Gynecol Surg (2006) 3: 103–109
DOI 10.1007/s10397-006-0189-x

ORIGINAL ARTICLE

Akihiro Takeda · Shuichi Manabe
Takashi Mitsui · Hiromi Nakamura

Laparoscopic management of selected solid adnexal tumors

Received: 24 December 2005 / Accepted: 28 February 2006 / Published online: 9 May 2006

Abstract Fourteen cases of laparoscopically treated solid/predominantly solid nonepithelial adnexal tumors are presented. Unilateral salpingo-oophorectomy was performed in seven cases [three fibromas, one thecoma, one strumal carcinoid, one Sertoli-Leydig cell tumor (G2), and one dysgerminoma], including a 21-year-old patient with fibroma with adnexal torsion. A 26-year-old patient with fibroma was treated by tumorectomy. Laparoscopic-assisted vaginal hysterectomy and bilateral salpingo-oophorectomy were done in five cases [three thecomas, one adult-type granulosa cell tumor, one Sertoli-Leydig cell tumor (G2)], including a 52-year-old patient with thecoma complicated by torsion. In a case of adenomatoid tumor of the fallopian tube, unilateral salpingectomy was done. Adjuvant platinum-based chemotherapy was added in a case of dysgerminoma, and there has been no sign of recurrence after 3 years.

Keywords Solid adnexal tumor · Fibroma · Adenomatoid tumor · Sertoli-Leydig cell tumor · Dysgerminoma · Laparoscopy

With the advent of surgical equipment and techniques in laparoscopy, ovarian tumors are now commonly managed by laparoscopy if conditions are suspected to be benign [1, 2]. However, laparoscopic surgery for either suspicious or malignant conditions remains controversial [3–5]. Cases of solid/predominantly solid adnexal tumor first diagnosed by ultrasonography include various natures of neoplastic conditions from benign to malignant [6, 7], and laparotomy is the usual approach to such cases. However, if cases are appropriately selected by careful evaluation of tumor characteristics [8, 9] before and during surgery, management of solid adnexal tumor by laparoscopic surgery becomes possible [10–12].

The present report describes the application of laparoscopic procedure to treat a select group of patients with solid/predominantly solid nonepithelial adnexal tumors under carefully evaluated conditions.

A. Takeda (✉) · S. Manabe ·
T. Mitsui · H. Nakamura
Department of Obstetrics and Gynecology,
Gifu Prefectural Tajimi Hospital,Maebata-cho,
Tajimi, Gifu, 507-8522, Japan
e-mail: reef@syd.odn.ne.jp
Fax: +81-572-251246

Materials and methods

Evaluation of tumor characteristics before and during surgery

Preoperative evaluation of adnexal tumor was first done by transvaginal and transabdominal ultrasound to categorize tumors according to the internal echogenic pattern [6, 7]. Tumors that showed a solid/predominantly solid pattern were further evaluated. In recently managed cases, blood flow in the tumor tissue was assessed by color Doppler ultrasound. Images were obtained by computed tomography (CT) or magnetic resonance imaging (MRI) for further evaluation of the tumor characteristics, and diagnosis was made by our hospital's diagnostic radiologists according to the criteria [8]. Tumor markers—including CA125, CA19-9, alpha-fetoprotein, carcinoembryonic antigen, and CA72-4—and nonspecific biochemical markers such as LDH were measured to assist the diagnosis. In cases that showed suspicious hormonal manifestation, serum hormones such as luteinizing hormone, follicle stimulating hormone, estradiol, progesterone, and testosterone were measured.

Patients with apparent malignant tumor diagnosed by ultrasonography and CT/MRI images were treated by laparotomy following the principles of classic oncologic surgery and were excluded from the present study. Patients with presumably benign tumors were treated by laparoscopic surgery. In suspicious cases, the tumor was carefully inspected under laparoscopic observation. Rapid cytological tests of peritoneal washings and pathological examination of frozen-sectioned tumor tissue obtained by needle biopsy were done, if necessary, and the final decision on

surgical procedure was made intraoperatively. Patients with tumors <10–12 cm that could be retrieved in an Endobag underwent laparoscopic surgery, and damage to the tumor capsule during the surgical procedure was minimized as much as possible. Preoperatively, informed consent, including a statement that conversion to laparotomy might be required if advanced malignant tumor were detected under laparoscopic observation or if the mass could not be managed by laparoscopy, was obtained from the patients and families.

Surgical procedures

Gasless laparoscopic surgery was performed as previously described [13] with modifications. Briefly, under general endotracheal anesthesia, the abdominal wall was lifted by a subcutaneous lift system (Mizuho, Tokyo, Japan), and a 5-mm Optiview port (Ethicon Japan, Tokyo, Japan) was placed at the superior edge of the umbilicus to introduce a 5-mm laparoscope (Olympus, Tokyo, Japan). Two other Optiview ports were placed laterally under direct vision: a 5-mm port on the left side and a 12-mm port on the right side at the level of the umbilicus. If necessary, a 20- to 30-mm suprapubic port was made by either Lap-Protector Mini (Hakko Medical, Tokyo, Japan) or Alexis Wound Retractor (Applied Medical, Rancho Santa Margarita, CA, USA). Adhesiolysis was done by Harmonic scalpel (Ethicon Japan, Tokyo, Japan). In cases of salpingo-oophorectomy, the utero-ovarian and infundibulopelvic ligaments were cut by either an Endo GIA or LigaSure Atlas (Tyco Health Care, Tokyo, Japan). In cases treated by laparoscopic-assisted vaginal hysterectomy (LAVH) and salpingo-oophorectomy, uterine artery and cervical ligaments were ligated and cut vaginally, and the upper ligaments were cut by either an Endo GIA or LigaSure Atlas. Tumor <6 cm was put into an Endopouch retriever (Ethicon Japan, Tokyo, Japan) to prevent spilling. Tumor >6 cm was retrieved in an Endocatch II (Tyco Health Care, Tokyo, Japan). Tumor tissue was cut into small pieces by scissors in the retriever bag and carried outside of the body. In cases of salpingo-oophorectomy, salpingectomy, or tumorectomy alone, tumor tissue was extracted either through a suprapubic port or posterior colpotomy. In cases of salpingo-oophorectomy with LAVH, adnexal tumor was transported through the vaginal stump after hysterectomy.

Results

Between 1994 and September 2005, 1,238 cases of adnexal tumor were surgically treated in our department (Fig. 1); 281 cases were treated by laparotomy and 957 cases by laparoscopic surgery. By image-diagnostic tumor pattern [6–8], the presence of massive ascites, and elevated serum tumor markers, 26 cases suggesting apparent epithelial malignancy were excluded; these cases were finally diagnosed as malignant by histopathological diagnosis. Twenty-eight cases were then diagnosed as solid/predominantly solid nonepithelial adnexal tumors by ultrasonography, and further evaluation of tumor characteristics was made by CT/MRI scan.

Twelve cases showing solid/predominantly solid tumor were treated by laparotomy alone, either because the tumor was large or because it was highly suspicious for advanced malignancy on preoperative diagnostic imaging procedures. Sixteen cases were initially examined by laparoscopy, and laparotomic conversion was made in two cases. A case of poorly differentiated Sertoli-Leydig cell tumor (G3) was converted to laparotomy because of large tumor size (120×74 mm) and high suspicion of malignancy on rapid pathological examination. A case of fibroma that was preoperatively diagnosed as subserosal myoma or solid adnexal tumor was converted to laparotomy because

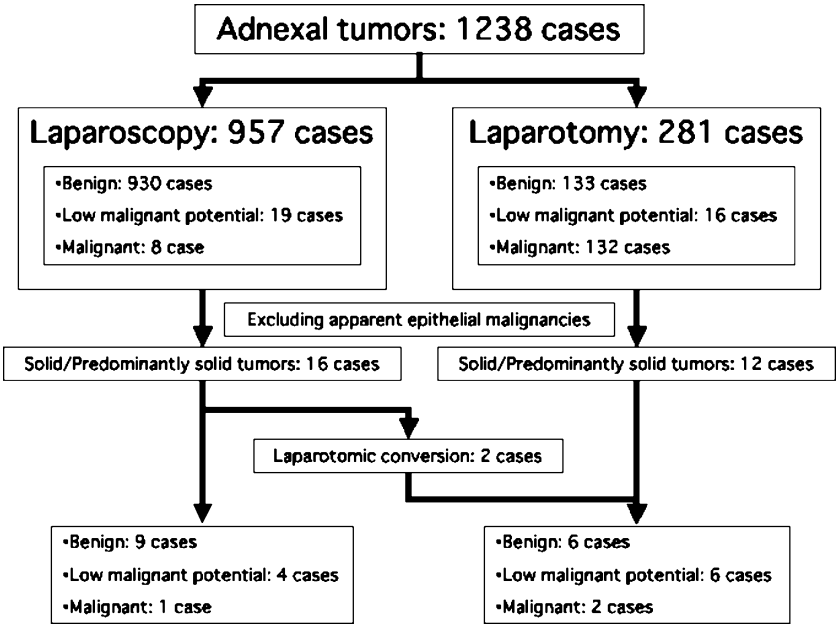

Fig. 1 Surgical management procedures for 1,238 adnexal tumors between January 1994 and September 2005 at Gifu Prefectural Tajimi Hospital

laparoscopic management was difficult due to large tumor size (130×84 mm) and poor expansion of the abdominal cavity.

Therefore, 14 cases of nonepithelial solid adnexal tumor were completely treated by laparoscopic/laparoscopic-assisted surgery, and these cases are retrospectively analyzed (Table 1). Preoperative differential diagnosis from subserosal/pedunculated myoma was difficult in four cases, and the correct diagnosis of adnexal tumor was made after laparoscopic observation in these four cases. Age distribution ranged from 21 to 66 years old. Tumor diameters varied from 47 to 109 mm. Elevation of preoperative serum CA125 value was noted in cases of fibroma with torsion (216 U/ml), fibroma with previous abdominal hysterectomy due to myoma (89 U/ml), and thecoma (88 U/ml). A 42-year-old patient with moderately differentiated Sertoli-Leydig cell tumor (G2) had slight hirsutism and elevated serum testosterone level (465 ng/dl). Benign ovarian tumors consisted of four fibromas and four thecomas. Cases of fibroma and thecoma were associated with adnexal torsion. One solid tumor of the fallopian tube was adenomatoid tumor. Low-malignant-potential cases consisted of one strumal carcinoid, one adult-type granulosa cell tumor, and two Sertoli-Leydig cell tumors (G2). There was one malignant case of dysgerminoma.

Unilateral salpingo-oophorectomy was performed in seven cases [three fibromas, one thecoma, one stromal carcinoid, one Sertoli-Leydig cell tumor (G2), and one dysgerminoma]. In a 26-year-old patient with fibroma, border between tumor and normal ovarian tissue was evident, and tumorectomy was done. Adenomatoid tumor of the fallopian tube was treated by salpingectomy. LAVH was done at the same time as bilateral salpingo-oophorectomy in four postmenopausal women (three thecomas and one adult-type granulosa cell tumor) and in a 42-year-old patient with Sertoli-Leydig cell tumor (G2) because of the presence of adenomyosis and at the patient's request. Homologous blood transfusion was not required in any case. The postoperative course was uneventful in all cases, and there were no recurrences noted in either the low-malignant-potential or the malignant cases.

Reports of four rare cases that showed characteristic clinical and laparoscopic findings are individually described below.

Torsion of fibroma in a young woman

A 21-year-old nursing-school student, gravida 0 para 0, was referred to our hospital because of lower abdominal tumor with a 2-week history of pain. Transabdominal ultrasonography showed the presence of a solid tumor in the lower abdomen (109×65 mm). MRI scans showed a complex internal pattern due to prominent ischemic change in the tumor (Fig. 2a). Under a diagnosis of solid adnexal tumor with torsion, urgent surgery was performed. Under laparoscopic observation, the tumor was necrotic (Fig. 2b), and the left adnexal tissue was twisted 720º. Because preservation of adnexal tissue seemed to be difficult, left salpingo-oophorectomy was performed using the LigaSure Atlas (Fig. 2c), and excised tissue was put into an Endocatch II. Tumor tissue was extracted through a suprapubic port after being cut into small pieces by scissors inside the retriever bag (Fig. 2d). The surgical duration was 53 min, and the estimated blood loss was 200 ml. Tissue weight was 273 g. The pathological diagnosis was fibroma with massive necrosis.

Adenomatoid tumor of fallopian tube

A 42-year-old woman, gravida 3 para 2, was referred to our hospital because of a pelvic tumor that was found during a regular check-up. Transvaginal ultrasonography (Fig. 3a) and abdominal MRI (Fig. 3b) showed the presence of a solid tumor in the left adnexal region (47×46 mm). Under a diagnosis of pedunculated myoma or solid adnexal tumor, laparoscopic surgery was performed. Under laparoscopic

Table 1 Solid adnexal tumors treated by laparoscopic surgery (*SO* salpingo-oophorectomy, *LAVH* laparoscopic-assisted vaginal hysterectomy)

Clinical classification	Postoperative diagnosis	Age	Tumor diameter (mm)	Surgical procedure
Benign	Fibroma	39	59×42	Unilateral SO
	Fibroma	21	109×65	Unilateral SO
	Fibroma	57	80×81	Unilateral SO
	Fibroma	26	50×45	Tumorectomy
	Thecoma	40	100×60	Unilateral SO
	Thecoma	66	94×79	LAVH + bilateral SO
	Thecoma	64	85×47	LAVH + bilateral SO
	Thecoma	52	80×60	LAVH + bilateral SO
	Adenomatoid tumor of fallopian tube	42	47×46	Unilateral salpingectomy
Low malignant potential	Strumal carcinoid	48	71×42	Unilateral SO
	Adult-type granulosa cell tumor	51	57×54	LAVH + bilateral SO
	Sertoli-Leydig cell tumor (G2)	31	48×31	Unilateral SO
	Sertoli-Leydig cell tumor (G2)	42	54×31	LAVH + bilateral SO
Malignant	Dysgerminoma	22	71×35	Unilateral SO

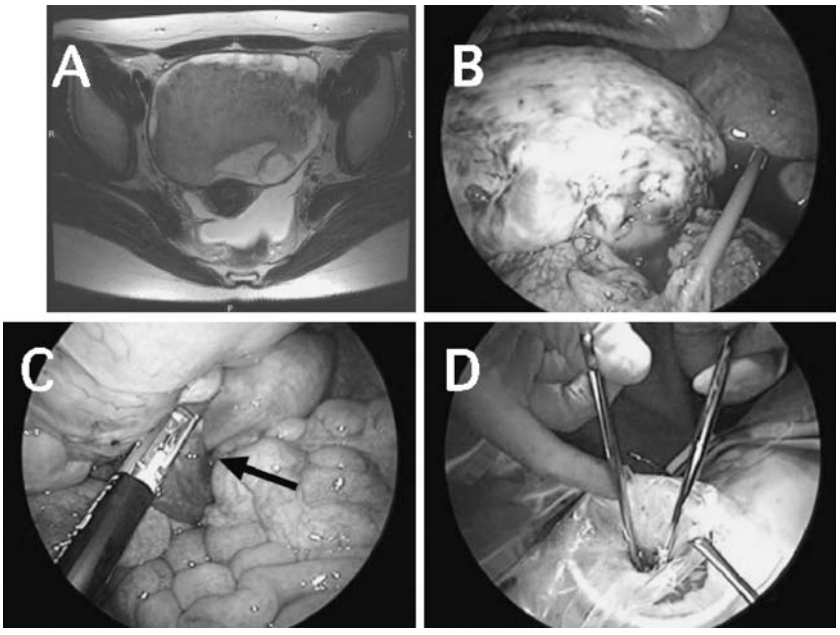

Fig. 2 **a** Cross-sectional T2-weighted magnetic resonance image of large ovarian fibroma showing a complex internal image due to necrotic change caused by adnexal torsion. **b** Laparoscopic view of fibroma of ovary with torsion. (**c** Left adnexal tissue with torsion (*arrow*) was excised by LigaSure Atlas. **d** Excised tissue was put into an Endocatch II and extracted through suprapubic port after being cut into small pieces by scissors

observation, the tumor was diagnosed as a solid fallopian tube tumor that occurred in the ampullary portion of the left tube (Fig. 3c), and left salpingectomy was performed using the EndoGIA. Excised tissue was put into an Endopouch retriever and extracted through a posterior colpotomy. The surgical duration was 60 min, and estimated blood loss was less than 20 ml. Tissue weight was 90 g. The pathological diagnosis was adenomatoid tumor of the fallopian tube (Fig. 3d).

Recurrent Sertoli-Leydig cell tumor (G2)

A 31-year-old woman, gravida 0 para 0, was referred to our hospital because of recurrent ovarian tumor. She had a history of laparotomic tumorectomy for ovarian tumor that had been initially diagnosed as sclerosing ovarian tumor 6 years earlier. This same tissue specimen was rediagnosed this time as Sertoli-Leydig cell tumor (G2) by pathologists at our hospital. Transvaginal ultrasonography and abdominal MRI showed the presence of a solid left adnexal mass (48×31 mm; Fig. 4a). Analysis by power Doppler ultrasound (Fig. 4b) showed abundant arterial blood flow with RI=0.55, MaxV=0.24 m/s, and MinV=0.11 m/s in the tumor tissue. There was no hormonal manifestation, with a

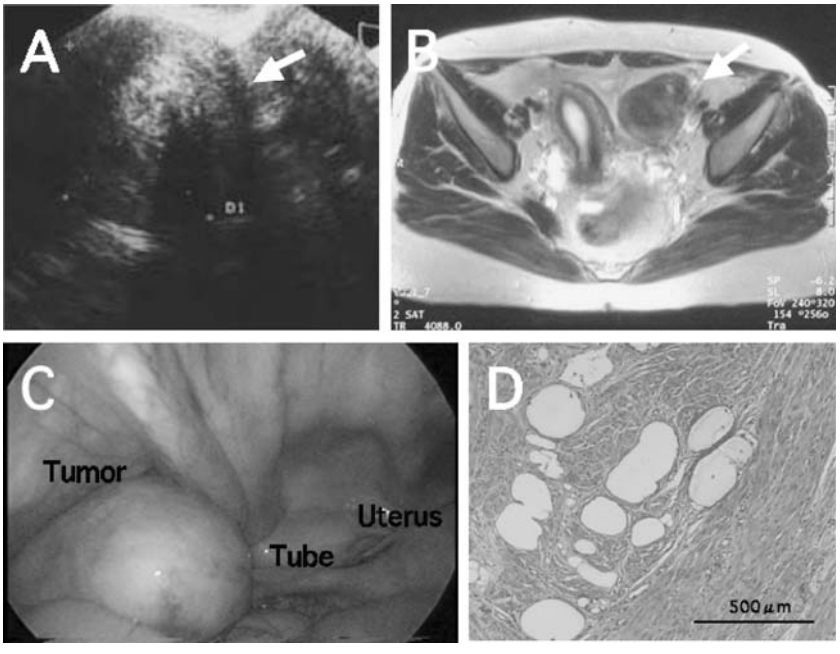

Fig. 3 **a** Transvaginal ultrasonographic finding of adenomatoid tumor of the fallopian tube showing a solid echogenic pattern (*arrow*). **b** Cross-sectional T2-weighted magnetic resonance image of adenomatoid tumor of the fallopian tube (*arrow*). **c** Laparoscopic observation of adenomatoid tumor in the distal portion of the left fallopian tube. **d** Histopathological findings of adenomatoid tumor of the fallopian tube (hematoxylin-eosin stain, scale bar: 500 μm)

normal testosterone level (30 ng/dl). Left salpingo-oophorectomy was done using the LigaSure Atlas (Fig. 4c), and excised tissue was put into an Endopouch retriever. Tumor tissue was carried out of the body through a posterior colpotomy. The surgical duration was 59 min, and estimated blood loss was 100 ml. Tissue weight was 26 g. The pathological diagnosis was recurrent Sertoli-Leydig cell tumor (G2) (Fig. 4d).

Dysgerminoma with pelvic inflammatory disease

A 22-year-old woman, gravida 0 para 0, with a disease history of recurrent chlamydia infection reported lower abdominal pain and vaginal discharge. Because *Chlamydia trachomatis* antigen was detected in her cervical canal, pelvic inflammatory disease was diagnosed and clarithromycin administered. At this time, transvaginal ultrasonography incidentally showed the presence of a solid tumor (71×35 mm) in the right adnexal region (Fig. 5a). Tumor marker values were not elevated, but serum LDH was elevated to 645 IU/l. Apparent metastatic lesions and lymphadenopathy were not detected by whole-body CT scan. Under the diagnosis of suspicious right ovarian tumor, laparoscopic surgery was performed. Dense pelvic adhesion due to chlamydia infection was noted around the uterus and adnexal tissue (Fig. 5b). Right adnexal tumor was attached to sidewall pelvic peritoneum. After adhesiolysis using the Harmonic scalpel, right salpingo-oophorectomy was done using the LigaSure Atlas. Rapid washing cytology of the peritoneal fluid was negative. Excised tissue was placed into an Endopouch retriever (Fig. 5c). Tissue specimen was obtained by needle biopsy, and the pathological diagnosis on frozen section was suspicious of dysgerminoma. Tissue was extracted through a suprapubic port. Because enlargement of the left ovary was not evident, tissue sampling from the left ovary was not done. The surgical duration was 59 min, and estimated blood loss was 70 g. Tissue weight was 76 g. The final pathological diagnosis was dysgerminoma (Fig. 5d). Postoperatively, the serum LDH level immediately decreased to normal. Because minor damage of the tumor capsule was noted during adhesiolysis, platinum-based adjuvant chemotherapy (bleomycin, etoposide, cisplatin) was performed. The patient currently remains disease-free 3 years postoperatively.

Discussion

Ovarian neoplasms are histopathologically classified according to their tissue of origin as epithelial, germ cell, sex cord-stromal, and metastatic tumors [14]. These tumors are clinically classified as benign, low-malignant-potential, and malignant tumors. By diagnostic imaging procedures, these same tumors are classified according to the internal echogenicity as cystic, solid, and mixed tumors [6, 7]. Ovarian tumors that show a solid/predominantly solid pattern on ultrasonography comprise 15-20% of all ovarian neoplasms. These solid ovarian tumors show a variety of histological appearances and malignant tendencies. The majority of these tumors are malignant epithelial ovarian neoplasms that are usually excluded from indications for laparoscopic management [1, 2]. However, benign tumors that can be managed by laparoscopic surgery exist in this group, and selected cases showing low-malignant-potential and malignant nonepithelial tumor without apparent metastatic lesions may also be laparoscopically managed.

Applications of laparoscopic surgery to benign cystic adnexal masses are well established as minimally invasive

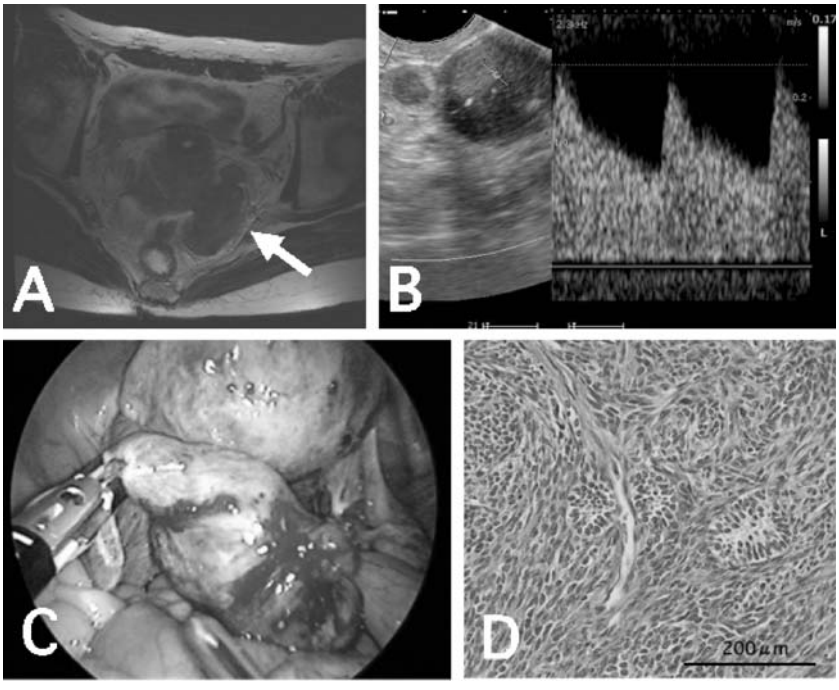

Fig. 4 **a** Cross-sectional T2-weighted magnetic resonance image of recurrent Sertoli-Leydig cell tumor (G2) of the ovary (*arrow*). **b** Power Doppler finding of recurrent Sertoli-Leydig cell tumor (G2) of the ovary showing abundant arterial blood flow with RI=0.55, MaxV=0.24 m/s, and MinV=0.11 m/s in the tumor tissue. **c** Left adnexal tissue was excised by LigaSure Atlas. **d** Histopathological findings of recurrent Sertoli-Leydig cell tumor (G2) of the ovary (hematoxylin-eosin stain, scale bar: 200 μm)

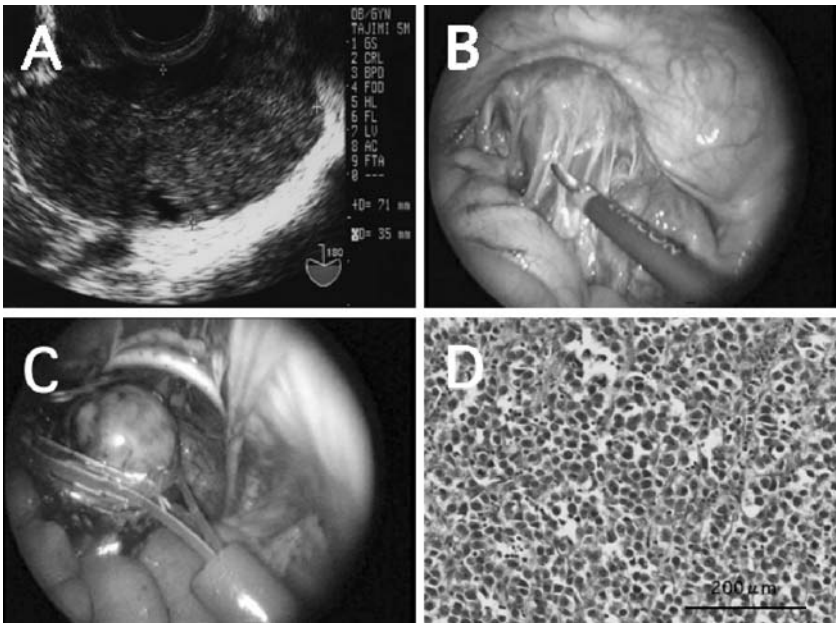

Fig. 5 **a** Transvaginal ultrasonographic finding of dysgerminoma showing solid echogenic pattern. **b** Adhesiolysis using Harmonic scalpel to demonstrate right adnexal tumor. **c** Excised tissue was put into an Endopouch retriever and extracted through a suprapubic port. **d** Histopathological findings of dysgerminoma (hematoxylin-eosin stain, scale bar: 200 μm)

treatment procedures [1, 2]. However, reports describing the application of laparoscopic surgery to solid adnexal tumors are limited [4, 10–12]. In contrast to a cystic adnexal mass that can be managed by laparoscopy after draining the cystic contents by aspiration, even when the tumor is large [15], in cases of solid adnexal tumor, the tumor size is a major limiting factor for laparoscopic manipulation of the tumor without damage. In the present study, we managed a case of fibroma measuring 130 mm in diameter by laparotomic conversion due to difficult laparoscopic manipulation. However, a fibroma measuring 109 mm in diameter could be managed by laparoscopy. Therefore, depending on the degree of expansion of the abdominal cavity, solid tumor exceeding 110–120 mm becomes difficult to manage by laparoscopic surgery and would be excluded from the indication.

Ovarian fibromas are benign neoplasms that arise from the connective tissue stroma of the ovary [14]. They constitute 3–5% of all ovarian neoplasms and are relatively common tumors in a group of solid/predominantly solid tumors. They are typically homogeneous, well-encapsulated masses that show decreased signal intensity on both T1- and T2-weighted MRI images [16]. However, once adnexal torsion occurs, these tumors develop hemorrhage and necrosis of the tumor tissue, showing areas of high signal intensity on both T1- and T2-weighted MR images due to passive congestion of the mass [17]. In the present study, cases of adnexal torsion caused by fibroma and thecoma, respectively, were treated by laparoscopic surgery. MRI findings that showed massive ischemic areas were helpful in diagnosing the probable torsion of the solid adnexal tumor in these cases. Although laparoscopic treatment of fibromas has previously been described [1, 2, 4, 5], management of relatively large fibroma with torsion occurring in a young woman has not previously been reported. The present study indicates that if any symptoms suggesting adnexal torsion due to a solid adnexal tumor exist and images on MRI show ischemic change in the tumor tissue, prompt laparoscopic surgery could be a treatment of choice for this type of tumor.

When solid/predominantly solid nonepithelial adnexal tumors are encountered, physicians should note that rarely encountered tumors exist in this category. Adenomatoid tumor of the fallopian tube is one such tumor. Pathologically, there is now agreement that it represents a unique variant of benign mesothelioma largely restricted to the genital region [14]. Adenomatoid tumors of the fallopian tube are usually found as incidental small (≤2 cm) lesions within the wall of the tube [18]. However, in the present case the tumor was relatively large, and the preoperative diagnosis was pedunculated myoma or solid adnexal tumor. Correct diagnosis of fallopian tube tumor was established after laparoscopic observation. Although laparoscopic management of adenomatoid tumor of the fallopian tube has not been reported, surgical intervention itself was not difficult by laparoscopy, as adenomatoid tumor of the fallopian tube is usually well circumscribed [18]. Therefore, knowledge about the existence of this type of tumor contributes to the consideration of the surgical approach to solid/predominantly solid adnexal tumors for successful laparoscopic management.

When solid/predominantly solid nonepithelial adnexal tumors are noted in younger women, preserving fertility is an important consideration. Especially in patients with low-malignant-potential and malignant tumors, the treatment strategy depends on the patient's age, the disease stage, and the degree of differentiation [19]. If there is no evidence of extension beyond the involved ovary, the tumor usually pursues a benign course, and conservative surgery with unilateral salpingo-oophorectomy is an accepted treatment [19]. In the present study, we encountered a 31-year-old patient with Sertoli-Leydig cell tumor (G2) and a 22-year-old patient with dysgerminoma. Because these women were young and gravida 0, preserving future fertility, in

addition to doing minimally invasive surgery, was strongly preferred by the patients and their families, and the treatment strategy was decided.

Sertoli-Leydig cell tumors of the ovary are classified as a sex cord-stromal cell tumor and are extremely rare tumors that constitute less than 0.5% of all ovarian tumors [14]. They are more commonly found in younger women and are frequently associated with virilization [19]. Previously, laparoscopic management of Sertoli-Leydig cell tumor was reported with favorable prognosis [4, 10, 11]. In the present study, we encountered a case of recurrent Sertoli-Leydig cell tumor that was initially treated by laparotomic tumorectomy. Since the border between tumor tissue and normal ovarian stroma is indistinguishable in most solid/predominantly solid tumors, salpingo-oophorectomy would be a safer approach considering the risk of tumor recurrence. Because the present case was treated in 2005, the follow-up duration is only 6 months to date. Although no recurrence has been noted, further close follow-up is required to assess the feasibility of the procedure in this type of case.

Dysgerminomas are classified as germ cell tumors and are uncommon tumors accounting for 1–2% of primary ovarian neoplasms [14]. Dysgerminomas are highly sensitive to radiation therapy and chemotherapy even at the advanced stage [20]. Since the majority of dysgerminoma cases occur in adolescence and early adult life and present as early-stage tumor with involvement of the unilateral ovary, it is important that therapy be optimized because of the young age of women affected and the threat that therapy may pose to fertility. For those wishing to preserve fertility, conservative surgery with close clinical, radiologic, and serologic follow-up is the treatment of choice, with chemotherapy for relapse [20]. Therefore, although laparoscopic management of dysgerminoma has not been reported in the literature except for a case that was incidentally found in the histological section of gonadoblastoma tissue in an adolescent with XY gonadal dysgenesis [21], laparoscopic surgery could be a rational alternative to laparotomy if procedures can be performed without severe damage to the tumor capsule, as in the present case. With platinum-based adjuvant chemotherapy, our patient shows no evidence of disease 3 years postoperatively. However, further close follow-up is necessary to confirm the feasibility of the laparoscopic procedure.

Conclusion

Laparoscopic surgery is a minimal-access procedure with the advantages of rapid recovery, minimal blood loss, decreased adhesion formation, shorter hospital stay, and minimal psychological trauma. All of these make laparoscopy a better approach in a selected series of cases showing solid/predominantly solid adnexal tumor. With proper selection, a group of patients with solid adnexal tumors can be managed by laparoscopic surgery. Further accumulation of cases is necessary to assess the feasibility and safety of laparoscopic procedures for solid adnexal tumors.

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