Acta Med. Okayama, 2013
Vol. 67, No. 3, pp. 191–195

Acta Medica Okayama
http://escholarship.lib.okayama-u.ac.jp/amo/

Case Report

Three Cases of Struma Ovarii Underwent Laparoscopic Surgery with Definite Preoperative Diagnosis

Nurliza Binti Md Nor, Tomoyuki Kusumoto*, Seiji Inoue, Keiichiro Nakamura, Noriko Seki, Atsushi Hongo, Junichi Kodama, and Yuji Hiramatsu

Department of Obstetrics and Gynecology, Okayama University Graduate School of Medicine, Dentistry and Pharmaceutical Sciences, Okayama 700–8558, Japan

Struma ovarii is a rare neoplasm that accounts for approximately 0.3% of ovarian tumors. Due to its ultrasound morphology, which is quite similar to that of malignant ovarian carcinoma, most struma ovarii cases are open operated with laparotomy rather than laparoscopy. We present 3 cases of struma ovarii, which were diagnosed preoperatively by imaging studies and removed by laparoscopic surgery. All patients were premenopausal women between ages 31–50. The magnetic resonance imaging (MRI) findings were complex masses composed of multiple cysts and solid components with T2-hypointense regions as well as multiple T1-hyperintense cystic areas, findings that are typical for struma ovarii. A combination of plain computed tomography (CT), positron emission tomography (PET)-CT, and scintigraphy was useful for diagnosis. Laboratory examination revealed elevated serum thyroglobulin, which led to the diagnosis of struma ovarii. Laparoscopic surgeries were performed without rupturing the tumors. Although it has been difficult to differentiate between struma ovarii and malignant tumors by conventional methods, recently MRI techniques appear make it possible to diagnose struma ovarii preoperatively from the abovementioned imaging characteristic, together with laboratory data. As for treatment, we think laparoscopy could be successful for struma ovarii, but the surgeon must be careful not to rupture the tumor intra-abdominally in order to prevent dissemination, which could lead to malignancy.

Key words: struma ovarii, ovarian neoplasms, MRI, laparoscopic surgery

Struma ovarii is a rare ovarian tumor characterized by the presence of ectopic thyroid tissue, which comprises more than 50% of the overall tumor mass. This neoplasm is generally considered to account for less than 5% of mature teratomas [1], approximately 0.3–1% of all ovarian tumors, and 2.7% of all dermoid tumors [2]. Malignant transformation rates of struma ovarii are reported to vary, ranging from 5–10% [3]. Therefore, most struma ovarii are considered benign tumors. However, most of cases are treated with laparotomy because struma ovarii resembles ovarian carcinoma. In general a differential diagnosis between this neoplasm and a malignant tumor is difficult due to the morphological similarities between these tumors.

According to ovarian cancer treatment guidelines in Japan, endoscopic surgery for malignant ovarian disease is not considered a standard surgical proce-

Received February 15, 2012; accepted January 10, 2013.
*Corresponding author. Phone:+81-86-235-7320; Fax:+81-86-225-9570
E-mail:tkusu@md.okayama-u.ac.jp (T. Kusumoto)

dure that can replace laparotomy at the present time [4].

Because of the difficulty of preoperative diagnosis, many benign ovarian surgeries have been treated with laparotomy, entailing a large scar and a long hospital stay.

We present 3 cases of struma ovarii, as identified on the basis of preoperative diagnostic imaging and blood testing, that were resolved laparoscopically.

Case Report

Cace 1. A 50-year-old woman presented with an ovarian tumor, which was identified during a medical check-up (gravida: 1, para: 1). Her medical history included a thyroid carcinoma at age 39, which had been treated at that time with subtotal thyroidectomy. A transvaginal ultrasound revealed a left ovarian tumor composed of 4-cm multilocular cysts and a 2-cm solid component. The cysts showed variable signal intensity on the T2-weighted magnetic resonance imaging (MRI) scan (Fig. 1A). ^{123}I scintigraphy showed accumulation in the left pelvis, and plain computed tomography (CT) revealed 4.5-cm cysts in the left ovary, with a solid component and calcification; further, an intracystic high-attenuation lesion was observed on precontrast CT images (Fig. 1B). Tumor markers were within normal ranges: carcinoembryonic antigen (CEA), 1.43 ng/ml; carbohydrate antigen (CA) 19-9, 20.2 U/ml; CA125, 7.4 U/ml; CA15-3, 8.9 U/ml; and CA72-4, 2.9 U/ml. A recurrence of thyroid cancer was suspected from the serum thyroglobulin level, which was elevated by 1,470 ng/ml. However, all other examinations ruled out this suspicion. Struma ovarii was then suspected, and laparoscopic bilateral salpingo-oophorectomy was performed. The tumor was collected using an ENDOPOUCH® (ETHICON, San Angelo, TX, USA) without rupture. Histopathological analysis revealed follicles of various sizes composed of thyroid tissue, indicative of struma ovarii.

Cace 2. A 31-year-old woman (gravida: 1, para: 1) presented with a thyroid tumor at age 28 and was under observation. A medical check-up revealed an elevation in CA19-9 by 235.2 U/ml and a slightly increased serum thyroglobulin, *i.e.*, by 99.5 ng/ml. The other tumor markers (squamous cell carcinoma (SCC), 1.6 U/ml; CEA, 2.09 ng/ml; CA125, 19.1 U/ml; CA72-4, 3.8 U/ml; alpha-fetoprotein (AFP), 3.3 U/ml) were within the normal ranges. Transvaginal ultrasound showed a multilocular cyst in the right ovary with a 2-cm solid component. An ovarian tumor on the right side was indicated from the pelvic MRI scan, which also showed 8.5-cm multilocular cysts containing a fatty component, a water component, and hair balls, together representing teratoma. Furthermore, there were multiple small cysts with thick septations, some of them filled with colloidal material, which exhibited low signal intensity on both T1 and T2-weighted images (Fig. 2). A diagnosis of struma ovarii with teratoma (and also right thyroid adenoma) was made.

We recommended a laparoscopic oophorectomy in order toavoid a rupture and to prevent dissemination of the contents, but the patient required us to preserve the normal portions of the bilateral ovaries. Laparoscopic enucleation of the right ovary was performed. We injected a small amount of saline under

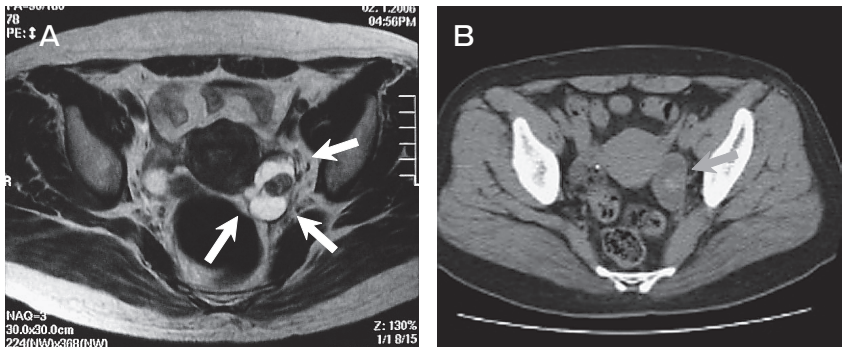

Fig. 1 A, Pelvic MRI (case 1): Variable signal intensity on T2-weighted image. Horizontal section; B, (case 1): Plain CT shows 4.5 cm cyst on the left ovary with intracystic high-attenuation lesion.

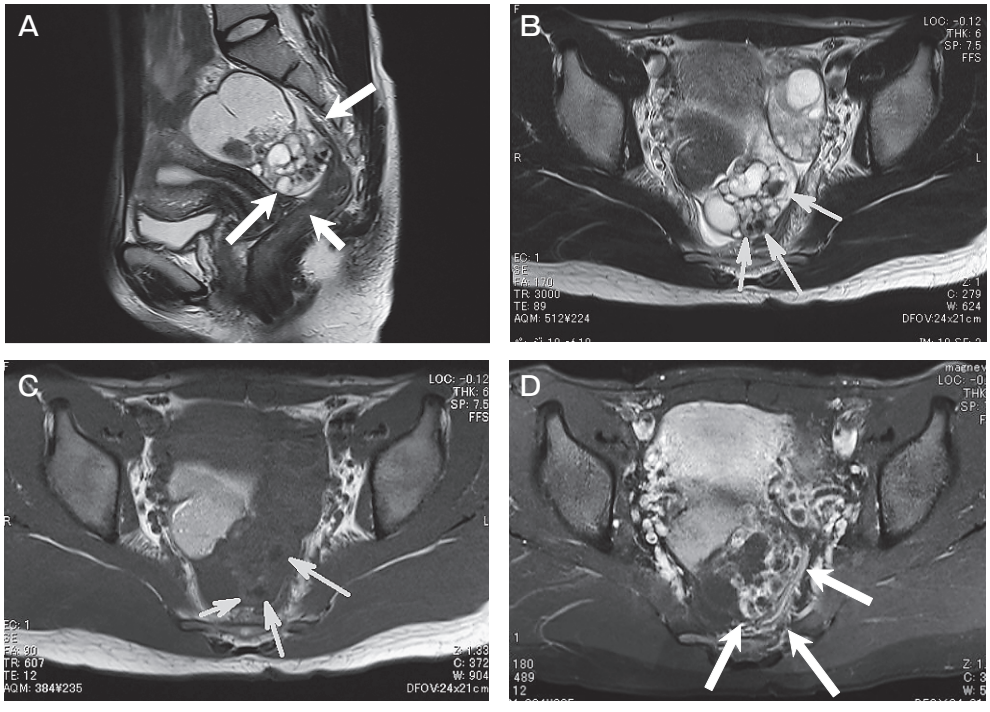

Fig. 2 (case 2): Pelvic MRI shows multiple small cysts with thick septations, some of them filled with colloidal material which show as low signal density in both T2- (A, B) and T1- (C, D) weighted images. arrow, ovarian tumor.

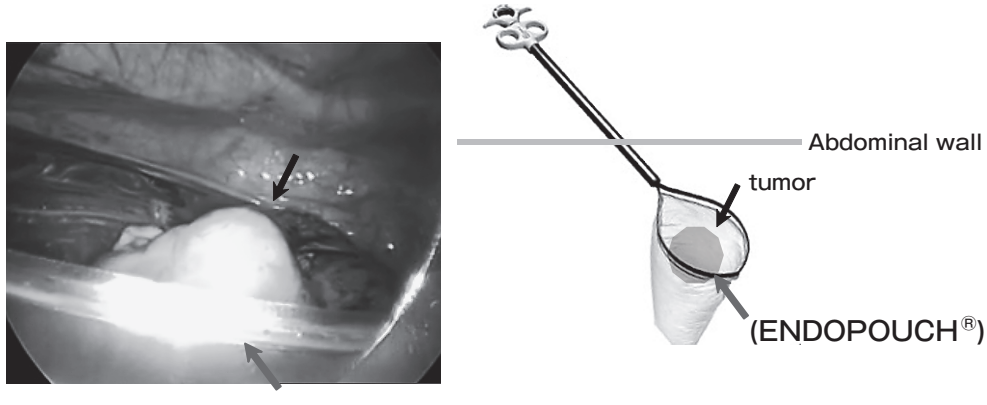

Fig. 3 (case 2): Laparoscopic findings and scheme: Ovarian tumor was collected without rupture using by ENDOPOUCH®.

the cortex before cutting the surface of the right ovary, then removed the cyst wall very carefully. The tumor was collected using an ENDOPOUCH® without rupture (Fig. 3). The fatty, hairball-containing loculus was diagnosed histologically as a mature teratoma. Other loculi containing serous fluids were composed of thyroid tissues. The postoperative diagnosis was maintained as struma ovarii.

Cace 3. A 31-year-old woman presented to the hospital for an evaluation of an ovarian tumor, which had been detected at age 29 by a routine pregnancy ultrasonography (gravida: 1, para: 1). She had no history of thyroid disorder. Delivery had been normal, but a follow-up ultrasound revealed a solid component in the ovary. The MRI scan showed 6-cm multilocular cysts, some of them containing fatty components. The

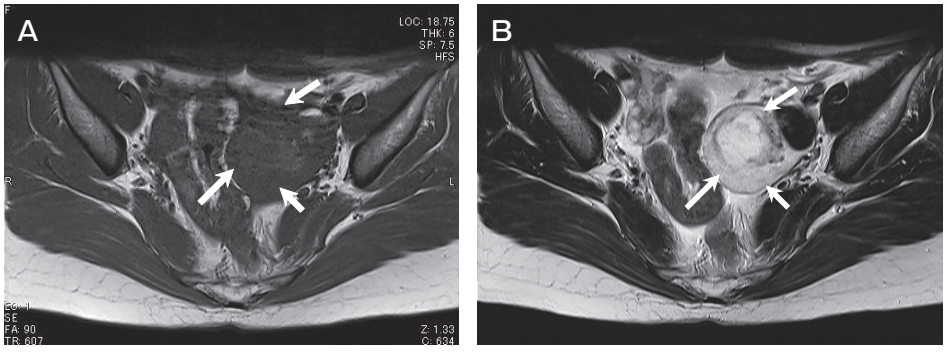

Fig. 4 (case 3): Pelvic MRI shows the left part of the mass (6 cm) producing a low signal intensity on T1-WI **A**, and a very low signal intensity on T2-WI **B**. At the center of the mass, a high-density area is seen on the T2-WI **B**.

left part of the mass produced a low signal intensity on T1-weighted images and a very low signal intensity on T2-weighted images, indicative of colloidal components. At the center of the mass, a non-uniform high-density area was noted on the T2-weighted MRI scans; this area was remarkably enhanced during the early phase of dynamic MRI (Fig. 4). Struma ovarii with mature teratoma was suspected. Although the serum thyroglobulin level had increased by 1,350 ng/ml, positron emission tomography (PET)-CT scans showed no abnormal accumulation. The other tumor markers were also within the normal ranges: CEA, 0.59 ng/ml; CA19-9, 16.9 U/ml; CA125, 17.2 U/ml; and CA72-4, 0.9 U/ml. Laparoscopic resection of the left ovary was performed, and the tumor was collected using an ENDOPOUCH® without rupture, the same as in cases 1 and 2. Histological analysis revealed the presence of follicles of various sizes; further, the follicular tissue consisted mainly of thyroid tissue and some cutaneous tissue, but not immature or malignant tissue; this suggested struma ovarii with a mature teratoma.

Discussion

Differential diagnosis between this neoplasm and malignant tumor is difficult due to their morphological similarities combined with the rarity of struma ovarii. All 3 of our patients were premenopausal women aged 31-50 years, and each of them had given birth. All 3 patients were asymptomatic; the findings indicative of struma ovarii were noted during medical check-ups. According to previous studies, most cases are asymptomatic, but thyroid hyperfunction is noted in 5-8% of patients [5, 6].

The ultrasonographic findings of struma ovarii are nonspecific, but it can occur when solid and cystic components are detected within the ovarian tumor on ultrasonography. We conclude the following from the results of imaging studies of struma ovarii: MRI findings show multilocular cysts with solid components of variable signal density within the loculi. Signal intensity is slightly high on T1-weighted images and very low on T2-weighted images without enhancement. This is because of hemorrhage or gelatinous (highly viscous) colloidal material. The solid parts are enhanced at the early phase of dynamic MRI and are markedly enhanced on enhanced T1-weighted images. Furthermore, an intracystic high-attenuation lesion can also be detected on plain CT [7], as in our first case. These findings correspond well with the low signal intensity on T1- and T2-weighted MRI scans, suggesting the presence of colloidal material. In mucinous cystadenocarcinoma, the T1- and T2-relaxation times are shortened on the basis of the protein concentration and the viscosity of the mucinous fluid, showing high and low signal intensity on T1- and T2-weighted images, respectively [6]. In the first case, ^{123}I scintigraphy revealed accumulation in the left pelvis. Although no functional thyroid tumor was detected, a previous report stated that preoperative ^{131}I scintigraphy of the minor pelvis could reveal active thyroid tissue [1, 8]. Therefore, serum thyroglobulin and scintigraphy (^{123}I or ^{131}I) could be useful for the diagnosis of struma ovarii, while PET-CT can be used to rule out the possibility of a malignant tumor, as happened in our third case. Histopathological examination, which reveals the presence of thyroid

follicles of various sizes, will provide an accurate diagnosis.

There are 2 surgical approaches to the resection of ovarian tumors. One is laparotomy or laparoscopic surgery, and the other is oophorectomy or enucleation. Laparoscopic surgery is less invasive than laparotomy. Enucleation can preserve the normal ovary, which can increase the chance of pregnancy. However, with this procedure it is difficult to avoid rupturing the tumor. Therefore, we first recommend laparoscopic oophorectomy unless the possibility of malignancy can be completely ruled out. If the patient wishes for pregnancy, then enucleation is more desirable.

In our cases, after deducing that the possibility of malignancy was low on the basis of various preoperative examinations, we performed laparoscopies. We think that oophorectomy or salpingo-oophorectomy is abetter choice than laparoscopic surgery, as the chances of rupturing the cyst are low, as seen in our first and third cases. However, for our second case, the patient requested minimal resection and consented to an enucleation of the tumor. Eventually we were able to perform laparoscopic enucleation without rupture of the tumor membrane, but it was not reproducible.

In all of the cases, the cysts were collected using an intra-abdominal ENDOPOUCH® and successfully taken out through a 12-mm cut at the navel without internal rupture. Laparoscopy is minimally invasive, but in cases of ovarian tumor, leakage of cystic fluid contents is still a matter of concern, as it would cause dissemination and also increase the tumor stage. The technique of putting the tumor into a bag in the abdominal cavity and removing it without leakage is very important in order to avoid abdominal dissemination and port site recurrences.

In conclusion, struma ovarii can be diagnosed preoperatively using MRI and serum thyroglobulin levels, without other forms of imaging.

Appropriate preoperative diagnosis will improve the patient's QOL. However, we must still be aware of the 5-10% possibility of malignancy [3]; the appropriate surgical procedure should be selected only after careful discussion with the patient and the obtainment of informed consent. Our nonrupture procedures can prevent the diffusion of malignant cells.

References

1. Mandic A, Rajovic J, Tesic M, Vujkov T, Krnojelac D and Komazec S: Benign and malignant struma ovarii: report of three cases and review of the literaturei. J BUON (2002) 7: 67-70.
2. Roth LM and Talerman A: The enigma of struma ovarii. Pathalogy (2007) 39: 139-146.
3. Dardik RB, Dardik M, Westra W and Montz F: Malignant struma ovarii: two case reports and a review of the literature. Gynecol Oncol (1999) 73: 447-451.
4. Ovarian cancer treatment guidelines; Japan Society of Gynecologic Oncology, 3rd Ed, Tokyo (2010), pp 42-43 (in Japanese).
5. Yoo SC, Chang KH, Lyu MO, Chang SJ, Ryu HS and Kim HS. Clinical characteristics of struma ovarii. J Gynecol Oncol (2008) 19: 135-138.
6. Matsuki M, Kaji Y, Matsuo M and Kobashi Y: Struma ovarii: MRI findings. Br J Radiol (2000) 73: 87-90.
7. Jung SI, Kim YJ, Lee MW, Jeon HJ, Choi JS and Moon MH: Struma Ovarii: CT findings. Abdom Imaging (2008) 33: 740-743.
8. Joja I, Asakawa T, Mitsumori A, Nakagawa T, Akaki S, Yamamoto M, Takeda Y, Ando M and Hiraki Y: I-123 uptake in nonfunctional struma ovarii. Clin Nucl Med (1998) 23: 10-12.