

Collision Tumor: Dermoid Cysts and Mucinous Cystadenoma in the Same Ovary and a Review of the Literature

Mehmet Sühha Bostanci¹, Ozge Kizilkale Yildirim², Gazi Yildirim², Murat Bakacak³, Isin Dogan Ekinci⁴, Sevgi Bilgen⁵ and Rukset Attar^{2*}

¹Sakarya University Medical School, Obstetrics and Gynecology, Turkey

²Yeditepe University Hospital, Obstetrics and Gynecology, Turkey

³Kahramanmaras Sütçü Imam University Medical School, Obstetrics and Gynecology, Turkey

⁴Yeditepe University Hospital, Pathology, Turkey

⁵Yeditepe University Hospital, Anethesiology and Reanimation, Turkey

*Corresponding author: Rukset Attar, Yeditepe University Hospital, Obstetrics and Gynecology, Istanbul, Turkey, Tel: 0902165784832/905378401900, E-mail: ruksetattar@hotmail.com

Abstract

Collision tumor is defined as the coexistence of two adjacent, but histologically distinct tumors without histological admixture in the same tissue or organ. Collision tumors involving ovaries are extremely rare. The coexistence of a mucinous cystadenoma with a dermoid cyst is infrequently reported. However, the most common histological combination of collision tumor in the ovary is the coexistence of teratoma with mucinous tumors. If a dermoid cyst accompanies a multiseptated cyst and if the multiseptalcyst contains fatty foci, these two components may be associated. Recognizing the potential for the coexistence of these two neoplasms in the same ovary is essential to be able to make a correct diagnosis.

Keywords

Collision tumor, Dermoid cyst, Mucinous cystadenoma

Introduction

Dermoid cyst is a type of germ cell tumor comprising welldifferentiated tissues and three germ cell layers: ectoderm, mesoderm, and endoderm, which is also known as mature cystic teratom [1]. With peak incidence in females aged 20–40–years, these tumors comprise 10–20% of all ovarian neoplasms [1]. These tumors are characterized as generally slow-growing and unilateral, with a reported bilateral incidence of 10% [2]. Multiple dermoid cysts within a single ovary are rare. Benign mucinous cystadenomas account for 80% of mucinous ovarian tumors and 20-25% of overall benign ovarian tumors [3].

A collision tumor is the coexistence of two adjacent, but histologically distinct tumors without histological admixture in the same tissue or organ. Such tumors have often been reported in various organs, but location in the ovary is rare. Some other collision tumors of the ovary have been described in literature as cystadeno carcinoma and granulosa cell tumor [4], teratoma with granulosa cell tumor [5], and serous adenocarcinoma and steroid cell tumor [6]. The juxtaposition with dermoid cysts has been reported as comprising approximately 5% of benign mucinous ovarian tumors and rare examples of proliferating mucinous tumors [7].

The case is here reported of a rare collision tumor in the ovary consisting of mucinous cystadenoma and two distinct dermoid tumors.

Case Presentation

A 27-year-old G1P1 Caucasian female presented with complaints of right-side pelvic pain and a sensation of fullness in her lower abdomen. A subsequent pelvic sonography showed a complex, right ovarian cyst of approximately $8 \times 4 \times 4$ cm in size; her carbohydrate antigen (CA) 125 level was 18.1U/mL (normal, 0–25U/mL, at our hospital) and CA 19.9 was 7.4 (normal range 0–37U/mL, at our hospital). The structure appeared cystic, anechoic with several septations and appeared to be a part of the right ovary. No pathological flow was noted and no free fluid in the Pouch of Douglas was seen. The left ovary appeared unremarkable and the uterus was noted to be within normal limits. Other abdominal organs, including the liver, pancreas, kidneys, adrenals, and GI tract, appeared normal. The patient had negative cytological findings on a vaginal/cervical smear.

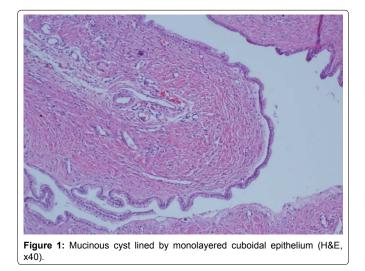
The patient was admitted for exploratory laparotomy. A right salpingo-oophorectomy was performed. Findings included a large simple-appearance right ovarian cyst occupying the entire right ovary with no normal residual ovarian stroma. An unremarkable left ovary, uterus, appendix, and liver were noted. The structure removed was diagnosed as a mucinous cystadenoma and 2 distinct mature cystic teratomas measuring totally $9 \times 5 \times 4.5$ cm and weighing 345gm.

The cut surface showed multi loculated cysts filled with cheese-

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like material, hair, and mucin, with a tan smooth inner lining (resembling a mucosal surface) and focal tan shiny areas. It was noted that the fimbriae were fused. The final pelvic wash was found to be negative for malignant cells and positive for reactive mesothelium cells.

The specimen was examined by light microscopy with hematoxylin-eosin stain. Microscopic examination of the right ovary revealed three tumor types: mucinous cystadenoma corresponded to the wall of the two cysts; mature cystic teratoma produced the skin and subcutaneous tissue, including sebaceous glands and hair containing mass.

Approval for this case report was granted by the Institutional Ethics Committee and informed consent was obtained from the patient (Figures 1-3).

Discussion

Ovarian neoplasms are thought to have three possible origins, surface epithelial-stromal tumors, sex cord-stromal tumors, and germ cell tumors. Germ cell tumors account for approximately 30% of primary ovarian tumors and of these 95% are mature cystic teratomas. Although mature cystic teratomas are generally unilateral, ovarian dermoid cysts may be bilateral and/or multiple in approximately 10-15% of cases. Long-term recurrence rate has also been reported [8,9]. Although rare, the possibility of additional neoplasms supervening in teratomas can be anticipated asteratomas are formed from a wide variety of ectodermal, mesodermal, and endodermal tissues. Surface epithelial-stromal tumors are classified as subgroups of serous tumors, mucinous tumors, endometrioid tumors, clear cell tumors, transitional cell tumors, and epithelial-stromal tumors. The most common mucinous tumors can be classified into 3 main categories of benign cystadenoma, borderline, and malignant. From the often large and usually cystic mucinous cystadenomas, multilocular rates have been reported as 76% and unilocular 24% [10]. Mucinous cystadenomas, which can progress to cystadeno carcinoma, are subdivided as benign, borderline, and malignant types [11]. However, of all mucinous ovarian neoplasms, 77% to 87% are benign [10,11].

Previous reports have detailed collision tumors in several organs other than the ovary, including the esophagus, stomach, liver, lung, thyroid and kidney [12,13]. The most common histological combination of collision tumor in the ovary is the coexistence of teratoma with mucinous tumors [14,15].

Various hypotheses have been suggested regarding the formation of collision tumors. The first hypothesis is that the coexistence of two primary tumors in the same tissue is due to a "chance accidental meeting". The second hypothesis proposed is that the presence of the first tumor creates changes in the microenvironment, engendering the development of the second primary tumor or the seeding of metastatic tumor cells. The third theory proposes that each primary tumor has its origins in a common stem cell [16].

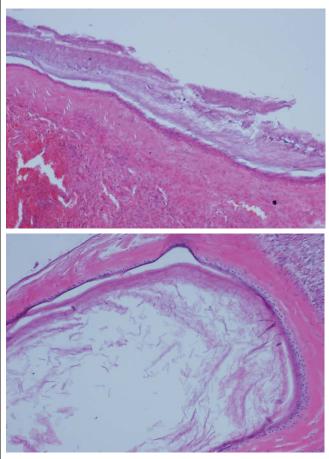


Figure 2,3: Dermoid cyst of the same ovary with squamous epithelium with keratinization (H&E, x100)

A clear diagnosis of collision tumor can be made when normal tissue intervenes between both tumors and there is no histological admixture at the interface. However, the presence of an equivocal intermediate transitional zone between the tumors may make it more difficult to differentiate between a collision tumor and a true mixed tumor [5,12]. The histological combination of teratoma and mucinous tumors is the most common form of collision tumours in the ovary [14].

The histogenesis of ovarian mucinous cystadenoma has not yet been fully clarified. There has been the suggestion of surface epithelial metaplasia origin or a teratomatous origin. Ultrastructure studies and mucin histochemical studies have supported the surface epithelial metaplasia theory [17,18]. The frequent co-existence of mature cystic teratoma and mucinous cystadenoma is the basis for the teratoma theory.

In a study by Vang et al. ovarian mucinous tumors associated with mature cystic teratomas were seen to exhibit morphological and immunophenotypic diversity [19]. Throughout the morphological spectrum, which ranges from cystadenomatous to carcinomatous, tumors of the surface epithelial-stromal type which resemble typical primary ovarian mucinous tumors may be seen and also lower intestinal tract type tumors of the kind usually encountered as secondary or metastatic tumors in the ovary [19].

Ovarian mucinous tumors, secondary or metastatic from a lower intestinal tract tumor are more often seen. Diagnosis is usually straightforward when such cases exhibit other characteristic features of metastatic disease (bilateral ovarian tumor, nodularity with surface involvement extra ovarian tumor, morphologically similar primary tumor elsewhere) [20].

Another study published in abstract form reported molecular findings (significant allelic imbalance for microsatellite markers, indicating homozygosity rather than heterozygosity for chromosomal polymorphisms) in some ovarian mucinous cystadenomas associated with mature cystic teratomas, which were consistent with a germ cell origin [21].

Although not all mucinous tumors are of germ-cell origin, mucinous elements in dermoid cysts may be of teratomatous origin as they are more likely to be intestinal rather than Mullerian in differentiation [22]. These coexisting ovarian tumors were reported by Kin et al. as rare collision tumors, the definition of which is a tumor with 2 adjacent but histologically distinct tumors with no histological admixture at the interface [14]. However, as there is some histopathological correlation between teratomas and mucinous tumors, coexistence may not be incidental.

In a study by Fuji et al. homozygous genetic patterns similar to those of the teratomatous components were shown in mucinous tumors arising together with mature cystic teratomas, and it was suggested that these mucinous tumors developed from pre-existing mature cystic teratomas [23].Evidence of a clonal relationship could be provided by additional molecular analyses demonstrating shared genetic changes in the mucinous tumor and adjacent conventional teratomatous elements to support the theory of a teratomatous origin for these mucinous tumors.

Okada et al. demonstrated that where the dermoid cyst accompanies a multiseptated cyst and if the multiseptated cyst contains fatty foci, there may be an association between these 2 components [24]. It was also stated that the recognition of the potential for coexistence of these 2 neoplasms in the same ovary is essential to be able to make a correct diagnosis [24].

Conclusion

In conclusion, to the best of our knowledge this is the first reported case of two mature cystic teratomas coexisting with mucinous cystadenoma in the same ovary. Collision tumors involving ovaries are extremely rare entities and coexistence of tumors with varying histogenesis may be diagnosed. During the intraoperative assessment of a dermoid cyst with unusual imaging findings, the intraoperative diagnosis cannot depend solely on macroscopy. The surgeon should establish good perioperative communication with the pathologists and if possible there should be intraoperative consultation with specialized gynaecology pathologists.

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