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GYNECOLOGIC ONCOLOGY

Struma ovarii with atypical features and synchronous primary thyroid cancer: a case report and review of the literature

Molly R. Siegel[1] · Rebecca J. Wolsky[2,3] · Edwin A. Alvarez[1] · Biftu M. Mengesha[1]

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Abstract

Purpose To present a case of struma ovarii with a typical features and synchronous primary thyroid carcinoma and review the available literature to guide diagnosis and management of these tumors.

Methods We present a case from our hospital of a 55-year-old woman who had an adnexal mass with features concerning for papillary thyroid carcinoma and was ultimately determined to be struma ovarii with atypical features. Subsequent thyroid imaging and biopsy revealed a primary cervical thyroid carcinoma. We performed a PubMed search of published English language articles using the search terms "malignant struma ovarii," "metastatic struma ovarii," "struma ovarii with malignant transformation," "struma ovarii papillary thyroid carcinoma," "struma ovarii follicular thyroid carcinoma," and "struma ovarii with concurrent primary thyroid carcinoma."

Results Literature review included 104 studies with a total of 195 patient cases. The average age at presentation was 44.9 years. 25.1% of patients had metastatic disease at presentation, and 6.2% had synchronous primary carcinomas; all of which were located in the thyroid.

Conclusions Thyroid carcinoma arising in struma ovarii or mature cystic teratoma should prompt clinical evaluation and imaging of the thyroid given the possibility of synchronous primaries, metastases, and recurrence.

Keywords Struma ovarii · Malignant struma ovarii · Struma ovarii with malignant transformation · Germ cell tumors

Background

Ovarian teratomas are germ cell tumors of the ovary that comprise approximately 20% of all ovarian tumors [1, 2]. These tumors may contain tissue from all three germ layers (ectoderm, mesoderm, and endoderm) commonly including hair, skin, teeth, bone, and thyroid [2, 3]. Approximately 20% of teratomas contain thyroid tissue, and 5% of thyroid-containing teratomas are classified as struma ovarii, defined as containing at least 50% thyroid tissue [1, 4]. There are three main types of thyroid tissue that arise in ovarian teratomas: tissue that is pathologically similar to normal thyroid, proliferative cellular lesions with hyperplastic or adenomatous features, and thyroid-type carcinomas [5].

Approximately 0.5–10% of struma ovarii are malignant, and 5–23% of those undergo metastasis [6–11]. The historic term "malignant struma ovarii" is now more appropriately called "thyroid carcinoma arising in struma ovarii" [6]. This term differentiates these carcinomas from the broader category of thyroid-type carcinoma in the ovary, which can develop from four precursors: mature cystic teratoma or

This work was performed at the Zuckerberg San Francisco General Hospital and the University of California, San Francisco.

✉ Molly R. Siegel
molly.siegel@ucsf.edu

Rebecca J. Wolsky
Rebecca.wolsky@ucsf.edu

Edwin A. Alvarez
Edwin.alvarez@ucsf.edu

Biftu M. Mengesha
Biftu.mengesha@ucsf.edu

[1] Department of Obstetrics, Gynecology, and Reproductive Sciences, University of California, San Francisco, Molly Siegel 550 16th Street, 7th Floor, Ob/Gyn Mailstop 0132, San Francisco, CA 94143, USA

[2] Department of Surgical Pathology, Zuckerburg San Francisco General Hospital and Trauma Center, San Francisco, USA

[3] Department of Anatomic Pathology, University of California, San Francisco, San Francisco, USA

struma ovarii; strumal carcinoid; metastases from primary thyroid carcinoma; or no clear precursor [12, 13]. Of the thyroid-type carcinomas arising from mature cystic teratomas and struma ovarii, the most common histological subtypes are papillary and follicular thyroid carcinoma [14]. Because these tumors are rare, guidelines for optimal treatment and surveillance are lacking [7, 15–17]. Here we present a case of a patient with struma ovarii with atypical features concerning papillary thyroid carcinoma and synchronous primary cervical thyroid carcinoma, followed by a literature review and discussion of the incidence, presentation, and management of such cases.

Case presentation

A 55-year-old postmenopausal P3 woman without any significant past medical history presented to our institution's Emergency Department with right-sided abdominal pain. She was found to have pyelonephritis complicated by a right psoas abscess on computerized tomography (CT) of the abdomen and pelvis, for which she was treated with intravenous antibiotics and imaging-guided drainage of the psoas abscess with drain placement. The CT also showed an incidental left adnexal mass characterized as heterogeneous with varying densities including fluid, nonenhancing and enhancing solid components, and densely calcified material, favored to represent a teratoma. A follow-up ultrasound was obtained which noted an 8 × 7 cm heterogeneous left adnexal mass with internal calcifications and solid and cystic components. She was referred to gynecology for further evaluation and management.

During the gynecologic consultation, the patient reported a 100-lb weight loss and intermittent fatigue over the past year, which she related to depression. She denied abdominal pain, bloating, night sweats, palpitations, hot or cold intolerance, hair or nail changes, voice changes, neck pressure or pain, or dysphagia. Her neck exam was notable for a thyroid goiter and the pelvic exam revealed a fixed, midline, nontender, solid mass palpated in the posterior cul-de-sac. Laboratory investigations were performed and notable for a CA-125 of 65 units/ml and thyroid function tests within normal limits (TSH 2.64 mIU/l, free T4 1.04 ng/dl). Magnetic resonance imaging (MRI) of the pelvis demonstrated a complex, multi-cystic left adnexal lesion measuring 7.9 × 7.9 × 8.8 cm with varying signal intensities containing fat and bone as well as numerous enhancing solid components, concerning for an immature teratoma of the ovary.

Given the fixed nature of the mass, concerning imaging features, and elevated CA-125 for a postmenopausal woman, the patient was referred to gynecologic oncology for consultation, after which she was recommended to undergo surgical removal of the mass and ovary with possible staging procedures. However, prior to her planned surgery, a follow-up pyelogram performed after removal of the psoas drain revealed nonfunctioning right kidney, and as such the patient's gynecologic surgery was delayed until after a right nephrectomy done with urology. She was unfortunately lost to follow-up for 6 months after her nephrectomy, and upon representation to gynecologic oncology, a repeat ultrasound showed an increased size of the left ovarian mass to 10.1 cm.

The patient then underwent an uncomplicated laparoscopic left salpingo-oophorectomy and pelvic washings. Intraoperative findings noted an 11-cm multi-cystic ovarian mass, which was removed from the abdomen without any spillage of contents. The final pathology report revealed a mature cystic teratoma comprised of approximately 60% thyroid tissue, consistent with struma ovarii. Selected images from the patient's pathology specimen are shown in Fig. 1. Additional elements such as urothelium, squamous epithelium, and bone were also present. Within the struma ovarii, a few microscopic foci displayed cytologic features concerning for papillary thyroid carcinoma. The features, which were noted on medium power but best appreciated on high power, included angulated, overlapping nuclei with fine powdery chromatin and nuclear grooves. Nuclear pseudo-inclusions were not appreciated. While the cytologic features were concerning, there was no distinct mass within the thyroid tissue, no infiltrative growth of the concerning cells, and no papillary architecture that would confirm an outright diagnosis of papillary thyroid carcinoma. The pathologist noted that by current endocrine tumor classifications, similar findings in a thyroid gland would be considered a microcarcinoma (< 1 cm papillary thyroid carcinoma) or non-invasive follicular thyroid neoplasm with papillary-like nuclear features (NIFTP), a benign diagnosis. The pathology was ultimately signed out as "struma ovarii with atypical features" with concern for papillary thyroid carcinoma. The pelvic washings were benign. Given the atypical features of the ovarian mass, the patient was referred to endocrinology for further evaluation.

Endocrinological consultation deemed the patient to be clinically euthyroid with normal thyroid and parathyroid studies. A thyroid ultrasound showed a 4-cm left heterogeneous nodule with internal vascularity and microcalcifications and a subcentimeter right superior thyroid nodule with suspicious features. She underwent fine needle aspiration for both nodules identified on ultrasound, and pathology demonstrated papillary thyroid carcinoma. She was then referred to Otolaryngology for surgical treatment and underwent an uncomplicated total thyroidectomy with cervical neck dissection; pathology revealed stage III (pT2N1a) papillary thyroid carcinoma. She was started on levothyroxine postoperatively, but was not able to have immediate postoperative radioactive iodine therapy as recommended due to lack of social support. Two months after surgery, a whole-body radioiodine scan showed significant radioactive uptake in the midline superior thyroid bed consistent with residual thyroid or thyroid carcinoma. The remnant

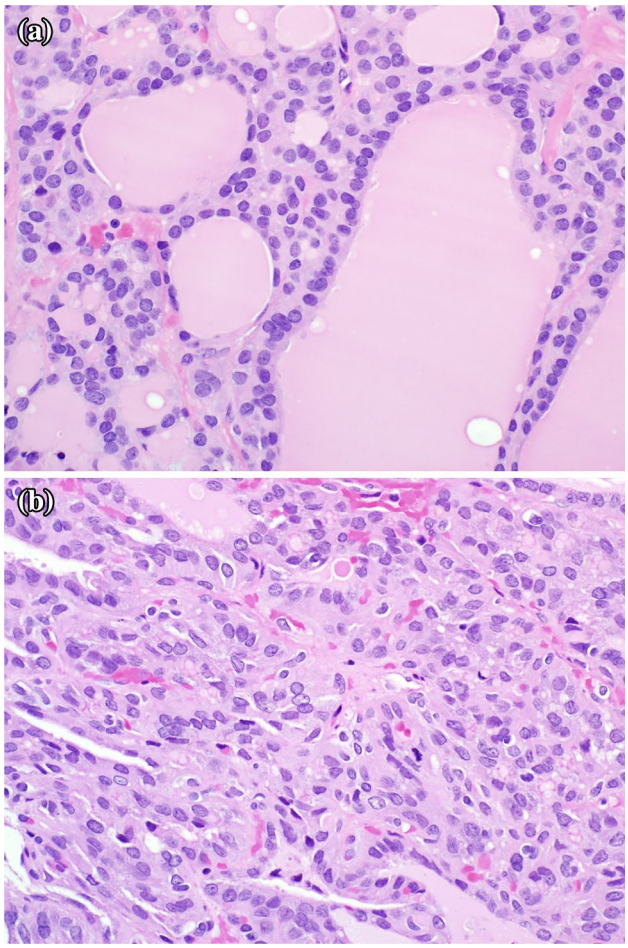

Fig. 1 Patient pathology images. **a** The majority of the thyroid tissue within the teratoma (struma ovarii) shows benign thyroid follicles; the cells' nuclei are evenly spaced with smooth nuclear chromatin (magnification 400×). **b** Focally on high-power microscopic review, the thyroid follicle cells demonstrate angulated, overlapping nuclei and nuclear groves, features concerning apillary thyroid carcinoma. No definite nuclear pseudoinclusions, papillary architecture, or mass-like growth are present (magnification 400×)

was excised, and final pathology was benign. The planned next step in her management is staged radioactive iodine therapy.

The patient was referred to medical genetics for possible genetic testing. With regard to surveillance of her ovarian mass, the patient will have semiannual clinical surveillance with gynecologic oncology after radioactive iodine thyroid ablation.

Literature review

Materials and methods

We performed a PubMed search of published English language articles using the search terms "malignant struma ovarii," "metastatic struma ovarii," "struma ovarii with malignant transformation," "struma ovarii papillary thyroid carcinoma," "struma ovarii follicular thyroid carcinoma," and "struma ovarii with concurrent primary thyroid carcinoma." We included cases of thyroid carcinoma arising in struma ovarii or mature cystic teratoma at initial ovarian pathologic evaluation.

Results

The initial search resulted in 308 articles for analysis. Figure 2 demonstrates the process by which studies were considered for review. We included 104 studies with a total of 195 patient cases. Characteristics of the cases are described in Table 1. The average age at presentation was 44.9 years. The most common presenting symptoms or signs were abdominal/pelvic pain ($N=41$), incidental imaging or exam findings ($N=29$), or abdominal mass with distension or bloating ($N=23$). Other presenting symptoms included abnormal or postmenopausal bleeding ($N=21$), thyroid dysfunction ($N=9$), gastrointestinal symptoms ($N=5$), urinary symptoms ($N=4$), pathologic fracture ($N=2$), ovarian torsion ($N=1$), back pain ($N=1$), and ascites ($N=1$). The most common pathologies were papillary thyroid carcinoma ($N=92$), follicular variant of papillary thyroid carcinoma ($N=47$), and follicular thyroid carcinoma ($N=34$). Metastatic disease was noted upon presentation in 49 cases (25.1%). The most common sites of metastases were ascites and peritoneal washings ($N=27$) and pelvic structures or pelvic peritoneum ($N=23$). Other metastatic sites included abdominal peritoneum, lung or chest, omentum, bone, liver, rectum, and pelvic or distant lymph nodes. Twelve patients (6.2%) had synchronous primary carcinomas; all of which were located in the thyroid [3, 6, 11, 18–26]. Eight of the synchronous primary thyroid carcinomas were papillary thyroid carcinomas, and four were follicular variant of papillary thyroid carcinoma. Six of the synchronous primary cervical carcinomas had histology that matched that of the ovarian tumor but was still determined to be a separate primary [3, 11, 19, 20, 23, 26].

Treatment was described in 139 patients (71.2%). 67 of these patients were treated with surgery alone (34.3%). The surgeries performed included unilateral salpingooophorectomy with or without staging procedures (19 patients); unilateral salpingoophorectomy with contralateral cystectomy with or without fertility-sparing staging (6 patients); total abdominal hysterectomy with bilateral salpingoophorectomy with or without staging procedures (37 patients); cystectomy (3 patients); and bilateral salpingoophorectomy with or without uterine-sparing staging (2 patients). The remaining patients were treated with surgery

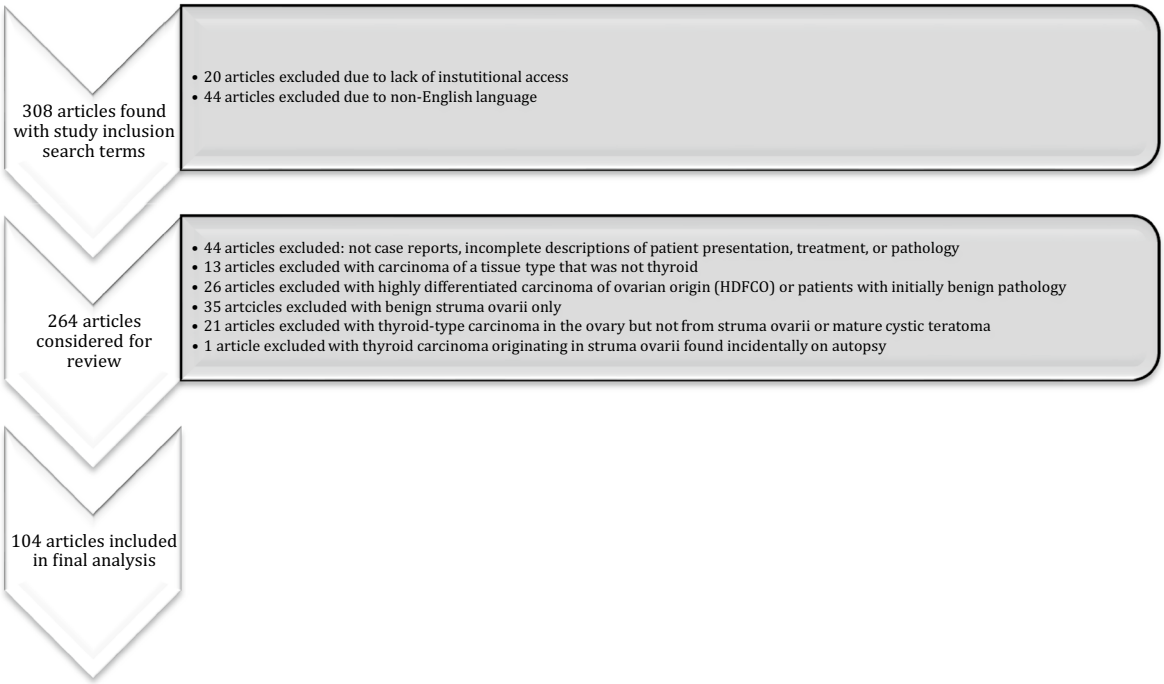

Fig. 2 Literature review inclusion process

followed by thyroidectomy alone (6 patients), surgery followed by radioiodine ablation alone (11 patients) or surgery followed by thyroidectomy and radioiodine therapy (50 patients). Four patients were treated with chemotherapy in addition to surgery and thyroid suppression. Eleven patients (5.6%) had a recurrence of carcinoma. The rest remained disease-free for the duration of study follow-up period.

Discussion

We present a case of struma ovarii with atypical features similar to papillary thyroid microcarcinoma with synchronous primary cervical papillary thyroid carcinoma and literature review on these cancers. To our knowledge, this is the largest review of thyroid-type carcinoma arising in struma ovarii or mature cystic teratoma available in current literature.

Papillary thyroid carcinoma, follicular variant of thyroid carcinoma, and follicular thyroid carcinoma appear to be the most common histologic subtypes of thyroid-type carcinoma arising in struma ovarii or mature cystic teratoma; this is consistent with prior studies on these tumors [17]. The rate of metastatic disease upon presentation found in our review (25.1%) is slightly higher than previously reported rates of metastases in thyroid-type carcinoma arising from struma ovarii or mature cystic teratoma [17, 27].

Only 12 prior case reports describe synchronous thyroid cancer arising in struma ovarii and primary cervical thyroid carcinoma [3, 6, 11, 18–26]. As with our patient and the majority of primary cervical thyroid cancers, the majority of synchronous thyroid carcinomas in our review were papillary thyroid carcinomas. Several mechanisms for synchronous carcinomas have been proposed. Activating BRAF mutations, which are commonly seen in primary cervical thyroid carcinoma, have been found in thyroid carcinomas in the ovary, suggesting a common pathway towards malignancy [28, 29]. Leong et al. suggested that a mutation event during embryogenesis may lead to loss of genetic stability, predisposing patients to multifocal synchronous cancers. They also suggest that repeated exposure to carcinogens may predispose thyroid tissue in all locations to malignant transformation [11].

Due to the rare nature of these tumors, there are no consistent guidelines for management after initial surgical diagnosis [17, 30]. Several algorithms for treatment have been proposed. Most commonly suggested are debulking surgery (with fertility sparing for those who desire future fertility) followed by total thyroidectomy and radioactive I-131 thyroid ablation; surveillance should include regular monitoring of serum thyroglobulin with or without whole-body I-131 scintigraphy [7, 29, 31, 32]. Others recommend a radioiodine body scan to evaluate for the presence

Table 1 Case reports from the literature

Study	# of cases	Average age (years)	Presenting symptoms	Pathology within struma ovarii	Metastases	Synchronous second primary?	Recurrence?
Kempers et al. [37]	5	49.8	Ascites (4), postmenopausal bleeding (1)	Follicular thyroid carcinoma (3), mixed papillary and follicular thyroid carcinoma (1), papillary thyroid carcinoma (1)	Yes (3): pelvis and chest (2); bone and liver (1)	No	Yes (2)
De Graaf et al. [38]	1	72	Dysuria, urinary frequency	Follicular thyroid carcinoma	No	No	No
Pardo-Mindan [39]	1	60	Abdominal mass	Mixed papillary and follicular carcinoma	Yes: contralateral ovary, peritoneum, omentum	No	Yes
Rosenblum [40]	1	37	Pelvic pain	Papillary thyroid carcinoma	Yes: contralateral ovary and ascites	No	No
Willemse [41]	1	36	Polyuria	Follicular thyroid carcinoma	Yes: contralateral ovary	No	Yes
Brunskill et al. [42]	1	38	Abdominal distension	Follicular variant of papillary thyroid carcinoma	No	No	No
Zakhem et al. [43]	2	41	Pelvic/abdominal pain (2), abnormal uterine bleeding (1)	Poorly differentiated follicular carcinoma (1); poorly differentiated mixed papillary and follicular carcinoma (2)	Yes (1): ascites	No	No
Devaney et al. [44]	13	50	Mass lesion, abdominal pain, hyperthyroidism	Papillary thyroid carcinoma (11); follicular thyroid carcinoma (2)	Yes (1); peritoneum	No	No
Brenner et al. [45]	1	49	Hyperhydrosis, weight loss	Follicular thyroid carcinoma	No	No	Yes
Berghella et al. [46]	1	32	Adnexal mass	Papillary thyroid carcinoma	No	No	No
Tennvall et al. [47]	1	50	Postmenopausal bleeding	Follicular thyroid carcinoma	Yes; peritoneal implants	No	No
Rose et al. [48]	1	42	Abdominal pain	Papillary thyroid carcinoma and follicular variant of papillary thyroid carcinoma (mixed)	No	No	No
Bhansali [49]	1	47	Abnormal uterine bleeding	Follicular thyroid carcinoma	No	No	No
Dardik et al. [4]	2	33.5	Abdominal pain	Follicular variant of papillary thyroid carcinoma (1); follicular thyroid carcinoma (1)	Yes (2): fallopian tubes and paraaortic lymph node (1); omental, cul-de-sac, bilateral adnexa, and hernia sac (1)	No	Yes (1)

Table 1 (continued)

Study	# of cases	Average age (years)	Presenting symptoms	Pathology within struma ovarii	Metastases	Synchronous second primary?	Recurrence?
Kano et al. [50]	1	50	Palpitations, weight loss	Follicular thyroid carcinoma	No	No	
Rotman-Pikielny et al. [51]	1	46	Abdominal pain, constipation	Follicular thyroid carcinoma	Yes; liver	No	No
Matsuda et al. [52]	1	48	Palpitations, hyperhydrosis	Papillary thyroid carcinoma	No	No	No
Kabukcuoglu et al. [53]	1	52	Abnormal uterine bleeding	Follicular variant of papillary thyroid carcinoma	No	No	No
Kostoglou-Athanassiou et al. [8]	1	38	Metorrhagia	Follicular variant of papillary thyroid carcinoma	No	No	No
Soto Moreno et al. [18]	1	31	First trimester bleeding	Papillary thyroid carcinoma	No	Yes; follicular variant of papillary thyroid carcinoma in thyroid	No
Sussman et al. [54]	1	53	Palpitations, tremors, weight loss	Follicular variant of papillary thyroid carcinoma	No	No	No
DeSimone [35]	1	32	Pelvic pain, abnormal uterine bleeding	Follicular thyroid carcinoma	No	No	No
Volpi et al. [55]	1	49	–	Papillary thyroid carcinoma	No	No	No
Griffiths et al. [56]	1	53	Postmenopausal bleeding	Papillary thyroid carcinoma	No	No	No
Makani et al. [27]	1	46	Incidental finding on gyn exam	Papillary thyroid carcinoma	No	No	No
Osmanagaoglu et al. [57]	1	45	–	Papillary thyroid carcinoma	No	No	
Zannoni et al. [58]	1	66	Fatigue, abdominal distension	Follicular variant of papillary thyroid carcinoma	No	No	No
Bolat et al. [59]	1	34	Abnormal uterine bleeding, pelvic mass	Papillary thyroid carcinoma	No	No	No
Garcia et al. [60]	1	22	Pelvic pain	Follicular thyroid carcinoma	No	No	No
Flavin et al. [61]	1	22	Pelvic pain	Papillary thyroid carcinoma	No	No	No
Mattucci et al. [62]	1	37	Pelvic pain	Follicular variant of papillary thyroid carcinoma	No	No	No
Doganay et al. [2]	1	53	Pelvic pain, abnormal uterine bleeding	Papillary thyroid carcinoma	No	No	No

Table 1 (continued)

Study	# of cases	Average age (years)	Presenting symptoms	Pathology within struma ovarii	Metastases	Synchronous second primary?	Recurrence?
Hatami et al. [63]	1	53	Abdominal pain	Papillary thyroid carcinoma	No	No	No
Janszen et al. [25]	3	39	Pelvic pain and stress incontinence (1); incidental exam finding (1); diarrhea (1)	Follicular variant of papillary thyroid carcinoma (1); follicular thyroid carcinoma (1); papillary thyroid carcinoma (1)	No	Yes (1); cervical thyroid	Yes; liver and lymph nodes (1)
Lao et al. [64]	1	36	Incidental finding on gyn exam	Follicular variant of papillary thyroid carcinoma	No	No	No
Roth et al. [12]	4	49.5	Pelvic mass (3); abnormal uterine bleeding (1)	Papillary thyroid carcinoma (3); poorly differentiated follicular-type carcinoma (1)	Yes (para-aortic lymph nodes	No	
Tomee et al. [65]	1	51	Postmenopausal bleeding	Papillary thyroid carcinoma	No	No	No
Yassa et al. [66]	1	25	Pelvic pain	Papillary thyroid carcinoma	No	No	No
Celestino et al. [67]	1	38	–	Follicular variant of papillary thyroid carcinoma	–	–	–
Garg et al. [36]	10	41.5	Incidental imaging finding (2);abdominal/pelvic pain (3); back pain (1); vaginal discharge (1); urinary frequency (1); history of complex masses (1)	Follicular variant of papillary thyroid carcinoma (6); papillary thyroid carcinoma (2); poorly differentiated carcinoma (2)	Yes (1); peritoneal implants	No	Yes (2)
Robboy et al. [68]	28	46	Abdominal swelling, pain, or mass, incidental exam or operative finding, vaginal bleeding, pathologic fracture	Follicular thyroid carcinoma (4); papillary thyroid carcinoma (20); follicular variant of papillary thyroid carcinoma (4)	Yes (17); peritoneal fluid and adhesions (13), ovarian capsule (4)	No	No
Wong et al. [69]	1	44	Weight loss; palpitations; sweating	Follicular variant of papillary thyroid carcinoma	No	No	No
Agrawal et al. [70]	1	36	Incidental finding on gyn exam	Papillary thyroid carcinoma	No	No	No
Coyne et al. [71]	1	38	Incidental imaging finding	Follicular variant of papillary thyroid carcinoma	No	No	No
Marcy et al. [72]	1	45	Abdominal pain	Follicular variant of papillary thyroid carcinoma	No	No	Yes; liver and pelvis

Table 1 (continued)

Study	# of cases	Average age (years)	Presenting symptoms	Pathology within struma ovarii	Metastases	Synchronous second primary?	Recurrence?
Roth et al. [73][a]	6	43	–	Follicular variant of papillary thyroid carcinoma (2); solid variant of papillary thyroid carcinoma (2); Follicular thyroid carcinoma (1), poorly differentiated carcinoma (1)	No	No	No
Salman et al. [74]	1	67	Abdominal distension	Papillary thyroid carcinoma	No	No	No
Sibio et al. [75]	1	74	Abdominal pain	Papillary thyroid carcinoma	Yes; peritoneal implants	No	No
Yucesoy et al. [76]	1	40	Pelvic pain	Papillary thyroid carcinoma	No	No	No
Alvarez et al. [77]	1	21	Incidental imaging finding	Papillary thyroid carcinoma	No	No	No
Kraemer et al. [15]	1	40	Incidental imaging finding	Papillary thyroid carcinoma	No	No	No
Menon et al. [78]	1	46	Abdominal pain	Follicular variant of papillary thyroid carcinoma	No	No	No
Tanaka et al. [79]	1	50	Incidental finding on gyn exam	Follicular variant of papillary thyroid carcinoma	No	No	No
Barrera et al. [80]	1	41	Abdominal pain, abdominal mass, vomiting	Papillary thyroid carcinoma	No	No	No
Collins et al. [81]	1	64	Adnexal mass; treated for adenocarcinoma for 3 years before path re-review	Papillary thyroid carcinoma	No	No	No
Gunasekaran et al. [82]	1	40	Abdominal distension	Follicular thyroid carcinoma	No	No	No
Hinshaw et al. [30]	1	74	Incidental imaging finding	Well differentiated strumal carcinoid, well differentiated follicular variant of papillary thyroid carcinoma, moderately differentiated mucinous adenocarcinoma arising in intestinal type mucinous cystadenoma	No	No	No

Table 1 (continued)

Study	# of cases	Average age (years)	Presenting symptoms	Pathology within struma ovarii	Metastases	Synchronous second primary?	Recurrence?
Jean et al. [7]	1	60	Incidental imaging finding	Papillary thyroid carcinoma	Yes; peritoneal washings	No	No
Lee et al. [83]	1	35	Incidental imaging finding	Follicular thyroid carcinoma	Yes; liver and peritoneum	No	No
Marti et al. [19]	4	44	Incidental finding on gyn exam (2); incidental imaging finding (1); abdominal pain (1)	Papillary thyroid carcinoma (3), follicular thyroid carcinoma (1)	No	Yes (1); papillary thyroid carcinoma in thyroid	No
Meringolo et al. [84]	1	62	Abdominal pain	Papillary thyroid carcinoma	No	No	No
Selvaggi et al. [85]	1	50	Incidental imaging finding	Follicular thyroid carcinoma	No	No	No
Shrimali et al. [32]	6	52	Abnormal uterine bleeding (3); dyspepsia (1); renal infection (1); abdominal distension (1)	Follicular thyroid carcinoma (3); papillary thyroid carcinoma (3)	No	No	No
Stanojevic et al. [86]	1	35	Incidental imaging finding	Follicular variant of papillary thyroid carcinoma	No	No	No
Krishnamurthy et al. [20]	1	51	Abdominal pain, distension	Follicular variant of papillary thyroid carcinoma	No	Yes; follicular variant of papillary thyroid carcinoma in thyroid	No
Leite et al. [21]	1	78	Pelvic pain, weight loss	Papillary thyroid carcinoma	No	Yes; follicular variant of papillary thyroid carcinoma in thyroid	
Leong et al. [11]	1	42	Enlarging abdominal mass	Papillary thyroid carcinoma	Yes; peritoneal washings	Yes; multifocal papillary thyroid carcinoma with perithyroidal lymph node metastases	No
Mardi et al. [87]	1	40	Pelvic pain	Tan-cell variant of papillary thyroid carcinoma	No	No	No
Matysiak-Grzes et al. [88]	1	20	Incidental imaging finding	Papillary thyroid carcinoma	No	No	No
Steinman et al. [34]	2	26.5	Abdominal pain (1); ovarian torsion (1)	Papillary thyroid carcinoma (1); follicular variant of papillary thyroid carcinoma (1)	Yes (1); bony pelvis	No	Yes (1)
Gonzalez Aguilera et al. [3]	3	51	Incidental imaging finding (3)	Papillary thyroid carcinoma (2); follicular thyroid carcinoma (1)	No	Yes (1); papillary microcarcinoma of thyroid	

Table 1 (continued)

Study	# of cases	Average age (years)	Presenting symptoms	Pathology within struma ovarii	Metastases	Synchronous second primary?	Recurrence?
Karagkounis et al. [89]	1	36	Amenorrhea	Tan-cell variant of papillary thyroid carcinoma	No	No	No
Kumar et al. [90]	1	52	Abdominal distension	Follicular variant of papillary thyroid carcinoma	No	No	No
Luo et al. [91]	1	46	Pelvic pain	Papillary thyroid carcinoma	Yes; contralateral ovary, uterus, omentum, nitestinal wall, bilateral oviduct, bilateral pelvic sidewall, pelvic lymph node	No	No
Ukita et al. [92]	1	45	Rib pain	Follicular thyroid carcinoma	Yes; rib bone, lungs	No	Yes
Brusca et al. [6]	1	30	Abdominal pain	Follicular variant of papillary thyroid carcinoma	No	Yes; papillary microcarcinoma of thyroid	No
Khunamornpong et al. [5]	1	22	Abdominal distension	Poorly differentiated thyroid carcinoma	No	No	No
Monti et al. [93]	1	43	Pelvic pain	Follicular variant of papillary thyroid carcinoma	No	No	No
Riaz et al. [94]	1	21	Abdominal distension	Follicular thyroid carcinoma	Yes; omentum and peritoneal nodules	No	No
Srbovan et al. [95]	1	62	Vaginal bleeding	Papillary thyroid carcinoma	No	No	No
Tan et al. [1]	3	42	–	Papillary thyroid carcinoma (3)	No	No	
Wei et al. [96]	15	46	–	Papillary thyroid carcinoma (10; follicular variant PTC in 8 cases), well-differentiated neuroendocrine stromal carcinoid (5)	Yes (1): round ligament	No	Yes (1)
Anagnostou et al. [97]	1	64	Abdominal pain, distension	Follicular thyroid carcinoma, clear cell variant	Yes: pouch of Douglas	No	No
Fukunaga et al. [98]	1	65	Abdominal pain	Anaplastic carcinoma	No	No	No
Lara et al. [99]	1	36	Abdominal pain	Papillary thyroid carcinoma	No	No	
Oudoux et al. [10]	2	58	Incidental finding on gyn exam (1), postmenopausal bleeding (1)	Follicular variant of papillary thyroid carcinoma (1); papillary carcinoma (1)	Yes (1): retrocaval lymph node, omental nodule	No	No

Table 1 (continued)

Study	# of cases	Average age (years)	Presenting symptoms	Pathology within struma ovarii	Metastases	Synchronous second primary?	Recurrence?
Williams et al. [100]	1	61	Abdominal pain	Insular carcinoma (poorly differentiated carcinoma) with adjacent papillary thyroid carcinoma	Yes: uterine serosa, cervix	No	No
Zhu et al. [101]	1	40	Incidental imaging finding	Papillary thyroid carcinoma	Yes; omentum	No	No
Boyd et al. [23]	1	30	RLQ pain	Papillary thyroid carcinoma	No	Yes; papillary thyroid carcinoma in thyroid	No
Gobitti et al. [9]	1	36	Incidental finding on gyn exam	Follicular variant of papillary thyroid carcinoma	Yes; uterus, parametria, peritoneum, pararectal, periappendiceal, perivesical, liver parenchyma	No	No
Llueca et al. [102]	1	43	Incidental finding on gyn exam	Papillary thyroid carcinoma	No	No	No
Middelbeek et al. [24]	1	55	Persistent elevated thyroglobulin after thyroidectomy	Papillary thyroid carcinoma	No	Yes; follicular variant of papillary thyroid carcinoma in thyroid	No
Pineyro et al. [103]	1	34	Abdominal pain	Follicular variant of papillary thyroid carcinoma	No	No	No
Al Hassan et al. [104]	1	42	Abdominal pain	Papillary thyroid carcinoma	Yes; contralateral ovary	No	No
Gomes-Lima et al. [26]	1	62	Incidental imaging finding	Papillary thyroid carcinoma	No	Yes; cervical thyroid	No
Iranparvar et al. [105]	1	10	Tachycardia	Papillary thyroid carcinoma	No	No	No
Lager et al. [106]	1	30	Left lower quadrant pain	Papillary thyroid carcinoma	Yes; bone and lungs	No	No
Moayerifar et al. [107]	1	11	Palpitation, abdominal pain	Follicular variant of papillary thyroid carcinoma	No	No	No
Wu et al. [108]	1	48	Constipation, hematochezia	Papillary thyroid carcinoma	Yes; rectum, liver, peritoneum	No	No
Yasutake et al. [109]	1	66	Incidental imaging finding	Papillary thyroid carcinoma	No	No	No
Seifert et al. [110]	1	67	–	Follicular variant of papillary thyroid carcinoma	Yes; bone	No	No

[a]Excludes eight cases previously reported in other studies quoted in this review and one case of adenoma-like follicular neoplasm

of metastatic disease after surgery or thyroid imaging to evaluate for suspicious lesions [15, 33]. Another option is to stratify patients based on risk; those that have tumors less than 2 cm that are confined to the struma ovarii with well-differentiated histology are offered thyroid suppression with thyroxine while those with higher risk disease are recommended to undergo thyroidectomy and ablation [30]. Those with metastatic disease or disease recurrence have been treated with chemotherapy and radiation [34].

Recurrence rates were low in our sample; previous reports have described recurrence rates from 7.5 to 38% [19, 23, 30, 35]. This may be due to the fact that prior studies included cases of patients with HDFCO or originally benign struma ovarii on ovarian pathology who later presented with metastatic disease, whereas we excluded these cases and included only those who had malignant pathology at initial diagnosis [36]. An initial diagnosis of malignancy would prompt further staging, treatment, and surveillance that may lead to fewer recurrences than an initial benign diagnosis or a missed initial diagnosis of malignancy.

While thyroid-type carcinomas arising in struma ovarii or mature cystic teratoma are rare, the high rate of metastatic disease and recurrence prompts thorough evaluation and close surveillance of patients after initial diagnosis and treatment. Struma ovarii cases with subtle cytologic features of papillary thyroid carcinoma, including cases such as ours in which the findings did not clearly warrant an outright diagnosis of thyroid carcinoma, should still be addressed [36]. The pathologist should communicate atypical features to clinician colleagues. Given the high rate (6.2%) of synchronous cervical thyroid carcinoma, a clinical evaluation and ideally ultrasound imaging of the thyroid in patients with even subtle features of thyroid-type carcinoma arising from struma ovarii or mature cystic teratoma is warranted.

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