Case Report

Adult-Type Granulosa Cell Tumor, an Unusual Site of Recurrent Tumor at The Sigmoid Colon: A Case Report and Literature Review

Thita Intralawan MD[1], Kanapon Pradniwat MD[2]

[1] Department of Surgery, Ratchaburi Hospital, Ratchaburi, Thailand

[2] Department of Pathology, Ratchaburi Hospital, Ratchaburi, Thailand

Granulosa cell tumor constitutes approximately 1% of all ovarian tumors. Extraovarian spread can occur as a late recurrence. The authors reported a case of 41-year-old woman with adult-type granulosa cell tumor at sigmoid colon. The patient visited the outpatient department for a yearly medical check-up with no specific complaints. She had a history of ovarian cystectomy 12 years prior. A mass in left pelvic cavity was revealed upon transvaginal ultrasonography and computer tomography (CT). At the time of laparoscopy, a well-defined subserosa mass was detected at the antimesenteric side of the sigmoid colon. The right and left ovaries appeared grossly normal. The tumor was successfully removed with laparoscopic sigmoidectomy. The pathologic findings were that of adult-type granulosa cell tumor, which was corresponding to the previous diagnosis made in the ovarian cystectomy specimen 12 years ago. The authors presented this case as an example of late recurrence of ovarian granulosa cell tumor in sigmoid colon, which could potentially be mistaken as other usual colonic tumors in the absence of previous history of ovarian tumor.

Keywords: Colonic granulosa cell tumor; Extraovarian granulosa cell tumor; Sex cord-stromal tumor

Received 6 June 2022 | Revised 19 August 2022 | Accepted 1 September 2022

J Med Assoc Thai 2022;105(10):1015-18

Website: http://www.jmatonline.com

Adult granulosa cell tumor is a type of ovarian sex cord-stromal tumors[1,2]. The tumor usually occurs in perimenopausal women. Recurrence is common, however, recognition of extragonadal recurrence can be challenging, especially after a long period following the first diagnosis. Here, the authors reported a case of a 41-year-old woman diagnosed with an adult granulosa cell tumor involving the sigmoid colon that was treated by laparoscopic sigmoidectomy.

Case Report

The authors reported a case of a 41-year-old female with an incidental finding of a pelvic mass by transvaginal ultrasonography during routine yearly medical check-up. The patient had a history of laparoscopic right ovarian cystectomy of adult-type granulosa cell tumor and left ovarian cystectomy of corpus luteal cyst 12 years prior.

Computed tomography (CT) revealed a 4.2×5.1 cm heterogeneous mass in the left pelvic cavity, which was initially suspected to be at the left adnexa (Figure 1). The basic blood chemistry results were all within normal limits. Tumor markers, including carcinoembryonic antigen (CEA), CA125, alpha-fetoprotein (AFP), and CA19-9, were not elevated. A preoperative diagnosis of recurrent granulosa cell tumor in the left adnexa was made, and the patient was scheduled for laparoscopic left salpingo-oophorectomy.

During the laparoscopy, a well-defined subserosal mass was visualized at the antimesenteric side of the sigmoid colon instead of an ovarian mass. No liver nodules, peritoneal nodules, or ascites were detected. Hence, the operation was converted to a colonoscopy, which was normal, followed by laparoscopic sigmoidectomy. A stapled side-to-side and functional end-to-end colorectal anastomosis was created. The patient's bowel function returned within two postoperative days, and she was discharged four days post-operatively with no immediate or delayed complications.

Correspondence to:

Intralawan T.

Department of Surgery, Ratchaburi Hospital, 85 Somboon Kun Road, Na Muang, Mueang Ratchaburi, Ratchaburi 70000, Thailand.

Phone: +66-87-9892898

Email: Thita_pie@hotmail.com

How to cite this article:

Intralawan T, Pradniwat K. Adult-Type Granulosa Cell Tumor, an Unusual Site of Recurrent Tumor at The Sigmoid Colon: A Case Report and Literature Review. J Med Assoc Thai 2022;105:1015-18.

DOI: 10.35755/jmedassocthai.2022.10.13673

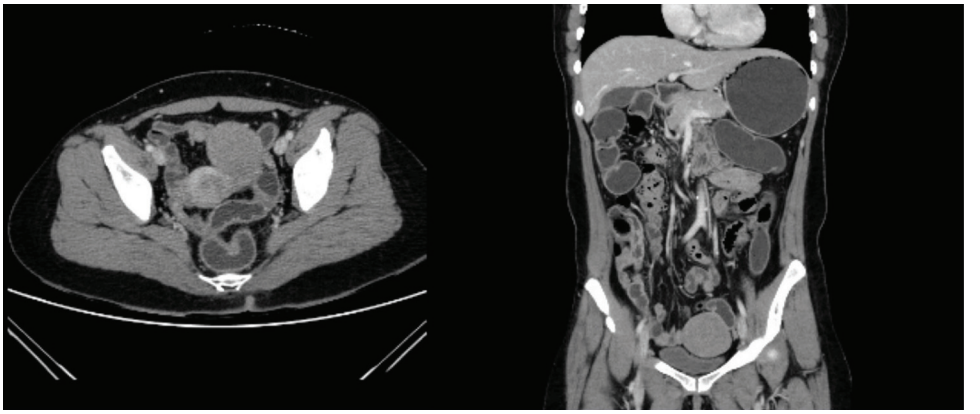

Figure 1. Axial and coronal view of computed tomography (CT) showing a 4.2×5.1 cm heterogeneous mass at the left adnexa.

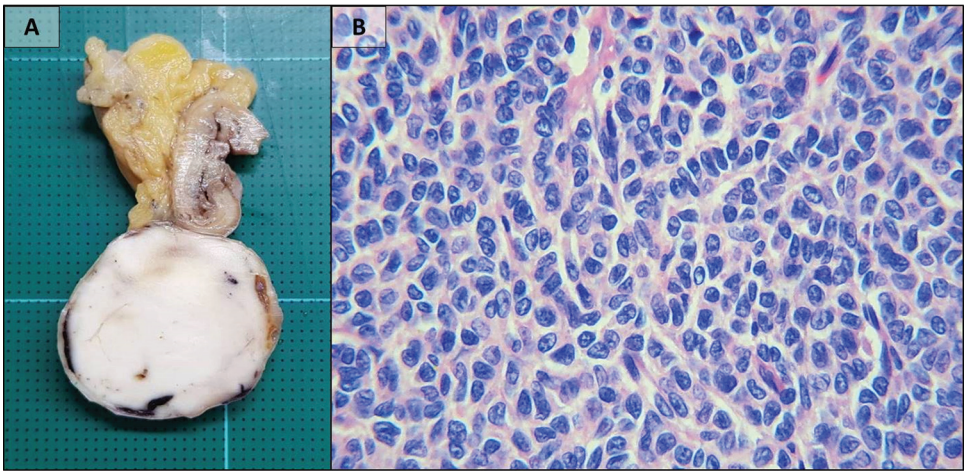

Figure 2. (A) The cut surfaces were soft, homogenous, and yellow-to-greyish white with focal cystic change. (B) Tumor cells show fine chromatin with irregular nuclear contour and coffee bean-like nuclear grooves.

Gross examination demonstrated a 5.4×4.7×4.6 cm well-circumscribed mass in the colonic subserosal area with its epicenter outside the colonic wall. The cut surfaces were soft, homogenous, yellow-to-greyish white with focal cystic change (Figure 2A). Apart from pressure effect of the mass against the colonic muscularis propria, the colonic segment was otherwise unremarkable. Histopathologic examination of the tumor revealed trabecular, microfollicular, and cord patterns of uniform oval-shaped cells with scant cytoplasm. These tumor cells showed fine chromatin with irregular nuclear contour and coffee bean-like nuclear grooves (Figure 2B). Mitotic figures were not seen. The findings were consistent with the previously diagnosed adult granulosa cell tumor in the right ovarian cystectomy.

Discussion

Ovarian granulosa cell tumor harbors 10% to 15% recurrence rate for stage I disease[3]. Because of its propensity for late recurrence, patients with adult-type granulosa cell tumors require long-term follow-up, with the pelvic peritoneum being reported as the most common location for recurrence[4,5]. The longest reported recurrence was 40 years after the initial diagnosis[6].

An unusual phenomenon of collision between granulosa cell tumor and other tumors is also established, including extra-ovarian sites such as colonic adenocarcinoma or uterine cervical lipoleiomyoma[7,8]. A reported recurrent tumor at the colonic site was found as a collision to colonic adenocarcinoma 16 years following initial treatment

of the ovarian granulosa cell tumor[6]. In English literature, the present case is the first reported extragonadal spread to the colon without co-existing or synchronous tumor. Considering the intraperitoneal site of tumor involvement, tumor spreading via the transcoelomic route is most likely, as with ovarian carcinoma. However, the true mechanism of peritoneal seeding of sex-cord stromal tumors is unknown. A study of ovarian cancer dissemination using an animal model (mice) described two major steps, adhesion to peritoneal mesothelium, and invasion into submesothelial organ parenchyma to acquire transcoelomic metastasis[9]. The authors hypothesize that penetration through the mesothelial layer allows tumor growth in the colonic subserosa. Local angiolymphatic entry of tumor cells to seeding in pelvic organs in the vicinity may also be postulated. However, lack of lymphadenopathy at the time of recurrence does not support this consideration. Besides, lymphatic metastases of the ovarian sex-cord stromal tumor, although possible, rarely occur[10] as opposed to epithelial ovarian tumors.

In the absence of a prior diagnosis of ovarian granulosa cell tumor, this colonic mass could have been misdiagnosed as an extraovarian granulosa cell tumor. This rare entity is believed to originate from ectopic gonadal tissue along the embryonic route of the genital ridge. Tumor sites reported in the literature include broad ligament, retroperitoneum, omentum, adrenal gland, and mesentery[2,4,5,11-17].

Granulosa cell tumors usually cause high estrogen levels. Symptoms depend on the patient's age and menstrual status. Fertile patients may have irregular cycles or menorrhagia, while uterine bleeding and abdominal pain may present in postmenopausal patients. Nevertheless, serum estradiol is not a marker for this condition, as 30% of cases have normal estradiol levels due to the lack of theca cells. Hence, the estradiol level is usually normal in extraovarian granulosa cell tumors[13]. The present patient showed no signs or symptoms of hyperestrogenism, and estradiol level was not collected preoperatively.

The mainstay treatment for granulosa cell tumor is surgery, and in the present case, the patient received laparoscopic sigmoidectomy. Laparoscopic surgery has been a safe and effective method[14], even for reoperations[15]. There is a paucity of information on postoperative therapy, but platinum-based chemotherapy may be a considerable option[16,17]. Moreover, the role of radiation therapy remains controversial[13].

Patients with granulosa cell tumors require long-term follow-up because the relapse can occur ten years after the initial treatment[18]. The longest reported period for recurrence was 40 years after initial diagnosis[6], with the pelvis being reported as the most common location for recurrence.

Prognostic factors of granulosa cell tumor, in decreasing order of strength, include stage, tumor size, nuclear atypia, mitotic activity, and sarcomatoid feature. Most granulosa cell tumors are stage I disease at the time of diagnosis with a 10-year survival rate of 85% to 90%. For stage II or higher, 5-year survival rate is less than 50%. Tumor size larger than 15 cm is the prognostic significance with only 34% absolute 5-year survival rate as opposed to 73% to 100% for stage I disease. Mitotic activity and sarcomatoid feature have been variably associated with higher mortality[19].

Conclusion

The authors reported the first case of adult granulosa cell tumor in the sigmoid colon. The tumor was removed by laparoscopic sigmoidectomy, and the patient was discharged without any complications. Laparoscopic surgery can be considered a safe and feasible approach for this condition.

What is already known on this topic?

Ovarian granulosa cell tumor is rare among all ovarian tumors. Late recurrence is not unusual, but when occurs, could cause confusion with other tumors of affected organs especially at extraovarian sites.

What this study adds?

This case report presents an unusual site of granulosa cell tumor recurrence in the sigmoid colon. Laparoscopic sigmoidectomy allows successful tumor removal.

Ethics approval

This present study was approved by the Human Research Ethics Committee of Ratchaburi Hospital (COE-RBHEC 006/2022).

Informed consent

Written informed consent was received from the patient to publish the data in the present study.

Authors' contributions

TI referred the case to KP, provided the clinical data, initiated writing the case report, and wrote the draft manuscript. KP made the final diagnosis, contributed photographs of gross pathology and

routine histology, was the co-author and edited the manuscript.

Conflicts of interest

The authors declare that they have no competing interests.

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