

Mucinous cystadenoma in an ovarian remnant

Ovariye remnant (over kalıntısı) zemininde gelişen müsinöz kistadenom

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ABSTRACT

The ovarian remnant syndrome, a complication of bilateral salpingo-oophorectomy, has progressively received more attention in gynecological surgery literature, in recent years. The syndrome is manifested by pelvic pain, and a palpable or sonographic pelvic mass. In rare cases, patients may present with large masses. Cystic masses or carcinomas are reported to develop in some of the ovarian remnant syndromes after surgery.

We hereby present a 69-year-old white female, complaining of abdominal pain, an enlarging abdominal mass, pollakuria, and urinary incontinence with duration of 1 year, 25 years after bilateral salpingo-oophorectomy. Clinical and radiological evidence of a mesenteric cyst was discovered. The final pathological diagnosis was mucinous cystadenoma in an ovarian remnant. Curiously, this patient had no history of endometriosis, previous pelvic or abdominal surgery excl. hysterectomy, pelvic inflammatory disease, inflammatory bowel disease, dense pelvic adhesions, or difficulty encountered during the previous hysterectomy. This tumor is the largest among all other ovarian remnants published in the international literature.

Women with complaints of abdominal or pelvic mass and /or pain with a history of total abdominal hysterectomy-bilateral salpingo-oophorectomy must be searched carefully and it must be kept in mind that ovarian remnant syndrome can develop without predisposing potential risk factors.

Key words: Mucinous cystadenoma, ovarian remnant syndrome, pelvic mass

ÖZET

Bilateral salpingooforektominin bir komplikasyonu olan overiyen remnant (over kalıntısı) sendromu jinekolojik cerrahi literatüründe artan ilgiyle dikkat çekmektedir. Sendrom, karın ağrısı, ele gelen veya ultrasonla fark edilen pelvik kitle ile kendini gösterir. Nadiren hastalar büyük kitle ile gelirler. Literatürde laparoskopik veya açık cerrahi girişim sonrasında gelişen overiyen remnant sendromu olguları ve bunların bazılarında benign kist veya karsinom geliştiği bildirilmiştir.

Altmış dokuz yaşında kadın hasta, total abdominal histerektomi ve bilateral salpingooforektomi ameliyatından 25 sene sonra, son 1 yıldır karında büyüyen kitle, karın ağrısı, sık idrara çıkma ve idrar kaçırma yakınmaları ile doktora başvurdu. Klinik ve radyolojik olarak en büyük çapı 35 cm olan mezenterik kist ile uyumlu bulguların saptanması üzerine ameliyat edildi. Patolojik tanısı "Ovariye remnant zemininde gelişen müsinöz kistadenom" olarak verildi. Beklenenden farklı olarak hastanın endometriyozis, geçirilmiş pelvik veya abdominal cerrahi, pelvik inflamatuvar hastalık, inflamatuvar barsak hastalığı, pelvik adezyon öyküsü yoktu veya gerçekleştirilen ilk total abdominal histerektomi-bilateral salpingooforektomi esnasında cerrahi olarak zorluk yaşanmamıştı. Bu tümör, uluslararası literatürde yayımlanan overiyen remnantlar arasında en büyük çapta olanıdır.

Daha önceden histerektomi olmuş kadınlarda, karında veya pelviste kitle ya da ağrı yakınması varsa, dikkatlice araştırılmalı ve potansiyel risk faktörleri olmasa bile remnant sendromunun gelişebileceği akıld tutulmalıdır.

Anahtar sözcükler: Müsinöz kistadenom, ovaryan remnant sendromu, pelvik kitle

INTRODUCTION

Ovarian remnant syndrome (ORS) refers to a condition occurring in women who have undergone bilateral salpingo-oophorectomy, with

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or without hysterectomy, in which a remnant ovarian tissue is left behind (1).

Ovarian remnant syndrome is not the same entity as residual ovarian syndrome in which an ovary intentionally left in place during gynecologic surgery, eventually causes pelvic pain (2).

An ovary can usually be removed without difficulty. However when it is attached to other pelvic organs, or the pelvic wall, some of the cortex may adhere to peritoneal surfaces of these structures (3). Risk factors associated with incomplete removal of an ovary and subsequent development of ORS include a history of endometriosis, pelvic inflammatory disease, previous multiple abdominal or pelvic surgeries and pelvic adhesive disease (2,4). In addition, intraoperative conditions (eg, intraoperative bleeding, anatomical variation, or deviation from proper surgical principles) may contribute to incomplete removal of the ovaries (1). Cortical tissue, which has been separated from its major blood supply, may undergo necrosis, cystic degeneration, or neoplastic changes, or it may remain functional, as Shemwell and Weed demonstrated in their experiments with cats in 1970 (5). The condition in which the detached ovarian tissue remains functional is called the ovarian remnant syndrome (3). Patients most frequently present with chronic pelvic pain, pelvic pain associated with a pelvic mass, or an asymptomatic pelvic mass (1).

CASE REPORT

A 69-year-old woman, Gravida: 3, Para: 2, and D&C: 1, comprised the study case. She had a clinical history of total abdominal hysterectomy and bilateral salpingo-oophorectomy which was performed for leiomyoma uteri, 25 years earlier. She had menopausal symptoms (vasomotor symptoms) for only two years after total abdominal hysterectomy and bilateral oophorectomy (TAH-BSO). After that period, the symptoms subsided spontaneously, despite the fact that she had not received hormone rep-

lacement therapy (HRT). She had no clinical history of endometriosis, pelvic inflammatory disease, previous pelvic and/or abdominal surgeries, inflammatory bowel disease, or pelvic adhesive disease. She presented to the gynecology clinic in July 2003 with complaints of abdominal pain, an enlarging abdominal mass, pollakuria, and also urinary incontinence, which had occurred in the previous year.

Laboratory findings: Normal blood biochemistry and a slight degree of anemia (Hb: 11.3 mg/dl, Htc: 34.1%) was detected. Tumor markers were not searched for.

Computerized tomography revealed a 28x35x20 cm, thin walled cystic mass between the portal hilus and symphysis pubis which filled the entire peritoneal cavity. This cystic mass had resulted in compression of the abdominal organs posteriorly. The urinary bladder was particularly compressed infero-posteriorly.

The pre-operative diagnosis was a mesenteric cyst.

Intraoperative findings: Status post-TAH-BSO. The pedunculated cystic mass originated from the rectosigmoid junction, and was not attached to visceral organs. The cyst contained 13 liters of yellow unclear fluid. A portion of the fluid was sent to the pathology laboratory for intra-operative cytological examination. Cytological examination revealed macrophages filled with mucinous material.

Pathological findings:

Macroscopical findings: A pedunculated, oligolocular, smooth, and thin walled cyst measuring 35x25x20 cm. Its content was drained during the surgery, and its residual content was mucoid material (Figure 1).

Microscopical findings: Mucinous cystadenoma with a single layer of mucinous epithelium. In the epithelium no crowding, stratification, hyperchromasia, or mitotic activity were seen (Figure 2). Mucin lakes were seen in luminal spaces. Focally ovarian stroma and corpus albicans were seen (Figure 3). The final histological diagnosis was a mucinous cystadenoma derived

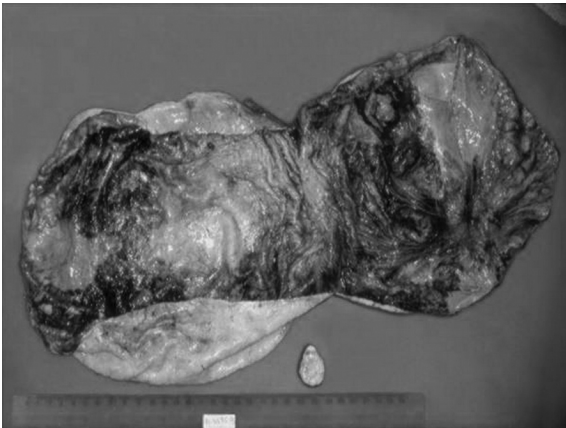


Figure 1. Macroscopic appearance of a cystic mass. Thin walled, unilocular cyst, 35 cm in its largest diameter.

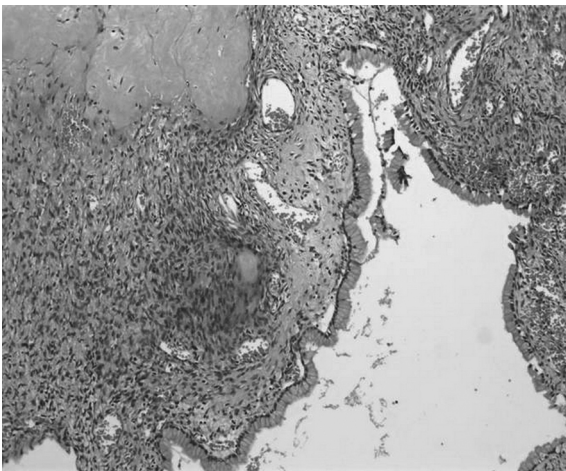


Figure 2. Single layer of columnar cells with abundant intracellular mucin and small basilar nuclei. There is no stratification, hyperchromasia, or mitotic activity (HE x40).

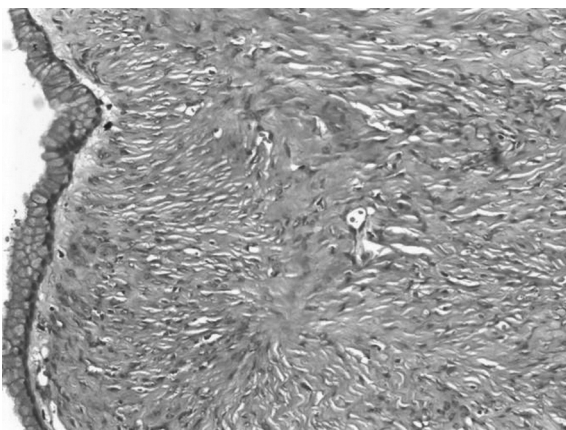


Figure 3. Ovarian tissue and corpus albicans, within wall of a cyst. Single layer of columnar cells with small basilar nuclei (HE x10).

from an ovarian remnant tissue.

DISCUSSION

Ovarian remnant syndrome is a fairly uncommon condition. However its incidence has probably increased in the past four decades (2).

The pre-operative and operative diagnosis of our case was mesenteric cyst. Primary tumors and cysts of the mesentery are quite rare (6). Reports of mesenteric cysts originated from ovarian tissue have been discussed under various titles. Wharton reported that in 1875 and 1887, reports had been published of women who continued to menstruate after bilateral oophorectomy (6). In 1903, Malcolm attributed this phenomenon to incomplete removal of the ovaries. In 1953, a case of intestinal obstruction due to the presence of a corpus luteal cyst within the mesentery of the terminal ileum was reported. This patient had undergone a bilateral oophorectomy (6).

Hormonally active remnants manifest with variable presentations, including pelvic and/or flank pain, dyspareunia, hydronephrosis due to ureteral compression, dysuria, bowel obstruction, painful defecation, and a palpable, tender pelvic mass.

The clinicopathological features of ovarian remnants reported to date in the literature are summarized in Table 1 (7,9,10,11,12,13,14). The largest series of ovarian remnants was reported from Mayo Clinic by Magtibay MP, et al (1). One hundred and eighty-six ORS patients were evaluated. Their mean age was 37.6 years (range, 20-73 years). The most common indications for BSO were endometriosis (57%), an ovarian mass (9%), PID (pelvic inflammatory disease) (7%), or other pathological conditions (28%) which were surgically managed during hysterectomy. The mean numbers of laparotomies and laparoscopies patients had undergone before BSO were 1.4 (range, 0-8) and 0.77 (range 0-10), respectively. In previously mentioned series 147 (79%) of the 186 patients, at least 1

Table 1. Clinicopathological features of ovarian remnants reported in the literature.

Case	Age	No. of previous pelvic surgeries	Clinical diagnosis	Months from prior oophorectomy	Mass size (cm)	Histological diagnosis
(9)1	40	3	Endometriosis	24	5	C. luteum
(9)2	34	2	Endometriosis	24	3	C. luteum
(9)3	38	3	Serous cystadenoma	6	5	C. luteum
(9)4	37	2	Endometriosis	26	3.5	Endometrial cyst
(9)5	46	4	Fibroid, Diverticulitis Ovarian cysts	1	10	Hemorrhagic cyst
(9)6	32	2	Endometriosis	30	2.5	C. luteum
(9)7	34	5	PID, ?, Ovary, ?	10	3.5	Follicular cyst
(9)8	36	6	Endometriosis	12	3.5	C. luteum
(9)9	37	1	Endometriosis	6	7, right 2.5, left	C. luteum bilateral
(9)10	25	1	PID	12	3	Follicular cyst
(9)11	41	2	Endometriosis, Endometrioma	36	3	?
(9)12	34	4	?, ?, ?, Follicular cyst	18	3.5	?
(10)13	32	6	Endometriosis	4	1.5	Follicular cyst
(10)14	33	4	Endometriosis	9	3.5	Ovarian tissue
(10)15	39	9	Adhesions	6	8	Follicular cyst
(10)16	35	5	Endometriosis	9	5	Ovarian tissue
(10)17	37	4	Endometriosis	7	2.1	Follicular cyst
(10)18	35	4	Endometriosis	3	3.2	Ovarian tissue
(10)19	37	5	Endometriosis	8	3.6	Ovarian stroma
(10)20	25	3	Endometriosis	4	2.6	Ovarian stroma
(10)21	31	2	Endometriosis	9	1.4	C. luteal cyst
(10)22	36	6	Endometriosis	8	1.5	Follicular cyst
(10)23	38	7	Endometriosis	12	1	C. luteal cyst
(10)24	48	3	Endometriosis	10	4	Ovarian tissue
(10)25	41	5	Adhesions	15	0.7	C. luteal cyst
(10)26	40	5	Endometriosis	13	3.2	Follicular cyst
(10)27	44	5	Adhesions	46	6.2	Follicular cyst
(10)28	54	3	Adhesions	49	0.8	Ovarian tissue
(10)29	26	6	Endometriosis	20	0.7	C. luteal cyst
(10)30	26	4	Endometriosis	4	1.5	Ovarian tissue
(10)31	41	7	Endometriosis	32	1.2	Ovarian stroma
(11)32	37	4	PID	12	*	?
(11)33	41	2	-	120	*	Ovarian tissue
(11)34	32	1	Endometriosis	?	*	Ovarian tissue
(11)35	42	3	-	12	*	Ovarian tissue
(11)36	34	5	-	12	*	Ovarian tissue
(11)37	42	2	-	24	*	Ovarian tissue
(11)38	24	6	-	24	*	Ovarian tissue
(11)39	44	2	-	12	*	Ovarian tissue
(7)40	70	1	Fibroids	288	?	Adenocarcinoma
(7)41	53	1	Endometriosis	108	12	Adenocarcinoma
(7)42	69	1	Endometriosis	264	8	Carcinoma
(7)43	42	1	PID	?	?	Borderline serous cystadenoma
(7)44	?	1	Dermoid cyst	?	?	Malignant change
(7)45	67	1	Fibroids, Endometriosis	168	26	Borderline mucinous tumor
(12)46	40	1	Fibroid, Endometriosis	12	3	Well differentiated adenocarcinoma
(13)47	37	1	Mucinous carcinoid tumor	12	3	C. luteum, Follicular cyst
(14)48	49	1	Endometriosis	12	7.5, right 3.8, left	Serous cystadenofibroma
(14)49	61	1	?	?	8.7	Serous cystadenoma
(14)50	44	5	Endometriosis	?	8.8	Serous cystadenoma
** 51	69	***0	-	300	35	Mucinous cystadenoma

• Each pelvic mass ranged from 3 to 10 cm (mean: 6 cm) in its largest diameter

** Our case.

*** TAH BSO for leiomyoma uteri. No other pelvic surgeries.

abdominal surgery had been performed before BSO. Histologically, remnant ovarian tissue

was associated with corpus luteum in 78 (42%), endometriosis in 54 (29%), follicular cyst in 12

(7%), simple cyst in 45 (24%), cystadenofibroma in 3 (2%), serous cystadenoma in 2 (1%), and cystadenoma in 1 (0.5%) case respectively.

Symptoms commonly present within 5 years after extirpative surgery (7). Our patient began to experience these symptoms 24 years after extirpative surgery. The majority of women have identifiable cystic structures on ultrasonography, commonly measuring between 3 and 4 cm in their largest diameters (6). To date the largest tumor reported in the literature was 26 cm which was conclusively diagnosed as invasive mucinous carcinoma (7,8). Our patient's tumor was 35 cm in its largest diameter.

Most patients with ORS are generally young and menopausal at the time of BSO. The ovarian tissue remnants in these women often remain functional and continue to secrete substantial concentrations of hormones, including estradiol. Therefore, a young woman with ORS who is not immediately started on estrogen replacement therapy (ERT) after a BSO, often lacks the typical menopausal symptoms observed after abrupt estrogen withdrawal. This clinical information may provide the clinician with clues to the diagnosis of ORS. In the recent series of Mayo Clinic, 37% of patients have not showed any symptoms of estrogen deprivation despite no ERT (1). Our patient had menopausal symptoms for only two years after TAH-BSO and those symptoms had resolved spontaneously, without receiving ERT.

In our case, the mucinous cystadenoma that developed from an ovarian remnant had the largest dimensions among all the tumors developed in ovarian remnant syndrome reported in the literature.

Tumors that have a preoperative diagnosis of mesenteric cyst in women with a history of TAH-BSO could be ovarian remnants. Therefore, these cysts must be examined carefully in the

pathology laboratory and proper sampling is necessary to show residual ovarian tissue.

Women with complaints of an abdominal or pelvic mass and/or pain with a history of TAH-BSO, must be evaluated carefully and it must be kept in mind that the ovarian remnant syndrome can develop without predisposing potential risk factors.

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