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PAPILLARY THYROID CARCINOMA WITHIN MATURE TERATOMA OVARIAN: A RARE CASE REPORT

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ABSTRACT:

Papillary thyroid carcinoma is a rare germ cell tumor. We report a case of a 50-year-old woman who presented with an enlarged abdomen since a year ago. From the physical and ultrasound examinations, the patient was then diagnosed with a suspected malignant ovarian cyst. Postoperative histopathology results showed papillary thyroid carcinoma arising within mature cystic teratoma. Then, the patient was examined for thyroid ultrasonography, which revealed a benign nodule on the left thyroid. This result was supported by normal results of thyroid function and anti-thyroglobulin antibody tests, which was a sign of benign condition. The anatomical pathologists then concluded that the primary was of ovarian origin. Differentiating the primary source of papillary thyroid carcinoma, either from the ovary or thyroid, is essential to plan further proper management. Here, the report aims to describe how to diagnose papillary thyroid carcinoma arising within mature teratoma of the ovary and its management strategy.

Keywords: Papillary Thyroid Carcinoma, Teratoma Ovarian, Thyroid Nodule

INTRODUCTION

Germ cell tumors are a rare type of ovarian tumor. Germ cell tumors account for 15-20% of all ovarian tumors, and most are mature teratomas (Gonet et al., 2020). Ovarian struma contributed for 2-5% of mature teratoma cases and 1% of all ovarian tumors (Kondi-Pafiti et al., 2011). Ovarian struma occurs most frequently in women in their fourth to sixth decade of life (Yoo et al., 2008). Although tumors are usually benign, about less than 5% of all ovarian struma

undergo neoplastic transformation, one of which becomes papillary thyroid carcinoma. Similar with thyroid tumors in general, the most malignant type of ovarian struma is well-differentiated thyroid cancer, and the most common type is papillary thyroid carcinoma (70%) (Ganly et al., 2009).

The clinical presentation of papillary thyroid carcinoma is non-specific and similar to the clinical features of other ovarian cancers. Tumors are often accidentally diagnosed during ultrasound or computed tomography (CT) scan examinations or during surgery (Zhang & Axiotis, 2010). Primary papillary thyroid carcinoma can originate from the ovary itself or metastatic from the thyroid gland. These two conditions are very important to differentiate because their prognosis and clinical management are different (Shaaban et al., 2014).

Therefore, this case report will discuss a case of papillary thyroid cancer in mature cystic teratoma of ovary in order to improve the knowledge about its diagnosis and management.

RESEARCH METHODS

Case report

A 50-year-old woman referred from a secondary hospital, presented with an enlarged stomach since ± 1 year ago, abdominal pain, weight loss of 5 kg in the last 6 months, eating and drinking well, infrequent defecation and urinating normally, no discharge and no history of post-coital bleeding. Stomach full quickly when eating and feel bloating. The patient was menarche aged 13 years, no menopause yet and has a parity of two.

On gynecological examination, solid mixed cystic mass size 20 x 20 cm, well defined, flat surface, limited mobility, lateralization is difficult to determine was palpable. Investigation with transabdominal ultrasound (TAS) showed a hypohyperechoic picture of the size of the probe, solid part (+), septa (+), papillae (-), intramass vascularization (score 1), lateralization difficult to determine, free fluid (-) on both ovarium. Pap smear results showed negative for intraepithelial lesion or malignancy (NILM). Laboratory examination of CA-125 was 576.6 U/mL and RMI was 1729.8. Then, the patient was suspected with malignant ovarian cyst.

The patient was planned for a total abdominal hysterectomy with bilateral salphyngooophrerectomy and frozen section. The results of the frozen section examination of right and left ovarian tissue showed the picture of papillary thyroid carcinoma arising within mature cystic teratoma. Then, this result was confirmed by biopsy and histopathological examination on the right and left ovaries (Figure 1). The results also showed the follicular type of papillary thyroid carcinoma arising within mature teratoma on both ovary's tissue, with infiltration of atypical cells between the connective tissue stroma of the peritoneum and omentum suggesting infiltrating papillary thyroid carcinoma.

The patient was then consulted to the endocrinology division. At this time, the patient did not complain of any thyroid

abnormalities and a physical examination of the neck did not reveal any lumps. The patient then underwent an ultrasound examination of the thyroid with the results of a complex cyst on the right thyroid lobe. Meanwhile, on left lobe showed an isoechoic nodule, oval in shape, with a hypoechoic halo at the edges, measuring ± 0.67 x 0.44 x 0.94 cm, which on CDUS showed vascularity at the edges and intra-lesion. There was no calcification with CDUS and no increase in vascularity in the parenchyma. An isoechoic nodule, oval in shape, with a hypoechoic halo at the margin, in the left thyroid lobe corresponds to TIRADS-3 (mildly suspicious). Thyroid function test results of TSHS and FT4 were 0.65 and 0.93, respectively, and thyroglobulin level was 17 ng/mL. Then, the anatomical pathologist confirmed that the primary origin was within an ovarian teratoma.

The final diagnosis of the patient was stage IIIC ovarian cancer (papillary thyroid carcinoma). The chemotherapy protocol was carried out with four series of BEP (bleomycin/etoposide/carboplatin). At the end of the IV series, the tumor markers were examined, and found a decrease in CA 125 level to 16.9 U/mL (0.0-35.0 U/mL) and LDH 268 U/L (8.00-23.00 U/L). Computed tomography (CT) scan with contrast examination showed no residue mass in the pelvic so the patient was declared to have Complete response (CR) (RECIST criteria). The patient is still on regular follow-up.

RESULTS AND DISCUSSION

Papillary thyroid carcinoma of the ovarian can be a case of metastases from primary thyroid carcinoma or papillary carcinoma arising in ovarian struma. It is important to distinguish between these two conditions because their prognosis and clinical management are different (Beyenburg et al., 2005). Previous study showed that women with early menarche (<12 years of age) and late menopause (>50 years of age) are at higher risk of developing ovarian cancer due to a higher number of ovulatory cycles (Koh et al., 2012). In this case, the patient was 50 years old, had menarche at 13 years old, had not menopause yet, and had a parity of two.

Symptoms of ovarian cancer are not specific, therefor the diagnosis of the disease is usually in late condition. Symptoms that appear include full stomach sensation, bloating, nausea, abdominal distension, fatigue, changes in bowel movements, urinary symptoms, back pain, dyspareunia, and weight loss (Koh et al., 2012). In this case, the patient had stomach symptoms and weight loss that has occurred for a year. In addition, the patient also complained of difficulty defecating, nausea, and the stomach was full quickly when eating. A thorough physical examination should be performed, including a rectovaginal examination of the empty bladder to look for pelvic and abdominal masses (Smith, 2017). In this case, we found a solid mixed cystic mass measuring 20 x 20 cm with well-defined margins, flat surface, limited

mobility, and lateralization was difficult to determine on physical examination.

Radiological examinations including transvaginal ultrasound and/or abdominal ultrasound are performed to support the diagnosis. This examination provides a good overview of the size, location, and complexity of ovarian mass. To determine the extent of the tumor, further imaging with computed tomography (CT) scan of the chest and abdomen, magnetic resonance imaging (MRI) of the pelvis, and/or positron emission tomography (PET) scan may be performed (Roth et al., 2008). In this case, a transabdominal ultrasound examination was performed and the results showed a hypohyperechoic mass, septa, solid sections, and intramass vascularity with a score of 1.

Measurement of CA-125 levels is usually done in conjunction with radiological examinations. The level of CA-125 also has a role to calculate the risk malignancy index (RMI), which is combined with the results of ultrasound examination and menopausal status. An RMI above 200 is associated with a high risk of malignancy, with a specificity of more than 96% (Renjen et al., 2018). In this case, only CA-125 and RMI were examined, and both of the results showed a risk of malignancy.

Cases of metastases papillary thyroid carcinoma from the thyroid gland are very rare and usually occurred in advanced thyroid carcinoma. Ovarian goiter containing thyroid-type carcinoma such as papillary carcinoma can also be distinguished from metastases from primary thyroid carcinoma by thyroid hormone examination and ultrasonography. Symptoms that may arise are progressive swelling in the anterior neck, usually > 1 cm. Other symptoms include hoarseness, dysphagia, and hemoptysis. The appearance of these symptoms indicates the possibility of malignancy in the thyroid (Jain, 2021). This patient did not have these symptoms and no palpable mass in the neck area on physical examination. Ultrasound examination of the thyroid gland revealed a complex cyst with a benign appearance in the right thyroid lobe and an isoechoic nodule with a hypoechoic halo at the edges with a midly suspicious impression on the left thyroid lobe. Fine needle aspiration biopsy (FNAB) is an examination to determine the risk of malignancy, the examination is done if the thyroid ultrasound found nodules measuring > 1cm. From the FNAB, cytopathological results were reported according to the Bethesda System for Reporting Thyroid Cytopathology.[10] Unfortunately, the FNAB examination could not be performed in this patient due to the small size of the thyroid nodule.

When an ovarian mass is found to contain cells with features of thyroid carcinoma, a differential diagnosis should be considered between thyroid cancer arising from ovarian goiter or ovarian metastases originating from thyroid carcinoma. In the case of ovarian metastases originating from thyroid carcinoma, they tend to have no teratomatous features (Brogioni et al., 2007). In this case, it was found a follicular variant on the right ovary and a follicular type on the left ovary of papillary thyroid carcinoma arising within mature teratoma.

The tumor extends to the peritoneum and infiltrates between the stroma of the omental tissue and the lymphatics. Based on the data above, the primary papillary thyroid carcinoma was from the ovary, neither a metastasis from thyroid carcinoma nor metastases from the ovary to the thyroid.

Surgical treatment of ovarian mass is the main modality. However, management after initial surgery is controversial. Mattucci et al[12], suggested that the management of malignancy in ovarian struma should be the same as for thyroid carcinoma. After surgical removal of ovarian neoplasms, patients are recommended to undergo thyroidectomy, radiotherapy with I-131, and levothyroxine suppression therapy. In the case of malignant ovarian struma with distant metastases, a more aggressive treatment approach is suggested (total hysterectomy with bilateral excision of the adnexa and ovaries, omentectomy, total thyroidectomy, and I-131 therapy) (Mattucci et al., 2007). Because the primary tumor came from the ovary and there was no clinical or supporting examination evidence of primary thyroid carcinoma, total thyroidectomy and I-131 therapy were not performed in this case.

Based on recommendations from the Royal College of Obstetric and Gynecology (RCOG), American Journal of Obstery and Gynecology (AJOG), European Society for Medical Oncology (ESMO), National Guidelines for Gynecological Cancer Medicine Services from HOGI 2018, and clinical practice guidelines at Prof Dr I.G.N.G Ngoerah Hospital, the advanced stage germ cell ovarian cancer was followed by BEP (bleomycin/etoposide/carboplatin) chemotherapy in 3-4 series after surgery. This patient was given four series of BEP chemotherapy.

Makani et al. recommended long-term monitoring for at least 10 years (Marti et al., 2012). Other source recommended regular follow-up of germ cells. Physical examination is carried out every month in the first year, every 2 months in the second year, every 3 months in the third year, every 4 months in the 4th year, and every 6 months thereafter (Berek et al., 2021). Based on practical guideline in our hospital, follow-up is done every 3-6 months for the first 2 years after therapy and every 12 months for the next 3 years. To determine the response to therapy that has been given was based on the RECIST (Response Evaluation Criteria in Solid Tumor) criteria: 1) complete response: the disappearance of all lesions within 4 weeks; 2) Partial response: at least 30% reduction in lesion size, within 4 weeks; 3) Stable disease: not included in the criteria for partial and progressive response; 4) Progressive disease: 20% increase with no complete, partial and stable response before disease progression or new lesions (Eisenhauer et al., 2009).In this case, a follow-up of 4 weeks and three months after therapy was carried out. Based on history, physical examination, and support examination obtained a complete response (RECIST criteria).

CONCLUSION

Papillary thyroid carcinoma is a rare form of mature teratoma. When an ovarian mass is found to contain cells with features of thyroid carcinoma, as in this case report, a differential diagnosis should be considered between thyroid cancer arising from ovarian goiter or ovarian metastases originating from primary thyroid carcinoma. Appropriate diagnosis and management will help increase the survival rate in patients with papillary thyroid type ovarian cancer. So, a multi-disciplinary collaboration is recommended in managing this case. After therapy, the patient should not be lost to follow-up. It is important to remind and contact the patient to keep control at the polyclinic.

BIBLIOGRAFI

Berek, J. S., Renz, M., Kehoe, S., Kumar, L., & Friedlander, M. (2021). Cancer of the ovary, fallopian tube, and peritoneum: 2021 update. *International Journal of Gynecology & Obstetrics*, *155*, 61–85.

Beyenburg, S., Mitchell, A. J., Schmidt, D., Elger, C. E., & Reuber, M. (2005). Anxiety in patients with epilepsy: systematic review and suggestions for clinical management. *Epilepsy & Behavior*, *7*(2), 161–171.

Brogioni, S., Viacava, P., Tomisti, L., Martino, E., & Macchia, E. (2007). A special case of bilateral ovarian metastases in a woman with papillary carcinoma of the thyroid. *Experimental and Clinical Endocrinology & Diabetes*, *115*(06), 397–400.

Eisenhauer, E. A., Therasse, P., Bogaerts, J., Schwartz, L. H., Sargent, D., Ford, R., Dancey, J., Arbuck, S., Gwyther, S., & Mooney, M. (2009). New response evaluation criteria in solid tumours: revised RECIST guideline (version 1.1). *European Journal of Cancer*, *45*(2), 228–247.

Ganly, I., Agrawal, N., Tuttle, R. M., Patel, S. G., & Shah, J. P. (2009). Management of Thyroid Cancer. *Principles and Practice of Head and Neck Surgery and Oncology. London*, 413–443.

Gonet, A., Ślusarczyk, R., Gąsior-Perczak, D., Kowalik, A., Kopczyński, J., & Kowalska, A. (2020). Papillary thyroid cancer in a struma ovarii in a 17-year-old nulliparous patient: a case report. *Diagnostics*, *10*(1), 45.

Jain, V. (2021). The role of imaging in the evaluation of hoarseness: A review. *Journal of Neuroimaging*, *31*(4), 665–685.

Koh, S. C. L., Huak, C. Y., Lutan, D., Marpuang, J., Ketut, S., Budiana, N. G., Saleh, A. Z., Aziz, M. F., Winarto, H., & Pradjatmo, H. (2012). Combined panel of serum human tissue kallikreins and CA-125 for the detection of epithelial ovarian cancer. *Journal of Gynecologic Oncology*, *23*(3), 175–181.

Kondi-Pafiti, A., Mavrigiannaki, P., Grigoriadis, C., Kontogianni-Katsarou, K., Mellou, A., Kleanthis, C. K., & Liapis, A. (2011). Monodermal teratomas (struma ovarii). Clinicopathological characteristics of 11 cases and literature review. *European Journal of Gynaecological Oncology*, *32*(6), 657–659.

Marti, J. L., Clark, V. E., Harper, H., Chhieng, D. C., Sosa, J. A., & Roman, S. A. (2012). Optimal surgical management of well-differentiated thyroid cancer arising in struma ovarii: a series of 4 patients and a review of 53 reported cases. *Thyroid*, *22*(4), 400–406.

Mattucci, M. L., Dellera, A., Guerriero, A., Barbieri, F., Minnelli, L., & Furlani, L. (2007). Malignant struma ovarii: a case report and review of the literature. *Journal of Endocrinological Investigation*, *30*, 517–520.

Renjen, P. N., Chaudhari, D. M., Shilpi, U. S., Zutshi, D., & Ahmad, K. (2018). Paraneoplastic cerebellar degeneration associated with ovarian adenocarcinoma: a case report and review of literature. *Annals of Indian Academy of Neurology*, *21*(4), 311.

Roth, L. M., Miller III, A. W., & Talerman, A. (2008). Typical thyroid-type carcinoma arising in struma ovarii: a report of 4 cases and review of the literature. *International Journal of Gynecological Pathology*, *27*(4), 496–506.

Shaaban, A. M., Rezvani, M., Elsayes, K. M., Baskin Jr, H., Mourad, A., Foster, B. R., Jarboe, E. A., & Menias, C. O. (2014). Ovarian malignant germ cell tumors: cellular classification and clinical and imaging features. *Radiographics*, *34*(3), 777–801.

Smith, C. G. (2017). A resident's perspective of ovarian cancer. *Diagnostics*, *7*(2), 24.

Yoo, S.-C., Chang, K.-H., Lyu, M.-O., Chang, S.-J., Ryu, H.-S., & Kim, H.-S. (2008). Clinical characteristics of struma ovarii. *Journal of Gynecologic Oncology*, *19*(2), 135–138.

Zhang, X., & Axiotis, C. (2010). Thyroid-type carcinoma of struma ovarii. *Archives of Pathology & Laboratory Medicine*, *134*(5), 786–791.

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