

Malignancy Arising in Dermoid Cysts: A Case Report and Literature Review

Maliheh Arab; Ph.D.¹, Azadeh Jafari Ashtiani; M.D.², Nafiseh Faghih; M.D.², Behnaz Ghavami; M.D.³, Behnaz Nouri; M.D.², Shahla Noori Ardabili; M.D.⁴

1 Department of Gynecology Oncology, Imam Hossein Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran

2 Department of Obstetrician and Gynecologist, Shahid Beheshti University of Medical Sciences, Tehran, Iran

3 Department of Obstetrics and Gynecology, Tehran University of Medical Sciences, Tehran, Iran

4 Department of Obstetrics and Gynecology, Atieh Hospital, Tehran, Iran

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Abstract

Objective: Malignant transformation in dermoid cysts is rare, and Squamous Cell Carcinoma (SCC) is the most common form. This event often occurs in large tumors and middle-aged women.

Case report: In this study, two cases are presented. They were menopause, and abdominal pain and adnexal mass was a common manifestation in both. Case 1 with adenocarcinoma arising in mature cystic teratoma had abnormal tumor markers and was diagnosed with a frozen section during surgery, but case 2 with SCC transformation had normal tumor markers, and the frozen section was not helpful in the first surgery. Both underwent complete staging surgery, and due to stage IC1 in case 1, she received chemotherapy, and in case 2, no adjuvant treatment was needed because of stage IA.

Conclusion: Considering the rarity of malignant transformation in the dermoid cyst, the best surgical approach and adjuvant therapy indications need further research.

Keywords: Dermoid Cysts; Ovarian Neoplasms; Malignant Transformation

Introduction

Dermoid cyst or cystic teratoma is the most common ovarian germ cell tumor and usually occurs at reproductive age. These tumors are often unilateral, and 10-12% of cases can be bilateral (1). In most cases, they are asymptomatic and may present with abdominal pain, torsion, infection, rupture, and malignant transformation (2). Malignant transformation is a rare event that occurs in 1-2% of

cases, and the most common type is squamous cell carcinoma (SCC) (1, 3). Other types include adenocarcinoma, carcinoid tumor, melanoma, thyroid carcinoma, adenosquamous carcinoma, transitional cell tumor, signet ring cell carcinoma, oligodendroglioma, and sebaceous carcinoma (4). Malignant degeneration often occurs in middle-aged women (>40 years) and large-sized tumors (>18 cm) (5, 6). Two patients with malignant transformation of SCC and adenocarcinoma who underwent surgery at Imam Hossein hospital, Tehran, Iran are presented.

Correspondence:

Dr. Maliheh Arab

Email: drmarab@sbmu.ac.ir



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Case report

Case 1: A 57-year-old multiparous and menopause female was referred to the gynecologic oncology clinic due to abdominal pain, left ovarian mass and abnormal tumor markers. A left ovarian mass (130×83 mm) containing fat and calcified foci as well as loculated ascites and possible tumor rupture was shown in the magnetic resonance imaging (MRI). The lung Computed tomography (CT) scan was normal. Tumor markers were abnormal (CA125>500, CEA>550, and CA19-9>1000). According to the results of tumor markers, gastroenterology consultation and endo-colonoscopy followed by biopsy were performed, and gastritis and hyperplastic polyps were reported. Laparotomy with midline incision was done. There were numerous fat loculated in the abdominal cavity. Three liters of ascites fluid were suctioned. Strong adhesion of the intestines to the abdominal wall, posterior uterus, and adnexa was released. The left adnexal mass of about 15 cm was ruptured. It contained hair and fat. The result of the frozen section was reported as differentiated intestinal-type adenocarcinoma in the background of mature cystic teratoma. The patient underwent total abdominal hysterectomy and bilateral salpingo-oophorectomy (TAH-BSO) plus staging including omentectomy and lymphadenectomy (LND). According to the final pathology result based on the international federation of gynecology and obstetrics (FIGO) system due to stage IC1; she underwent six courses of chemotherapy using paclitaxel/carboplatin. Tumor markers and abdominal and pelvic CT were normal at the end of treatment. So far, she has no evidence of recurrence for 1.5 years of follow-up.

Case 2: A 48-year-old multiparous and menopause female was referred to the gynecologic oncology clinic due to abdominal pain and ultrasound and MRI results suggesting a 10-cm multi-loculated mass containing calcification in favor of dermoid cyst. All tumor markers were normal. The patient underwent laparotomy and the 10-cm right ovarian mass was sent for frozen section. Otherwise abdominal exploration was normal. The frozen section reported dermoid cyst and TAH-RSO plus Left salpingectomy was performed with left ovary conservation. The final pathology report of the right ovarian mass was differentiated non-keratinizing SCC arising on mature teratoma. She underwent second surgery, including left oophorectomy, omentectomy, and bilateral lymphadenectomy, all of

them were free of tumor. According to the FIGO system, she was in Stage IA, and the plan was followed up. There was no evidence of recurrence during the two years of follow-up.

Discussion

Malignant transformation in the dermoid cyst is considered a challenging issue for gynecologic oncologists due to the lack of specific diagnostic and treatment protocols (7). In the study by Blake et al. (2015), all patients with mature cystic teratoma (MCT) were investigated from 2004 to 2012 (6). Results showed that 1.2% of these patients had a malignant transformation. These patients' most common clinical symptoms included bloating and abdominal pain, the mean age of 53.7 years, and an average mass of 18 cm (6). According to the results of this study, large ovarian masses suspected to be cystic teratoma in older women should be managed aggressively. The present study included menopausal patients assessed due to abdominal pain and a 10-13 cm pelvic mass.

Tazo Yuki et al. (2016) investigated the test results of two patients and suggested that it is challenging to diagnose malignant transformation in MCT patients preoperatively, especially if the levels of tumor markers are close to the normal cut-off point (8). They also considered the role of an intraoperative frozen section to help diagnose malignancy. In the present study, tumor marker levels were abnormal in Case 1 and the frozen section confirmed evidence of malignancy, but Case 2 had normal tumor markers before surgery, and the intraoperative frozen section did not report malignancy.

Hurwitz et al. (2007) study also found that preoperative diagnosis of malignancy in MCT is not easy by serum tumor markers or imaging modalities and MRI might be more valuable imaging modality (9). In our study, MRI findings were not helpful in Case 2, and there were no definitive symptoms of malignancy in Case 1. Although SCC is the most common form of malignancy in the dermoid cyst, other types of malignancy were also found in the literature review (10, 4). Akazawa et al. (2018) investigated 155 SCC and non-SCC cases and found no significant differences between these groups regarding surgery, adjuvant therapies, and general survival (4). The primary surgical treatment varies from unilateral cystectomy to total abdominal hysterectomy, bilateral salpingo-oophorectomy, and debulking surgery.

Table 1: Previous study case reports of malignant transformation in mature cystic teratoma

Author	Number of patients	Age*	The largest size of tumor* (mm)	Imaging findings	Abnormal tumor markers	Type of surgery	Stage	Pathology	Adjuvant treatment	Follow up*
Our study	2	57	130	Fat and calcified foci	CA125, CA19-9, CEA	TAH-BSO, LND, Omentectomy	IC1	Intestinal type adenocarcinoma	Chemotherapy	1.5y
		48	100	Calcification	-	TAH-BSO LND, Omentectomy	IA	SCC	-	2y
Tazo Yuki et.al 2016 (8)	2	45	59	Dermoid cyst	SCC Ag	TAH-BSO	IA	SCC	Chemotherapy	22m
		35	135	Cyst and focal thickening	SCC Ag, CA19-9	TAH-BSO, Omentectomy	IA	SCC	Chemotherapy	21m
Tehrani et.al 2021 (7)	2	51	125	Solid cystic mass	CA125, CEA	TAH-BSO	IA	SCC	Chemotherapy	20m
		53	198	Solid cystic mass with fat foci and omental thickening	CA125, Roma	TAH-BSO, Omentectomy	IIIC	SCC	Chemotherapy	9m dead
Qin Li et.al 2021 (13)	14	51.3	140	-	SCC Ag=13	CRS=12 subop surgery=2	>IA=7	SCC=13 AC=1	Chemotherapy=7	31.2m
Mahtate et.al 2021(14)	1	53	68	Well defined mass and fat intensity	No	TAH-BSO, Omentectomy	-	SCC	Chemotherapy	-
Rathore et.al 2018(15)	8	44.2	117	-	-	-	-	SCC:4, TCC:2 AC:1, Melanoma:1	-	-
Shimada et.al 2019(16)	1	48	70	-	-	TAH-BSO, Partial omentectomy	-	SCC	Chemotherapy	7m dead
Jitsumori et.al 2017(17)	1	67	210	Solid component	CEA, CA125, CA19-9, SCC Ag	-	IA	SCC	No	2y
Shafizade et.al 2021(18)	1	54	125	Solid component, Thickened wall	-	TAH-BSO LND, partial omentectomy	IA	SCC	No	1y

*in more than 2 cases indicated as mean. CEA: carcinoembryonic antigen, TAH-BSO: total abdominal hysterectomy and bilateral salpingo-oophorectomy, LND: lymphadenectomy, SCC: squamous cell carcinoma, y: year, m: month, Ag: antigen

Unilateral adnexectomy is recommended in patients of reproductive age and the early stages, but in menopausal patients, more extensive surgery is performed regardless of the disease stage (3, 10). There is no agreement regarding adjuvant therapy and the effect of chemotherapy or radiotherapy (7). In a study of 155 patients, Congcog et al. (2019) found that hysterectomy and platinum-based chemotherapy are associated with better survival (11).

Akazawa et al. (2018) also mentioned that adjuvant therapy in both SCC and non-SCC groups was first-line chemotherapy, as in patients with epithelial ovarian cancer (4). In Xiaohog's study (2021), the successful role of immunotherapy in a chemotherapy-resistant patient suffering from SCC was reported (12). Both patients of the present study underwent complete debulking surgery, including TAH-BSO and LND, and Case 1 received chemotherapy according to stage IC1, and Case 2 was in stage IA, and follow-up was enough. Various studies acknowledged that earlier diagnosis and optimal surgery ensure better survival and treatment outcomes (4, 7, 10, 12).

The present study showed no evidence of recurrence during the follow-up period up to now. Previous studies showed that most patients with malignant transformation in the last five years were over 45 years of age and had a large mass size (10 cm or more). Preoperative imaging findings did not help to predict malignancy. Tumor markers, including SCC antigen (Ag), can be helpful and were abnormal in many cases. In the present study, SCC Ag was not performed in two patients, but in Case 1, other tumor markers were distorted, and no specific imaging findings suggesting malignant dermoid cyst were reported. Summarizing the reports, regarding the surgical approach, the minimal procedure included TAH-BSO and omentectomy or omental biopsy. Regarding adjuvant therapy, Taxol-Carboplatin-based chemotherapy has been regarded as the preferred treatment, and even in some studies, chemotherapy has been performed for stage IA patients. Mortality was more in patients with higher stages (Table 1).

Conclusion

We add 2 cases of malignant transformation in dermoid cyst to medical literature, 57 and 48 years old presenting with 13- and 10-centimeter ovarian masses. Considering the rarity of these cases, the best surgical approach and adjuvant therapy indications

need further research.

Conflict of Interests

Authors declare no conflict of interests.

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References

1. Pradhan P, Thapa M. Dermoid Cyst and its bizarre presentation. *JNMA J Nepal Med Assoc.* 2014;52(52):837-44.
2. Moussaoui E, Closon F, Brichant G, Kotzampassakis N, Nisolle M. Chemical peritonitis complicating the spontaneous rupture of a dermoid cyst of the ovary. *Rev Med Liege.* 2018;73(7-8):413-8.
3. Bonahy AA, Sabbah H, Vadell AH, Baba NE. Malignant teratoma: about a case and a review of the literature. *The Pan African Medical Journal.* 2017;27:61.
4. Akazawa M, Onjo S. Malignant transformation of mature cystic teratoma: is squamous cell carcinoma different from the other types of Neoplasm?. *Int J Gynecol Cancer.* 2018 ;28(9):1650-1656.
5. Esterson YB, Gaballah M, Grimaldi GM, Raj MH, Pellerito JS. Ovarian dermoid cyst complicated by small bowel obstruction, entero-ovarian fistula formation, and malignant degeneration. *Clin Imaging.* 2019;56:47-51.
6. Black JD, Roque DM, Pasternak MC, Buza N, Rutherford TJ, Schwartz PE, et al. A series of malignant ovarian cancers arising from within a mature cystic teratoma: a single institution experience. *Int J Gynecol Cancer.* 2015 ;25(5):792-7.
7. Tehranian A, Ghahghaei-Nezamabadi A, Seifollahi A, Kasraei S, Dehghani-Nejad H, Maleki-Hajiagha A. Ovarian mature cystic teratoma with malignant transformation: two case reports. *J Med Case Rep.* 2021;15(1):23.
8. Tazo Y, Yoshimura Y, Shoda T, Kyushima N, Okada T, Yamazaki H. Relevance of frozen sections and serum markers in invasive squamous cell carcinoma arising from ovarian mature cystic teratoma: two case reports. *J Med Case Rep.* 2016; 10: 20.
9. Hurwitz JL, Fenton A, McCluggage WG, McKenna S. Squamous cell carcinoma arising in a dermoid cyst of the ovary: a case series. *BJOG.* 2007;114(10):1283-7.
10. Al-Rayyan ES, Duqoum WJ, Sawalha MS, Nascimento

- MC, Pather S, Dalrymple CJ, et al. Secondary malignancies in ovarian dermoid cyst. *Saudi Med J*. 2009;30(4):524-8.
11. Li C, Zhang Q, Zhang S, Dong R, Sun C, Qiu C, et al. Squamous cell carcinoma transformation in mature cystic teratoma of the ovary: a systematic review. *BMC Cancer*. 2019;19(1):217.
12. Li X, Tang X, Zhuo W. Malignant transformation of ovarian teratoma responded well to immunotherapy after failed chemotherapy: a case report. *Annals of Palliative Medicine*. 2021;10(7):8499-505.
13. Qin L, Zhao T, Liu X, Wang H, Gu X, Chen D, et al. Malignant transformation arising from mature ovarian cystic teratoma: A case series. *Medicine (Baltimore)*. 2021;100(13):e24726.
14. Mahtate M, Talib S, Slaoui A, Zerai N, Lakhdar A, Rhrab B, et al. Malignant Degeneration of a Mature Ovarian Teratoma. *Case Rep Obstet Gynecol*. 2021; 2021:5527467.
15. Rathore R, Sharma S, Agarwal S. Malignant transformation in mature cystic teratoma of the ovary: a retrospective study of eight cases and review of literature. *Prz Menopauzalny*. 2018;17(2):63-8.
16. Shimada T, Higashijima A, Fukushima A, Komatsu N, Noguchi M, Ohashi K, et al. Malignant transformation from mature cystic teratoma of the ovary. *J Obstet Gynaecol Res*. 2019;45(9):1957-1960.
17. Jitsumori M, Munakata S, Yamamoto T. Malignant Transformation of Mature Cystic Teratoma Diagnosed after a 10-Year Interval. *Case Rep Obstet Gynecol*. 2017;2017:2947927.
18. Shafizadeh F, Babazadeh S, Nasiri Amiri F, Shafizadeh A. Squamous Cell Carcinoma in the Shadow of the Mature Cystic Teratoma of the Ovary: A Case Report. *Journal of Obstetrics, Gynecology and Cancer Research*. 2021;6(4):233-8.

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