Int J Clin Exp Pathol 2016;9(8):8741-8744
www.ijcep.com /ISSN:1936-2625/IJCEP0022761

Case Report

Mature cystic ovarian teratoma with thyroid papillary carcinoma and strumal carcinoid: a case report and review of literature

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Received December 26, 2015; Accepted March 8, 2016; Epub August 1, 2016; Published August 15, 2016

Abstract: Mature cystic teratoma of the ovary is a frequent event, but thyroid papillary carcinoma and strumal carcinoid appear in mature cystic ovarian teratoma at the same time is extremely rare. The present case is the first patient of mature cystic ovarian teratoma with thyroid papillary carcinoma and strumal carcinoid reported from China in English. A review of the literature is performed, and the clinical and pathological characteristics of the patients about mature cystic ovarian teratoma with elements of both thyroid papillary carcinoma and strumal carcinoid of the reported cases before are analyzed.

Keywords: Mature cystic ovarian teratoma, thyroid papillary carcinoma, strumal carcinoid

Introduction

Mature cystic teratoma (MCT) is not a rare occurrence, accounting for about 20% of ovarian tumors, but malignant transformation of MCT is infrequent [1]. Many components of the MCT can undergo malignant transformation, and the most frequent tumor is squamous cell carcinoma [2], and less common malignancies include thyroid papillary carcinomas, adenocarcinomas, carcinoid tumors and so on [2, 3]. However, mature cystic ovarian teratoma contains components of thyroid papillary carcinoma and strumal carcinoid is extremely rare. Here we report a case and review literature.

Case report

A 43-year-old woman underwent an abdominal pain for one year and presented with a mass in left side of her ovary. The enhanced computed tomography (CT) scan noted strong enhancement and magnetic resonance imaging of the mass was performed, which revealed the mass measuring 11.6 × 7.7 cm (**Figure 1**). The patient underwent surgical resection in our hospital and the tumor was completely resected.

Gross examination

Pathologic gross examination revealed a 9*7*5 cm mass. On cut surface, the mass was cystic, filled with solid tan-white to tan-yellow tissue, grease and hair was also identified. No normal appearing ovarian parenchyma was seen grossly (**Figure 2**).

Histology and immunohistochemistry

Mature cystic teratoma with struma ovarii component arising from the left ovary with multiple malignant foci. The mature cystic teratoma components consist of thyroid tissues (**Figure 3A**), strumal carcinoid and papillary thyroid carcinoma. The well differentiated strumal carcinoid tumor (**Figure 3B**) was confirmed positive for synaptophysin (**Figure 3C**), and well differentiated papillary thyroid carcinoma (**Figure 3D**) arising in struma ovarii was tested positive for Thyroid Transcription Factor 1 (TTF-1) (**Figure 3E**), HBME-1 (**Figure 3F**), Galectin-3 and Cytokeratin 19 (CK19) by IHC. Additional findings included a benign mature cystic teratoma of the contralateral right ovary. All lymph nodes were negative for metastatic disease.

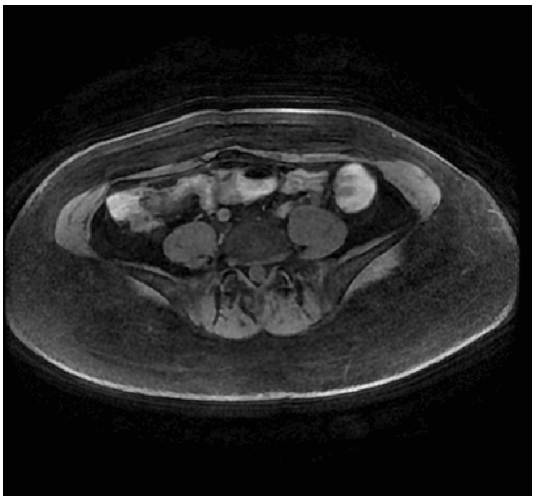

Figure 1. Magnetic resonance imaging of the mass was performed, which revealed the mass measuring 11.6 × 7.7 cm.

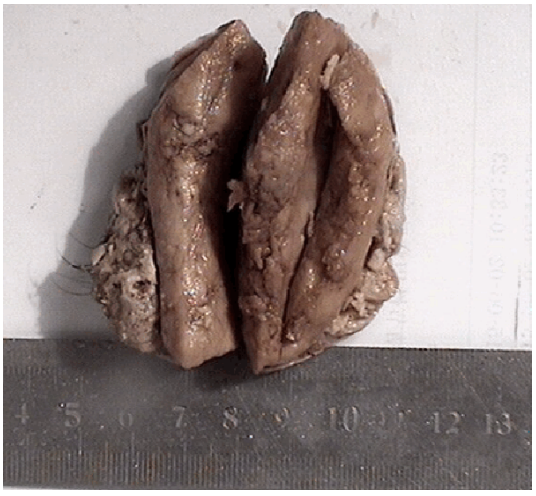

Figure 2. The mass was cystic on cut surface, filled with solid tan-yellow to tan-white tissue and a minimal amount of grease and hair.

Discussions

Mature cystic teratoma of ovarian is a frequent event. However, mature cystic ovarian teratoma with components of thyroid papillary carcinoma and strumal carcinoid at the same time is extremely rare. To the best of our knowledge, mature cystic ovarian teratoma with elements of both thyroid papillary carcinoma and strumal carcinoid has been documented in 2 prior reports [4, 5]. We summarize the prior reported cases and the clinicopathological features of these cases are detailed in **Table 1**. In our review, there are 3 cases in total including our case. All patients are adults, with the median age of 62. The symptoms occurred in 33.3% (1/3) of patients. After surgical resection, all 3 patients got good effects.

The computed tomography (CT) scan and ultrasound examination are helpful for the preoperative diagnosing but nonspecific. Therefore, pathological examination is usually necessary in order to reach a precise diagnosis and a definitive treatment.

The pathologic gross examination revealed a smooth outer surface and the cut surface was cystic, but filled with solid tan-yellow to tan-white tissue. A minimal amount of hair was also identified. Histologically, the tumor was characterized by the presence of thyroid papillary carcinoma and strumal carcinoid. Besides, our case showed thyroid papillary carcinoma and goiter coexist in the lesion, suggesting that the malignant transformation of a mature cystic teratoma into thyroid papillary carcinoma [4]. Immunohistochemical study is important for the diagnosis and differential diagnosis. Immunohistochemistry staining showed the component of thyroid papillary carcinoma was positive for HBME-1, Galectin-3 and CK19, and negative for CD56, and the strumal carcinoid was positive for Syn, CgA and CD56. These markers are useful for the differential diagnosis of MCT with thyroid papillary carcinoma and strumal carcinoid from other tumors such as sex cord-stromal tumors, malignant mesothelioma and ovarian serous papillary adenocarcinoma. Given the histopathologic and immunohistochemical nature, the present case was consistent with the diagnosis of MCT with thyroid papillary carcinoma and strumal carcinoid.

Likely due to the rarity and resulting inability to design management trials, there is no consensus on the treatment of MCT with thyroid papillary carcinoma and strumal carcinoid. Various case studies recommend different treatment options. Surgical resection is necessary for the treatment. Adjuvant treatment possibilities include radiation, traditional chemotherapy, thyroidectomy and ablation, and thyroxine suppression therapy. No recurrences were noted in the patients receiving adjuvant therapy after complete response to initial surgery [6, 7]. So we suggested that appropriate adjuvant therapy could be a good choice for this patient.

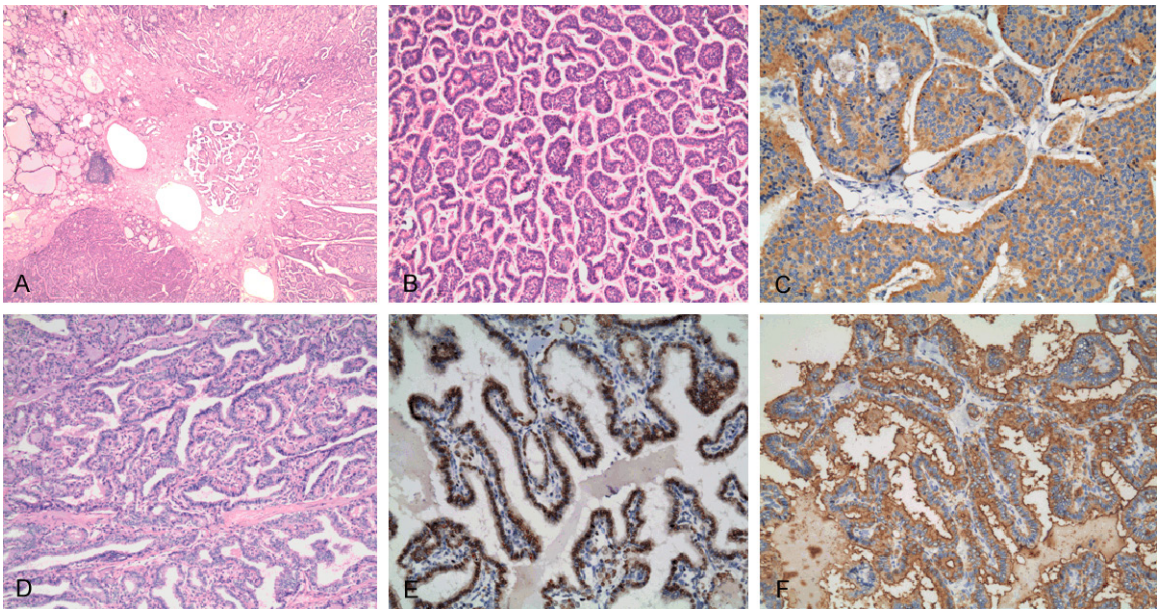

Figure 3. The mature cystic teratoma components consist of thyroid tissues, strumal carcinoid and papillary thyroid carcinoma (The central field of lesion) (A). The malignant components consist of well differentiated strumal carcinoid tumor (B) confirmed with positive immunohistochemistry (IHC) for synaptophysin (C) and papillary thyroid carcinoma (D) arising in struma ovarii tested positive for Thyroid Transcription Factor 1 (TTF-1) (E) and HBME-1 (F).

Table 1. Clinical and pathological features of MCT with thyroid papillary carcinoma and strumal carcinoid

Case no	Age	Presentation	Size	Treatment	Histology (components)
1	74	Asymptomatic	4 cm	Enucleation of the lesion	Thyroid carcinoma
					Strumal carcinoid
					Mucinous adenocarcinoma
2	69	Asymptomatic	21 cm	Enucleation of the lesion	Thyroid carcinoma
					Strumal carcinoid
3	43	bellyache	9 cm	Enucleation of the lesion	Thyroid carcinoma
					Strumal carcinoid

Some advocate a post-operative total-body scintiscan to assess for residual malignant disease and trending serum thyroglobulin levels to monitor for recurrence and adjuvant therapy only if these were positive, and this should be used with caution if not in the setting of thyroidectomy since most malignant struma ovarii have poor iodine uptake and thyroid hormone synthesis [8, 9].

In summary, we presented a case of mature cystic ovarian teratoma with thyroid papillary carcinoma and strumal carcinoid. To our best of knowledge, the occurrence of the tumor is exceedingly rare. The characteristic findings of imaging studies, the histological features and immunohistochemical staining are helpful for the diagnosis of MET with thyroid papillary carcinoma and strumal carcinoid. The patient got good effect after surgical resection and receiving adjuvant chemotherapy post-operation. For the time being, the follow-up is necessary.

Acknowledgements

We thank Dr. Ping Xie (Department of Pathology, Shanghai Pudong Hospital of Fudan University, Shanghai, China) for valuable assistance with English translation.

Disclosure of conflict of interest

None.

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