CASE REPORT

Struma ovarii recurrence with peritoneal strumosis: A case report

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Abstract

Background: Malignant transformation of thyroid tissue in struma ovarii and metastasis are uncommon. Benign thyroid tissue may spread to the peritoneal cavity in rare cases, and pathologic examination of the peritoneal implants shows multiple nodules of varying sizes of mature thyroid tissue similar to struma ovarii. This condition is termed “peritoneal strumosis”. It is very rare and therefore, both the criteria for diagnosis and its management are under discussion. The recommended treatment of metastatic struma ovarii should contain, in addition to local resection of the extraovarian tumor, total thyroidectomy in conjunction with radioiodine scanning and radioiodine ablation.

Case report: A 38 year-old woman had presented with benign struma ovarii 12 years previously. She was referred to our department because of a right ovarian tumor. After confirming the diagnoses of metastatic recurrent benign struma ovarii, we performed debulking surgery with the aim of resecting all tumor masses. The uterus, left adnexa, and pelvic peritoneum were surgically extracted “en bloc”. No macroscopic tumor was left post-operatively. The first post-operative ^{123}I scintography with15 MBq iodine-123 one month post-operatively showed a small but suspicious focus in the front left perivesical pelvic area. In the ventral visual presentation multiple discrete foci, located in right perivesical, supravesical and in the ventral upper abdomen, were seen. We then referred the patient for thyroidectomy and radioiodine ablation. The following periodic follow-ups of the patient did not show any signs of recurrence or progression of the disease.

Conclusion: Patient with benign metastatic struma ovarii (also known as highly differentiated follicular carcinoma of ovarian origin - HDFCO) may affectively be treated with local resection of the extraovarian tumor and subsequent thyroidectomy followed by radioactive iodine ablation.

Key words

Struma ovarii, Highly differentiated follicular carcinoma of ovarian origin, Peritoneal strumosis, Radioiodine ablation, Extraovarian tumor

1 Introduction

Twenty percent of ovarian tumors are teratomas, and up to 20% of teratomas contain thyroid tissue. Struma ovarii consisting of at least 50% of thyroid tissue [1] is less common and accounts for 1-4% of benign ovarian teratomas. Bilateral struma ovarii is very rare and has not been reported frequently [2]. Struma ovarii usually shows up as a pelvic mass and presents as pseudo-Meigs' syndrome in about 5% of cases [3]. However, preoperative diagnosis of struma ovarii is rarely reported and may occur in patients with symptoms of hyperthyroidism but this association is observed in less than 5% of cases [4].

Malignant transformation of thyroid tissue in struma ovarii and metastasis is uncommon. Benign thyroid tissue may spread to the peritoneal cavity in rare cases, and pathologic examination of the peritoneal implants shows multiple nodules of varying sizes of mature thyroid tissue similar to struma ovarii [5]. This condition is termed “peritoneal strumosis”. The differential diagnosis of peritoneal dissemination of struma includes highly differentiated follicular carcinoma (HDFCO) [6] or typical thyroid carcinoma metastatic to the ovary, which has also been reported [7]. Because of its harmless histological appearance, this form of follicular carcinoma cannot be diagnosed until the neoplasm spreads beyond the ovary thus showing evidence of aggressive behavior [6]. Histological features of thyroid carcinoma are found in 5–37% of struma ovarii (referred to as malignant struma ovarii). The nuclear features of papillary carcinoma are usually used as the criteria for diagnosis of malignant struma ovarii, and most cases reported are papillary carcinoma [8, 9, 10]. Immunohistochemical staining with HBME-1 (Hector Battifora mesothelial [cell] 1) and galectin-3, often expressed in papillary thyroid carcinoma, can also help to confirm the diagnosis [11].

2 Case report

A 38 year-old woman with korean ancestry, para II, gravida II was referred to our department with a pelvic mass. She presented with a good performance status but had right lower abdominal pain. She had a regular menstrual cycle, her history revealed two unremarkable cesarean sections. Medical history revealed that she had laparoscopic right ovarian cystectomy for mature teratoma with benign struma ovarii 12 years previously. The histological report was available and reviewed. The patient did not receive any follow-up for the struma ovarii after her initial operation and levels of thyroid hormone were not reported.

On pelvic examination the vulva, vagina, portio and uterus showed no abnormality, but a 5 cm mobile mass in the Douglas pouch was diagnosed. An ultrasonographic study showed the right ovary (55 × 42 mm) was partly cystic with many septums and weak vascularisation. Free fluid was not detectable. Laboratory testing revealed normal levels of CA 125, CEA, β-HCG, and α-FP; with the exception of thyroglobulin all thyroid parameters were normal: TSH was 1.17 μIU/mL (normal range: 0.27-4.2 μIU/mL), FT4=1.20 ng/dL (0.93-1.7 ng/dL), FT3 = 2.64 pg/mL (2.04-4.4 pg/mL), Thyroglobulin=108.2 ng/mL (1.4-78.0 ng/mL), Thyroglobulin-antibodies=12.87 IU/mL (< 115 IU/mL). Computed tomography scan study reported a hyperdense irregular lesions in the right parametrium (4.3 × 4.6 cm) and in the pelvis (5.1 × 4 cm partly nodular, partly confluent), with a strong suspicion of infiltration to the rectum and with multiple nodules in the pelvic cavity. Ascites was not reported. Chest x-ray showed no pleural effusion or any metastases.

We performed a diagnostic laparoscopy and observed that the liver, diaphragm, stomach, transverse colon, and the right para-colon gutter appeared normal. The greater omentum showed some isolated white cherry-size nodules. Multiple cherry-sized nodes were detected in the area of the small pelvis and the left para-colon gutter, in the great omentum as well as on the bladder's peritoneum and the peritoneum of the small pelvis. In addition, a large tumor with a diameter of 5cm was detected on the right pelvic wall, which adhered to the caecum. Another tumor with a diameter of 7cm and with a relatively smooth surface texture was located in the Douglas pouch and was attached to the rectum. Samples of the right ovarian mass and peritoneum surfaces were taken for histological examination.

Histological diagnosis reported a benign appearing struma ovarii (Figure 1). Therefore, debulking surgery was planned taking into account the extent of surgery necessary to remove all visible tumors. The patient received a sagittal laparotomy. Intraoperatively, we found no ascites, the greater omentum showed knotty tumors with diameters up to 2 cm, the right diaphragm showed numerous knotty tumors of 2 cm thickness; the para-colon gutter, left diaphragm, liver, spleen, gall bladder, and lesser omentum were inconspicuous. Further tumors up to 1cm diameter were found in the iliocoecal region and the mesentery of the appendix. A conglomeration of the uterus, left adnexa and tumor cysts adherent to rectum and sigma were identified in the pelvis. All tumor masses, the uterus, left adnexa, and pelvic peritoneum were surgically resected “en bloc”. No macroscopic tumor was left post-operatively.

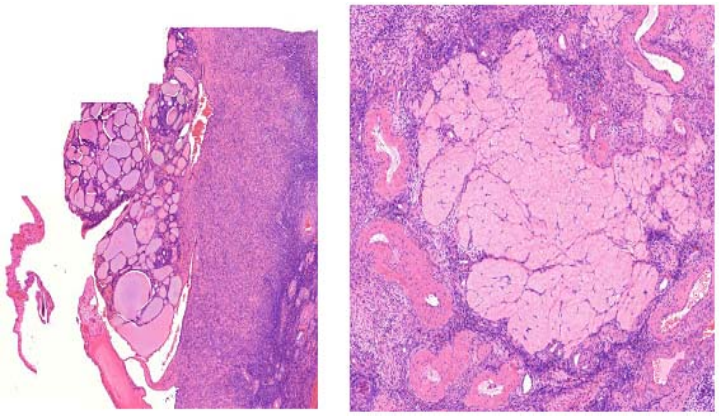

Figure 1. Presence of mature thyroid tissue in multiple specimen sections

The final pathological evaluation revealed struma ovarii without any evidence of malignancy in both the right adnexa and the tumor nodules from all over the abdomen and pelvis. The histological examination of the left adnexa did not show any abnormality.

The first post-operative ^{123}I scintography with 15 MBq iodine-123 one month post-operatively showed a small but suspicious focus in the front left perivesical pelvic area. In the ventral visual presentation multiple discrete foci, located in right perivesical, supravesical and in the ventral upper abdomen, were seen. The different postoperative possibilities of treatment were discussed with the patient in detail and she decided not to follow our recommendation of thyroidectomy and radioiodine ablation and preferred to undergo regular screening for recurrence and for thyroid function including scintigraphies. A radioactive iodine uptake test with12 MBq iodine-123 8 months post-operatively showed three side-by-side foci in the pelvic area located cranial of the bladder. These findings showed increased activity compared to the post-operative investigation.

At that point, the patient agreed to have a total thyroidectomy followed by radioiodine ablation which was performed in our institution one year after debulking surgery. The first screening afterwards included thyroid scintigraphy with 107.6 MBq Tc-99m and was performed two weeks after thyreoidectomy. The ablative radiotherapy was administered one month later.

The periodic follow-ups of the patient over 3 years have not shown any signs of recurrence or progression of the disease.

3 Discussion

Metastatic struma ovarii or peritoneal strumosis is very rare and therefore, both the criteria for diagnosis and the management are under discussion. The knowledge about the course of malignant struma ovarii is based on single case

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reports and small case series [1]. A review of 39 patients with malignant struma ovarii showed only 9 patients (23%) with metastasis. The predominant sites were adjacent pelvic structures including the contralateral ovary, but some patients had distant metastases to the lungs, bone, liver, and brain [12]. Roth et al. had analysed the relationship of HDFCO and cases reported as peritoneal strumosis and finally doubted the existence of the latter as a distinct clinicopathologic entity. Therefore, this group recommended an approach similar to our strategy: they suggest both local resection of the extraovarian tumor and subsequent thyroidectomy followed by radioactive iodine ablation for these patients [6]. McGill et al. performed a review on 40 patients with struma ovarii and concluded also that the recommended treatment of metastatic struma ovarii should contain total thyroidectomy in conjunction with radioiodine scanning and radioiodine ablation [13]; another two case reports came to similar conclusions [14, 15].

Follow up may include the use of thyroglobulin as a tumor marker, and diagnostic radioiodine scans may be performed to screen for residual or recurrent disease [16]. Repeat ablative radioiodine treatment may still have a curative potential or at least can provide extended disease free survival. Recombinant human TSH (rhTSH) is occasionally used before ^{131}I administration to achieve a concentration of ^{131}I activity in the tumour high enough for a significant cytotoxic effect, because ^{131}I uptake by most of thyroid carcinomas is quite low in absence of high levels of endogenous TSH [17, 18]. Follow-up may last for at least 15 years; Zekri et al. had described a patient with struma ovarii that was initially diagnosed as benign and presented 10 years later with distant metastases [19]. In addition, our patient presented with recurrence and peritoneal metastasis of a benign struma ovarii 12 years after the first diagnosis.

Conflict of interests

The authors declare that there are no conflicts of interest.

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