



Objective

Up to our knowledge, this is the first reported case of synchronous ovarian and vulva cancer.

Background

The Surveillance, Epidemiology, and End Results (SEER) Program came to agreement to restrict the use of the term “synchronous” multiple primary cancers to when the occurrence of the second primary cancer was **within 2 months**, and if later than 2 months it should be termed “metachronous”.

Ovarian clear-cell carcinoma (OCCC) represents 5%–10% of epithelial ovarian cancers and is recognized as a more aggressive disease with a distinct biologic behavior.

Bartholin gland carcinoma comprises approximately 0.1%–5% of all vulvar carcinomas and less than 1% of female genital malignancies.

Adenoid cystic carcinoma of Bartholin gland is a rare variant, comprising 15% of all Bartholin gland carcinomas. About 350 cases of Bartholin gland adenoid cystic carcinoma have been reported in the literature.

Case Presentation

61 years old, presented by post menopausal bleeding. On clinical examination, a non-tender **2-cm right Bartholin gland mass** was found. An ultrasound showed **a complex multiloculated left adnexal mass** measuring approximately 15 × 8 × 10 cm containing a 7 × 5 × 5 cm solid component. **Serum CA 125** was 59 IU/ml.

CT showed a larger (approximately 32 × 13.5 × 22.5 cm) complex mass arising out of the pelvis along with a right-side Bartholin mass with suspicious right inguinal nodes. **MRI** confirmed the same findings.

Surgery performed included a total abdominal hysterectomy, bilateral salpingo-oophorectomy, infracolic omentectomy, pelvic peritoneal biopsies, and peritoneal washings. At the same time, the right Bartholin gland mass was excised. **Final histology** revealed **Stage 2B left OCCC** and a right **Bartholin gland** adenoid cystic carcinoma with lymphovascular invasion, incompletely excised, staged at least FIGO **Stage 1B**.

MDT to decide the best route of care. **PET** was performed that did not detect any nodal or distant disease. The MDT decided to **proceed with three cycles of chemotherapy** so that the ovarian cancer had some therapy and **then remove the groin nodes** before completing the remaining three cycles. After three cycles (around 4 months after the initial surgery), excision of the Bartholin gland scar site and bilateral inguino-femoral lymph node dissection were performed. Recovery was uneventful.

Case Presentation (continue)

Histopathology reported no residual carcinoma in the scar excision site, but **1/5 lymph nodes** on the right side and **1/8 lymph nodes** on the left side had metastatic adenocarcinoma.

All overall morphologic and immunohistochemical features were otherwise consistent with **metastatic OCCC**. Further adjuvant chemotherapy cycles were advised following MDT discussion

Discussion

Balancing the need for chemotherapy while addressing the **requirement for inguinal lymphadenectomy** was the **challenge faced**, as well as how to prioritize care.

Bartholin gland adenoid cystic carcinomas are very rare and are considered more indolent and have a lower rate of groin node metastasis compared with vulvar squamous cell carcinomas. Nevertheless, there were concerns regarding the CT/MRI scans, suggesting suspicious inguinal lymph glands. Although the **PET scan was negative, the false-negative rates** for detecting nodal disease in vulvar cancer are reported to be **10% or higher**.

However, removing the inguinal glands would delay chemotherapy, considering the high rates of postoperative wound infection and breakdown.

It was **not anticipated** that the groin nodes would prove to have metastases from the ovarian cancer considering the previous early-stage disease. Even in **advanced ovarian cancer** the **incidence of** inguinal node involvement is reported **at around 3%**.