A case of extremely rare ovarian tumor: Primary ovarian adenomyoma

Shoji KAKU[1)], Takuya MORIYA[2)], Naoki KANOMATA[2)], Tsuyoshi ISHIDA[1)]
Yangsil CHANG[1)], Norichika USHIODA[1)], Yuichiro NAKAI[1)]
Koichiro SHIMOYA[1)], Takafumi NAKAMURA[1)]

1) Department of Obstetrics and Gynecology, 2) Department of Pathology 2, Kawasaki Medical School, 577 Matsushima, Kurashiki, 701-0192, Japan

ABSTRACT Smooth muscle tumors of the ovary are rare, and ovarian adenomyoma are extremely rarer. Only 5 cases of primary ovarian adenomyoma have been reported in the world. Herein, we report a case of unilateral ovarian adenomyoma which was diagnosed in a premenopausal woman with the preoperative diagnosis of a thecoma of the left ovary.

(Accepted on September 30, 2011)

Key words：**Ovarian tumor, Adenomyoma, Case report**

INTRODUCTION

Smooth muscle tumors of the ovary are rare, and ovarian adenomyoma are extremely rare. Adenomyomas are benign tumors and they typically originate within the uterus, but only 5 cases of primary ovarian adenomyoma have been reported in the world(1-4). Herein, we report a case of unilateral ovarian adenomyoma. She underwent total abdominal hysterectomy and unilateral salpingo-oophorectomy with the preoperative diagnosis of an uterine myoma and a thecoma of the left ovary.

CASE REPORT

A 42-year-old, gravida 1, para 1, woman was admitted to our hospital with a complaint of lower abdominal pain. Upon abdominopelvic examination, a solid, movable mass was noted in the left lower quadrant. A 6.0x5.5x5.0cm solid tumor (Fig.1-A, B) was detected in the pelvis by magnetic resonance imaging (MRI). The solid tumor had no continuity with the uterus or the digestive tract. Most parts of the tumor demonstrated low intensity in plane T1 and T2-weighted MRI, but a few parts did demonstrate high intensity. And the tumor was stained as a result of the use of contrast media (Fig.1-A, B). Moreover, 7.0x5.5x5.0cm uterine myoma was also detected (Fig.2).

The serum levels of CA125, CA19-9, and CEA were 91.0U/mL, 159.0U/mL, and 1.1ng/ml respectively. We were not able to completely deny a malignant tumor because the inside of solid tumor was heterogeneous indicating a partial old bleeding by MRI (Fig.1-A, B). However, we thought the preoperative diagnosis of the left ovarian tumor was

Corresponding author
Shoji Kaku
Department of Obstetrics and Gynecology, Kawasaki Medical School, 577 Matsushima, Kurashiki, 701-0192, Japan.
Phone : 81 86 462 1111
Fax : 81 86 464 1135
E-mail : kaku@med.kawasaki-m.ac.jp

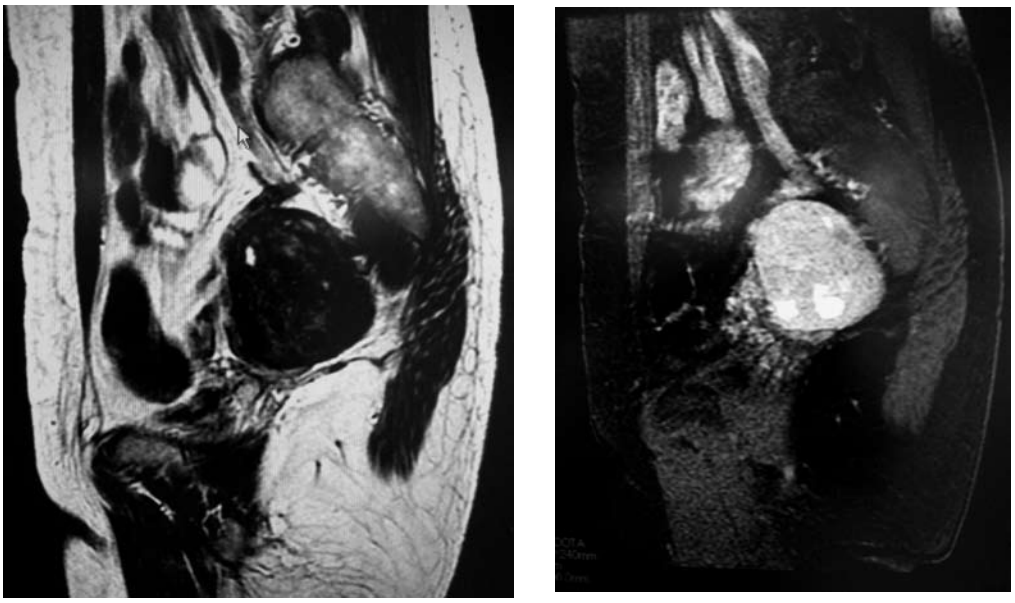

Fig. 1. Plane T2-weighted MR imaging (A) and contrast-enhanced T1-weighted fat suppression MR imaging using contrast media (B). The MRI showed left ovarian tumor which was stained as a result of the use of contrast media.

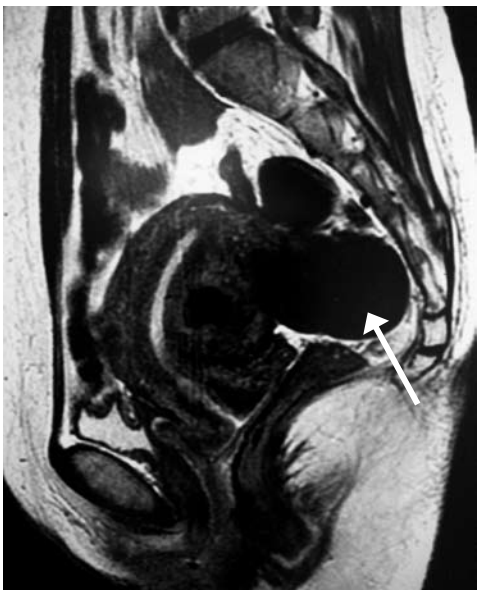

Fig. 2. Plane T2-weighted MR imaging. The MRI showed uterine myoma (white arrow).

thecoma.

Upon laparotomy, a left ovarian tumor and uterine myoma was found and a standard total abdominal hysterectomy and left salpingo-oophorectomy was subsequently performed. The right ovary appeared normal and no peritoneal dissemination was observed.

Macroscopically, the left ovary was 6.2x5.5x5.2 cm and firm (Fig.3). The capsule was intact, showing no invasion. Incision of the tumor revealed that the cut surface is smooth and the color is light yellow, with dark brownish bleeding in one portion (Fig.4).

Microscopically, the tumor was composed of benign glands and smooth muscle cells (Fig. 5). These smooth muscle bundles were without atypia and significant mitotic activity (Fig.6). Immunohistochemical staining showed a strong positive for alpha-smooth muscle actin and desmin (Fig.7-A, B), but these cells were slightly positive for Ki-67 (Fig.7-C). The final pathological diagnosis was adenomyomas.

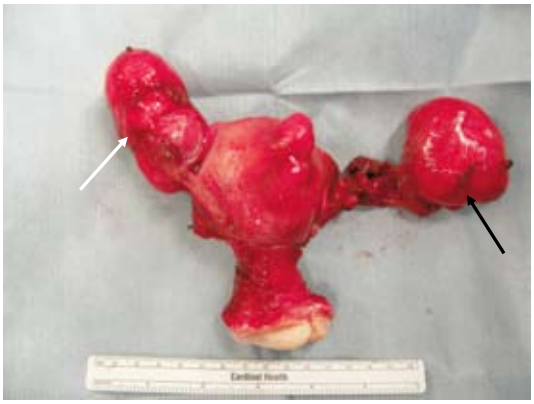

Fig. 3. The uterus and left ovary resected by operation. A 7.0x5.5x5.0 cm uterine myoma (white arrow) and 6.2x5.5x5.2 cm solid tumor (brack arrow) was detected.

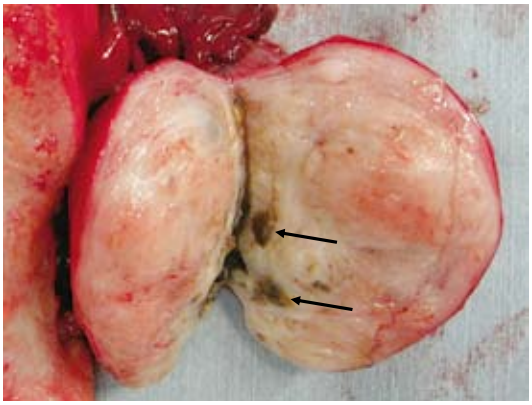

Fig. 4. Macroscopic appearance of the cut surface of the tumor.
The cut surface is smooth and the color is light yellow, with apoplexy in a portion (black arrow).

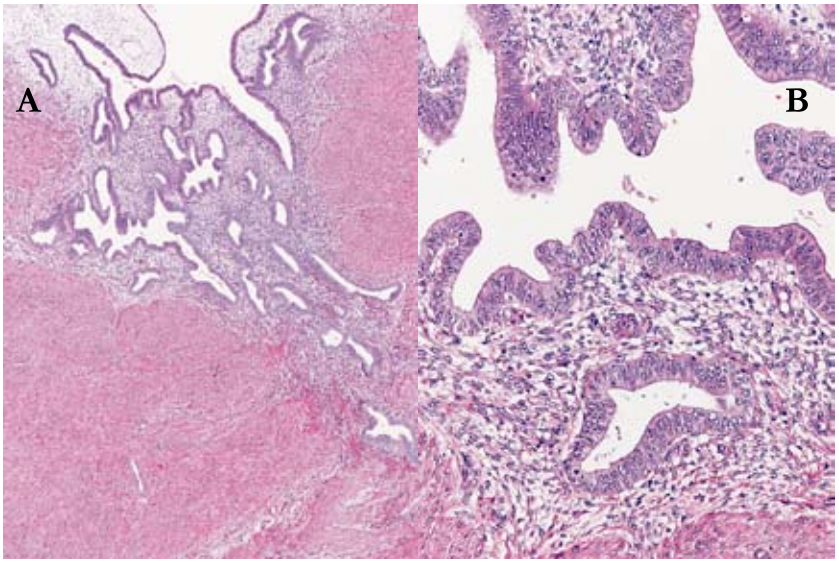

Fig. 5. Histological findings of the left ovary. (H.E. A:x40, B: x100)
The tumor is composed of benign glands and smooth muscle cells.

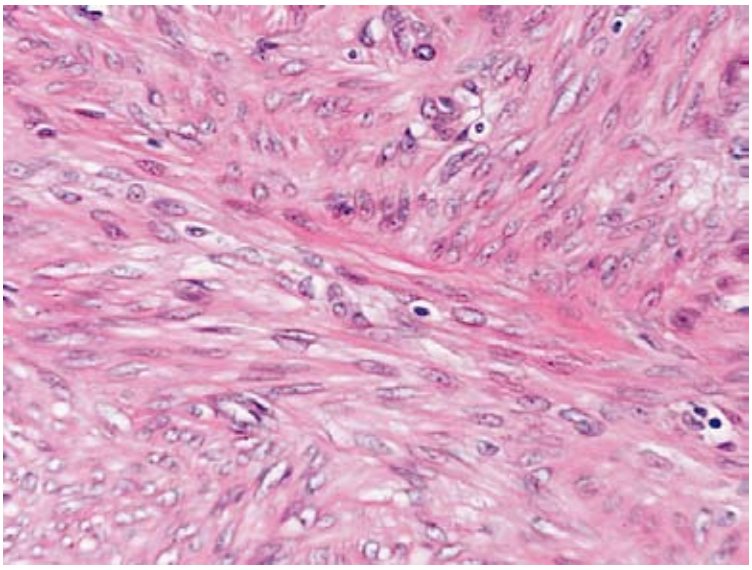

Fig. 6. Histological findings of the left ovary. (H.E. x400)
These smooth muscle bundles were without atypia and significant mitotic activity.

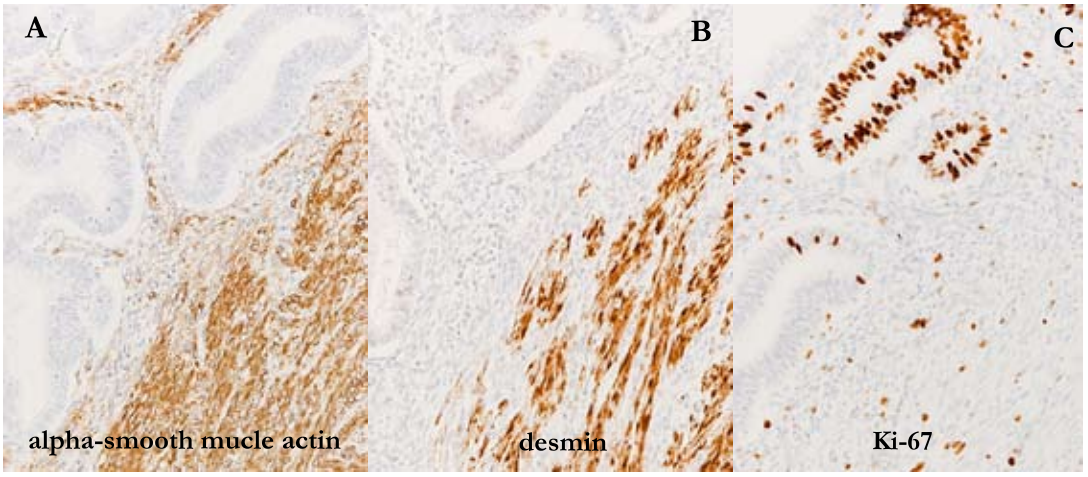

Fig. 7. Immunohistochemical staining of the left ovary. (H.E. x400)
It showed strong positive for alpha-smooth mucle actin (A) and desmin (B), but slightly positive for Ki-67 (C).

DISCUSSION

Adenomyomas are benign tumors and they typically originate within the uterus. On the other hand, extrauterine adenomyomas are rare tumors. They may arise from the broad ligament, from the fallopian tube, or from the ovary. Particularly, in the case of primary ovarian adenomyoma only 5 cases have been reported in the world (1-4). In 1986, McDougal *et al.*, reported the first case of ovarian adenomyoma(1), it was found in an endometriotic cyst of the right ovary. The ages of these five cases were from 36 to 60 years old. The age of our case was also in this range.

Due to their low prevalence, very few imaging studies of ovarian smooth muscle tumors have been published(5). The main tumors in differential

diagnosis are fibroma and thecoma of the ovary (4, 6). In our case, we also thought preoperative diagnosis of the ovarian tumor was a thecoma, but we were not able to completely deny a malignant tumor because the inside of the solid tumor was heterogeneous indicating a partial bleeding and degeneration. Adenomyoma is a benign tumor. So it is thought to be very significant preoperatively to distinguish this from a malignancy. About the image diagnosis, further accumulation of such cases is necessary in the future.

In conclusion, adenomyoma of the ovary is a very rare tumor. But we were not able to completely exclude the possibility of a malignant tumor because of the solid tumor. For example, when we observed the image of the myoma with partial bleeding, we thought that this tumor should also be taken into consideration.

REFERENCES

1）McDougal RA, Roth LM: Ovarian adenomyoma associated with an endometriotic cyst. South Med J 79: 640-642, 1986

2）Bayar U, Demirtas E, Usubutun A, Basaran M, Esinler I, Yarali H: Ovarian adenomyoma following gonadotrophin treatment for infertility. Reprod Biomed Online 13: 676-679, 2006

3）Api O, Ergen B, Gul AE, Ergen C, Unal O, Turan C: Primary ovarian adenomyoma in a woman with endometrial polyp: a case report and review of the literature. Arch Gynecol Obstet 280: 445-448, 2009

4）Mandal S, Mahajan D, Khurana N: Ovarian adenomyoma mimicking an ovarian malignancy: a case report with literature review. Int J Surg Pathol 17: 38-40, 2009

5）San Marco L, Londero F, Stefanutti V, Costa L, Rocco M: Ovarian leiomyoma. Case report. Clin Exp Obstet Gynecol 18: 145-148, 1991

6）Guney M, Ozsoy M, Oral B, Mungan T, Kapucuoglu N: Unilateral primary ovarian leiomyoma in adolescent: a case report. Arch Gynecol Obstet 275: 507-510, 2007