

Coexistence of endometriosis in women with mature cystic ovarian teratoma may not be rare.

Heesuk Chae (✉ hschae@jbnu.ac.kr)

Research

Keywords: mature cystic teratoma, endometriosis, concomitant diseases

Posted Date: January 28th, 2020

DOI: <https://doi.org/10.21203/rs.2.21971/v1>

License:  This work is licensed under a Creative Commons Attribution 4.0 International License.

[Read Full License](#)

Version of Record: A version of this preprint was published at Journal of Gynecology Obstetrics and Human Reproduction on November 1st, 2020. See the published version at <https://doi.org/10.1016/j.jogoh.2020.101786>.

Abstract

Background

We investigated the incidence of endometriosis in women with mature cystic ovarian teratoma and analyzed the clinicopathologic features of this occurrence.

Methods

From January 2017 through December 2018, we retrospectively studied 71 women who had undergone cystectomy for mature cystic ovarian teratoma (n = 55, teratoma group) and coexistence of endometriosis and mature cystic ovarian teratoma (n = 16, complex group). Serum anti-Müllerian hormone (AMH) levels were measured preoperatively and one month after surgery.

Results

Sixteen (22.54%) patients had coexistence of endometriosis and mature cystic ovarian teratoma (complex group); 55 patients had mature cystic teratoma alone (teratoma group). Early-stage endometriosis (stage I) was present in eight patients and advanced-stage endometriosis (stage III or IV) was present in eight. In five cases (31.25%), the coexistence of endometrioma and mature cystic teratoma in the same ovary was observed. The mean operation time was significantly shorter in the teratoma group than in the complex group (61.02 ± 22.74 vs. 86.31 ± 35.35 min, $p = 0.007$). The complex group had more dysmenorrhea (43.8% vs. 7.3%, $p = 0.002$) and a significantly higher rate of decrease in serum anti-Müllerian hormone (AMH) levels (33.06 ± 24.92 vs. $16.31 \pm 28.17\%$, $p = 0.048$).

Conclusion(s)

The prevalence of coexisting endometriosis and mature cystic ovarian teratoma may be underestimated. Patients with this rare concurrence may present with worsening dysmenorrhea and damage to ovarian reserve after surgery may be greater in patients with coexisting endometriosis than in patients with mature cystic teratoma alone.

Background

Mature cystic ovarian teratomas (MCTs) or dermoid cysts are the most common ovarian tumor in women of reproductive age and account for about 20% of ovarian tumors [1, 2]. MCTs account for approximately 70% of benign ovarian tumors in women under 30 year of age and 50% of pediatric tumors [3, 4]. They occur bilaterally in 10–15% of cases [1]. These tumors are usually asymptomatic and are often discovered incidentally during routine physical or during radiologic examinations [5, 6] The proportion of asymptomatic tumors has been reported to range from 6–64.5% [7, 8, 9, 10]. In addition, MCTs have a tendency to grow slowly with an estimated growth rate of 1.67 mm/year [1]. Therefore, recently, expectant management of MCTs as an alternative to surgical management has been proposed [1, 3].

Endometriosis is a chronic inflammatory condition defined by the presence of endometrial tissue with glands and stroma outside the uterus [11]. Endometriosis is estimated to affect 10% of women in the reproductive age group [12]. The incidence of endometriosis in women with dysmenorrhea is approximately 40–60%, whereas it is up to 20–30% in women with subfertility [11, 13].

Although MCTs and endometriosis are common gynecologic disorders occurring in women of reproductive age, concomitant MCTs and endometriosis are notably rare and have been reported in few case reports. However, the author has observed some patients with unusual concomitant MCTs and endometriosis.

The aim of this study was to determine the incidence of endometriosis in patients with a diagnosis of MCT who underwent laparoscopy and to clarify further the clinical manifestations of this condition.

Methods

Patients

All patients operated on for MCTs at the Departments of Obstetrics and Gynecology of the Chonbuk National University Hospital from January 2017 through December 2018 were included in this retrospective study. This study was approved by the Institutional Review Board of Chonbuk National University Hospital (File number: 2018-05-018-006). The requirement for obtaining informed consent from the patients was not applicable owing to the retrospective nature of the study.

The inclusion criteria were as follows: (1) mature cystic ovarian teratoma confirmed by pathology and (2) endometriosis diagnosed by a pathology specimen and/or a visual confirmation of endometriosis lesions. The exclusion criteria were as follows: (1) patients with a history of previous adnexal surgery, and (2) any hormonal treatment, such as oral contraceptives, within six months prior to surgery. All patients were divided into the following groups: patients with MCTs alone (teratoma group) and patients with concomitant MCTs and endometriosis (complex group).

Data regarding the patient's age, parity, body mass index (BMI), preoperative serum cancer antigen (CA) 125 and carbohydrate antigen (CA) 19 – 9 level, hemoglobin (Hb) level, serum anti-Müllerian hormone (AMH) level, site and size of cyst, stage of endometriosis and pathology reports were retrieved from operative reports and medical records. Blood samples were preoperatively measured using radioimmunoassay for CA 125 and CA 19 – 9. The cutoff values for CA 125 and CA 19 – 9 was 35 U/mL. The Hb concentration was determined both preoperatively and 24 hours after surgery.

The rate of decrease of Hb concentrations was calculated using the following formula:

$$\left[\frac{\text{preoperative Hb concentration} - \text{postoperative Day 1 Hb concentration}}{\text{preoperative Hb concentration}} \right] \times 100(\%).$$

The serum AMH concentrations were measured preoperatively and one month after the surgery. The serum AMH level was determined using an enzyme-linked immunoassay kit, AMH Gen II (Beckman Coulter, USA).

The rate of decrease of serum AMH levels was calculated using the following formula:

$$\frac{[(\text{preoperative serum AMH level} - \text{postoperative month 1 AMH level}) / (\text{preoperative serum AMH level})] \times 100(\%)}.$$

Cyst size was determined by preoperative imaging studies including ultrasonography, computed tomography (CT), and magnetic resonance imaging (MRI). In the case of bilateral ovarian cysts, the sum of their largest diameters was defined as the cyst size. The stage of endometriosis was defined according to the revised American Fertility Society classification [14].

Statistical analysis

R language ver. 3.3.3 (R Foundation for Statistical Computing, Vienna, Austria) and T&F program ver. 3.0 (YooJin BioSoft, Korea) were used for all statistical analyses. Data was expressed as mean \pm standard deviation for continuous variables. The Mann-Whitney U test or Kruskal-Wallis H test were used to test the difference of continuous clinical measurements between the teratoma group and the complex group composed of ovarian endometrioma and peritoneal endometriosis. For post hoc analysis, the Bonferroni method was applied to adjust p value. For categorical variables, data was expressed as sample number and percentage, N (%), and p values were computed using the chi-squared test or Fisher's exact test.

Results

A total of 71 patients were included, of whom 55 had teratoma (teratoma group) and 16 had a coexistence of endometriosis and MCTs (complex group). Table 1 presents their demographic and clinical characteristics, including serum AMH levels and surgery data.

Table 1

Demographics and clinical characteristics of patients with mature cystic teratoma alone (teratoma group) and with coexisting mature cystic teratoma and endometriosis (complex group)

Variable	Subgroup	N(%)	Teratoma	Complex	p value
Sample No (%)		71 (100)	55 (77.46)	16 (22.54)	
Age (years)		71 (100)	26.44 ± 6.82	27.38 ± 5.60	0.436
BMI (kg/m ²)		71 (100)	23.67 ± 5.81	22.74 ± 4.48	0.685
Parity (n)		71 (100)			0.758 [§]
	0	55 (77.5)	41 (74.5)	14 (87.5)	
	1	6 (8.5)	5 (9.1)	1 (6.2)	
	2	5 (7)	5 (9.1)	0 (0)	
	3	3 (4.2)	2 (3.6)	1 (6.2)	
	4	1 (1.4)	1 (1.8)	0 (0)	
	5	1 (1.4)	1 (1.8)	0 (0)	
CA125 (U/mL)		71 (100)	11.97 ± 7.30	19.03 ± 16.49	0.066
CA19-9 (U/mL)		71 (100)	41.88 ± 92.45	71.51 ± 121.67	0.218
Largest diameter of cyst (cm)		71 (100)	5.85 ± 3.23	7.73 ± 3.19	0.020*
Laterality of cyst (n)		71 (100)			0.368
	Unilateral	63 (88.7)	50 (90.9)	13 (81.2)	
	Multilateral	8 (11.3)	5 (9.1)	3 (18.8)	
Locularity of cyst (n)		71 (100)			0.054

Variable	Subgroup	N(%)	Teratoma	Complex	p value
	Unilocular	51 (71.8)	43 (78.2)	8 (50)	
	Multilocular	20 (28.2)	12 (21.8)	8 (50)	
Preop Hb level (g/dl)		71 (100)	13.21 ± 0.90	13.14 ± 0.78	0.689
Postop Hb level (g/dl)		71 (100)	11.89 ± 0.95	11.58 ± 0.84	0.298
Decrease of Hb level (g/dl)		71 (100)	1.31 ± 0.85	1.56 ± 0.64	0.253
Rate of decrease of serum Hb levels (%)		71 (100)	9.82 ± 6.30	11.85 ± 4.69	0.186
Operation time (min)		70 (100)	61.02 ± 22.74	86.31 ± 35.35	0.007**
Stage of EMS		16 (100)			NA
	I	8 (50)	0 (0)	8 (50)	
	III	7 (43.8)	0 (0)	7 (43.8)	
	IV	1 (6.2)	0 (0)	1 (6.2)	
Preop AMH level (ng/mL)		66 (100)	5.38 ± 3.80	4.55 ± 4.08	0.209
Postop AMH level (ng/mL)		65 (100)	3.97 ± 2.52	3.01 ± 2.92	0.077
Decrease of AMH level (ng/mL)		63 (100)	0.99 ± 1.86	1.54 ± 1.82	0.180
Rate of decrease of serum AMH levels (%)		63 (100)	16.31 ± 28.17	33.06 ± 24.92	0.048*
Clinical presentation (n)		71 (100)			
	Abdominal pain	12 (16.9)	9 (16.4)	3 (18.8)	>0.99 [#]
	Dysmenorrhea	11 (15.5)	4 (7.3)	7 (43.8)	0.002 ^{#**}
	Incidental finding	29 (40.8)	24 (43.6)	5 (31.2)	0.550 [#]

Variable	Subgroup	N(%)	Teratoma	Complex	p value
	Irregular vaginal bleeding	19 (26.8)	18 (32.7)	1 (6.2)	0.074 [#]
Continuous variables are expressed as mean ± SD.					
Categorical variables are expressed as sample number and %.					
p-values for continuous variables are computed using Mann-Whitney U test (i.e. Wilcoxon rank-sum test).					
p-values for categorical variables are computed using Fisher's exact test, chi-squared test or two sample proportion test.					
p-value [§] computed using chi-squared test					
p-value [#] computed using two sample proportion test					
N (%): computed in the total sample or subgroups excluding missing data					
BMI: body mass index					
CA 125: cancer antigen 125					
CA 19-9: carbohydrate antigen 19-9					
Hb : hemoglobin					
AMH: anti-Müllerian hormone					
NA: not available					
* p <0.05, ** p <0.01					

No other parameters concerning age, parity, body mass index, laterality of cyst, and locularity of cyst differed significantly between the two groups. Serum CA 125 and CA 19 - 9 values also showed no significant differences between the two groups. The diameter of the cyst was significantly larger in the complex group than in the teratoma group ($p = 0.02$). The mean operation time in the teratoma group was significantly shorter with a mean of 61.02 ± 22.74 min vs. 86.31 ± 35.35 min in the complex group ($p = 0.007$). However, there was no significant difference in intraoperative blood loss between the two groups. The rate of decrease in serum AMH levels was significantly higher in the complex group, with a median of 33.06 ± 24.92 vs. 16.31 ± 28.17 for the teratoma group ($p = 0.048$).

Further, a comparison of the initial clinical presentation in each group during the visit to doctor is shown in Fig. 1. The majority of patients in the teratoma group were asymptomatic (24 cases, 43.6%) and the lesions were discovered incidentally during routine ultrasound examination, whereas the most common

symptom in the complex group was dysmenorrhea (7 cases, 43.8%). Dysmenorrhea was more common in the complex group than in the teratoma group (43.8% vs. 7.3%, respectively; $p = 0.002$).

The complex group was divided into two subgroups according to lesion type of endometriosis: ovarian endometrioma and peritoneal endometriosis. To compare further which pairs of groups were significantly different, post hoc comparison analysis was performed among the three groups: teratoma group, ovarian endometrioma subgroup, and peritoneal endometriosis subgroup (Table 2). Significant differences in the largest diameter of cyst ($p = 0.047$), operation time ($p = 0.02$), postoperative serum AMH level ($p = 0.045$) were found among the three groups. However, the post hoc comparison analysis showed that the postoperative serum AMH level alone was significantly lower in the ovarian endometrioma subgroup than in the teratoma group ($p = 0.039$). The difference in the rate of decrease in the serum AMH levels among the three groups was a borderline level of statistical significance ($p = 0.059$), but in the post-hoc comparison analysis, the differences in each subgroups were not statistically significant although the difference was the largest between the teratoma group and the ovarian endometrioma subgroup ($p = 0.065$).

Table 2

Mean comparison analysis among teratoma group and the complex group (ovarian endometrioma and peritoneal endometriosis)

Variable	Teratoma (N = 55, 77.5%)	Ovarian endometrioma (N = 5, 7%)	Peritoneal endometriosis (N = 11, 15.5%)	p value	p value [1]	p value [2]	p value [3]
Age (years)	26.44 ± 6.82	28.2 ± 5.54	27 ± 5.85	0.659	> 0.99	> 0.99	> 0.99
BMI (kg/m ²)	23.67 ± 5.81	24.01 ± 7.12	22.17 ± 2.94	0.885	> 0.99	> 0.99	> 0.99
CA125 (U/mL)	11.97 ± 7.3	22.08 ± 13.42	17.65 ± 18.15	0.120	0.193	0.791	> 0.99
CA19-9 (U/mL)	41.88 ± 92.45	112.77 ± 177.63	52.75 ± 91.35	0.262	0.328	> 0.99	0.856
Largest diameter of cyst (cm)	5.85 ± 3.23	8.98 ± 3.45	7.16 ± 3.06	0.047*	0.112	0.349	> 0.99
Preop Hb level (g/dl)	13.21 ± 0.9	12.56 ± 0.66	13.41 ± 0.7	0.132	0.210	> 0.99	0.147
Postop Hb level (g/dl)	11.89 ± 0.95	11.04 ± 0.76	11.83 ± 0.77	0.147	0.153	> 0.99	0.294
Decrease of Hb level (g/dl)	1.31 ± 0.85	1.52 ± 0.54	1.58 ± 0.71	0.515	> 0.99	> 0.99	> 0.99
Rate of decrease of serum Hb levels (%)	9.82 ± 6.3	12.1 ± 4.27	11.73 ± 5.06	0.388	0.821	> 0.99	> 0.99
Operation time (min)	61.02 ± 22.74	86.8 ± 22.15	86.09 ± 40.97	0.020*	0.089	0.137	> 0.99
Preop AMH level (ng/mL)	5.38 ± 3.8	2.75 ± 1.8	5.67 ± 4.79	0.167	0.176	> 0.99	0.480
Postop AMH level (ng/mL)	3.97 ± 2.52	1.48 ± 0.94	3.96 ± 3.37	0.045*	0.039*	> 0.99	0.245
Decrease of AMH level (ng/mL)	0.99 ± 1.86	1.27 ± 0.93	1.71 ± 2.26	0.391	0.855	0.995	> 0.99
Rate of decrease of serum AMH levels (%)	16.31 ± 28.17	43.55 ± 15.29	26.49 ± 28.33	0.059	0.065	> 0.99	0.574

Variable	Teratoma (N = 55, 77.5%)	Ovarian endometrioma (N = 5, 7%)	Peritoneal endometriosis (N = 11, 15.5%)	p value	p value [1]	p value [2]	p value [3]
Variables are expressed as mean ± SD, and p values are computed using Kruskal–Wallis H test							
p-values ^[1] : computed between the teratoma group and the ovarian endometrioma subgroup using a post-hoc analysis algorithm of Bonferroni							
p-values ^[2] : computed between the teratoma group and the peritoneal endometriosis subgroup using a post-hoc analysis algorithm of Bonferroni							
p-values ^[3] : computed between the ovarian endometrioma subgroup and the peritoneal endometriosis subgroup using a post-hoc analysis algorithm of Bonferroni							
BMI: body mass index							
CA 125: cancer antigen 125							
CA 19-9: carbohydrate antigen 19-9							
Hb : hemoglobin							
AMH: anti-Müllerian hormone							
* p <0.05							

The individual clinicopathologic features of the 16 patients with coexisting MCTs and endometriosis are shown in Table 3. In five patients, the coexistence of MCTs and ovarian endometrioma in a single ovary was observed. In case 5 and case 13, the coexistence of MCTs and ovarian endometrioma with histologic admixture in an ovarian cyst was observed, whereas in the other cases, separate teratomatous and endometrial lesions in a single ovary were observed. Further, case 2 had a combination of serous cystadenoma and teratoma, whereas, case 6 had teratoma with coincidental ovarian endometrioma and serous cystadenoma as a separate pathology.

Table 3

Summary of the clinicopathologic characteristics of the 16 patients with coexistent endometriosis and mature cystic ovarian teratoma.

Case no.	Age (yr)	CA125 (U/mL)	CA19-9(U/mL)	Presentation	Stage	Pathology
1	26	20.1	47.1	Dysmenorrhea	I	Teratoma, pelvic endometriosis
2	25	12.2	321.79	Dysmenorrhea	I	Teratoma, pelvic endometriosis, serous cystadenoma
3	26	5.8	26.44	Dysmenorrhea	I	Teratoma, pelvic endometriosis
4	36	70.2	71.04	Incidental findings	IV	Teratoma, pelvic endometriosis
5	27	19.2	52.19	Abdominal pain	IV	Teratoma, ovarian endometrioma
6	25	24.2	18.17	Abdominal pain	III	Teratoma, ovarian endometrioma, seous cystadenoma
7	29	12.9	26.44	Dysmenorrhea	I	Teratoma, pelvic endometriosis
8	22	17.4	12.61	Dysmenorrhea	I	Teratoma, pelvic endometriosis
9	38	9.2	2.69	Dysmenorrhea	II	Teratoma, pelvic endometriosis
10	26	19.9	3.98	Dysmenorrhea	I	Teratoma, pelvic endometriosis
11	19	12.3	28.94	Incidental findings	I	Teratoma, pelvic endometriosis
12	25	29.5	429.19	Incidental findings	III	Teratoma, ovarian endometrioma
13	38	1	16.8	Incidental findings	III	Teratoma, ovarian endometrioma
14	26	36.5	47.49	Incidental findings	III	Teratoma, ovarian endometrioma

CA 125: cancer antigen 125

CA 19-9: carbohydrate antigen 19-9

Case no.	Age (yr)	CA125 (U/mL)	CA19-9(U/mL)	Presentation	Stage	Pathology
15	29	8.3	24.82	Abdominal pain	III	Teratoma, pelvic endometriosis
16	21	5.8	14.41	Irregular vaginal bleeding	I	Teratoma, pelvic endometriosis
CA 125: cancer antigen 125						
CA 19-9: carbohydrate antigen 19-9						

Discussion

The present study, to the author's knowledge, is the first attempt to investigate the incidence of the coexistence of MCTs and endometriosis and to evaluate the differential clinical characteristics of patients with this complex condition.

Although MCTs and endometriosis are benign diseases that commonly affect women of reproductive age, the coexistence of MCTs and endometriosis, especially in a single ovary, is rare [6, 15, 16]. However, in this study, this coexistence was not rare. Although this study included a relatively small number of patients, the incidence of coexisting MCTs and endometriosis was 22.54% and its incidence in a single ovary was 7.04%. Recently, Matalliotaki et al. proposed that endometriosis is linked with an increased risk of benign gynecological tumor, such as ovarian cyst, adenomyosis and uterine leiomyomas [17]. In that study, dermoid cysts (1.5%) were observed in women with endometriosis. This data and the present study's findings suggest that coexisting MCTs and endometriosis may be more common than previously reported.

MCTs are often discovered incidentally in asymptomatic women [1, 10]. Patients with MCTs develop symptoms such as discomfort or pain when MCTs are complicated by torsion, rupture, and infection [6]. However, various types of pain are associated with endometriosis: dysmenorrhea, deep dyspareunia, and pelvic pain unrelated to intercourse or menstruation, such as pain during defecation or urinating [18]. Sonographic examination is the initial method for identifying adnexal cystic structures, but MCTs can be occasionally difficult to distinguish from endometriomas, hemorrhagic cyst, and mucinous cystic neoplasm based on ultrasonography alone [19], and additional modalities including CT or MRI may be needed. However, the complexity and rarity of coexisting MCTs and endometriosis make it more difficult to diagnose. This study demonstrated a high prevalence of coexisting MCTs and endometriosis in those presenting with dysmenorrhea. The occurrence of dysmenorrhea was more common among women with this complex condition, whereas most MCTs occurred without clinical symptoms. The diagnosis of concomitant endometriosis in patients with MCTs rests on a high index of suspicion. Thus, gynecologic symptoms such as worsening dysmenorrhea may be a diagnostic clue.

Further, in this study, the operative time was significantly shorter in patients with MCTs alone than in patients with coexisting MCTs and endometriosis. In addition, in one patient with a histologic admixture of MCTs and endometriosis in an ovarian cyst, the procedure was converted to laparotomy because of dense adhesions and a larger ovarian cyst (exceeding 10 cm in diameter). These observations indicate that concurrent endometriosis with MCTs makes surgery much more difficult.

The recommended management of MCTs is generally surgical removal because of a large cyst size, risk of torsion, and concern for occult malignancy. Traditionally, MCTs have been removed by elective surgery and this procedure accounts for 20–35% of the surgical removal of ovarian tumors [1, 20]. MCTs are the most common germ cell tumors of the ovary in women of reproductive age, and account for approximately 70% of benign ovarian neoplasms in women under 30 years of age [21, 22]. The major concern among these younger women is future fertility. Therefore, the appropriate treatment of MCTs in children and adolescents remains unclear. A retrospective study by Hoo et al. found that the success rate of expectant management of MCTs was high and suggested that this approach be considered as a viable alternative to surgical management [1]. O'Neill et al. suggested that close follow-up without intervention should be considered to preserve ovarian function and future fertility in children and adolescents with MCTs [3]. However, endometriosis found in conjunction with MCTs makes justifying the expectant approach even more challenging. Endometriosis is a common disorder in adult women. Goldstein et al. reported a 47% prevalence of endometriosis in adolescents undergoing laparoscopy for pelvic pain [23,24]. Several studies reported a reduced number of retrieved oocytes for in vitro fertilization and premature ovarian failure after surgery for endometriosis [25,26]. The present study's results showed that the rate of decline of serum AMH levels was significantly higher in patients with coexisting MCTs and endometriosis than in patients with MCTs alone. The study's small sample size makes generalizing the conclusion difficult, but this result suggests that surgical cystectomy for this condition in a single ovary could cause more damage to the ovarian reserve.

The malignant transformation of MCTs is an uncommon complication, with an incidence of 0.17–2% [3, 4]. However, endometriosis is present in about 10–15% of ovarian cancers and is usually associated with endometrioid carcinoma or clear cell adenocarcinoma [27]. Moreover, Anteby et al. reported that the recurrence rate of MCTs following cystectomy was 3–4% [28]. Recurrence rates of endometrioma after surgical excision vary considerably, ranging from 6 percent through 30 percent [29,30]. Consequently, patients with coexisting MCTs and endometriosis may have an increased risk of malignancy and recurrence.

Further, a collision tumor is defined as the coexistence of two adjacent, but histologically distinct tumor components [31]. Collision tumors have been reported in various organs including the esophagus, stomach, liver, bone, kidney, central nervous system and lung, and so on, but such tumors are relatively rare in the ovary [32]. In addition, the most common histologic combination of a collision tumor in the ovary is the coexistence of teratoma and mucinous tumors; therefore a combination of teratoma with serous cystadenoma is rare [33].

This study had several limitations. First, not all patients had pathology-confirmed endometriosis. Second, the reverse case—the presence of MCTs in patients with endometriosis— was not considered. In addition, the number of patients analyzed was small.

Conclusions

The findings of this study suggest that the coexistence of MCTs and endometriosis is not as rare as previously thought. This study also showed that among women with MCT, those with coexisting endometriosis had marked dysmenorrhea compared to women with MCT alone and that damage to ovarian reserve after cystectomy in women with coexisting endometriosis may be greater than in women with MCT alone. These results can not only help categorizing this complex disease, but can also be used to standardize its treatment. Nevertheless, further prospective studies are required to verify whether the coexistence of MCTs and endometriosis is indeed not rare.

Declarations

Ethics approval and consent to participate

This study was approved by the Institutional Review Board of Chonbuk National University Hospital (IRB File No. 2018-05-018-006). Due to the retrospective design of the study an informed consent is not applicable.

Consent for publication

Not applicable

Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests.

Funding

We did not receive any funding for this study.

Authors' contributions

HC performed data collection, data analysis, and manuscript writing and editing. The author read and approved the final manuscript.

Acknowledgements

Not applicable

Abbreviations

BMI

body mass index

CA 125

cancer antigen 125

CA 19 – 9

carbohydrate antigen 19 – 9

Hb

hemoglobin

AMH

anti-Müllerian hormone

References

1. Hoo WL, Yazbek J, Hollland T, Mavrelou D, Tong EN, Jurkovic D. Expectant management of ultrasonically diagnosed ovarian dermoid cysts: is it possible to predict outcome? *Ultrasound Obstet Gynecol* 2010;36:235-40.
2. Yan L, Li M, Zhang B, Xu X, Xu Z, Han T, et al. Effect of ovarian dermoid cyst excision on ovarian reserve and response: Insights from in vitro fertilization. *Gynecol Minim Invasive Ther* 2016;5:161-5.
3. O'Neill KE, Cooper AR. The approach to ovarian dermoids in adolescents and young women. *J Pediatr Adolesc Gynecol* 2011;24:176-80.
4. Ayhan A, Bukulmez O, Genc D, Karamursel BS, Ayhan A. Mature cystic teratomas of the ovary: case series from one institution over 34 years. *Eur J Obstet Gynecol Repro Biol.* 2000;33:153-7.
5. Stany MP, Hamilton CA. Benign disorders of the ovary. *Obstet Gynecol Clin North Am* 2008;35:271-84.
6. Chae H, Rhee C. Endometriosis coexisting with mature cystic teratoma in the same ovary and ectopic pregnancy of left fallopian tube: a rare coexistence. *Clin Case Rep* 2015;3:315-8.
7. Kim MJ, Kim NY, Lee DY, Yoon BK, Choi D. Clinical characteristics of ovarian teratoma: age-focused retrospective analysis of 580 cases. *Am J Obstet Gynecol* 2011;205:32 e1-4.
8. Peterson WF, Prevost EC, Edmunds FT, Hundley JM, Morris FK. Benign cystic teratomas of the ovary; a clinic-statistical study of 1,007 cases with a review of the literature. *Am J Obstet Gynecol* 1955;70:368-82.
9. Lakkis WG, Martin MC, Gelfand MM. Benign cystic teratoma of the ovary: a 6-year review. *Can J Surg* 1985;28:444-6.

10. Comerci JT Jr, Licciardi F, Bergh PA, Gregori C, Breen JL. Mature cystic teratoma: a clinicopathologic evaluation of 517 cases and review of the literature. *Obstet Gynecol* 1994;84:22-8.
11. Selcuk I, Bozdog G. Recurrence of endometriosis; risk factors, mechanisms and biomarkers; review of the literature. *J Turk Ger Gynecol Assoc* 2013;14:98-103.
12. Chandra A, Rho AM, Jeong K, Yu T, Jeon JH, Park SY, et al. Clinical experience of long-term use of dienogest after surgery for ovarian endometrioma. *Obstet Gynecol Sci* 2018;61:111-7.
13. Farquhar C. Endometriosis. *BMJ* 2007;334:249-53.
14. American Society for Reproductive: Revised American Society for Reproductive Medicine classification of endometriosis: 1996. *Fertil Steril* 1997;67:817-21. .
15. van der Merwe JL, Siebert I, van Wyk AC. Rare case of perplexing ovarian endometriosis. *Fertil Steril* 2010;94:1910.e17-9.
16. Caruso ML, Pirrelli M. A rare association between ovarian endometriosis and bilateral ovarian teratoma. Case report. *Minerva Ginecol* 1997;49:341-3.
17. Matalliotaki C, Matalliotakis M, Ieromonachou P, Goulielmos GN, Zervou MI, Laliotis A, et al. Co-existence of benign gynecological tumors with endometriosis in a group of 1,000 women. *Oncol Lett* 2018;15:1529-32.
18. Kinkel K, Frei KA, Balleyguier C, Chapron C. Diagnosis of endometriosis with imaging: a review. *Eur Radiol* 2006;16:285-98.
19. Patel MD, Feldstein VA, Lipson SD, Chen DC, Filly RA. Cystic teratomas of the ovary: diagnostic value of sonography. *Am J Roentgenol* 1998;171:1061-5.
20. Nezhat C, Winer W, Nezhat F. Laparoscopic removal of dermoid cysts. *Obstet Gynecol* 1989;73:278-81.
21. Templeman CL, Fallat ME, Lam AM, Perlman SE, Hertweck SP, O'Connor DM. Managing mature cystic teratomas of the ovary. *Obstet Gynecol Surv* 2000;55:738-45.
22. Savasi I, Lacy JA, Gerstie JT, Stephens D, Kives S, Allen L. Management of ovarian dermoid cysts in the pediatric and adolescent population. *J Pediatr Adolesc Gynecol* 2009;22:360-4.
23. Smorqick N, As-Sanie S, Marsh CA, Smith YR, Quint EH. Advanced stage endometriosis in adolescents and young women. *J Pediatr Adolesc Gynecol* 2014;27:320-3.
24. Goldstein DP, De Cholnoky C, Emans SJ. Adolescent endometriosis. *J Adolesc Health Care* 1980;1:37-41.
25. Chen Y, Pei H, Chang Y, Chen M, Wang H, Xie H, et al. The impact of endometrioma and laparoscopic cystectomy on ovarian reserve and the exploration of related factors assessed by serum anti-Mullerian hormone: a prospective cohort study. *J Ovarian Res* 2014;7:108.
26. Busacca M, Vignail M. Endometrioma exision and ovarian reserve: a dangerous relation. *J Minim Invasive Gynecol* 2009;16:142-8.
27. Takahashi K, Kurioka H, Irikoma M, Ozaki T, Kanasaki H, Miyazaki K. Benign or malignant ovarian neoplasms and ovarian endometriomas. *J Am Assoc Gynecol Laparosc* 2001;8:278-84.

28. Anteby E, Dubuisson J, Samouth N, Shimonovitz S, Ariel I, Hurwitz A. Germ cell tumors of the ovary arising after dermoid cyst resections: A long term follow-up study. *Obstet Gynecol* 1994;83:605-8.
29. Busacca M, Marana R, Caruana P, Candiani M, Muzil L, Calia C, et al. Recurrence of ovarian endometrioma after laparoscopic excision. *Am J Obstet Gynecol* 1999;180:519-22.
30. Sesti F, Capozzolo T, Pietropolli A, Marziali M, Bollea MR, Piccione E. Recurrence rate of endometrioma after laparoscopic cystectomy: a comparative randomized trial between post-operative hormonal suppression treatment or dietary therapy vs. placebo. *Eur J Obstet Gynecol Reprod Biol* 2009;147:72-7.
31. Singh AK, Singh M. Collision tumours of ovary: a very rare case series. *J Clin Diagn Res* 2014;8:FD14-6.
32. Choudhary S, Adishesha S. Collision tumors of ovary: a rare phenomenon. *Int J Case Rep Imag* 2012;3:68-70.
33. Bige O, Demir A, Koyuncuoglu M, Secil M, Ulukus C, Saygili qu. Collision tumor: serous cystadenocarcinoma and dermoid cyst in the same ovary. *Arch Gynecol Obstet* 2009;279:767-70.

Figures

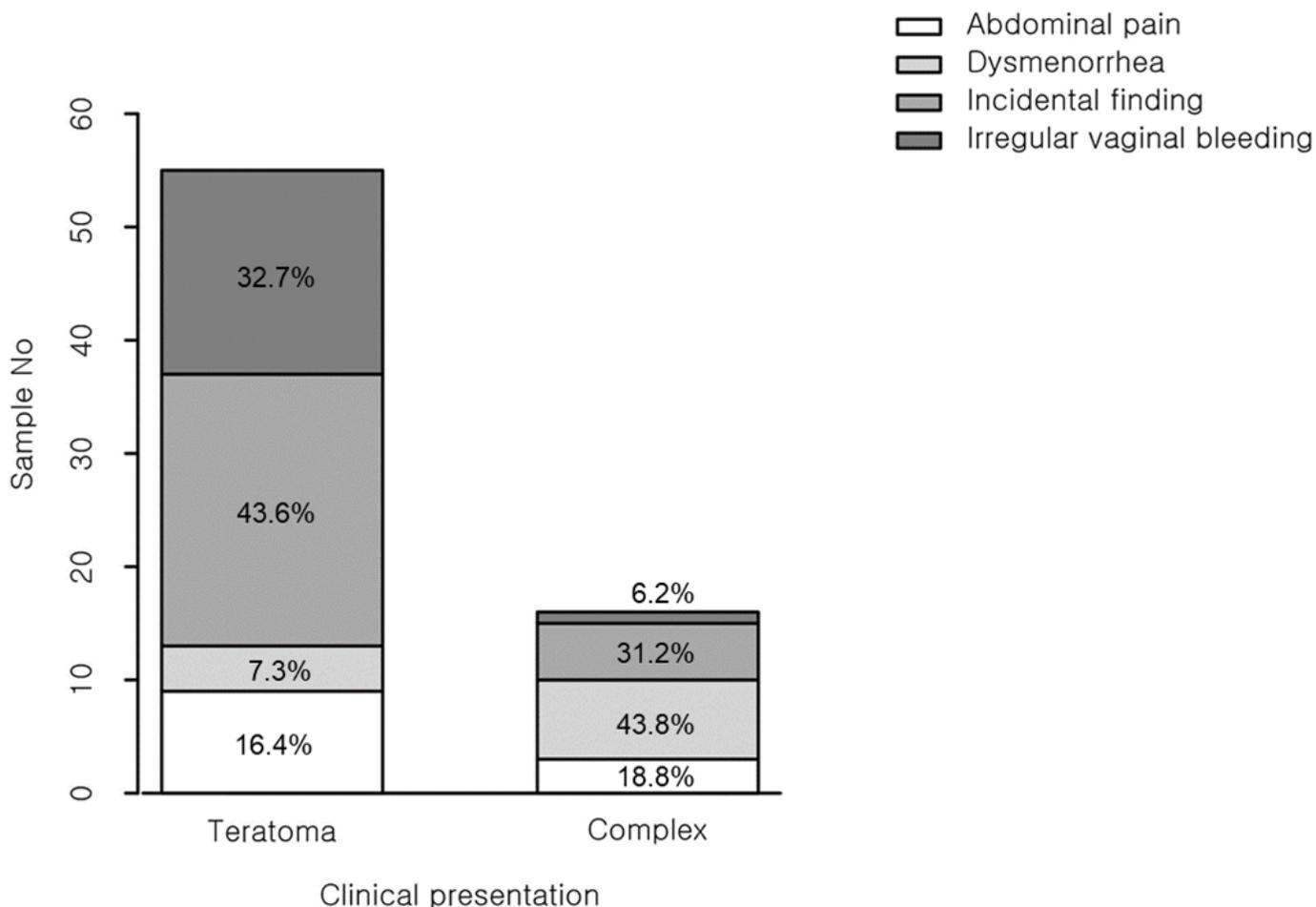


Figure 2

Bar plot representing sample distribution of clinical presentation between the teratoma group and the complex group. Y axis presented as sample number. Sample percentage (%) of each clinical presentation presented inside each bar.