Papillary Thyroid Microcarcinoma Arising Within a Mature Ovarian Teratoma: Case Report and Review of the Literature

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ABSTRACT: Mature cystic teratoma is the most common kind of ovarian germ cell tumor. Malignant transformation is uncommon, with thyroid cancer rarely found. Papillary thyroid microcarcinoma has rarely been described as associated with ovarian teratomas. We report a case of a 34-year-old woman who presented with abdominal pain and an ovarian mass. After surgery, the patient was diagnosed with a follicular variant papillary thyroid microcarcinoma that arose within a mature cystic ovarian teratoma. Based on the small size of the primary lesion and patient preferences, no further treatment was performed. To our knowledge, this is the third reported case of papillary thyroid microcarcinoma arising within a mature ovarian teratoma without struma ovarii. There is no consensus on the surgical approach and postoperative management of this condition. Whether further therapy with total thyroidectomy and radioiodine ablation may be beneficial is unknown. In conclusion, papillary thyroid microcarcinoma can also arise within mature ovarian teratomas. Although a favorable prognosis is anticipated, there is limited information about its history or prognosis.

KEYWORDS: Mature ovarian teratoma, thyroid carcinoma, papillary thyroid microcarcinoma

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Introduction

Mature cystic teratoma is the most common kind of ovarian germ cell tumor, accounting for approximately 10% to 20% of tumors of this organ. Malignant transformation is uncommon, with an estimated risk of 0.17% to 2%.[1] When malignant transformation does occur, in 80% of cases, a squamous cell carcinoma is found.[2,3] Less common malignancies include sarcomas, adenocarcinomas, malignant melanomas, basal cell carcinomas, carcinoid tumors, and thyroid carcinomas. We present the case of a patient with a follicular variant of papillary thyroid microcarcinoma arising within a mature cystic ovarian teratoma (MCT).

Patient

A 34-year-old woman presented with abdominal pain and a left pelvic mass. Family history is positive for mother diagnosed with differentiated thyroid carcinoma. Ultrasonography revealed a right cystic ovarian mass measuring 99 mm × 72 mm. In addition, a left 127 mm × 77 mm mass was reported.

During laparotomy, a right ovarian cyst was excised. Furthermore, during left cystectomy, a rupture of the cyst occurred, and as a result, a left adnexectomy was performed. Histopathology study of the left ovary revealed an 80 mm × 55 mm × 50 mm MCT (Figure 1), as well as a 4-mm single follicular variant of papillary thyroid carcinoma (PTC) (Figure 2). Moreover, a 55 mm × 44 mm × 35 mm MCT in the right ovary was found. The resection margins were clear and there were no lymph nodes in the specimen.

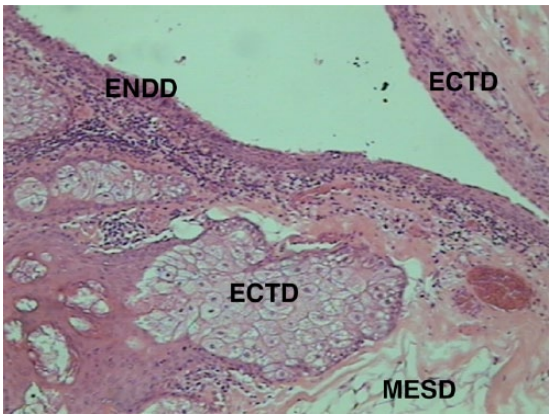

Figure 1. Mature cystic teratoma. ECTD: ectodermal derivates: squamous epithelium and adnexal structures. MESD: mesodermal derivatives: adipose tissue. ENDD: endodermal derivatives: respiratory epithelium (hematoxylin-eosin, original magnification ×200).

The thyroid function tests were normal. A thyroid ultrasound showed a 4 mm × 4 mm × 5 mm right hypoechoic nodule, with irregular margins, microcalcifications, and peripheral vascularity. In addition, a 4 mm × 2 mm × 4 mm isthmic hypoechoic nodule with peripheral vascularity was encountered. Fine-needle aspiration (FNA) of both nodules yielded insufficient material. Repeat FNA revealed benign thyroid nodules,

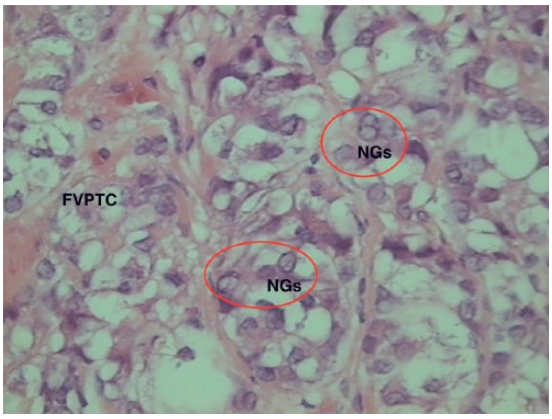

Figure 2. High-power magnification of follicular variant papillary thyroid microcarcinoma. The nuclei are clear, large, and oval, and there are intranuclear inclusion and NGs (hematoxylin-eosin, original magnification ×200). FVPTC indicates follicular variant papillary thyroid carcinoma; NGs, nuclear grooves.

classifying it as Bethesda category II. Based on the small size of the primary lesion and patient preferences, no further treatment was performed. An abdominal computed tomographic scan, a thyroid ultrasound, and a ^{131}I diagnostic whole body scan were planned, but the patient was lost to follow-up.

Discussion

We report a case of incidental papillary thyroid microcarcinoma (PMC) follicular variant within a MCT. Although histologic characteristics of thyroid carcinoma are found in approximately 5% to 37% of struma ovarii–type tumors,[4,5] they rarely occur in mature teratomas. Differentiated thyroid carcinoma arising from an MCT is exceptional, with an estimated incidence of 0.1% to 0.2%.[6] These are typically found incidentally in histopathology.

Dane et al[7] reviewed 15 cases of differentiated thyroid carcinoma arising in a mature ovarian teratoma. Since then, 4 additional cases have been reported.[8–11] Most patients, as with our case, presented with abdominal pain. Only 2 patients did not report any symptoms. Papillary thyroid carcinoma was the most frequent histopathologic type (53%), followed by follicular variant of PTC (42%) and follicular carcinoma (5%).

Only 2 cases presented with thyroid tumor size ⩽1 cm. Ryder et al[12] reported a 0.9-cm follicular variant PTC within a 4.6-cm MCT. Thyroid ultrasound was normal, as was a ^{131}I diagnostic whole body scan. No further treatment was performed in this patient. In addition, Dias et al[11] reported on 2 foci of follicular variant PTC (the largest of 3 mm) within a 4.5-cm mature ovarian teratoma. Thyroid ultrasound was also normal and no additional treatment was done.

Optimal treatment of thyroid carcinoma arising within MCT is unclear due to the rarity of the disease. Moreover, no data on recurrence are available. In some of the reported cases, thyroidectomy was performed. Some authors[7,9] support thyroidectomy as it allows for thyroglobulin monitoring as well as radioiodine treatment if needed. Moreover, it enables differentiation of thyroid carcinoma with ovarian metastases from thyroid carcinoma within MCT. However, in some cases, no thyroidectomy was performed.[8,13–15] In those cases, no primary thyroid carcinoma was clinically apparent in further follow-up.

Furthermore, there is a monodermal variant of ovarian teratoma known as struma ovarii that is predominantly composed of mature thyroid tissue. This variant is diagnosed when thyroid tissue constitutes 50% or more of the overall tissue. Struma ovarii accounts for 5% of ovarian teratomas.[4,5] Malignant struma ovarii is diagnosed when the histopathologic criteria of thyroid carcinoma are found.

Differentiated thyroid carcinomas appearing in struma ovarii can rarely present with a locally invasive disease and even with metastases.[16] Metastases can occur in approximately 20% of the patients.[6] Moreover, ovarian metastases from a primary thyroid carcinoma may occasionally occur as well. In such cases, the ovarian mass does not present with teratomatous characteristics.[17]

Multiple molecular abnormalities have been reported in thyroid cancer arising from ovarian teratomas, primarily in malignant struma ovarii. These include point mutations in BRAF (V600E and K601E),[18–20] ret/PTC rearrangements,[21] and point mutations in HRAS[22] and NRAS.[23] In thyroid carcinomas arising within MCT without struma ovarii, no molecular markers have been reported. Molecular genetics may help to differentiate benign from malignant lesions.[20] However, it is uncertain if they have a significant impact on cancer prognosis in this type of tumors.

After surgical resection of malignant struma ovarii, subsequent therapy depends on the extent of the primary lesion and disease stratification. There is no consensus on the optimal treatment of malignant struma ovarii. Treatment recommendations are based on single case reports or case series. Further therapy may include total thyroidectomy and radioiodine ablation. This allows for thyroglobulin monitoring, as well as radioiodine treatment if needed. In a review of 68 patients with malignant struma ovarii, 9% of patients had documented primary thyroid carcinomas, and most of them were invasive.[24] However, this study reported an excellent survival rate regardless of the management approach, with only 1 disease-specific death.

Papillary thyroid microcarcinomas have been infrequently reported as arising within struma ovarii.[25–27] In these cases, after surgical resection of ovarian tumor no further therapy involving the thyroid was completed.

Papillary thyroid microcarcinoma arising from and confined to the thyroid has an almost 100% survival rate at 30 years, with current guidelines favoring partial thyroidectomy without further radioiodine ablation in the absence of other adverse features.[28] It may be that papillary thyroid microcarcinoma is a nonprogressive subclinical disease with no impact on morbidity and mortality.[29]

To our knowledge, this is the third reported case of PMC arising within a mature ovarian teratoma without struma ovarii. There is scant information on the natural history or prognosis of PMC arising within ovarian tumors. Consequently, there is no consensus on the surgical approach and postoperative management of this entity.

Pathologists should be aware of this entity and exclude it in the workup of ovarian teratomas. It is important to have a multidisciplinary team including an endocrinologist and gynecologist to manage thyroid cancer arising within ovarian teratomas. As this is a rare entity, treatment should be individualized. We believe that a long-term follow-up of these cases is needed to know more about the prognosis of this uncommon disease.

In conclusion, PMC can also arise within mature ovarian teratomas. Although a favorable prognosis is anticipated, there is limited information about the history or prognosis of PMC within the ovary. The report of additional cases is important to understand the cause, pathogenesis, and natural history of this rare disease as well as the optimal treatment for this condition.

Author Contribution

MMP wrote the first draft of the manuscript. JP, PS, and KdlS contributed to the writing of the manuscript. JP, PS, SdlP, and BC made contributions to the acquisition of the clinical data. MMP, JP, PS, KdlS, SdlP, BC, and RP agreed with the manuscript results and conclusions. MMP, JP, and PS jointly developed the structure and arguments of the paper. MMP and RP made critical revisions and approved the final version of the manuscript. All authors reviewed and approved the final manuscript.

Disclosures and Ethics

As a requirement of publication, author(s) have provided to the publisher signed confirmation of compliance with legal and ethical obligations including but not limited to the following: authorship and contributorship, conflicts of interest, privacy and confidentiality, and (where applicable) protection of human and animal research subjects. The authors have read and confirmed their agreement with the ICMJE authorship and conflict of interest criteria. The authors have also confirmed that this article is unique and not under consideration or published in any other publication and that they have permission from rights holders to reproduce any copyrighted material. Any disclosures are made in this section.

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