

Case Report

Follicular variant of papillary thyroid carcinoma arising from a dermoid cyst: A rare malignancy in young women and review of the literature

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Abstract

Objective: Benign or mature cystic teratomas, also known as dermoid cysts, are composed of mature tissues, which can contain elements of all three germ cell layers. Malignant transformation of a mature cystic teratoma is more common in postmenopausal women, however, it can also, rarely, be identified in younger women. We present a case of a 19-year-old woman with malignant transformation of an ovarian mature cystic teratoma.

Case Report: Our case was a 19-year-old woman, who was diagnosed postoperatively with follicular variant of papillary thyroid carcinoma in a mature cystic teratoma. She underwent right cystectomy for adnexal mass. Postoperative metastatic workup revealed a non-metastatic disease and the patient did not undergo any further treatment. After 2 months, a near-total thyroidectomy was performed. Serum thyroglobulin levels were monitored on follow-up and the patient is asymptomatic.

Conclusion: Malignant transformation of a dermoid cyst is a rare ovarian neoplasm. We believe that unilateral oophorectomy or cystectomy is a reasonable treatment option for cases in which there is no evidence of capsular invasion, vascular invasion or gross metastasis, and preservation of fertility is desired. Total thyroidectomy was diagnosed in selected cases. Serial serum thyroglobulin should be used as a tumor marker for follow-up.

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Keywords: carcinoma; cystectomy; dermoid cyst; follicular variant papillary thyroid mature cystic teratoma; malignant transformation

Introduction

The term teratoma is derived from the Greek root “teratos” which means monster [1]. Mature cystic teratoma (MCT), commonly referred to as a dermoid cyst of the ovary, is the most common type of ovarian teratoma and germ cell neoplasm comprising 10–20% of ovarian tumors [2]. In its pure form, MCT is always benign, however, malignant transformation of MCT has been reported, rarely, with a rate = 1–2% [3]. Malignant transformation is most common in postmenopausal women, however, it can also be rarely identified in younger women. The most common malignant tumor in mature cystic

teratomas is squamous cell carcinoma, whereas, papillary thyroid carcinoma caused by mature cystic teratomas is extremely rare. Malignant transformation is rarely diagnosed pre-operatively, due to its rarity [4]. Hence, in most cases, malignant transformation is most commonly diagnosed only by post-operative pathological examination.

In this paper, we report a rare case of follicular variant papillary thyroid cancer arising from a mature cystic teratoma of the ovary, which was unexpectedly discovered during histopathologic examination.

Case report

A 19-year-old woman was admitted to our gynecology polyclinic with a complaint of 3 months duration of amenorrhea

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and abdominal distension. She was married 7 months prior to the examination and had no history of pregnancy. Her physical examination revealed a mass in the abdominal region. Transvaginal ultrasonography revealed a mass = 15 cm × 16 cm, containing hyperechogenic areas. From MRI findings, the tumor was considered to be an ovarian cystic tumor containing a fatty structure (Fig. 1). A laparotomy was performed and the findings were as follows: the right dermoid ovarian cyst was hairy and had a solid component, size = 14 cm × 15 cm; the size and shape of the uterus, the left ovary and the tubes were normal. No other pelvic pathology was observed and liver and mesenteric examinations were normal. The mass appeared to have originated from the right ovary, was mobile and had a smooth surface. An ovarian cystectomy was performed. Macroscopic appearance of the lesion resembled a dermoid cyst involving adipose like tissue, hair and tooth. No palpable lymph nodes were seen. Histopathologic evaluation of the lesion demonstrated a follicular variant papillary thyroid carcinoma in a mature cystic teratoma as the microscopic focus, surrounded by normal ovarian tissue. Microscopic examination showed the thyroid tissue in the mature cystic teratoma to be 2.5 cm. Thyroid tissue composed of follicles, lined with cuboidal epithelium and filled with colloid was seen at one focus containing a 0.2 cm sized encapsulated follicular variant of thyroid papillary carcinoma. Immunohistochemistry demonstrated focal membrane staining with HBME-1 (Fig. 2). The tumor was limited to the thyroid gland and did not impact the ovarian capsule or vascular spaces. The



Fig. 1. MR images show the mass with high-signal-intensity due to its mucinous contents. Low-signal-intensity central nodule is also seen.

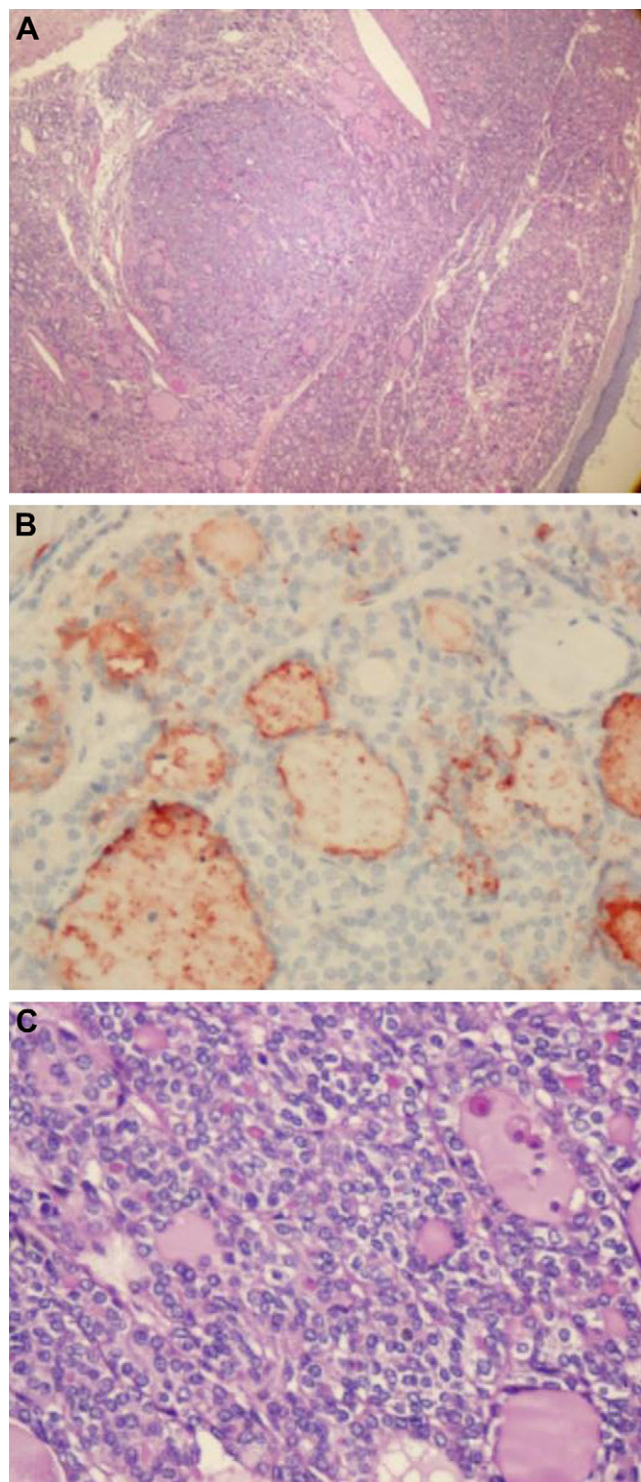


Fig. 2. (A) follicular variant papillary thyroid carcinoma within an ovarian teratoma. Normal ovarian tissue is seen on the right. On the right, normal-appearing thyroid tissue merges with a focus of follicular variant papillary thyroid carcinoma (H&E, × 40); (B) membrane positivity for HBME-1 (H&E × 400); and (C) follicular variant of papillary thyroid carcinoma, exhibiting clear nuclei, prominent nucleoli, occasional nuclear grooves, and follicles with scalloping (H&E × 400).

histologic features of the specimen were consistent with follicular variant papillary thyroid carcinoma arising in a mature cystic teratoma. There was no immature tissue inside the teratoma. Peritoneal washing showed benign mesothelial cells. Thyroid ultrasonography was necessary to eliminate any primary thyroid malignancy. Multiple nodes were detected at both the right and left lobes of the gland. Fine needle aspiration biopsy revealed benign thyroid nodules. The serum thyroglobulin level = 73.4 ng/mL (normal range 0.73–84 ng/mL). Thyroid-stimulating hormone = 0.62 mIU/mL (normal range = 0.35–5 mIU/mL), free thyroxine = 1.37 ng/dL (normal range = 0.7–1.9 ng/dL) and free triiodothyronine = 3.18 pg/mL (normal range = 1.57–4.71 pg/mL) (1 month after surgery). After 2 months, a near-total thyroidectomy was performed and pathological examination of the thyroid gland was normal. The patient was started on exogenous thyroid hormone. Serum thyroglobulin levels were monitored on follow-up of the patient. Five months after surgery and taking levothyroxine, thyroglobulin = 3.65 ng/dL, TSH = 0.64 mIU/L and FT4 = 1.31 ng/dL. The patient was under surveillance for 6 months, with no evidence of recurrence.

Discussion

A mature cystic teratoma of the ovary or dermoid cyst is composed of well-differentiated derivatives of the three germ

layers, the ectoderm, the mesoderm and the endoderm, while ectodermal elements usually predominate. Complications of cystic teratomas include torsion (16%), malignant degeneration (2%), rupture (1–2%), and infection (1%) [5]. Malignant transformation is usually diagnosed postoperatively, from the histological findings. In 1957, Peterson reviewed 229 cases of malignant transformation of mature cystic teratomas [6]. He reported malignant transformation rates of 1.8%; 81.1% of the tumors were squamous cell carcinomas and 6% were adenocarcinomas. The other malignant transformations include malignant melanoma, sarcoma, basal cell carcinoma, carcinoid tumor, adenocarcinoma of intestinal epithelium, and thyroid carcinoma [7,8].

Malignant transformation of thyroid tissue in MCT can be classified histopathologically into three types [9]. The first and most common type is papillary carcinoma (44%). The malignant features of these tumors include optically clear or ground-glass overlapping nuclei with nuclear grooves. The second type is follicular carcinoma, which is diagnosed when cells containing mitotic figures begin forming follicles and capsular or vascular invasion is noted. Follicular carcinoma accounts for approximately 30% of malignant degeneration in MCT. Lastly, there is a follicular variant of a papillary carcinoma, which accounts for 26% of malignant degeneration. This follicular variant is unique because it shares similar nuclear cytological features of a papillary carcinoma; it has,

Table 1
Clinical details of the patients with papillary thyroid carcinoma on thyroid tissue in an ovarian teratoma (excluded struma ovarii cases).

Patient	Study	Age (y)	Symptoms at diagnosis	Tumor size (cm)	Intervention	Pathology	Thyroidectomy
1	Chadha [12]	71	Abdominal pain	18	TAH + BSO	Follicular thyroid carcinoma	Not reported
2	Doldi [13]	58	Abdominal swelling and discomfort	4	Tumor resection	Papillary carcinoma of the thyroid	Not reported
3	Krnojelac [14]	43	During c/s	8	Oophorectomy	Papillary carcinoma of the thyroid	Yes
4	Zergeroglu [15]	28	Abdominal discomfort, pain	9 × 8	TAH + BSO	Papillary carcinoma of the thyroid	Not reported
5	Sayhan [16]	71	Abdominal pain	12 × 10 × 8	TAH + BSO	Papillary carcinoma of the thyroid	Unknown
6	Soto-Moreno [17]	31	Episode of metrorrhagia	7 × 5	Tumor resection	Papillary carcinoma	Yes
7	Bal [1]	60	Post-menopausal bleeding	7	TAH + BSO	Papillary carcinoma	Unknown
8	Ryder [18]	49	Abdominal pain and emesis	4.6	Unilateral oophorectomy	Follicular variant papillary thyroid carcinoma	Unknown
9	Lee [19]	35	Intermittent low abdominal discomfort	13 × 11	LSO followed by TAH + USO	Papillary carcinoma, follicular variant	Unknown
10	Quadri [20]	54	Lower abdominal pain	8 × 7 × 4	TAH + BSO	papillary carcinoma, follicular variant	Yes
11	Al-Nawafleh [21]	36	Left lower abdominal pain	6 × 5	LSO	Papillary carcinoma	Unknown
12	Lataifeh [22]	39	Incidental	4.5	LSO	Papillary thyroid cancer	Yes
13	Bougerra [23]	44	Abdomino pelvic mass- painless	20 × 13	TAH + BSO	Papillary thyroid carcinoma	Yes
14	Tanaka [24]	50	Hypermenorrhea	8	TAH + BSO	Follicular variant thyroid-type papillary carcinoma	Unknown
15	Our case	19	Abdominal distension	15 × 16	Cystectomy	Follicular variant papillary thyroid carcinoma	Yes

LSO = laparoscopic salpingo-oophorectomy; TAH = total abdominal hysterectomy; BSO = bilateral salpingo-oophorectomy; USO = unilateral salpingo-oophorectomy.

however, a follicular architecture. In our case, a follicular variant papillary thyroid carcinoma was found. It is a teratoma with no predominance of thyroid tissue, where we found a limited area with typical histological characteristics of a follicular variant papillary thyroid carcinoma and positive thyroglobulin on immunohistochemical analysis. Thus, it is considered a papillary carcinoma on thyroid tissue in an ovarian teratoma.

The presence of thyroid tissue in the ovaries was first described by Von Kahlden in 1895 [10]. Papillary thyroid cancer arising from ovarian teratomas is a rare occurrence, with an estimate of 0.1–0.3% [11]. To date, only 15 cases of papillary thyroid carcinoma arising in a mature cystic teratoma of the ovary have been reported. Herein, we report the clinicopathologic features of one such case. A review of the literature is also presented (Table 1) [1,12–24]. The average age was 45 years. These patients predominantly presented with abdominal pain (50%), a pelvic mass (12%) and menstrual irregularities (12%). Papillary carcinoma was the most common (60%) histopathological finding followed by follicular variant of papillary carcinoma (33%) and follicular carcinoma (6%).

The survival outcome of patients with malignant transformation of the ovarian mature cystic teratomas is dependent on the stage. These tumors arising from ovarian teratomas can occasionally present with locally invasive disease, or with distant metastases, and may be associated with recurrences in some patients. In the present case, a cystic lesion was found on the right ovary, but no suspicious clinical features such as rupture, tumor dissemination, ascites or adhesions were observed. The optimal treatment for malignant transformation of MCT is uncertain because of the rarity of the disease [21]. This rarity suggests a low likelihood of future prospective studies to determine the best treatment options. Conservative treatment is often offered to younger patients who wished to retain their fertility and it consists of unilateral salpingo-oophorectomy or cystectomy without adjuvant therapy. Conservation of ovarian tissue on the affected side is acceptable. In the 19-year-old patient, cystectomy was performed in order to preserve fertility. The patient was treated by cystectomy with the intact removal of the cyst without spillage.

Subsequently, thyroidectomy was performed to rule out primary thyroid papillary carcinoma, because the thyroid ultrasound scan showed multinodular goiter, and the patient was started on exogenous thyroid hormone. These patients must undergo a thyroidectomy, which serves two purposes [25]: (1) it ensures that the patient's thyroid is normal and rules out the possibility of metastasis to the ovaries; and (2) it potentiates iodine therapy, because the removal of all normal thyroid tissue allows the radioactive iodine to concentrate in the accessory thyroid tissue. Thyroglobulin, produced by both normal and abnormal thyroid tissue, has been used as a post-operative tumor marker. Serum thyroglobulin levels are checked every 4 months during the 1st year after surgery. If a patient's thyroglobulin level is unexpectedly high, a radioactive iodine scan is carried out, in order to evaluate for recurrence.

After thyroidectomy, we followed up the patient by measuring serum thyroglobulin levels, with periodic transvaginal ultrasonography and abdominal MRI. During a 6 month long follow up, we did not find any evidence of recurrence or metastasis.

In conclusion, we describe follicular variant papillary thyroid carcinoma arising from the thyroidal tissue of an ovarian dermoid cyst.

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