

Лучевые методы в диагностике первичной и рецидивной злокачественной струмы яичников: клинический случай

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АННОТАЦИЯ

Представлено редкое клинико-диагностическое наблюдение первичной и рецидивной злокачественной струмы яичников.

Злокачественная струма правого яичника выявлена через 2 года после хирургического лечения первичной доброкачественной струмы левого яичника. Спустя полгода у пациентки выявлен рецидив заболевания, визуализируемый исключительно по данным радиоизотопных методов исследования. Визуализация рецидивных очагов по брюшине на ультразвуковом исследовании была отмечена на четвёртом году противоопухолевого лечения. По данным ультразвукового исследования, по брюшине малого таза на всём протяжении визуализировались множественные солидные опухолевые очаги изо-гипоэхогенной структуры с наличием локусов низко- и умеренно скоростного кровотока даже в мелких очагах: скорость кровотока (peak systolic velocity, PS) была в диапазоне от 2 до 9 см/сек, максимальный индекс сосудистого сопротивления (resistivity index, RI max) — 0,53. На протяжении 4 лет пациентке проводилась радиойодтерапия ¹³¹ I активностью 6,0 ГБк. Состояние пациентки на фоне проводимого лечения удовлетворительное.

Ключевые слова: ультразвуковое исследование; трёхмерная ангиография; ультразвуковая томография; энергетическое допплеровское картирование; МРТ; рецидив; злокачественная струма яичника; перитонеальный струмоз.

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Radiation methods in the diagnosis of primary and recurrent malignant ovarian struma: A case report

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ABSTRACT

We provide a rare clinical and diagnostic observation of primary and recurring malignant ovarian struma.

Malignant struma of the right ovary was detected 2 years after surgical treatment of primary benign struma of the left ovary. Six months later, the patient was diagnosed with a disease relapse, visualized exclusively according to radioisotope research methods. In the fourth year of anticancer treatment, ultrasonography revealed recurring foci along the peritoneum. According to the ultrasound data on the pelvic peritoneum and the projection of the removed right ovary, multiple solid nodes with high blood flow were visualized. Peak systolic velocity ranged from 2 to 9 cm/s in minor lesions from 4 to 12 mm, with an RI max of 0.53. For 4 years, the patient underwent radioiodine therapy with ¹³¹I with an activity of 6.0 GBq; the patient's condition during the treatment was satisfactory.

Keywords: 3D angiography; magnetic resonance imaging; malignant struma ovarii; peritoneal strumosis; power Doppler mapping; relapse; ultrasound; ultrasound tomography.

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放射治疗方法在原发性和复发性恶性卵巢甲状腺肿诊 断中的应用:临床病例

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简评

本文介绍了一个罕见的原发性和复发性恶性卵巢甲状腺肿的临床诊断观察。

右侧卵巢的恶性甲状腺肿是在左侧卵巢的原发性良性甲状腺肿手术治疗2年后发现的。6个 月后,患者出现了只有放射性同位素技术数据才能观察到的疾病复发。在抗肿瘤治疗的第 四年,超声检查发现了复发的腹膜病灶。根据超声检查,在全体盆腔腹膜上可以看到多个 等渗及低回声结构的实体肿瘤病灶,存在低到中等速度的血流病灶,即使是小病灶:血流 速度(peak systolic velocity, PS)在2到9cm/s之间,最大血管阻力指数(resistivity index, RI max)为0.53。患者曾接受放射性碘治疗¹³¹I,活性为6.0GBq,治疗了4年。在治疗 的背景下,患者的情况是令人满意的。

关键词: 超声检查; 三维血管造影; 超声层析成像; 能量多普勒成像; MRI; 复发; 恶性卵 巢甲状腺肿; 腹膜甲状腺肿。

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INTRODUCTION

Malignant struma ovarii is a rare disease referred as monodermal teratomas and somatic-type tumors (WHO Classification of Tumours, 2020) [1].

Major contributions to the knowledge about this condition were made in the late 19th century. Richard Böttlin was the first to describe thyroid tissue in the ovary in 1889, whereas struma ovarii was first characterized by Clemens von Kalden in 1895. The term "struma ovarii colloides" came into common use thanks to Julius Robert von Meyer. At about the same time, Ludwig Pick supposed that thyroid elements in "ovarian goiter" may be transformed. Over time, the whole range of thyroid disorders was characterized for struma ovarii, including nodular goiter, toxic goiter, auto-immune thyroiditis, adenoma, and carcinoma [2, 3].

Moreover, 90%–95% of struma ovarii cases are benign, and carcinoma is very rare in its setting. Available clinical and diagnostic data on struma ovarii is primarily descriptive [3–11].

The timely diagnosis of struma ovarii plays a key role in patients of reproductive age who plan to preserve fertility because the extent of surgical treatment varies based on the histological subtype of this condition. Although laparoscopic oophorectomy or ovarian resection may be considered for benign struma ovarii, malignant struma ovarii may require the removal of the uterus and its appendages and resection of the greater omentum [8]. A study showed that the average time from the initial diagnosis of struma ovarii (both benign and malignant) to advanced malignancy ranged from 2 to 9 years [2].

Considering that ovarian goiter is a neoplasm rather than hypertrophy of the ovarian stroma, as it is the case in the thyroid gland, its dissemination and metastatic potential to other organs support the point made in the latest 2020 World Health Organization Classification of Female Genital Tumors: "Peritoneal implants well-differentiated thyroid tissue in a patient with a histologically benign struma ovarii, known as "peritoneal strumosis," is currently considered to be a metastasis of a well-differentiated follicular carcinoma originating in the struma ovarii" [1, 12]. The 5-year survival rate of patients treated with radioactive iodine (iodine-131, ¹³¹I) was higher than that of untreated patients (94.9 vs. 64.8%) [4, 10].

Medical database (PubMed and Medline) search identified studies of ultrasound (US) or magnetic resonance imaging (MRI) patterns of malignant struma ovarii and recurrent peritoneal strumosis; however, descriptions of the disease and its perfusion features are still unclear, which was the reason for publishing this study.

CASE REPORT

Patient

This study presents the features of a case of T3cN0M0 primary recurrent malignant struma ovarii in patient R. aged 25 years.

One month after childbirth, the patient underwent a left oophorectomy for a benign struma of the left ovary and right ovarian resection for a dermoid cyst of the right ovary. She was event-free for 2 years. In 2015, diagnostic laparoscopy and biopsy of peritoneal lesions were performed for ascites and ovarian enlargement in another medical institution. The Federal State Budgetary Institution "Russian Research Center for Radiology" of the Ministry of Health of Russia (RRCR) reviewed the tissue specimen slides and revealed poorly differentiated malignant struma ovarii. In blood tests performed in 2015, thyroglobulin level was 35.156 (normal, 0-50) ng/mL; anti-thyroglobulin antibodies, 0.52 U/mL; thyroid-stimulating hormone, 0.76 ng/mL (normal, <0.2); thyroxine, 0.69 pmol/L; CA-125, 1,339.5 U/mL; human chorionic gonadotropin, 1.2 mIU/mL; and alpha fetoprotein, 2.25 IU/mL.

Investigations

After diagnostic laparoscopy and biopsy of lesions in the pelvic peritoneum performed in another medical institution, complex ultrasonography was performed at the RRCR. An inhomogeneous isohypoechoic mass with areas of an anechoic structure of irregular and rounded shape was found in the projection of the right appendages. Pronounced pathological blood flow in the solid component of the mass, ascites, and carcinomatosis of the peritoneum of the small pelvis were also found. Figure 1 shows a 3D angiogram of the primary malignant struma ovarii on the right ovary.

eSaote Pro machine US was used; the transvaginal transducer and volume transvaginal transducer were used. Ultrasonography was performed in a standard 2D mode, and high-tech US methods were then used: power Doppler mapping, US computed tomography (CT), and 3D angiography.

Figure 2 shows uneven thickening of the pelvic peritoneum covering the anterior wall of the uterus. The structure of the blastomatously altered peritoneum of the small pelvis is isohypoechoic, with ill-defined edges on the interface with the body of the uterus.

The intensity of the blood flow in the tumor lesion in the retrouterine pouch is remarkable, as well as its smoothness in the thickened (up to 5–7 mm) peritoneum of the small pelvis (Fig. 3).

In addition to US, a multiparametric MRI of the pelvic organs was performed using a high-field tomograph with a magnetic field strength of 1.5 Tesla. The MRI examination was in line with the guidelines of the European Society of Urogenital Radiology (ESUR Guidelines, 2019) and included several modes: T2-weighted imaging (T2-WI), T1-weighted imaging (T1-WI), diffusion-weighted imaging (DWI; b = 800, b = 1,400), and T1 with dynamic contrast enhancement [13].

The MRI of the malignant struma ovarii demonstrated a space-occupying predominantly solid mass of the right ovary with uneven poorly defined bumpy edges, areas of multiple cystic inclusions ranging in size from 0.7 to 3 cm with a heterogeneously increased MR signal in T1-WI,



Fig. 1. 3D angiography of a poorly differentiated malignant struma ovarii on the right (arrows).



Fig. 2. Dopplerography in the energy mode. Malignant struma ovarii. Carcinomatosis of the pelvic peritoneum anterior to the uterus (arrow). The visualized tumor lesion is 6 mm thick.

heterogeneous (from hyper- to hypo-) MR signal in T2-WI, which was consistent with colloid nodules with highprotein contents of different viscosities. Solid areas of the tumor were characterized by different rates of diffusion restriction, from 0.7 to 1.4 mm²/s \times 10⁻³. Multiple nodular masses along the peritoneum of the small pelvis and diffuse thickening of the peritoneum were also detected. The paramagnetic accumulation in the tumor node along the peritoneum and nodes of the peritoneum was sharply increased in T1-WI, with suppression of the signal from the adipose tissue (Fig. 4). Meanwhile, sagittal postcontrast images clearly show the absence of paramagnetic accumulation in the colloid nodules of the malignant struma of the right ovary (Fig. 4f).

Three months after the extirpation of the uterus with the right appendages, omentectomy, and thyroidectomy, the US showed 5–10 mL of free fluid in the small pelvis. Based on the comprehensive US findings, the free fluid persisted for 4 years despite ongoing radioiodine therapy (¹³¹I; 6.0 GBq; a total of 11 cycles); however, no lesions were detected. The



Fig. 3. 3D angiography of the malignant struma lesion along the peritoneum in the retrouterine space (arrow).

first recurrent lesion of the primary tumor along the pelvic peritoneum, visualized by US, appeared 4 years after the start of the combination therapy.

US demonstrated that the recurrent lesion of malignant struma ovarii along the pelvic peritoneum, identified in the presence of ascites, had an iso-hypoechoic structure with



Fig. 4. Pelvic magnetic resonance images of patient R. with malignant struma of the right ovary: (*a*) T2-FS-WI in the axial plane; (*b*) T1-WI in the axial plane; (*c*) diffusion-weighted imaging (b = 1,000); (*d*) apparent diffusion coefficient map; (*e*) T1-FS-WI + contrast in the axial plane; (*f*) T1-FS-WI + contrast in the sagittal plane. The solid arrow indicates colloid nodules in the malignant struma of the right ovary. The dotted arrow shows lesions along the pelvic peritoneum with increased paramagnetic accumulation and restricted diffusion similar to the solid component of the primary tumor.

fuzzy contours and small size (4–12 mm). The peritoneum of the small pelvis outside the lesion was <4 mm thick; however, 3D angiography and US-CT revealed that even small blastomatous lesions were well vascularized. Blood flow was also present in the 4–5 mm thick structure of the pelvic peritoneum (Figs. 5–7).

To identify specific US signs of malignant struma ovarii, recurrent tumor lesions along the peritoneum of the malignant struma in one patient were compared with recurrent lesions of serous ovarian adenocarcinoma in 12 patients. When comparing the tumor lesions in the retrouterine space, a more pronounced neoangiogenesis was noted in the tumor lesion of the malignant struma ovarii, where the peak systolic velocity (PS) was recorded in small lesions (4–12 mm) and ranged from 2 to 9 cm/s. The maximum vascular resistivity index (RI max) was 0.53. No blood flow was noted in recurrent lesions along the pelvic peritoneum of the serous adenocarcinoma up to 9 mm, and in lesions up to 15–20 mm, PS varied from 2 to 4 cm/s or was <2 cm/s (Figs. 5 and 8).



Fig. 5. Dopplerography in the energy mode. The arrows show recurrent lesions of the malignant struma ovarii.



Fig. 6. US-CT of the recurrent lesions of the malignant struma ovarii.



Fig. 7. 3D angiography of the recurrent lesions of the malignant struma ovarii in the pelvic peritoneum in the presence of ascites.



Fig. 8. Dopplerography (energy mode) of the tumor lesion along the peritoneum in the retrouterine space in a patient with stage IIIC serous ovarian cancer.

Thus, this case demonstrated that the combined MRI findings of the thyroid tissue (colloid nodules), restricted diffusion areas, and increased paramagnetic accumulation in its solid component may indicate a malignant struma ovarii. Recurrent lesions of malignant struma, even small ones (up to 4–5 mm), are well vascularized. Malignant struma of the right ovary was detected 2 years after surgical treatment of primary benign struma of the left ovary.

The somatic condition of the patient was not aggravated by ascites and persistent recurrent tumor lesions in the small pelvis.

DISCUSSION

In this study, we analyzed US and MRI signs of primary recurrent malignant struma ovarii. Our data are generally

consistent with those of individual case reports of this neoplasm in the literature. US visualizes a malignant struma ovarii as a multicystic tumor with irregular septa and heterogeneous echogenic solid components. Complex US (power Doppler mapping, US-CT, and 3D angiography) show pronounced neoangiogenesis in both primary malignant struma ovarii and its small recurrent lesions. Its characteristic feature is the so-called struma pearls, i.e., well-defined rounded solid areas, which are consistent with the colloidrich thyroid tissue [8, 14–17].

In the presented case, the tumor did not show typical MRI signs for a malignant struma ovarii; however, the MRI pattern allows suspicion of the inclusions of colloid nodules in the thyroid tissue of the solid component of the ovarian tumor. Varying degrees of restricted diffusion were observed in the solid tumor component and lesions along

the pelvic peritoneum. Extremely pronounced paramagnetic accumulation in the solid component of the tumor visualized in the post-contrast series and the absence of accumulation in the colloid nodules were pathognomonic. In the study by Gil et al. [14], MR images revealed struma ovarii as multicystic tumors with solid components, often with high T1-WI signal intensity, whereas cysts had different T2-WI signal intensities depending on fluid viscosity. Tamura et al. [17] analyzed 18 cases of struma ovarii, and their MRI study revealed a solid contrast-accumulating area that corresponded to a malignant variant of the tumor only in 54% of cases; meanwhile, restricted diffusion was established only in 11% of cases.

In the present study, restricted diffusion was heterogeneous but indicated malignant features, given the perfusion parameters in the tumor. Data similar to ours were reported by Yamauchi et al. [7]: the solid component in the malignant struma ovarii was characterized by the increased MR signal in diffusion-weighted imaging and a reduced signal in apparent diffusion coefficient maps. Thus, when compared with a pathomorphological study, the areas of the papillary carcinoma in struma ovarii corresponded to the area of true diffusion restriction in MRI.

Herein, we compared recurrent lesions of malignant struma and serous ovarian cancer. The recurrent lesions of malignant struma were similar in structure to serous ovarian cancer and had an isohypoechoic structure; however, US angiography and US-CT did not record blood flow in 4-5 mm lesions of the serous ovarian cancer. In this case, we did not register blood flow in the 4-mm lesion of serous ovarian cancer in the pelvic peritoneum. Moreover, Ranade et al. [19] reported peritoneal strumosis (struma peritonei) by US in a patient 6 years after surgical treatment of stroma ovarii, and the lesion had a mixed structure with calcifications.

Brogsitter et al. [20] reported a case of peritoneal strumosis and highlighted the role of histological examination of conditions similar to those of ovarian carcinoma. Many aspects of peritoneal strumosis remain unexplored; however, the authors suggest a connection with either rupture of mature ovarian teratoma or ovarian resection for mature ovarian teratoma. In addition, the authors noted a "slow" course of

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CONCLUSION

Preoperative diagnosis of malignant struma ovarii is quite challenging. Although struma ovarii is difficult to differentiate from ovarian adenocarcinoma, US and MRI may be instrumental for the differential diagnosis.

In summary, the combination of the thyroid tissue and areas of restricted diffusion in the solid tumor component visualized in MRI may be a sign of malignant stroma ovarii. Compared with serous ovarian cancer, small (up to 4–5 mm) recurrent lesions of malignant struma ovarii along the pelvic peritoneum are even well vascularized and can be detected using US volumetric imaging in the angio mode.

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