Non-Hodgkin Lymphomas in Mature Cystic Teratomas: A Case Report and Review of the Literature

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Abstract

Background: Mature teratomas, better known as dermoid cysts, are the most common ovarian neoplasms in women in the second and third decade of life. They are invariably benign, and most women are asymptomatic. Ovarian cystectomy is the preferred therapeutic option.

Case: A 24-year-old woman was planned for elective laparoscopic cystectomy for a suspected teratoma; operative findings led to a unilateral oophorectomy. Pathological analysis of the specimen revealed a focus of large cell lymphoma of unknown lineage arising in a mature cystic teratoma. A total body positron emission tomography (PET) scan revealed no other disease, and the patient was managed conservatively with regular follow-up.

Conclusion: Lymphoma in a teratoma is an excessively rare finding with only five previously reported cases. A review of the literature revealed very different theories as to its pathogenesis and management.

Résumé

Contexte : Le tératome mature, mieux connu sous le nom de kyste dermoïde, est le néoplasme de l'ovaire le plus répandu chez les filles et les femmes de 10 à 29 ans. Il est toujours bénin et généralement asymptomatique. L'approche de traitement privilégiée est la kystectomie.

Cas : Une femme de 24 ans devait subir une kystectomie par laparoscopie en raison d'un tératome soupçonné; or, les observations faites durant l'opération ont mené à la réalisation d'une ovariectomie unilatérale. L'analyse pathologique des tissus extraits a révélé la présence d'un lymphome à grandes cellules de lignée inconnue à l'intérieur d'un tératome kystique mature. La tomographie par émission de positons (TEP) de l'ensemble du corps n'a décelé aucune autre anomalie. La patiente a fait l'objet d'une prise en charge conservatrice et d'un suivi régulier.

Conclusion : La découverte d'un lymphome dans un tératome est extrêmement rare : seuls cinq cas avaient été rapportés avant celui-ci. Une revue de la littérature a montré qu'il existait des théories très variées sur la pathogenèse et la prise en charge de cette affection.

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Key Words: Teratoma, ovarian neoplasm, lymphoma

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INTRODUCTION

Mature cystic teratomas are very common benign tumours that account for approximately 10% to 20% of all ovarian neoplasms.[1] Very rarely, unexpected findings on pathology are discovered in an otherwise benign-appearing teratoma. We report herein a case of an unexpected non-Hodgkin large cell lymphoma of unknown lineage in a mature cystic teratoma and briefly review the current literature.

THE CASE

A nulliparous, previously healthy 24-year-old Caucasian woman was referred to our gynaecology clinic after a previously resolved episode of severe right lower abdominal pain 6 months prior. She was asymptomatic at the time of first encounter. A transabdominal ultrasound revealed a heterogeneous, cystic ovarian mass measuring 7.5 cm × 4.7 cm, with a predominance of adipose tissue suggestive of a cystic teratoma. She was scheduled for a laparoscopic cystectomy to prevent further occurrence of what was presumed to have been, in retrospect, an ovarian torsion.

The patient underwent an elective laparoscopic cystectomy. The right ovary and fallopian tube appeared necrotic and were found to be amputated from their vascular attachment at approximately half of the length of the tube. This seemed to indicate a possible previous ovarian torsion and leakage of fluid, causing extensive adhesions. After releasing complex adhesions, we noted the patient had sustained an almost complete salpingo-oophorectomy due to torsion, and the residual necrotic fallopian tube and ovary were removed. On gross examination, the ovary comprised sebaceous material, teeth, and hair, consistent with a mature cystic teratoma. The left ovary and fallopian tube were unremarkable.

Gross examination of the specimen revealed a cystic structure comprising sebaceous material, teeth, and hair, consistent with a mature cystic teratoma; part of the wall of the cyst contained adipose tissue, with areas of bone and cartilage. Microscopic examination revealed a classic mature cystic teratoma, except for one slide that showed the cyst focally lined by a population of large atypical cells (Figure 1) that expressed CD45 and CD30 (Figure 2); the proliferation rate was 80% to 90% as determined by immunohistochemical staining for Ki67. Other immunostains were negative, including CD2, CD3, CD5, CD20, CD25, and perforin. A diagnosis was made of large cell lymphoma, precise subtype unclear.

The patient was seen in clinic postoperatively to discuss the results of the pathology. On questioning, she denied any B-symptoms (night sweats, fever, or weight loss) that might suggest a systemic lymphoma. She was referred to the hematology-oncology clinic for management. Physical examination and blood work were negative. A whole body positron emission tomography scan showed no lymphoma. The patient was discussed at Tumour Boards; it was concluded that there was insufficient evidence of a systemic lymphoma, and she did not receive systemic treatment. A follow-up ultrasound done 6 months postoperatively showed a questionable remnant of right ovary with no obvious lesion. The patient will continue to be followed closely.

ABBREVIATIONS

CT	Computed tomography
DLBCL	diffuse large B-cell lymphoma
DLBCL-CI	diffuse large B-cell lymphoma associated with chronic inflammation
PET	positron emission tomography
R-CHOP	rituximab, cyclophosphamide, hydroxydaunomycin, oncovin, prednisone

DISCUSSION

Mature cystic teratomas represent approximately 95% of ovarian germ cell tumours.[2] They are composed of tissue originating from any of the three germ cell layers (i.e., ectoderm, mesoderm, and endoderm).[3] They are commonly composed of sebaceous material, hair, teeth, cartilage, bone, thyroid, and neural tissue but have the potential to include any type of tissue.[4] The presence of lymphoma is a very rare occurrence in mature cystic teratomas. To our knowledge, there have been only five reports of an incidental finding of lymphoma in a mature cystic teratoma (Table).[5–9] We will review herein the theories postulated by the authors of these reports.

Primary Ovarian Lymphoma

In three of these cases, a possible primary ovarian lymphoma was suggested as an explanation for their findings.[5–7] Primary ovarian lymphoma is a relatively rare occurrence in itself, constituting approximately 0.5% of extranodal non-Hodgkin lymphomas and 1.5% of all ovarian malignancies.[10] Criteria for the diagnosis of primary ovarian lymphoma were proposed by Fox et al.[11]: (1) tumour is confined to the ovarian regional lymph nodes or adjacent organs at the time of diagnosis, (2) lymphoma cells are absent from the bone marrow and peripheral blood, and (3) extraovarian disease appears only several months after the ovarian presentation.

In the first case report published by Seifer et al.,[5] the patient was a 24-year-old woman found to have a right adnexal mass on routine prenatal examination. Postpartum oophorectomy revealed a diffuse large B-cell lymphoma in a mature cystic teratoma on pathological analysis.[5] A primary ovarian lymphoma was hypothesized but did not meet the aforementioned criteria, and the patient was lost to follow-up.[5] McKelvey et al.[6] reported a 52-year-old woman presenting with abnormal uterine bleeding and an ovarian mass found on transvaginal ultrasound. She underwent a total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy, and peritoneal washings.[6] The finding of DLBCL in a mature cystic teratoma on pathological examination prompted a computed tomography and ultrasound of the chest and abdomen and a bone marrow aspiration, all negative.[6] The patient was subsequently treated with six cycles of cladribine, mitoxantrone, dexamethasone chemotherapy.[6] A repeat bone marrow aspiration and CT scan showed no evidence of disease at 8 months.[6] Ghandi et al.[7] reported a 36-year-old woman with a cystic ovarian mass found on follow-up ultrasound for uterine fibroids. After a total abdominal hysterectomy and bilateral salpingo-oophorectomy, DLBCL was found

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