical management. Am J Case Rep 23: e935707, 2022.
7. Alsan CI, Vinh-Hung V, Eren F and Abacioğlu U: Adenoid cystic carcinoma of the Bartholin's gland: Case report and systematic review of the literature. Eur J Gynaecol Oncol 32: 567-572, 2011.
8. Hou JL, Wu LY, Zhang HT, Li N and Yu GZ: Clinicopathological characteristics of six patients with adenoid cystic carcinoma of the Bartholin gland. Zhonghua Zhong Liu Za Zhi 32: 290-293, 2010 (In Chinese).
9. Yoon G, Kim HS, Lee YY, Kim TJ, Choi CH, Song SY, Kim BG, Bae DS and Lee JW: Analysis of clinical outcomes of patients with adenoid cystic carcinoma of Bartholin glands. Int J Clin Exp Pathol 8: 5688-5694, 2015.
10. Topolovec Z, Blažičević V, Šijanović S and Vidosavljević D: Squamous cell carcinoma of Bartholin gland coexistent with human papillomavirus. Eur J Gynaecol Oncol 36: 482-484, 2015.
11. Wang W, Chen H, Guo H, Chen L, Zhu M and Zhu Y: Adenoid cystic carcinoma of the Bartholin's gland is easily misdiagnosed: A case report and literature review. Medicine (Baltimore) 101: e31744, 2022.
12. Togashi Y, Dobashi A, Sakata S, Sato Y, Baba S, Seto A, Baba S, Seto A, Mitani H, Kawabata K and Takeuchi K: MYB and MYBL1 in adenoid cystic carcinoma: Diversity in the mode of genomic rearrangement and transcripts. Mod Pathol 31: 934-946, 2018.
13. Nakamura K, Aimono E, Tanishima S, Nomura H, Imai M, Hayashi H and Nishihara H: Genetic profiling of patients with adenoid cystic carcinoma of the Bartholin's glands reveals potential new routes for targeted therapies: A case report. Diagn Pathol 15: 64, 2020.
14. KurmanRJ,EllensonLHandRonnettBM(eds):Blaustein'sPathology of the Female Genital Tract. Springer, Cham. Available: https://link.springer.com/referencework/10.1007/978-3-319-46334-6.
15. Finan MA and Barre G: Bartholin's gland carcinoma, malignant melanoma and other rare tumours of the vulva. Best Pract Res Clin Obstet Gynaecol 17: 609-633, 2003.
16. Akbarzadeh-Jahromi M, Aslani FS, Omidifar N and Amooee S: Adenoid cystic carcinoma of Bartholin's gland clinically mimics endometriosis, A case report. Iran J Med Sci 39: 580-583, 2014.
17. Santiago AE, Teunissen N, Ricardo BFP, Cândido EB, Furtado RS and Filho ALD: High-grade transformation in adenoid cystic carcinoma of the Bartholin gland: Case report. Rev Bras Ginecol Obstet 43: 980-984, 2021.
18. Oonk MHM, Planchamp F, Baldwin P, Mahner S, Mirza MR, Fischerová D, Creutzberg CL, Guillot E, Garganese G, Lax S, *et al*: European society of gynaecological oncology guidelines for the management of patients with vulvar cancer-update 2023. Int J Gynecol Cancer 33: 1023-1043, 2023.
19. Takatori E, Shoji T, Miura J, Takeuchi S and Sugiyama T: Chemoradiotherapy with irinotecan (CPT-11) for adenoid cystic carcinoma of Bartholin's gland: A case report and review of the literature. Gynecol Oncol Case Rep 4: 16-19, 2012.
20. Evin C, Just PA, Borghese B, Fabiano E, Bennani S, Canny E, Marisa L, Derive N, Laurent-Puig P, Alexandre J and Durdux C: Adenoid cystic carcinoma of Bartholin's gland, a case report with genomic data and literature review. Cancer Radiother 27: 328-336, 2023.
21. Güral Z, Yücel S and Agaoglu F: Bartholin's gland adenoid cystic carcinoma: Report of three cases and the review of literature. Indian J Cancer 61: 346-349, 2024.
22. Johnson LR, Nair RP, Sambasivan S, Mony RP, Gangadharan J, Kumar A and Ahamed IM: Adenoid cystic carcinoma of vulva-11 years' single-institution experience. J Obstet Gynaecol India 67: 196-201, 2017.
23. Samba S, Guerrouaz MA, Bensghier A, Berhili S, Moukhlissi M and Mezouar L: Adenoid cystic carcinoma of the vulva: A case report. Cureus 16: e56048, 2024.
24. Alcedo JC, Fábrega JM, Arosemena JR and Urrutia A: Imatinib mesylate as treatment for adenoid cystic carcinoma of the salivary glands: Report of two successfully treated cases. Head Neck 26: 829-831, 2004.

25. Faivre S, Raymond E, Casiraghi O, Temam S and Berthaud P: Imatinib mesylate can induce objective response in progressing, highly expressing KIT adenoid cystic carcinoma of the salivary glands. J Clin Oncol 23: 6271-6273, 2005.
26. Nieuwenhuyzen-de Boer GM, Dasgupta S, Ewing-Graham PC and Van Bockstal MR: Adenoid cystic carcinoma of the Bartholin gland is not HPV-related: A case report and review of literature. Pathol Res Pract 216: 152968, 2020.
27. Doutel D, Venda D, Silva F, Martins C, Félix A and Ferreira J: Adenoid cystic carcinoma of the vulva and vagina: A clinicopathologic, immunohistochemical, and molecular characterization of five cases. Int J Gynecol Pathol 43: 637-645, 2024.
28. Zhu J, Yin L, Wang Q, Wu X and Ke G: Clinicopathologic and survival analysis of patients with adenoid cystic carcinoma of vulva: single-institution experience. Int J Clin Oncol 25: 2144-2150, 2020.
29. Barcellini A, Gadducci A, Laliscia C, Imparato S, Vitolo V, Preda L and Valvo F: Adenoid cystic carcinoma of Bartholin's Gland: What is the best approach? Oncology 98: 513-519, 2020.
30. Irfan H, Ben-Tarifite Y and Mahmood R: Recurrent adenoid cystic carcinoma of the vulva: A case report of salvage stereotactic ablative radiotherapy to the pudendal nerve. Radiol Case Rep 20: 5148-5155, 2025.