CASE REPORT

Oncocytic and tall columnar type papillary thyroid carcinoma arising on a mature cystic teratoma: A case report and literature review

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Abstract

Mature cystic teratoma (MCT) is the most common germ cell ovarian tumour, which accounts for 15-20% of all ovarian neoplasms. The frequency of MCT cases undergoing malignant transformation ranges from 0.17% to 2%. Our aim for presenting this case is to contribute to formation of an algorithm in the literature for the treatment and follow-up of MCT undergoing malignant transformation. A 38-year-old female patient presented to the emergency service with acute abdomen. The patient underwent salpingo-oophorectomy due to a prediagnosis of ovarian torsion with a dermoid cyst. Postoperative pathological examination reported oncocytic and tall columnar type papillary thyroid carcinoma arising on a mature cystic teratoma. During the follow up no local recurrences or metastases were identified in one-year. The rarity of MCT cases undergoing papillary type thyroid carcinoma transformation hinders the establishment of an algorithm for treatment and follow-up in literature.

Keywords: Mature cystic teratomas, Papillary thyroid carcinoma.

Introduction

Mature cystic teratoma (MCT), also called dermoid cysts, is the most common germ cell ovarian tumour, which accounts for 15-20% of all ovarian neoplasms.[1] Although MCT is generally benign, foci of malignant transformation are reported in literature ranging from 0.17% to 2% as determined by histopathological examination.[2] Malignant transformation is common in postmenopausal women, however, it is rare in premenopausal period. Squamous cell carcinoma is the most common histopathological type (80%) of MCT undergoing malignant transformation.[3] Of MCT cases undergoing malignant transformation, papillary thyroid carcinoma is the rarest type[2] with ranges varying from 0.1% and 0.2%, and it is usually diagnosed postoperatively.[4] The majority of patients with MCT present with signs of pelvic mass and/or torsion, which is the most common complication of these neoplasms.[5]

In this case report, we present a very rare case in literature, a case of postoperatively diagnosed oncocytic and tall columnar type papillary thyroid carcinoma arising on a mature cystic teratoma without any natural thyroid tissue. The study was approved by the Institutional Ethics Committee and the consent of the patient was taken prior to the writing of the manuscript and for publishing the case.

Case Presentation

A 38-year-old patient who had inguinal and lumbar pain for approximately 1 year, presented to the Emergency Service of Izmir Atatürk Training and Research Hospital in April 2016 with the complaints of a sharp pain in lower abdomen, with accompanying nausea, and vomiting. Adjacent to the left side of the umbilicus, a painful mass and signs of acute abdomen were detected by abdominal examination during Obstetrics and Gynecology consultation. Cervix was hyperaemic and hypertrophic during speculum examination. No diagnostic features were identified in the cervico-vaginal smear specimen taken for cytological inspection and for detection of HPV. There was pain on the left adnexal lodge and on moving the cervix during the manual

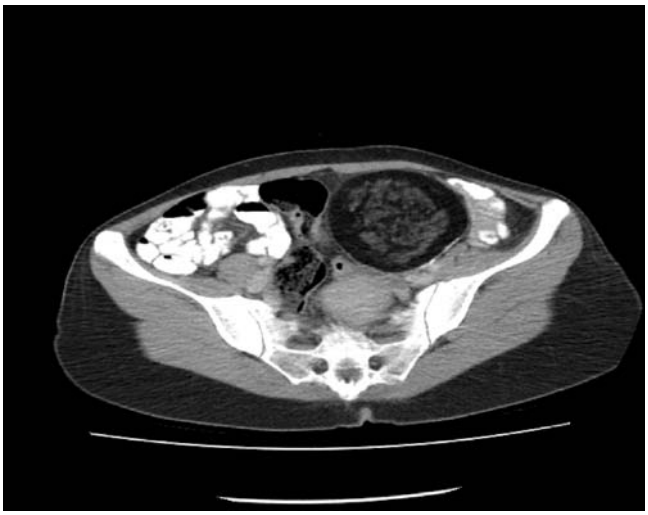

Figure-1: Cross section of the mass obtained by the lower abdominal CT.

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examination of the vagina.

The laboratory examination results were negative for β-hCG. Haemogram and biochemistry test results were normal except a leukocytosis of 11.87 K/μL. Doppler Ultrasonography of the pelvis identified a 10x8.5x6.5 cm sized, uniformly contoured space occupying lesion in the left adnexal lodge with cystic components, and with a heterogeneous internal structure, demonstrating images compatible with hyperechogenicity and calcification. These findings were reported to have a potential to be compatible with torsion of a teratodermoid tumour. A computerized tomography (CT) imaging revealed "a typical mature cystic teratoma of 10 cm in diameter" (Figure-1).

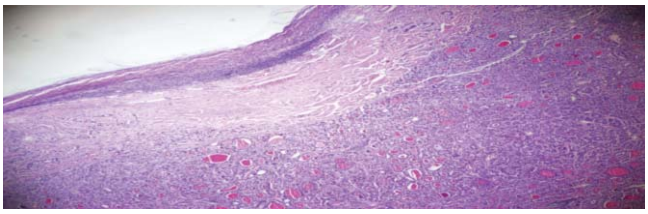

Figure-2: Ovary tissue and papillary thyroid carcinoma tissue.

Upon obtaining the patient's consent for salpingo-oophorectomy, the abdomen was exposed by a Pfannenstiel incision under emergency conditions. A cystic mass of approximately 11x7 cm size, with a smooth surface, originating from the left ovary and extending to the umbilicus was observed to be torsioned twice around itself. No adhesions were observed between the surface of the cyst and other intraabdominal tissues. A left salpingo-oophorectomy was performed because no natural ovarian tissue was identified.

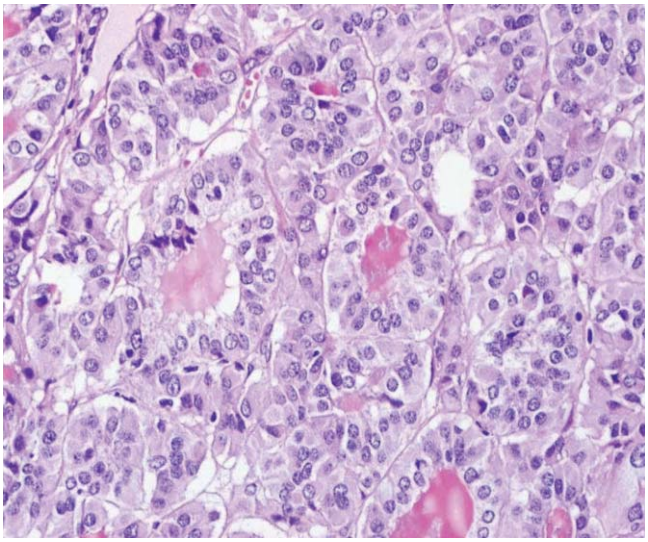

Figure-3: Oncocytic variant.

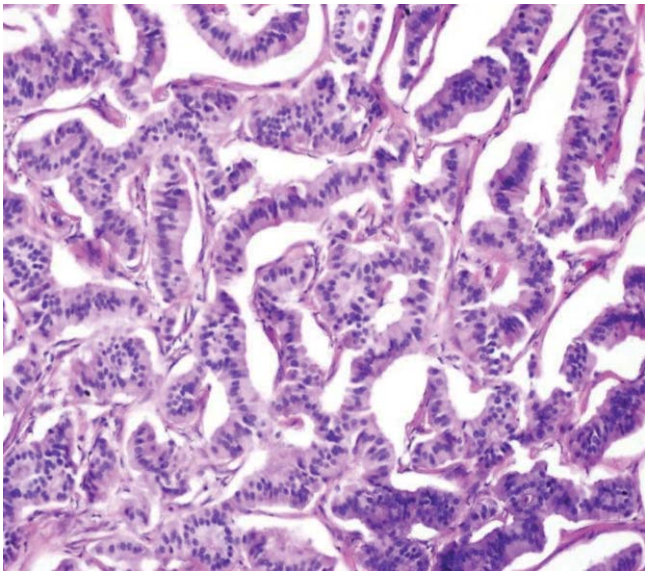

Figure-4: Tall cell variant.

The pathological examination reported a cystic mass with a smooth surface and with a diameter of 11cm, containing a 5.5x0.6 cm sized tube. On its cross section, some material, featuring like hair and sebum was drained and some solid structure of 2.7 cm size was observed on its wall. Microscopic examination revealed thyroid tissue, which was solid and papillary (Figure-2). It was well defined at some areas, contained colloid and demonstrated a trabecular growth pattern. Some of the cells had abundant eosinophilic cytoplasm and transparent nuclei, and were observed to have oncocytic features. Lipogranulomatous reaction was also noted in specimens taken from the cyst wall. Immunohistochemical (IHC) examination revealed negative for calcitonin, cyclin D1, HMBE1, Galectin 3 and m CEA; and positive for thyroglobulin, TTF1, synaptophysin, and cytokeratin 19. KI67 proliferation index was assessed as 1%.

With these findings, diagnosis of a "mature cystic teratoma with malign transformation" which contains two different morphologic types (oncocytic and tall columnar) of thyroid papillary carcinoma was made (Figure-3,4).

Thyroid hormone plasma levels and thyroglobulin level of the patient were normal, however anti-thyroglobulin level was > 500 U / mL (0-60 U/mL). A normal parenchymal vasculature was identified by colour Doppler ultrasonography of the thyroid gland, whereas a hypoechoic solid nodule of 17x8 mm size was detected in

the medial right lower zone. The fine needle aspiration biopsy of the nodule was compatible with chronic lymphocytic thyroiditis. The patient was decided to be followed up without any diagnostic features reported by abdominal and thoracic CT examinations.

Discussion

Although MCT is generally benign, malignant transformation is reported in the literature at rates ranging from 0.17% to 2%.[3] The frequency of the malignant transformation component to be identified as a differentiated thyroid carcinoma varies from 0.1% to 0.2%.[5] This differentiated thyroid carcinoma tissue is histopathologically examined in 3 classes with the most common type (44%) being the papillary carcinoma. Other types are follicular (30%) and follicular variant of papillary carcinoma (26%).

There is no consensus in literature on the treatment and follow-up of MCT cases undergoing malignant transformation.[5-7] However, total abdominal hysterectomy together with opposite salpingo-oophorectomy is considered to be the most appropriate surgical option for postmenopausal women or for women, who have no intention for further childbirth if the specimens of these women reveal MCT by frozen section examination. In fertility-sparing surgery, unilateral oophorectomy is performed if there is no capsular invasion or metastasis. The presented 38-year-old case had one living child and had intention for further fertility. Because of this, and because the capsule was intact and there were no signs in favour of invasion preoperatively, the unilateral salpingo-oophorectomy under emergency conditions for the prediagnosis of dermoid cyst and torsion was found sufficient and either complementary or staging surgery was not deemed necessary.

In order to determine metastatic or recurrent disease, in MCT cases undergoing malignant transformation, follow-up of thyroglobulin (Tg) levels is recommended.[8] The only source of circulating Tg is the thyroid tissue and ovarian teratomas containing thyroid tissue, which is a very rare condition.[9] However, the potential of Tg levels being high in benign thyroid diseases hampers determination of it as a convenient tumour marker in MCT, who did not undergo thyroidectomy and who contain thyroid tissue with malignant transformation. On the other hand, the high levels of antithyroglobulin antibody (anti-TgAb) may cause Tg levels to be erroneously low. For this reason, the follow-up of Tg levels is favourable for patients, who underwent thyroidectomy only and for patients left with no or very little thyroid tissue. In order to evaluate Tg levels correctly, follow-up of Tg levels together with anti-TgAb levels is advisable as persisting high levels of anti-TgAb indicate a persistent disease. In this presented case, Tg measurements performed by the endocrinology outpatient clinic at 6 month intervals were 0.261 and 0.606 ng/mL (1.6 - 59.9 ng/mL), and the anti-TgAb test results corresponding to these two consecutive measurements were > 500 U / mL and 434 U / mL (0-60 U / mL), respectively.

In the literature, follow-up of suspected thyroid nodules of patients with malignant thyroid carcinoma (MCT)16 is controversial. Management of these cases varies from follow-up by conventional ultrasonography together with Tg levels, to total thyroidectomy with or without radioactive iodine ablation.[10]

In this case, the absence of natural thyroid tissue arising on MCT was commented in favour of a metastatic lesion originating from thyroid gland. Therefore, doppler ultrasonography of the thyroid gland was performed. Fine needle aspiration biopsy of the detected thyroid nodule was conducted, resulting in the diagnosis of lymphocytic thyroiditis. Total thyroidectomy is recommended in literature since it enables follow-up of cases with persistent high anti-TgAb and Tg levels, however, it has been reported that MCT cases with papillary thyroid carcinoma transformations are followed-up uneventfully without total thyroidectomy.[11] Our patient was followed-up for malignancy without thyroidectomy, and no malignancy was detected by the fine needle aspiration biopsy performed at the end of the one year. She is still being followed-up by the endocrinology and gynecology outpatient clinic at 6 months intervals. A case report of a dermoid cyst undergoing papillary thyroid carcinoma transformation with follicular variant, accompanied with a literature review of 15 cases by Dane C. et al.;[12] considered one-sided oophorectomy to be a sufficient option in cases when there is no evidence of invasion. In this presented case, the intention for fertility, the absence of local invasion as determined by the pathological examination, and the smooth capsule of the cyst lead to the conclusion that the unilateral salpingo-oophorectomy performed under emergency conditions due to torsion was a sufficient intervention. The follow-up of the case performed at the gynaecology and endocrinology clinic at 3-month intervals was found adequate. No pathological features were detected except lymphocytic thyroiditis by anti-TgAb levels and by fine needle aspiration biopsy as evaluated by the endocrinology clinic and by

gynaecological examination and pelvic ultrasonography revealing normal features.

Conclusion

We described a case of Oncocytic and Tall Columnar Type Papillary Thyroid Carcinoma Arising On a Mature Cystic Teratoma. The rarity of this casesinders the establishment of an algorithm for treatment and follow-up. Therefore, decision making should be multidisciplinary during treatment and follow-up. For our case no metastasis, residual disease or recurrence was detected; so that unilateral salpingo-oophorectomy was a sufficient intervention.

Disclaimer: None to declare.

Conflict of Interest: None to declare.

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