

# Adenoid Cystic Carcinoma of the Bartholin Gland a Rare Tumor: A Case Report

Yassine Bouhtouri<sup>1\*</sup>, Ihssan Elouarith<sup>2</sup>, Fatima EL Mangoub<sup>1</sup>, Saad Benali<sup>1</sup>, Moulay Abdelah Baba Habib<sup>1</sup>, Moulay Mehdi El Hassani<sup>1</sup>, Hafid Hachi<sup>3</sup>, Basma El Khannoussi<sup>2</sup>, Jaouad Kouach<sup>1</sup>

<sup>1</sup>Department of Gynecology and Obstetrics, Military Hospital of Instruction Mohamed V Rabat, Rabat, Morocco

<sup>2</sup>Department of Anatomy and Pathology at the Institute of National Oncology, Moulay Abdellah Hospital, Rabat, Morocco

<sup>3</sup>Gynaecological-mammary Pole at the Institute of National Oncology Moulay Abdellah Hospital Rabat, Morocco

DOI: 10.36348/sijog.2022.v05i03.013

| Received: 14.02.2022 | Accepted: 20.03.2022 | Published: 30.03.2022

\*Corresponding author: Yassine Bouhtouri

Department of Gynecology and Obstetrics, Military Hospital of Instruction Mohamed V Rabat, Rabat, Morocco

## Abstract

**Introduction:** cystic adenoid carcinoma of the Bartholin gland is a very rare tumor with a low incidence of 1% of female genital malignancies; it is characterized by its aggressive nature and frequent recurrence. **Case report:** We report a case of a cystic adenoid carcinoma of Bartholin's gland in a 38-year-old female patient who was treated surgically with good evolution. **Conclusion:** Adenoid cystic carcinoma is a very rare tumor of the Bartholin gland, whose diagnosis is based on the anatomopathological study.

**Key words:** Bartholin gland carcinoma, genital tract carcinoma, Adeno cystic carcinoma.

**Copyright © 2022 The Author(s):** This is an open-access article distributed under the terms of the Creative Commons Attribution 4.0 International License (CC BY-NC 4.0) which permits unrestricted use, distribution, and reproduction in any medium for non-commercial use provided the original author and source are credited.

## INTRODUCTION

Adenoid cystic carcinoma of Bartholin's gland is a very rare tumor; its incidence is 1% of all female genital malignancies and 1-7% of all vulvar carcinomas [1]. It occurs mostly in women over 40 years of age and is characterized by its aggressive nature with prolonged clinical symptoms and frequent local recurrence [2].

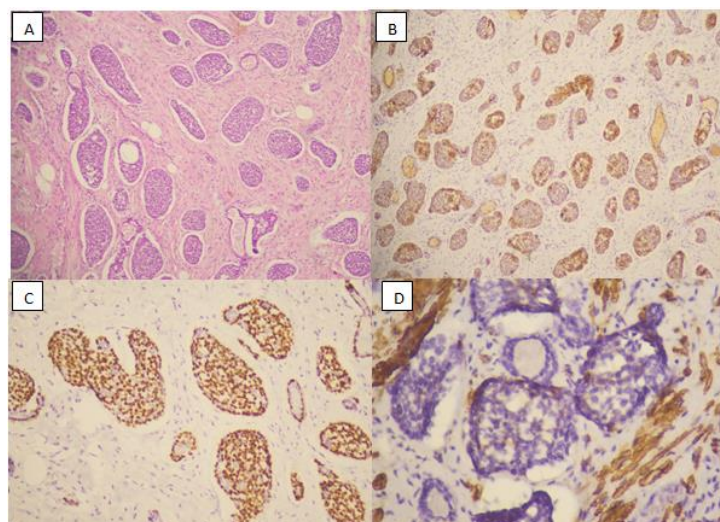
## CASE PRESENTATION

We report the case of a 38-year-old female patient, with no notable pathological history. She presented in our structure for a painful swelling of the Bartholin's gland evolving for 5 days. The clinical examination revealed a patient in good general condition, afebrile, and hemodynamically stable. Genitally, swelling of the right Bartholin gland with a suspicious palpable induration.

The patient underwent an excision of the Bartholin gland and was referred for anatomopathological study. The operation was simple

and the patient was discharged from the hospital 48 hours after the procedure. The result of the anatomopathological study showed a carcinomatous proliferation made of basal cells with dense angular nuclei showing moderate cytonuclear atypia and with rare mitoses. These cells are mainly arranged in clusters, with cribriform structures in places. They are bordered by myoepithelial cells. The stroma is hyalinized fibroblastic. The immunohistochemical study showed positive labelling of the luminal tumor cells by anti-CK7 antibodies and positive labelling of the myoepithelial tumor cells by anti-P63 and anti-AML antibodies. Because of these morphological and immunohistochemical aspects, the diagnosis of a cystic adenoid carcinoma of Bartholin's gland was retained with healthy margins of resection. The patient underwent a thoracoabdomino-pelvic scan which did not reveal any secondary localization.

The patient has not presented any recurrence to date.



**Fig-1: Histopathological findings of the tumor**

#### Immunostaining findings of the tumor

- A. Tubular pattern contains simple tubules composed of inner ductal and outer myoepithelial cells (HE x 10).
- B. CK7 staining is positive in the tumor cells of epithelial component (x10)
- C. P63 staining (x20)
- D. AML staining is positive in the tumor cells of myoepithelial component (x40)

## DISCUSSION

Adenoid cystic carcinoma of Bartholin's gland is an extremely rare tumor in clinical practice. It usually occurs in women over 40 years of age and often presents as a painful nodule [2]. The tumor grows slowly with local infiltration and a tendency to invade the nerve. Recurrences are frequent. It can metastasize to the lung, bone, liver, and other organs, as well as to the inguinal or pelvic lymph nodes. Abdominal and pelvic CT scans have been described to assess the local and lymph node extension of the disease, but no imaging modality has been able to prove its superiority to date [3].

The final diagnosis can only be made after histopathological examination, as in our case. Histopathologically, adenoid cystic carcinoma of Bartholin's gland is a characteristic biphasic glandular tumor composed of ductal and myoepithelial cells. The characteristic feature is the presence of tubular, cribriform, and solid patterns. The most recognizable architectural pattern is the cribriform pattern, characterized by nests of tumor cells interrupted by sharply cut spaces filled with basophilic matrix. The myoepithelial cells have dark, angular nuclei and sparse cytoplasm, giving a basaloid appearance. Immunohistochemically, the ductal component is typically positive for CD117 and CK7 while the myoepithelial component is positive for myoepithelial markers such as p63 and SMA [4, 5].

The rarity of the pathology does not allow for a consensus for management. For small unilateral lesions far from the midline, simple surgical excision with negative resection margins is sufficient, which is the case of our patient; however, extensive radical vulvectomy is required if the lesion is extensive. Dissection of inguinofemoral and pelvic lymph nodes should be performed only if they are clinically and radiologically involved [6, 7]. Adjuvant radiotherapy is recommended in case of positive surgical resection margins or in case of perineural invasion. The local recurrence rate is 9.5% after adjuvant radiotherapy, compared to 37.5% for those who did not receive adjuvant radiotherapy [8].

Neoadjuvant chemotherapy or in combination with radiotherapy is indicated for non-operable or metastatic tumors and also significantly reduces the relapse rate [9].

The average survival time of patients with adenoid cystic carcinoma of the Bartholin gland is 5 to 10 years. Overall survival after surgery depends on the stage of the disease and the resection margins, less than 07 years in case of affected margins and between 15 years and 30 years in case of healthy margins [8].

## CONCLUSION

Adenoid cystic carcinoma is a very rare tumor of the Bartholin gland which is characterized by its slow development, aggressive character and very high recurrence rate. The recommended treatment remains surgery with negative resection margins for operable tumors. Radiation therapy is considered if the margins are positive, and chemoradiotherapy for patients with advanced stages of disease or non-operable.

## REFERENCES

1. Yang, S.Y.V., Lee, J.W., Kim, W.S., Jung, K.L., Lee, S.J., Lee, J.H. (2006). Adenoid cystic carcinoma of the Bartholin's gland: Report of two cases and review of the literature. *Gynecol Oncol*, 100:422-5.
2. Hung, F.Y., Wang, K.L., Wang, T.Y. (2005). Adenoid Cystic Carcinoma of Bartholin's Gland: A Case Report. *Taiwan J Obstet Gynecol*, 44; 72-4.
3. Di Donato, V., Casorelli, A., Bardhi, E. (2017). Bartholin gland cancer. *Crit Rev Oncol Hematol*, 117; 1–11.
4. WHO Classification of tumors of female genital tumours Edited by the who classification of tumours editorial board, 5th ed. 2020
5. WHO Classification of head and neck, 4th ed. Lyon : IARC Press ; 2017
6. Anaf, V., Buxant, F., Rodesch, F., Simon, P., Van de Stadt, J., Noel, J.C. (1999). Adenoid cystic carcinoma of Bartholin's gland: what is the optimal approach? *Eur J Surg Oncol*, 25; 406-9.
7. Abrao, F.S., Marques, A.F., Marziona, F., Abrao, M.S., Junqueira, L.C.U., Torloni, H. (1985). Adenoid cystic carcinoma of Bartholin's gland: review of the literature and report of two cases. *J Surg Oncol*, 30; 132-7.
8. Alsan, C.I., Vinh-Hung, V., Eren, F. (2011). Adenoid cystic carcinoma of the Bartholin's gland: case report and systematic review of the literature. *Eur J Gynaecol Oncol*, 32; 567–572.
9. Ouldamer, L., Chraïbi, Z., Arbion, F., Barillot, I., Body, G. (2013). Bartholin's gland carcinoma: Epidemiology and therapeutic management. *Surg Oncol*, 22; 117-22.