JCOMS eISSN:2783-3127 https://journalofcoms.com

Journal of

Current Oncology and Medical Sciences

Vol. 3, No.4

Case report Free Access

Papillary thyroid carcinoma arising from mature cystic teratoma ovary: a case report

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Abstract

Introduction: Mature cystic teratoma is a kind of ovarian germ cell tumour. Malignant transformation in it is uncommon with thyroid cancer being rarely found. Given its rarity and nonspecific symptoms, misdiagnosis and indifference when compared to other ovarian lesions is very common.

Case presentation: Herein we report a case of a 58-year-old post-menopausal female who presented with a history of abdominal distension and loss of appetite. She was found to have an abdominopelvic mass on examination and a raised CA125 levels for which she underwent an MRI pelvis which was suggestive of an O-RADS 5 lesion for which she underwent a staging laparotomy. The final histopathology and immunohistochemistry were suggestive of papillary thyroid carcinoma arising from a mature ovarian teratoma. After a multidisciplinary tumour board analysis, she was planned to be kept under follow–up with regular serum thyroglobulin monitoring. She has no signs of disease recurrence to date.

Discussion: Struma ovarii is one type of monodermal ovarian teratoma in which the tumour contains more than 50 % thyroid tissue. Diagnosis in such cases is difficult due to the lack of typical symptoms. In most of the cases, the diagnosis is incidental. Optimal treatment is still unclear given the rarity of the disease. In a few cases, thyroidectomy was done whereas in a few others it was omitted. Further therapy may include radioiodine treatment if needed..

Conclusion: To the best of our knowledge there is very scant information available on the natural history, prognosis and management of papillary thyroid carcinoma arising from mature cystic teratoma ovary. Hence, a multidisciplinary treatment approach may be needed for the same.

Keywords: Papillary thyroid carcinoma, Mature cystic teratoma, Germ cell tumour

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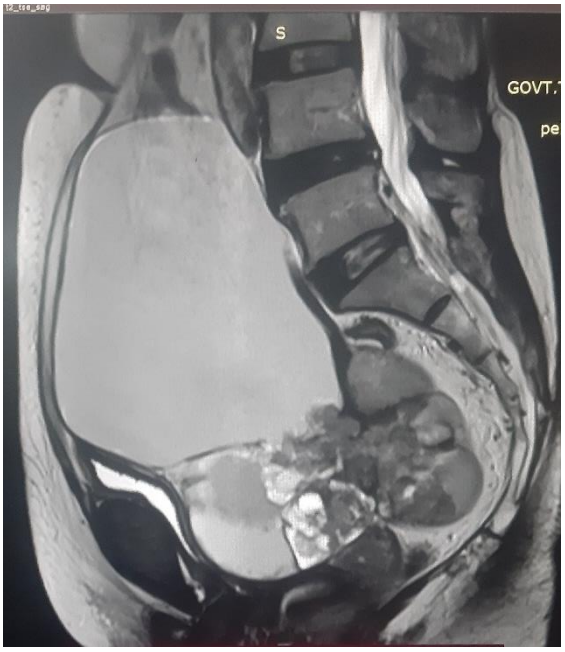
Receive: 2023.11.12, Accepted: 2023.9.10

Introduction

Mature cystic teratomas comprise 20% of all ovarian neoplasms and are considered to be the most common type of germ cell tumors of the ovary (1). They can be either unilateral or bilateral and commonly appear in reproductive age, but have also been reported in postmenopausal women and children (2). Malignant transformation is uncommon, with an estimated risk of 0.17% to 2% (3). When malignant transformation occurs, in most cases (80%) it is squamous cell carcinoma as histology (4). Less common ones include sarcomas, adenocarcinomas, malignant melanomas, basal cell carcinomas, carcinoid tumors, and thyroid carcinomas (5). Struma ovarii is a rare ovarian lesion that is characterized by the presence of thyroid tissue in at least half of the overall ovarian mass. This mass comprises less than 1 % of ovarian tumors and also upto 2 to 5 % of all ovarian teratomas. The patients usually are asymptomatic with pelvic mass and pain being the common presenting symptoms, making it usually diagnosed post-operatively based on histopathology (6). A small proportion of struma ovarii may undergo malignant transformation, with papillary carcinoma the most common type of malignancy seen. The criteria used to identify a malignant change in struma ovarii are identical to those used to evaluate the thyroid gland (7). Only 5–8 % of these patients usually have clinical hyperthyroidism (8). Owing to the rarity of the tumor, there are no specific clinical, radiological, or serum markers that distinguish struma ovarii in the absence of thyroid hormone abnormalities. Thus, a definitive diagnosis is made by histopathological examination (7). Herein we present a case of papillary thyroid carcinoma arising within a mature cystic ovarian teratoma in a 58 year old post menopausal female.

Case presentation

A 58 year old post menopausal female presented with a two months history of abdominal distension and loss of appetite. On examination she had a palpable mass per abdomen which was of 18 week size felt more towards the left side . An ultrasonography of the abdomen was done which revealed a large abdominopelvic multilocular cystic lesion which was likely of pelvic origin. An MRI pelvis followed which revealed a large abdominopelvic cystic lesion of 12.8 x11.7x 15.2 cm with multiple internal septation (Ovarian-Adnexal Reporting and Data System - O-RADS 5) with mild ascites and no evidence of pelvic lymphadenopathy (Figure1).



Figire 1. MRI pelvis showed an abdominopelvic cystic lesion.

Her serum tumour markers showed a raised CA – 125(Cancer Antigen 125) level (157.8 U/L). Serum CEA(Carcinoembryonic Antigen) and CA19.9 (Cancer Antigen 19.9) were within the normal limits. She then underwent a total abdominal hysterectomy with bilateral salpingoophorectomy with omental biopsy. Intraoperatively she was found to have a large cystic lesion of around 15x 15 cm replacing the whole of the left ovary with smooth surface and no papillary excruscences. The right ovary was adherent to the mass and uterus was found to be atrophic . The omentum was found to have nodularity and a biopsy was taken from it and ascitic fluid was sent for cytology. The pathology findings showed an ovarian mass of size 19x13x 8 cm with the bosselated surface which was solid and cystic with multiple rents on the surface. The microscopic examination showed papillary carcinoma with no immature component (Figure 2).

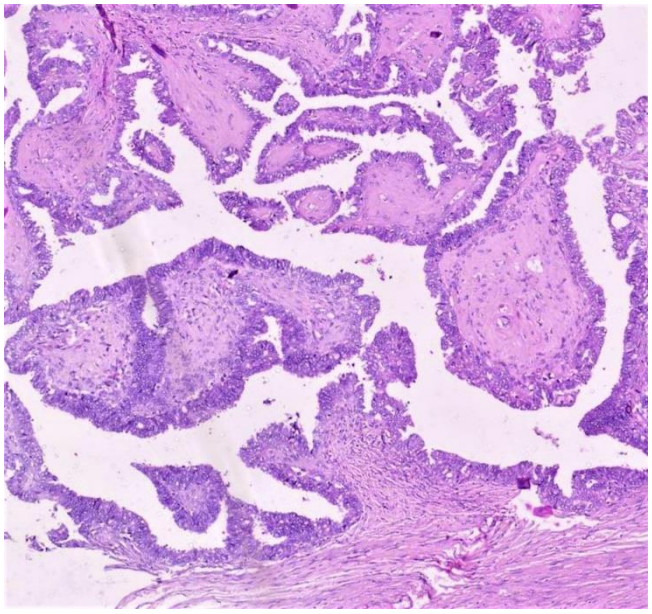


Figure 2. Histopathology image [40 X magnification] showing papillary thyroid carcinoma in a mature cystic ovarian teratoma.

Immunohistochemistry showed diffuse strong positivity for PAX8 (Paired Box 8), TTF 1(Transcription Termination Factor 1), thyroglobulin (Figure 3) and negativity for WT1 (Wilms' tumour gene 1) thus confirming the final diagnosis of papillary thyroid carcinoma in a mature cystic ovarian teratoma. The omental biopsy was suggestive of congestion only. Ascitic fluid cytology was doe which was found to be negative. Thyroid function tests were done which were found to be normal. Serum alpha-fetoprotein [AFP] was found to be 7.46 and beta HCG (human chorionic gonadotrophin) to be 5.55. Post-operative CA-125 was 13.9 U/L. An ultrasound neck was done which showed a small solid nodule measuring 2.7 x 1.7 mm in the midpole of right lobe with no calcification. A fine needle aspiration was done which was negative for malignancy. She was planned to be kept on follow–up with serum thyroglobulin check every 6 months. At present, she has completed about 6 months of follow-up with no signs of disease recurrence anywhere.

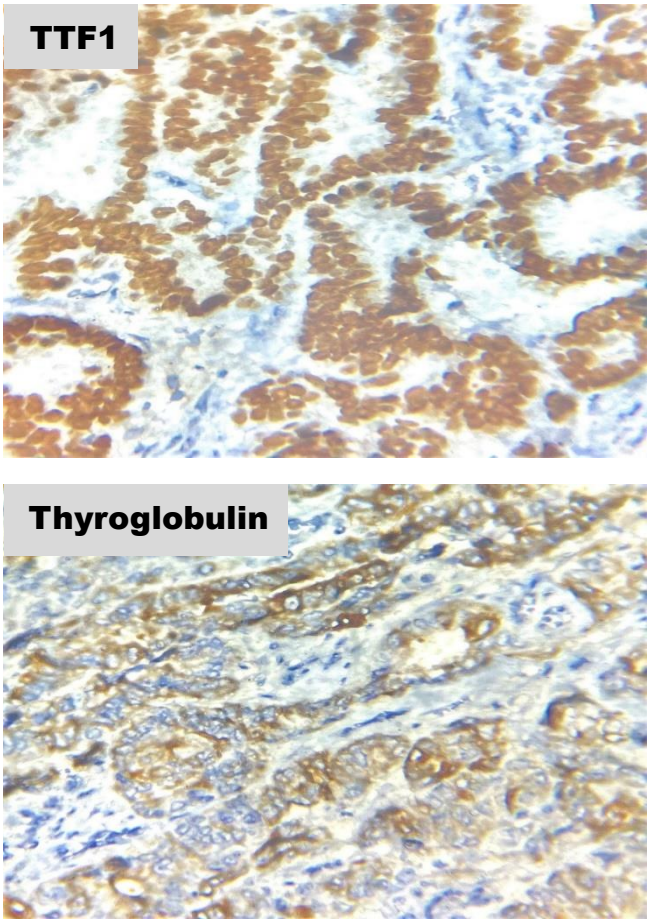


Figure 3. Immunohistochemistry images showing TTF-1 and thyroglobulin positivity.

Discussion

Struma ovarii is an ovarian germ cell tumour. It comprises of more than 50% thyroid tissue and can be differentiated from a mature teratoma, which contains only a small component (less than 50%) of benign thyroid tissue. Struma ovarii typically arises unilaterally, with 5% of cases seen bilaterally. A small proportion of struma ovarii may undergo malignant transformation (7). Malignant struma ovarii was first described by Wetteland in 1956 (9). Malignancy in struma ovarii is diagnosed based on histopathological criteria and guidelines for primary thyroid gland disease. Papillary and follicular carcinoma are the common histologies seen (10)0 Differentiated thyroid carcinoma arising from an MCT is rare with an estimated incidence being that of 0.1% to 0.2% (8). It is typically found incidentally in histopathology (5).

Multiple molecular abnormalities have been reported in thyroid cancer arising from ovarian teratomas, primarily in malignant struma ovarii. 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. However, it is uncertain if they have a significant impact on cancer prognosis in this type (5).

Struma ovarii may mimic the clinical symptoms of ovarian malignancy, presenting with ascites, a complex ovarian cyst, and an elevation of CA-125 (7). A case of pseudo-Meigs syndrome which includes ascites in the setting of hydrothorax, and elevated CA 125 levels has been described in malignant struma ovarii. The associated symptoms disappear, and the elevated CA 125 levels return to normal postoperatively usually without adjuvant therapy (11). Metastasis of malignant struma ovarii is seen in approximately 5 to 23 % of cases and is mainly intra-abdominal, although blood-borne metastasis can occur in the liver, lung, brain, bone, vertebra, and the contralateral ovary (8). Follicular carcinoma is more likely to metastasize to the lung, liver, and central nervous system whereas papillary carcinoma is said to involve the abdominal cavity and lymph nodes and occasionally the liver (12).

Dane *et al* (13) reviewed 15 cases of differentiated thyroid carcinoma arising in a mature ovarian teratoma and since then, 4 additional cases have been reported. (14 -17) Most patients, as in our case, presented with abdominal pain, only 2 patients did not report any symptoms. Papillary thyroid carcinoma (PTC) was the most common histopathologic type (53%), followed by follicular variant of PTC (42%) and follicular carcinoma (5%). Only 2 cases presented with thyroid tumor size $\leqslant$1 cm (5).

Ryder *et al* (18) reported a 0.9-cm follicular variant PTC within a 4.6-cm mature cystic teratoma (MCT). Thyroid ultrasound and ^{131}I diagnostic whole body scan were normal. No further treatment was performed on this patient. Dias *et al* (17) reported 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.

The optimal treatment of thyroid carcinoma arising within MCT is unclear because of the rarity of the disease. Moreover, no data on recurrence are available. In some of the reported cases, thyroidectomy was performed (5). whereas in some others, no thyroidectomy was performed (19-21). In these cases, no primary thyroid carcinoma was clinically apparent in further follow-up.

Differentiated thyroid carcinomas seen in struma ovarii can rarely present as a locally invasive or metastatic disease (22). Ovarian metastases from a primary thyroid carcinoma may occasionally occur and in such cases, the ovarian mass does not present with teratomatous characteristics (23).

After surgical resection 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 either single case reports or case series. Further therapy may include total thyroidectomy and radioiodine ablation which needs thyroglobulin monitoring, as well as radioiodine treatment if needed (24).

Conclusion

To our best knowledge, there is very scant information on the natural history and prognosis of papillary thyroid carcinoma arising on a mature cystic ovarian teratoma. Currently, there is no management consensus on this entity. It is important to have a multidisciplinary approach in such cases with an individualized approach toward treatment. We believe that a long-term follow-up is needed to comment on the natural course and prognosis of this disease.

Author contribution

PJN, **NN**, and **VSH** contributed to the conception, design, and definition of intellectual content, literature search, data acquisition, data analysis, statistical analysis, manuscript preparation, editing and review.

Conflict of interest

The authors declare no conflict of interest.

Funding

There is no funding.

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