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CASE REPORT

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Ovarian apocrine carcinoma arising from a mature cystic teratoma: a case report

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Abstract

Background: Mature cystic teratomas of the ovary are common; however, their malignant transformations are rare. Their transformation into apocrine carcinoma has been reported in only a few cases. Herein, we report our experience with this rare cancer. **Case**: In a 44-year old woman, bilateral ovarian tumor were incidentally discovered during preoperative positron emission tomography for right breast cancer. Laparotomy revealed ovarian tumors and enlarged para-aortic lymph nodes. Therefore, total hysterectomy, bilateral salpingo-oophorectomy, partial omentectomy, and para-aortic lymph node dissection were performed. Pathological examination revealed that the primary left ovarian tumor was a mature cystic teratoma with malignant transformation into apocrine carcinoma. Metastasis was observed in the resected lymph nodes. Subsequently, a mastectomy was performed for right breast cancer, which was determined as invasive ductal carcinoma. Postoperative recovery was favorable. No adjuvant chemotherapy was administered for ovarian cancer; follow-up was continued. **Conclusion**: This case involved synchronous primary ovarian and breast cancers. It was difficult to determine whether the ovarian cancer was primary or metastatic until the surgery was performed. Ovarian apocrine carcinoma is rare, and no established effective therapy exists, therefore complete surgical excision remains critical for effectively managing this rare cancer.

Keywords

Apocrine carcinoma; Breast cancer; Malignant transformation; Mature cystic teratoma

1. Introduction

Mature cystic teratomas are the most common benign ovarian tumors in women. However they rarely undergo malignant transformation, with an incidence of approximately 1.5–2% [1]. Squamous cell carcinoma is the most frequent form, whereas its transformation into adenocarcinoma or sarcoma is relatively uncommon [1]. Malignant transformation into apocrine carcinoma has been reported only a few times [2–4]. In this case, an ovarian tumor was incidentally discovered during the preoperative evaluation for breast cancer; a diagnosis of apocrine carcinoma arising from a mature cystic teratoma was confirmed following laparotomy. Metastasis to the retroperitoneal lymph nodes was observed, and breast cancer was diagnosed separately as invasive ductal carcinoma. This rare case involved simultaneous occurrence of two distinct primary cancers.

2. Case presentation

A 44-year-old nulliparous, with a regular menstrual cycle (28-day cycle), Jehovah's Witness woman without relevant medical or family history, had been experiencing discomfort in her right breast lump for 5 years. Although she did not have specific medical history and her breast lump was initially diagnosed as a sebaceous cyst at another hospital, she was later diagnosed with breast cancer. Positron emission tomography-computed tomography (PET-CT) was performed for preoperative staging, which revealed bilateral ovarian tumors and abnormal uptake in the left ovarian tumor and para-aortic lymph nodes (Fig. 1a). Although ovarian metastasis from breast cancer was considered, a primary malignant ovarian tumor could not be ruled out; she was referred to our Department of Gynecology. On pelvic examination, a large pelvic mass was palpated down to the umbilicus and freely mobile. Transvaginal ultrasonography revealed a multilocular mass with high internal echoes. Contrast-enhanced magnetic resonance imaging indicated that the left ovarian tumor was a mature cystic teratoma with solid components, suggesting malignant transformation (Fig. 1b). Blood test results revealed slightly elevated levels of tumor markers: Squamous cell carcinoma antigen (SCC): 1.9 ng/mL, carbohydrate antigen 19-9 (CA19-9): 66.9 U/mL and carbohydrate antigen (CA125): 50.5 U/mL.

The patient underwent exploratory laparotomy, which revealed a smooth-surfaced left ovarian tumor (the largest diam-

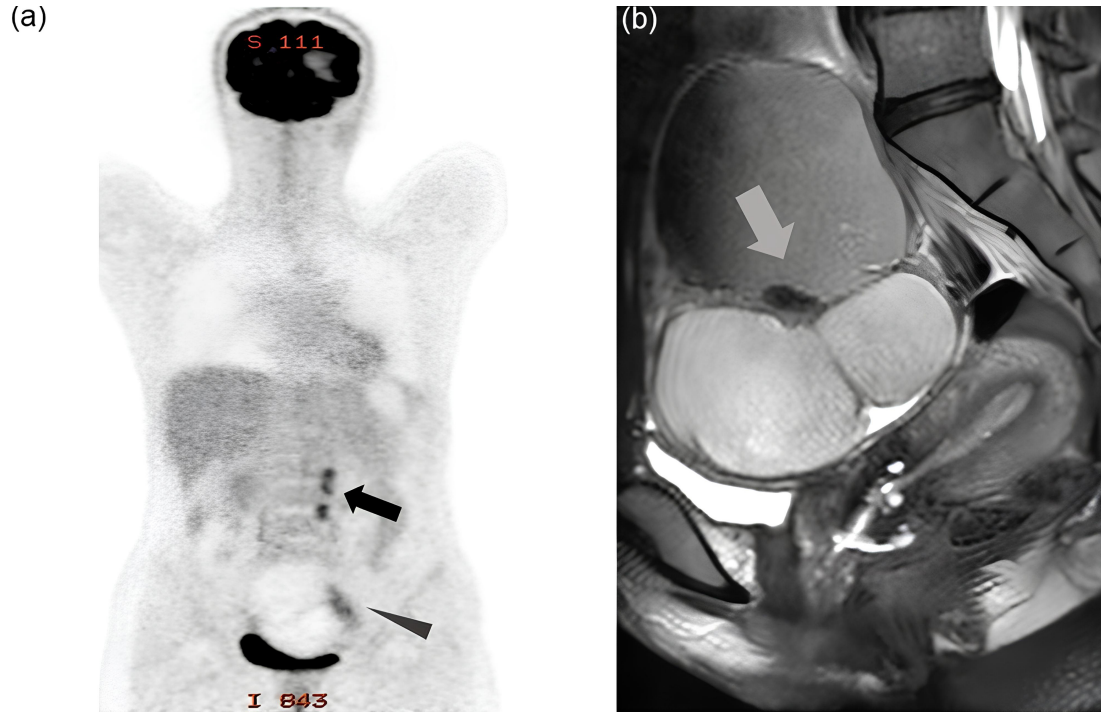

FIGURE 1. Medical imaging. (a) Accumulation observed in the para-aortic lymph node (←) and the left ovarian tumor (◀). (b) Contrast-enhanced magnetic resonance imaging (T2-weighted imaging): The left ovarian tumor shows liquid components, including fat and hemorrhagic areas, with suspected nodular components (←), indicating possible malignant transformation.

eter was 14 cm) and a right ovarian tumor (the largest diameter was 4 cm), both of which were mobile, and an enlarged para-aortic lymph node. Rapid intraoperative pathological diagnosis confirmed the malignancy of the left ovarian tumor. Therefore, total hysterectomy, bilateral salpingo-oophorectomy, partial omentectomy dissection, and para-aortic lymph node (left: 326B1, B2; right: 326B2) biopsy were performed. Additionally, the presacral and left internal iliac lymph nodes, which were palpable and hard, were biopsied. Given the patient's Jehovah's Witness faith (inability to receive blood transfusion owing to religious beliefs), systematic lymph node dissection was avoided to minimize blood loss. Pathological findings of the left ovarian tumor (Fig. 2a) revealed a mature teratoma containing the skin; gastrointestinal epithelium; respiratory epithelium; serous and mucinous glands; and adipose tissue, along with adenocarcinoma characterized by abundant eosinophilic cytoplasm. The tumor showed features of apocrine carcinoma, with tumor cells containing cytoplasmic vacuoles proliferating in cord-like structures within the fibrous connective tissue, and apocrine-type decapitation secretion was observed (Fig. 2b). Immunohistochemical staining revealed an Estrogen receptor (ER)-negative, Progesterone receptor (PgR)-negative, Human epidermal growth receptor type2 (HER2)-negative, Androgen receptor (AR)-positive and Epidermal growth factor receptor (EGFR)-positive tumor, characteristics of apocrine carcinoma (Fig. 2c). In the right ovary, a benign mature cystic teratoma was identified. Moreover, the resected lymph nodes showed histological findings similar to those in the left ovary. No metastases were observed in the omentum or appendix.

Postoperative recovery was favorable; the patient was discharged on postoperative day 10. One month later, surgery was performed for right breast cancer, which was identified as invasive ductal carcinoma (T1b N0 M0, Stage IB, ER-positive and PgR-positive). The pathological findings of ovarian and breast cancers were distinct, confirming the presence of dual cancers. No adjuvant chemotherapy was administered for ovarian apocrine carcinoma. One month after discharge, compared with before surgery, blood test results revealed decreased levels of tumor markers: SCC: 0.7 ng/mL, CA19-9: 49.8 U/mL and CA125: 18.1 U/mL. Given the patient's Jehovah's Witness faith, we concerned that bone marrow suppression could be critical due to her inability to receive blood transfusion, no adjuvant chemotherapy was conducted. She continues to be monitored in the outpatient department.

3. Discussion

Malignant transformation of mature cystic teratomas is rare, occurring in approximately 1.5–2% of cases [1]. The median age for this transformation is 55 years; the tumor diameter is often greater than 10 cm [1]. Squamous cell carcinoma is the most frequent type of transformation, accounting for 85–90% of cases, followed by adenocarcinoma and sarcoma [1]. Most cases involve peritoneal dissemination or direct invasion, with lymph node metastasis being the least common [5]. In this case, the patient was relatively young (44 years) and tumor size was similar without evidence of peritoneal dissemination but with lymph node metastasis. Reports of apocrine carcinoma arising from a teratoma are rare; however, previous cases have also been reported in younger patients with lymph node metastasis (Table 1, Ref. [2–4]). Upon reviewing four cases, including the present case, we observed that tumor sizes varied significantly, ranging from over 10 cm [2], as seen in this case, to approximately 5 cm [3, 4].

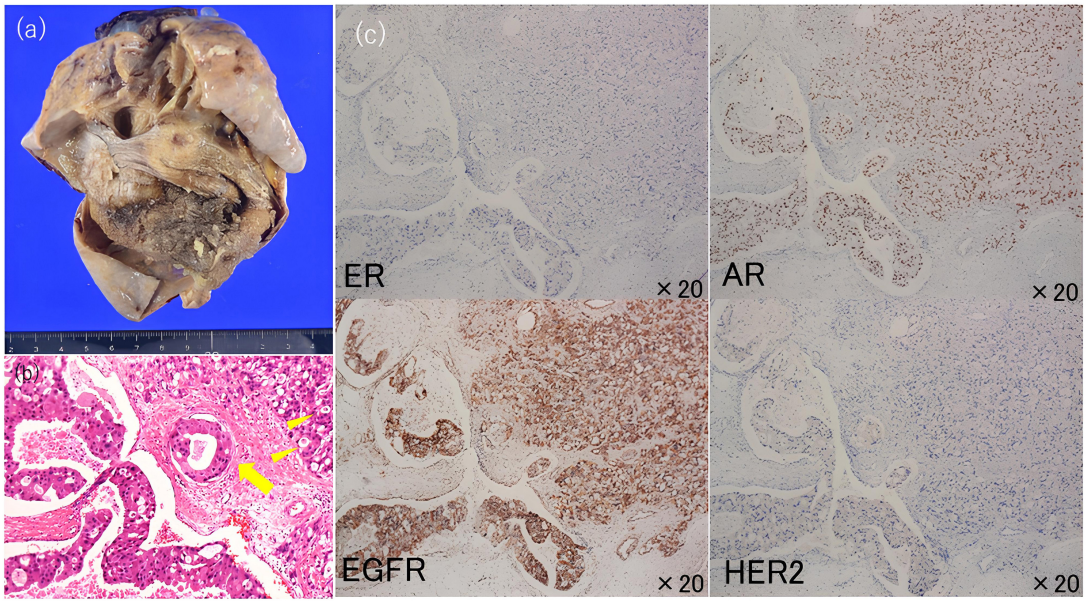

FIGURE 2. Pathological findings. (a) The tumor, approximately the size of a child's head, contains fat, bone and hair components. (b) Hematoxylin and eosin staining (×20 magnification): Adenocarcinoma with abundant eosinophilic cytoplasm is observed, depicting fence-like proliferation within fibrous connective tissue. Tumor cells exhibit cytoplasmic vacuoles (◀), and decapitation secretion is noted within the glandular lumen (←). (c) Immunohistochemical staining reveals that the tumor is ER-negative, AR-positive, EGFR-positive and HER2-negative. ER: Estrogen receptor; AR: Androgen receptor; EGFR: Epidermal growth factor receptor; HER2: Human epidermal growth receptor type2.

While malignant transformation of teratomas is often characterized by dissemination or invasion into surrounding tissues, no adhesion was observed in this case. Similarly, there is a case report underwent surgical treatment due to pain caused by torsion without adhesions [3], whereas there is a case with adhesions to the rectum [2]. Larger tumors were found to be multilocular with solid components [2], whereas the two smaller tumors were unilocular [3, 4]. These findings suggest that an ovarian apocrine carcinoma may have characteristics distinct from the more common malignant transformation of teratomas to squamous cell carcinoma. Additionally, it may be difficult to preoperatively diagnose apocrine carcinoma based on imaging findings alone. Whether age and lymph node metastasis are characteristics of apocrine carcinoma in patients with teratomas remains unclear, and further studies are needed. No chemotherapy regimen has been established for this condition. In a report by Xu *et al*. [2], although platinum-based agents were administered in several cases of recurrent ovarian cancer, none were effective.

Apocrine carcinoma typically originates from the breast or skin apocrine glands, accounting for <1% of cutaneous malignancies [6] and 1% of breast cancers [7]. It is characterized by abundant eosinophilic or vacuolated cytoplasm, nuclear pleomorphism and decapitation secretion [8]. Immunohistochemical analysis often shows ER and PgR negativity, AR positivity, and positivity for HER2 or EGFR [8].

This case is unique because breast and ovarian cancers simultaneously occurred. Among the infrequent apocrine carcinomas, those originating from the breast and skin are common. Hence, ovarian metastasis from breast cancer was considered in the differential diagnosis; however, based on the pathological findings, the patient was diagnosed with synchronous primary cancers. Regarding the metastasis, the lymph node metastasis from the breast cancer is approximately 33% [9]. However, peritoneal and retroperitoneal metastases are rare at 0.6% [10]. Lymph node metastasis from the malignant transformation of mature cystic teratomas is also rare. Initially, metastasis from the breast cancer was also considered. However, ultimately, the para-aortic lymph node metastasis was diagnosed as originating from the ovarian carcinoma. Regarding onset, although patients reported by Xu *et al*. [2] and Morimitsu *et al*. [3] underwent surgery owing to pain, this patient did not experience any pain, allowing the tumor to grow into a large mass. It was only incidentally discovered on preoperative PET-CT performed to evaluate right breast cancer.

4. Conclusion

Herein, we report a rare case of malignant transformation of a mature cystic teratoma into apocrine carcinoma. The patient continues to be monitored without postoperative adjuvant chemotherapy. Given the absence of established chemotherapeutic regimens for this rare cancer, complete surgical excision remains critical for effective management.

5. Limitation

Ovarian Apocrine Carcinoma Arising from a Mature Cystic Teratoma is a quite rare tumor. There is no established standard of care yet. The efficacy of anticancer therapy is also unknown. Further case accumulation and study are important.

TABLE 1. Past reports of ovarian apocrine carcinoma.

Author	Reported year	Age	Delivery history	Tumor size/right or left	Tumor characteristics	Metastatic site
This Case	2024	44	nulliparous	14 cm left ovary	multilocular cystic tumor with solid components, no adhesion	Pelvic/Para-aortic Lymph node
Xu L *et al*. [2]	2018	46	multiparous	12 cm left ovary	multilocular cystic tumor with solid components, adhered to rectum	Pelvic Lymph node
Holmes M *et al*. [4]	2017	32	unknown	4 cm unknown	unilocular cystic tumor	Supraclavicular Lymph node 6 months later after surgery
Morimitsu Y *et al*. [3]	1993	41	unknown	5.5 cm right ovary	unilocular cystic tumor, torsion of its pedicle	None

AVAILABILITY OF DATA AND MATERIALS

Data sharing is not applicable to this article because this is a case report.

AUTHOR CONTRIBUTIONS

HK—conception; manuscript writing; acquisition of data and editing. RY, YF—acquisition of data. MN—analysis and interpretation of data. MK—supervision.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

This study was performed in line with the principles of the Declaration of Helsinki. It is not a clinical trial, NHO Kagoshima Medical Center has confirmed that ethics approval is not required. Written informed consent was obtained from the patient for publication of this case report and accompanying images.

ACKNOWLEDGMENT

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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