Papillary Thyroid Carcinoma Arising within Mature Ovarian Teratoma: A Case Report

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Abstract

Mature cystic teratoma is the commonest ovarian germ cell tumor. Though malignant transformation is uncommon, papillary thyroid carcinoma has rarely been described as associated with ovarian teratomas. We report a case of a 34-years old multiparous woman who presented with acute abdominal pain and an ovarian mass. After salphingo-oophorectomy, the patient was diagnosed as papillary thyroid carcinoma that arose within a mature cystic ovarian teratoma. To our knowledge, this is the first reported case of papillary thyroid carcinoma arising within a mature ovarian teratoma in this tertiary health care center in Chattogram. We recommend long term follow up to see any metastatic possibility.

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Keywords: Mature cystic teratoma, papillary thyroid carcinoma, struma ovarii.

Introduction

Among the ovarian germ cell tumors Mature Cystic Teratoma (MCT) is the most common and comprises 10–20% of all ovarian tumors. However, malignant transformation of MCT is not common and the incidence is 1–3%.[1] Squamous Cell Carcinoma (SCC) is the commonest type, found in 80% of cases.[2] Papillary thyroid carcinoma (PTC) within teratoma is one of the rarest types with ranges varying from 0.1% and 0.2%, and usually diagnosed postoperatively.[3] The synchronous development of malignant struma ovarii and primary thyroid carcinoma is extremely rare, though a handful of cases were reported.[4] So, presence of thyroid tissue in teratoma, should proceed with further work up to confirm the diagnosis and to explore the possibility of a malignant lesion in the mass- either primary or metastasis.[5] Here we present a case of a patient with a PTC arising within a ovarian MCT.

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Case Presentation

A 34-year-old multiparous woman who had abdominal pain, distention and irregular menstrual bleeding for approximately for 6 months, presented to the emergency service of Chittagong Medical College Hospital in May 2019 with the complaints of a sharp pain in lower abdomen, with accompanying vomitting. There was no previous medical or surgical history.

On abdominal examination, a tender mass adjacent to the left side of the umbilicus were detected. Manual examination of the vagina revealed tenderness and mass in left adnexal region. Paps smear was done with no abnormality detected. Haemogram and biochemical test results were normal except a hemoglobin level of 9.2 gm/dl. CA-125 was within normal limit and β-HCG was normal. Ultrasonography of the lower abdomen revealed a complex solid cystic mass measuring 82x62x55 mm, with mixed echotexture, compatible with dermoid cyst.

At laparotomy, a cystic mass of approximately 9x7cm size, with a white, smooth glistening surface, originating from the left ovary was observed. Opposite ovary was apparently normal and no adhesion or intra-abdominal deposit was observed. Left sided salphingo-oophorectomy was performed preserving the uterus and right ovary and sent to Department of Pathology, Chittagong Medical College, Chattogram for histopathological evaluation. On gross pathological examination, a cystic mass of 9 cm in diameter (fig-1) with 3cm fallopian tube were noted. On cross section, hair, sebum & fatty materials were come out and some thick greenish fluid was drained and some solid structure was observed on its wall (fig-2).

Microscopic examination revealed mature teratomatous component represented by skin with associated adnexal structures, muscles, fat, benign glands lined by mucin containing columnar epithelium (fig-3,4) and thyroid tissue, within the thyroid tissue foci papillary thyroid carcinoma (fig-5,6) was found. Lining cells had oval nuclei showing nuclear overlapping, grooving and intranuclear cytoplasmic inclusions. Follicles also contained amorphous eosinophilic thick colloid. The fallopian tube was unremarkable. Immunohistochemical (IHC) examination revealed positivity for TTF-1 (fig-8,9). With these findings, diagnosis of a "mature cystic teratoma with malignant transformation to papillary thyroid carcinoma" was made. Postoperatively plasma levels of T_3, T_4, TSH and thyroglobulin of the patient were normal. A normal parenchymal vasculature was identified by postoperative ultrasonography of the thyroid gland.

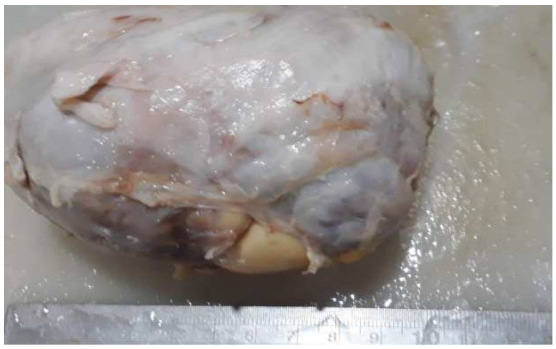

Figure 1. Gross appearance of teratoma

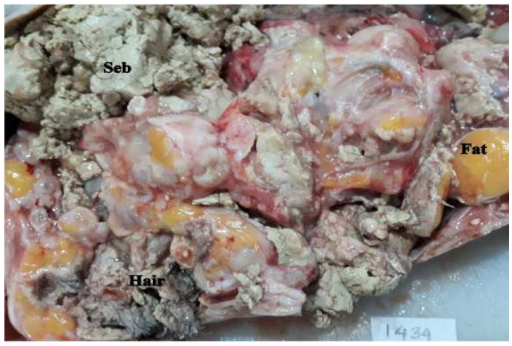

Figure 2. Cut opened teratoma (Sebum, hair, fat)

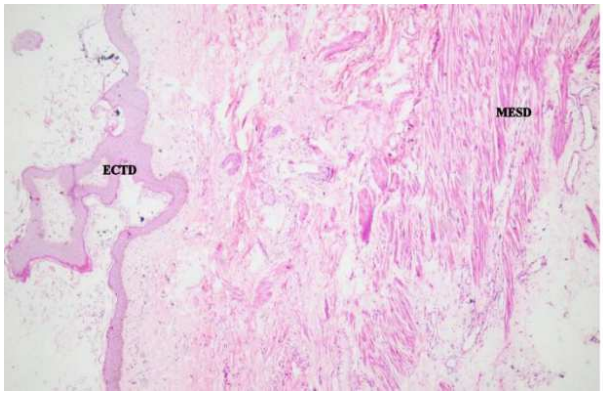

Figure 3. Ectodermal & mesodermal derivatives (H&E stain, 40X)

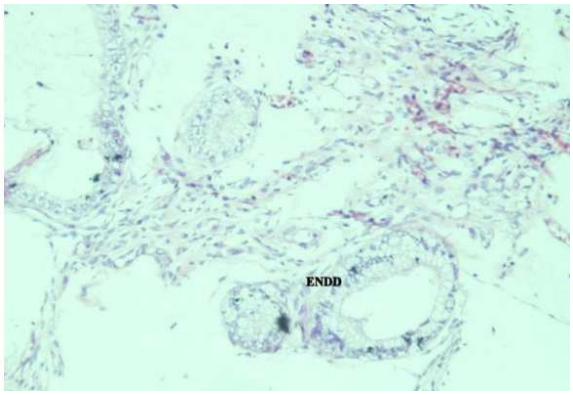

Figure 4. Endodermal derivatives (H &E stain, 400X)

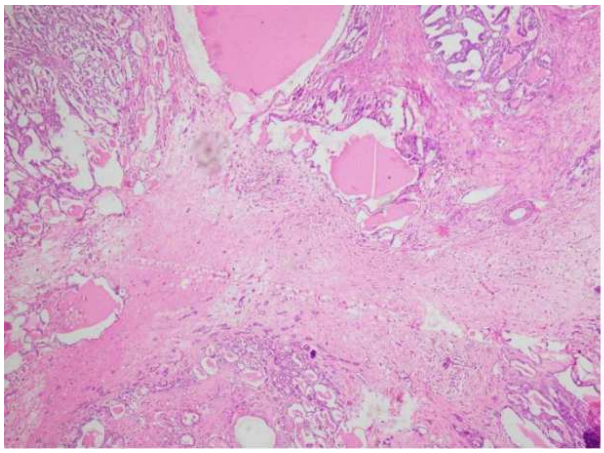

Figure 5. Teratoma showing thyroid tissue (H&E, 40X)

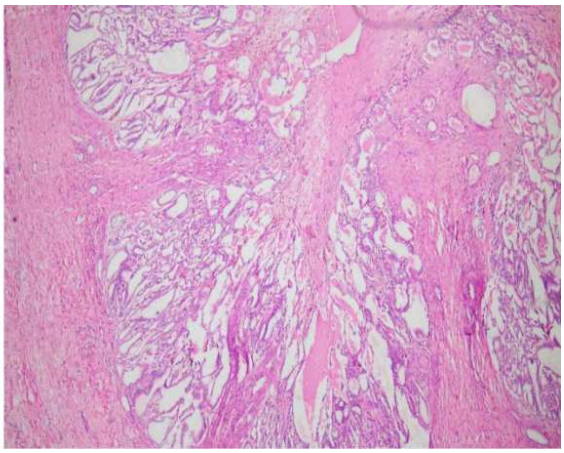

Figure 6. Thyroid tissue having papilla (H&E, 100X)

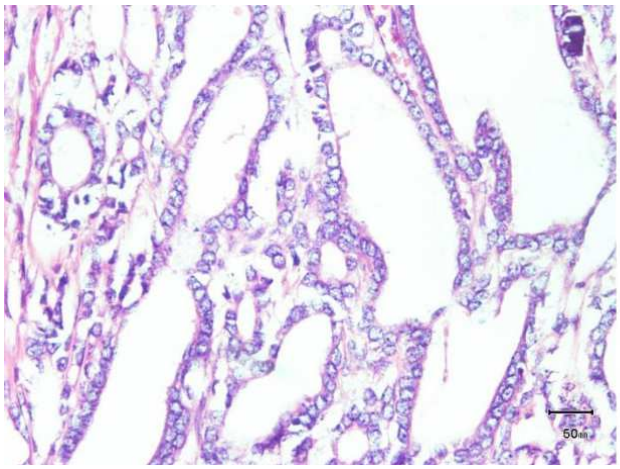

Figure 7. Orphan Anie eye nuclei (H & E, 400X)

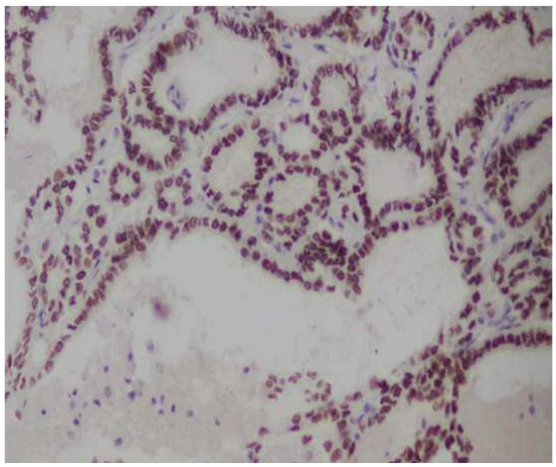

Figure 8. Thyroid tissue (TTF-I immunoreactivity (100X)

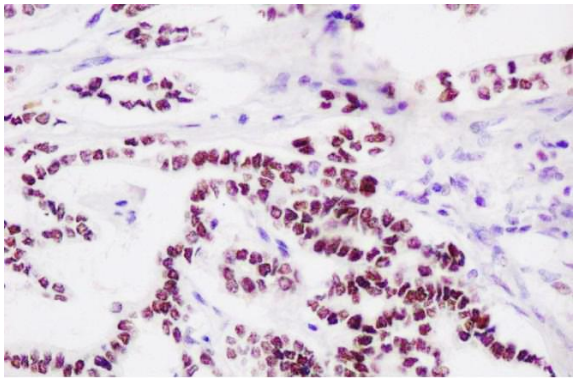

Figure 9.TTF-I immunoreactivity (200X)

Discussion

Mature cystic teratomas are also known as dermoid cysts, because they are mostly cystic, skin & skin appendages are the most common structures. Thyroid tissue is present in 10% of the all cases.[6] Teratomas containing more than 50% of thyroid tissues are called struma ovarii, often presented as monodermal teratoma.[7] Malignant transformation of MCT is rare; however, several types of malignancy can develop from any one of three germ-cell layers. Squamous cell carcinoma, derived from ectoderm is the commonest type; less common malignancies include soft tissue sarcomas, adenocarcinomas, malignant melanomas, basal cell carcinomas, carcinoid tumors, and thyroid carcinomas.[2] Among thyroid carcinomas the most common histological type is the papillary carcinoma (44%), other types are follicular carcinoma (30%) and follicular variant of papillary carcinoma (26%).[3]

The malignant change of an initially benign cystic teratoma is detected in patients between 40 and 60 years of age, older than its benign counterpart. Although the cancer occurs at any age, most patients are postmenopausal.[1] The tumor may present as pelvic discomfort, with a pelvic mass on abdominal imaging (USG, CT, MRI) or during laparotomy for any other reason. Preoperative definitive diagnosis of stroma ovarii or papillary thyroid carcinoma is not possible. The only possibility of preoperative diagnosis is by radioactive iodine scan (not done routinely).[8] Various case reports have been published over the past few years regarding the histological diagnoses and treatment options. The diagnoses of thyroid carcinomas arising in teratomas should be made following the guidelines for diagnosing carcinomas in thyroid gland. Disease is treatable with good out come in most cases. Only 7% and 14% of patients with papillary carcinoma and typical follicular carcinoma, respectively died of disease. Due to rarity of disease no consensus on treatment has been made, however treatment options include oophorectomy, additional thyroidectomy, radioactive iodine and long term follow up with serum thyroglobulin measurement.[9]

In order to determine metastatic disease, in MCT cases undergoing malignant transformation, follow up of thyroglobulin (Tg) levels is recommended. The only source of circulating Tg is the thyroid tissue and ovarian teratomas containing thyroid tissue, which is a very rare condition. However, high Tg level in benign thyroid diseases hamper 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 anti-thyroglobulin antibody (anti-Tg Ab) 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-Tg Ab levels is advisable as persisting high levels of anti-Tg Ab indicate a persistent disease.[3] In our case, Plasma T_3, T_4, TSH and Tg level were normal and normal parenchymal vasculature was identified by

ultrasonography of the thyroid gland. Anti-Tg Ab level can't be performed due to patient's refusal. Logani *et al.,* 2001 was commented the absence of normal thyroid tissue and features of teratoma, in favour of a metastatic lesion originating from thyroid gland.[10] In the presented case, histologic evidence of mature teratoma, normal thyroid tissue along foci of papillary thyroid carcinoma, and positive immunohistochemical stain for thyroid transcription factor-1(TTF-1) indicates primary thyroid carcinoma arising within MCT.

Conclusion

Whether further therapy with total thyroidectomy and radioiodine ablation may be beneficial is unknown. The rarity of MCT cases undergoing PTC transformation impedes the establishment of a protocol for treatment and follow-up. We recommend that a long-term follow-up of these cases is needed to know more about the prognosis and to see any local recurrence or metastasis.

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