A Rare Triple Coexistence Of A Collision Tumor, A Benign Mature Cystic Teratoma And A Hemorrhagic Follicular Cyst Of The Ovaries

B Papaziogas, A Souparis, M Grigoriou, P Tsiaousis, E Kogia, K Panagiotopoulou, K Atmatzidis

Citation

Abstract
Ovarian collision tumors are rare entities and most commonly consist of a benign mature ovarian teratoma and an ovarian cystadenoma or cystadenocarcinoma. Serous cystadenomas are the most common ovarian neoplasms, representing 20% of them. Benign mature ovarian cystic teratomas are often diagnosed in women of child-bearing age (12-15% of ovarian neoplasms). Benign mature ovarian solid teratomas are less common than the cystic ones. Follicular cysts are cysts of unruptured mature ovarian follicles. We present a rare case of a triple coexistence of a large collision tumor (consisting of a serous cystadenoma and a benign mature cystic teratoma) in the right ovary and a benign mature solid teratoma and a hemorrhagic follicular cyst in the left ovary of a young-aged woman, with a palpable mass in the lower abdomen. Right-sided salpingo-oophorectomy and left ovarian cystectomy preserving as much intact ovarian tissue as possible, took place. The possible existence of an ovarian collision tumour should carefully be examined pre- and postoperatively and histologically, so as to avoid misdiagnosis of a possible malignancy.

INTRODUCTION
Collision tumors represent a coexistence of two adjacent but histologically distinct tumors, without histologic admixture in an organ [1]. Collision tumors have been reported in various organs, but they are rare in ovaries. Ovarian collision tumors are most commonly composed of teratoma and cystadenoma or cystadenocarcinoma, but other histologic combinations have been also reported [2, 3], with the mechanism of origin still uncertain.

Ovarian serous cystadenomas represent 20% of all ovarian tumors. They usually appear during the climacteric age as unilocular cysts, with a diameter less than 15 cm and containing serum-like fluid [4].

Ovarian teratomas are the most common germ cell neoplasms and, in many series, the most common excised ovarian neoplasms [5]. The most common of these tumors are the benign mature cystic teratomas (also known as dermoid cysts), representing 12-15% of the ovarian neoplasias [5]. Mature cystic teratomas are bilateral in about 10% of cases [5]. The mature solid teratomas are another type of ovarian teratomas, which do not meet the criteria for an immature teratoma. They are benign teratomas, just like the cystic ones [6].

The follicular cysts are solitary or multiple cysts of unruptured ovarian follicles, which appear in the ovarian surface as pale cystic masses, with a diameter rarely exceeding 4 cm [4].

We present a rare case of a non child-bearing young-aged woman, who was hospitalized for the removal of a large cystic mass, probably arising from the right ovary. The eventual pathology exam revealed a collision tumor of a benign serous cystadenoma and a mature solid teratoma in the right ovary, and a mature cystic teratoma and a hemorrhagic follicular cyst in the left ovary.

CASE REPORT
A 29-year-old non child-bearing female, presented to our department complaining of a palpable mass in the lower abdomen, with a chronic mild pelvic pain and menstrual
irregularity. The patient denied fever or chills and, except for pain, had no significant gastrointestinal symptoms. The patient, on admission, had a menstrual period, the onset of which was delayed for two weeks. She had no significant medical or surgical history. Family history was negative.

Abdominal examination revealed a large, smooth, palpable and mobile mass, extending from the pelvis to approximately 3-4 cm below the umbilicus. The lower border of this mass was palpable through a vaginal digit exam. There was no guarding or rebound tenderness. Abdominal ultrasound revealed a unilocular cystic mass of 17 cm in diameter, probably originating from the right ovary, with a thin cystic wall and without hyper-echogenic content. There was no free fluid detected between the intestinal loops or in the Douglas pouch (Fig.1).

**Figure 1**

Figure 1: The cystic mass of the lower abdomen, probably of ovarian origin, as seen in the ultrasound.

Laboratory tests, both hematological and biochemical, did not exceed normal ranges, as well as the tumor markers CEA and CA-125 (1.61 ng/ml and 14.63 IU/ml, respectively). Lastly, the urine pregnancy test was negative.

The patient underwent an exploratory laparotomy, under general anesthesia and a Pfannenstiel incision. Upon entering the abdominal cavity, a large cystic mass (Fig.2), under a relative tension, protruded out of the minor pelvis. The organ of origin was indeed the right ovary, which was compressed by the cystic mass and macroscopically fully replaced by it. The cyst's content was about 300 cc, serum-like and lucid. The fluid was aspirated, avoiding any intraperitoneal spillage. A part of its wall was sent for frozen section, which revealed the presence of a layer of cubic epithelium, without any malignancy evidence, placing the diagnosis of a serous cystadenoma. With the surgical field now open to inspection, a careful observation of the rest of the internal genitalia was performed, thus disclosing two cystic masses in the left ovary, one of which contained skin adnexes (Figure 3). A right salpingo-oophorectomy with suture of the stump and excision of the two cysts of the left ovary, preserving as much residual ovarian tissue as possible, was performed.

**Figure 2**

Figure 2: Intraoperative photo. The serous cystadenoma of the right ovary, upon entering the abdominal cavity.
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Figure 3
Figure 3: Intraoperative photo of the left ovary. The cystic mass containing skin adnexes (1) and the hemorrhagic follicular cyst (2).

The permanent pathology report showed: a) a collision tumor of the right ovary, forming from i) a unilocular serous cystadenoma, with a smooth internal and external surface and a maximum diameter of 13 cm, and ii) a mature solid teratoma, containing fatty tissue and hair follicles, which was situated in a thickening of the serous cystadenoma wall, b) a mature cystic teratoma of the left ovary, containing sebum and hair follicles, and c) a hemorrhagic follicular cyst of the left ovary (Figures 4 and 5). The cytological examination of the large cystic mass fluid of the right ovary showed no malignancy evidence, confirming the benignity of the cystic mass.

The patient was discharged on the fifth postoperative day in a good general condition. A thorough periodic imaging follow-up of the abdomen and especially the residual internal genitalia was recommended.

Figure 4
Figure 4: Pathology findings. Ovarian collision tumor. a) The wall of the serous cystadenoma, consisting of unilayer cubic epithelium and a psammoid body. b) The thickening of the serous cystadenoma wall, which revealed the existence of the mature solid teratoma (fatty tissue and hair follicles).

Figure 5
Figure 5: Pathology findings. Left ovary: a) The benign mature cystic teratoma (dermoid cyst). b) The hemorrhagic follicular cyst.

Figure 6
Figure 5: Pathology findings. Left ovary: a) The benign mature cystic teratoma (dermoid cyst). b) The hemorrhagic follicular cyst.
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DISCUSSION

Collision tumors have been described in various organs other than the ovaries, including esophagus, stomach, liver, bone, kidneys, brain and lungs. Most of them were reported in the form of case reports. Collision tumors involving ovaries are rare and only a few cases have been described \[8, 9, 10\]. The most common histologic combination of collision tumors of the ovary consists of teratoma and mucinous tumors (cystadenomas or cystadenocarcinomas) \[11\].

In one third of the cases, serous cystadenomas are bilateral \[4\]. On the other hand, Morgante et al. \[12\] reported a bilateral localization of ovarian dermoid cysts in 7.9% of cases, associated with cysts of other histotypes homolaterally in 8.1% and contralaterally in 15.7%. The associated cysts were of a functional nature (17.2%), endometriotic (4.5%), serous cystadenomas (5.3%) and mucinous cystadenomas (1.2%). Bournas et al. \[13\] presented a case of bilateral ovarian dermoid cysts, three of them in the right ovary and one in the left, while Thomsen and Jochumsen \[14\] reported a case of bilateral ovarian dermoid cysts and an extra-ovarian dermoid cyst in the pouch of Douglas.

In our case, we present the rare triple coexistence of an ovarian collision tumor, together with a mature cystic teratoma of the ovaries and a hemorrhagic follicular cyst, a coexistence that has never been reported in the English-speaking literature, as far as we know.

The standard treatment of all the neoplastic masses of the ovaries, either benign or malignant, is surgical excision. The operation is either of a conservative approach, with or without ovarian excision, or of a radical approach, with total hysterectomy and bilateral salpingo-oophorectomy. In the case of a large serous cystadenoma, which compresses the ovarian parenchyma, unilateral salpingo-oophorectomy is inevitable. In the case of benign cystic or solid teratomas, the surgical excision with preservation of as much intact ovarian tissue as possible is mandatory \[1\].

The laparoscopic treatment of the ovarian dermoid cysts, first described in 1989 \[15\], is preferred by most surgeons nowadays. The advantages of the laparoscopic excision versus the laparotomic ones have been clearly established through a series of prospective randomized trials \[16, 17\]. The risk of unexpected malignancy of the mass is reduced by accurate preoperative diagnosis and staging \[18\] and an absence of intraoperative spillage of the cyst's contents \[19\]. In our case, though, we proceeded with an elective laparotomy, instead of a laparoscopic approach, because a definitive preoperative diagnosis and staging was impossible. Ultrasound identification criteria of mature cystic teratomas are relatively difficult to obtain, especially if malignancy is to be eliminated, thus the sensitivity and the specificity of the method are reported to be 58% and 98%, respectively \[19\].

Although the implications of intraperitoneal spillage of dermoid cysts (mainly, adhesion formation and chemical peritonitis) are controversial, the standard practice is to avoid the spillage of the cystic content. The post-spillage risk of chemical peritonitis is low \[16\], but existing, while, on the other hand, it seems to increase in neglected ruptures of dermoid cysts \[23\], or in failing to perform a peritoneal lavage immediately after an intraoperative rupture \[16\]. Spillage is reported to be as high as 80% of laparoscopic procedures and 10% of the laparotomic ones \[24, 25\].

Malignant transformation of mature cystic teratomas is a rare complication, reported to occur in 1-2% of cases in the older literature \[26\], mainly (85% of cases) in women aged over 40. The most common malignant histologic type is squamous cell carcinoma (75%), followed by adenocarcinoma (7%) and ovarian carcinoid (7%). There are also reports of sarcomas, melanomas and thyroid gland carcinomas development. Anteby et al. \[27\] reported that women with bilateral or multiple dermoid cysts have a relatively higher risk of developing ovarian neoplasms, therefore their postoperative follow-up should be thorough (with ultrasound and/or computed tomography of the abdomen).

In conclusion, ovarian collision tumors are rare, but even rarer is the coexistence with other benign ovarian tumors, as
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in our case. The preoperative suspicion of the existence of such tumors will, on the one hand, increase the surveillance level of the surgeon during the operation and, on the other hand, will force the pathologist to perform a thorough examination of the excised mass, so as to avoid misdiagnosis of a second type of tumor (usually a mucinous one), which might prove especially important in the subsequent treatment and outcome of the patient.

CORRESPONDENCE TO

Dr. B. Papaziogas Fanariou str. 16 551 33 Thessaloniki/ Greece Tel: 0030-2310-992562, Fax: 0030-2310-992563 e-mail: papaziog@med.auth.gr

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Author Information

B. Papaziogas
2nd Surgical Department, Aristotle University of Thessaloniki School of Medicine, “G.Gennimatas” Hospital

A. Souparis
2nd Surgical Department, Aristotle University of Thessaloniki School of Medicine, “G.Gennimatas” Hospital

M. Grigoriou
2nd Surgical Department, Aristotle University of Thessaloniki School of Medicine, “G.Gennimatas” Hospital

P. Tsiaousis
2nd Surgical Department, Aristotle University of Thessaloniki School of Medicine, “G.Gennimatas” Hospital

E. Kogia
2nd Surgical Department, Aristotle University of Thessaloniki School of Medicine, “G.Gennimatas” Hospital

K. Panagiotopoulou
Institute of Pathology, “G.Gennimatas” Hospital

K. Atmatzidis
2nd Surgical Department, Aristotle University of Thessaloniki School of Medicine, “G.Gennimatas” Hospital