CASE REPORT

doi: 10.5455/medarch.2020.74.73-76

MED ARCH. 2020 FEB; 74(1): 73-76

RECEIVED: JAN 12, 2020 | ACCEPTED: FEB 12, 2020

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Papillary Cystadenofibroma of Fallopian Tube: Case Report with a Literature Review

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ABSTRACT

Introduction: Tumors of Fallopian tubes are rare in general, and they are the rarest tumors of female genital tract. According to classification of World health organisation (WHO), papillomas, cystadenoma, adenofibroma, cystadenofibroma (CAF), metaplastic papillary tumors and endometrioid polyps belong to group of benign tumors. Serous papillary cystadenofibroma (SPCAF) is rare tumor and it is usually located on fimbrial end of the tube and it is considered that it has „Müllerian“ origin. **Aim:** The aim of this article is to show a rare case of cystadenofibroma of Fallopian tube which was found as random sample during histopathological analysis of specimen which was extracted during laparotomy due to the large tumor mass in abdomen. **Case report:** A 43-year-old patient underwent surgery for a large tumor mass in the abdomen, unknown lesions and pathohistology, which was radiologically verified four years before hospitalization. We removed the specimen that made up the tumor, along with the uterus and adnexes, weighing 14 kg and sent for histopathology. A large tumor mass is made up of a giant uterine myoma, and in the analysis of the other preparation, in addition to endometrial adenocarcinoma, there is also a rare Fallopian tube cystadenofibroma. On the sixth day of hospitalization, the patient is discharged home. At control after three months, the patient was without problems, with ongoing adjuvant brachytherapy. **Conclusion:** Improvement of prevention measures and work on the continuing education of patients and physicians at the primary care level are needed to ensure that patients receive the best treatment in a timely manner. Cystadenofibroma is a rare tumor in general and gynecologic oncology, and as authors it is a great honor for us to contribute to the world literature and to present the twentieth case of this tumor.

Keywords: Cystadenofibroma, Fallopian tube, Giant myoma, Surgery.

1. INTRODUCTION

Tumors of Fallopian tubes are rare in general, and they are the rarest tumors of female genital tract. According to classification of World health organisation (WHO), papillomas, cystadenoma, adenofibroma, cystadenofibroma (CAF), metaplastic papillary tumors and endometrioid polyps belong to group of benign tumors (1). Serous papillary cystadenofibroma (SPCAF) is rare tumor and it is usually located on fimbrial end of the tube and it is considered that it has „Müllerian“ origin (2). Preoperative diagnosis is not usually made. Only in the case of benign cystadenofibromas, the cyst can be extracted with the genital organs preserved, especially in younger patients of fertile age. Most women do not have any symptoms and most are accidentally detected during systematic examinations or surgery for other gynecological or abdominal

diseases (3). In our case, the reason why the patient underwent laparotomy is a large tumor of the abdomen of unknown etiology, which later turned out to be a uterine myoma at the pathohistological diagnosis.

Uterine myomas are the most common tumors that arise from the female reproductive tract. However, giant fibroids weighing more than 11,4 kg are extremely rare. They can cause pressure on the surrounding organs, heart and lungs, which can be life-threatening (4).

2. AIM

The aim of this article is to show a rare case of cystadenofibroma of Fallopian tube which was found as random sample during histopathological analysis of specimen which was extracted during laparotomy due to the large tumor mass in abdomen.

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Authors	Year of publication	Age (in years)	Clinical findings	Treatment	Site and Size	Microscopy
Kanbour <i>et al.</i>	1973	63	Incidental finding during surgery for prolapse	Vaginal hysterectomy	Intramural part of left uterine cornua, 2 cm	Cystic papillary adenofibroma
Silverman <i>et al.</i>	1978	36	Incidental finding during tubal ligation following termination of pregnancy	Bilateral partial salpingectomy	Cystic mass at fimbrial end of left tube, 3.5 cm	Serous cystadenofibroma
de la Fuente	1982	73	Incidental finding during surgery for uterine leiomyomas	TAH with BLSO	Fimbrial end of right fallopian tube, 2.5 cm × 2 cm × 2 cm	Mixed mullerian tumor-adenofibroma
Casasola and Mindan	1989	32	Incidental finding during operation for multiple uterine leiomyomas	Hysterosalpingo-oophorectomy	NA	Cystadenofibroma
Chen	1994	24	Primary infertility	BL tubal cystectomy with wedge biopsy of the ovary	Fimbrial end of right and left fallopian tube, 2.5 cm × 2 cm and 0.3 cm × 0.2 cm respectively	Bilateral papillary adenofibroma
Sills <i>et al.</i>	2003	NA	Incidental finding during IVF embryo transfer	Laparoscopic tubal cystectomy	Distal end of right fallopian tube, 5.5 cm	Serous cystadenofibroma
Gürbüz and Ozkara	2003	48	Irregular vaginal bleeding with uterine leiomyomas	TAH with BLSO	Serosal surface of right fallopian tube, 0.4 cm	Serous cystadenofibroma
de Silva <i>et al.</i>	2010	19	Pain in right iliac fossa	Right SO	8 cm	Cystadenofibroma
Mondal	2010	27	Ectopic pregnancy	Left salpingectomy	Fimbrial end of left fallopian tube, 2 cm × 1.5 cm	Adenofibroma with ectopic pregnancy
Erra and Costamagna.	2012	50	Incidental finding during operation for leiomyomas	TAH with BLSO	Fimbrial cyst, 3 cm	Serous cystadenofibroma
Pandey <i>et al.</i>	2012	20	Incidental finding during emergency LSCS with BLTL	LSCS with BLTL	Cystic mass in the fimbrial end of left tube, 4 cm × 3 cm	Serous cystadenofibroma
Fukushima <i>et al.</i>	2014	32	Incidental finding during operation for a suspected case of ectopic pregnancy	Linear salpingoostomy and ovisceration	Solid cystic mass near ampule of left fallopian tube, 20 mm	Papillary cystadenofibroma
Khatib <i>et al.</i>	2015	30	Incidental finding during operation for a suspected case of ovarian neoplasm	Left tubal cystectomy	Solid cystic mass on the serosal aspect of left fallopian tube, 12 cm × 10 cm	Serous papillary cystadenofibroma

TAH with BLSO: Total abdominal hysterectomy with bilateral salpingo-oophorectomy, LSCS with BLTL: Lower segment cesarean section with bilateral tubal ligation, NA: Not available, IVF: In vitro fertilization

Table 1 Reported cases of cystadenofibroma available in medical literature (2,3, 6-16)

3. CASE REPORT

A 43-year-old patient was admitted to the General and Abdominal Surgery Clinic for surgical treatment of high abdominal tumor mass. She states that the first subjective problems in terms of feeling of heaviness in the abdomen and difficulty breathing occurred one year before admission, and from medical records we learn that the tumor formation was first mentioned four years before admission on the abdominal ultrasound. Menstrual history was neat, had no births or abortions. Aunt died of breast cancer and her sister suffered from breast cancer. The examination shows that the abdomen is above the plane of the thorax, tense, palpator painfully insensitive, with palpable mass of irregular shape, mostly hard, and sometimes softer consistency, extending from the small pelvis to the xyphosternum, with limited mobility, and gives the impression that it permeates the entire abdominal cavity. MRI analysis of abdominal organs is dominated by extensive inhomogeneous, lobulated, expansive tumor formation that represses and dislocates intra- and retroperitoneal organs. All routine examinations were in the reference values, as were the tumor markers AFP, CEA, CA-19.9 and CA-125.

During explorative laparotomy, a large tumor mass, irregular appearance, smooth surface is found, mostly solid consistency with several cystic formations, two of them are the size of a handball. The described tumor formation is easily separated anteriorly and laterally from the anterior abdominal wall, and further exploration reveals that the mass suppresses the liver, and with its extreme proximal border upwards, it encloses the gall-bladder and hepatoduodenal ligament, the left liver lobe,

and medially pushes the left lobe and the diaphragm, laterally suppresses the spleen, like all the small intestine. The small pelvis is also filled with tumor formation. Further preparation separates the tumor mass from the posterior abdominal wall and retroperitoneum without signs of infiltration of the organs of this area. The uterus and adnexa are dislocated by the tumor to the lateral right and cannot separate from the tumor. The left Fallopian tube with a linked ovary is of a clear macroscopic appearance. The right Fallopian tube is with a cystic formation of dimensions 180x55x23mm and a linked ovary of dimensions 70x15x10mm, gray-white in color and swirling structure. A hysterectomy with bilateral adnexectomy was performed in collaboration with an intraoperatively consulted gynecologist. The tumor mass weighs 14 kg with a dimension of 540x20x140mm. The patient is post-operatively placed in the Intensive Care Unit, and after two days of hospitalization returns to the abdominal surgery department where she resides for four days before being discharged home.

Microscopic samples of large tumor mass correspond to uterine myoma with no signs of increased mitotic activity or tumor type of necrosis. Endometrial snippets show an image of a minimally invasive endometrial adenocarcinoma, of a medium differentiated type, "G2", which engages the endometrium with minimal invasion of the myometrium and without noticeable lymphovascular invasion, pT1a, FIGO IA. The rest of the endometrium shows hyperplastic endometrial polyp and focuses on simple and complex hyperplasia. Microscopic analysis of the vaginal portion of the uterus reveals a picture of slight dysplastic changes (CIN1). The cervical canal is

dominated by the image of chronic cervicitis. Cystic dilated follicles are seen in the sections of both ovaries. In the left Fallopian tube, a picture of chronic salpingitis is seen.

Snippets from the right Fallopian tube at the fimbrial end show a microscopic image of serous cystadenofibroma: "The tumor shows expansive growth and clearly narrows the lumen of the Fallopian tube. There are many small cystic spaces containing papillary protuberances with glomeruloid features in some areas. The papillary compartments of different sizes and shapes have a single-layered cuboidal epithelium showing a clear cytoplasm and a pronounced cellular border. Tumor cells with eosinophilic cytoplasm have no cilia relative to ciliated epithelial cells. The base of the papilla is composed of fibrous stroma of dense connective tissue, with no signs of invasion or atypia. " The intraluminal fibrotic altered fringes are imbued with a mixed inflammatory infiltrate corresponding to chronic exacerbative salpingitis.

Due to the association of this tumor with Von Hippel Lindau syndrome, we indicate that brain MRI, which was satisfactory (5). At control after three months, the patient had no subjective problems, with ongoing adjuvant brachytherapy due to the histopathologically verified endometrial adenocarcinoma of FIGO IA.

4. DISCUSSION

Giant tumors are not a rare occurrence in abdominal surgery. A look at our database shows that most of them are found in women and are of genital tract origin. In this case, we found a large intra-abdominal mass that occupies the entire abdomen, weighing 14 kg. The whole tumor was removed en block with the uterus and adnexa without damaging the archetypes of the same, with minimal blood loss and duration of surgery of 1h and 45min. At the histopathological diagnostics we obtained a varied finding. The large tumor mass corresponds to the uterine myoma, the endometrium showed the presence of early-stage adenocarcinoma, and, incidentally, one of the most rare tumors in general and gynecologic oncology was found at the fimbrial end of the left tube: serous cystadenofibroma. According to earlier reports, our case should be the twentieth in the world literature, unique in its finding: found intraluminally, not cystic, and associated with the most common tumor of the female genital tract.

Cystadenofibroma was first diagnosed by Iwanow in 1909, who called it papileferum CAF (3). Since then, only 19 cases have been reported in the world literature. The age of patients ranges from 19 to 73 years.

The last, nineteenth case of this tumor was reported in 2015 in a thirty-year-old woman who underwent surgery for a suspected ovarian tumor (16). Analyzing these cases, we find that CAF is most often in the form of a round solitary mass and that most are present at the fimbrial end of the tube. They are usually small and have a diameter of about 0.5-3 cm. Most are cystic, with a rough papillary outgrowth. The tumor appears to have a benign course and no malignant potential has been described.

Differential diagnosis of tubular origin tumors include tube cancer, serous papillary tumor of low malignant potential (STLMP) and borderline tumor. Most occur in postmenopausal women. Unless it is impossible to achieve adequate radicality, tubular cystectomy is sufficient and appropriate therapy. In our case, a hysterectomy with bilateral adnexectomy was unavoidable.

We hope that this case report will find readers among general and abdominal surgeons and gynecologists for unusual intraoperative findings, pathologists because of "colorful" microbial findings, and other involved physicians of different specialties of secondary and tertiary level of health care. We consider that the message of this case is a message of big importance for the doctors of primary health care to the extent that it encourages them to improve accessibility and communication with the patient, to be good health managers in a way to "protect the patient from the health system" and to be persistent in continuous self and patient education.

5. CONCLUSION

Continuous patient education and improvement of preventive measures, which are the primary task of primary care, are needed to ensure that patients receive the most appropriate treatment in a timely manner. We believe that having a 14kg tumor, which was first radiologically verified four years before surgery, is a failure for both doctors and patients.

Seorus cystadenobiroma of Fallopian tube is rare lesion which is usually small and asymptomatic. Therefore, it is usually diagnosed during surgery for other gynecological disorders. It is an honor for us to contribute to the overall database relating to this tumor through the presentation of this case and to present the twentieth recorded case in the world literature in its full diversity and circumstances.

- **Author's contribution:** Each author gave a substantial contribution in acquisition, analysis, and data interpretation. Each author had a part in preparing article for drafting and revising it critically for important intellectual content. Each author gave the final approval of the version to be published and agreed to be accountable for all aspects of the work, ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.
- **Conflicts of interest:** There are no conflicts of interest to declare.
- **Financial support and sponsorship:** Nil.

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