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A Rare Serous Papillary Cystadenofibroma of the Fallopian Tube

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ABSTRACT

Tumors of the fallopian tube consist an uncommon and rare entity of the female genital tract. The benign tumors of the fallopian tube are endometrioid polyp, metaplastic papillary tumor, cystadenofibroma, adenofibroma, cystadenoma and papilloma, according to the classification held by the World Health Organization (WHO). It is essential we report that the great majority of women with these tumors are asymptomatic and these tumors are mostly diagnosed incidentally during a gynecological examination or a pelvic ultrasound scan or during an operation for other surgical reasons.

INTRODUCTION

Serous papillary cystadenofibroma is an extremely rare benign tumor of the fallopian tube. Until now, only 19 cases in the world literature have been described [1-5] The focus of this paper is a case study concerning a 37-year-old woman with pelvic pain, whose gynecological examination revealed a pelvic tumor confirmed with transvaginal ultrasound and further assessed with magnetic resonance imaging. At that point the differential diagnosis included para-ovarian tumor or a GIST tumor and subsequently was scheduled for surgical management. A histopathological exam revealed a serous papillary cystadenofibroma of the fallopian tube.

CASE REPORT

A 37-year-old Caucasian female, presented to the Outpatient Clinic complaining of intermittent pelvic pain, located on the right iliac fossa and radiating to the right abdomen and lumbar area. It occurred every other month for a year, with no alleviation of the symptoms, which lasted 24 h despite positioning or the use of common analgesics. Her menstrual history was regular and she had two full-term caesarian deliveries with the last childbirth 2 years ago. Speculum examination did not demonstrate any vaginal discharge and bimanual examination revealed a small mass arising from the pelvis. The cervix was not sensitive on her bimanual examination, while there was discomfort in the right adnexa. The transvaginal ultrasound demonstrated a mass arising from the right adnexa having a size of 5.3cm x 2.2cm. The uterus and the left adnexa have no pathology at the moment. There was no free fluid in the pouch of Douglas. Magnetic resonance imaging showed a small oval structure of mixed magnetic signal comprising both of cystic and compact areas, but also islets of fat with a size of 4,5 cm x 6cm, which required differential diagnosis between a para-ovarian tumor and a stromal GIST tumor.

Patient's surgical history included a right ovarian cystectomy 3 years ago. Hypothyroidism treated with T4 100μg and a mild Von Willebrand's deficiency was reported from the patient, but not requiring any further treatment at the time.

The blood test exams were normal and the tumor markers were: cancer antigen - 125 = 7.6 u/ml (normal 0-35 u/ml), lactate dehydrogenase = 155 u/l (normal 135-214 u/l), carcinoembryonic antigen = 0.69 ng/ml (normal 0-5 ng/ml), cancer antigen - 19.9 = 23.1 u/l (normal < 37 u/l), and alpha-feto protein = 2.3 ng/ml (normal 0-6 ng/ml).

She was scheduled for admission to the hospital. A decision for an exploratory laparotomy was made. Intraoperative findings revealed a firm tumor with a cauliflower-like surface arising from the right fallopian tube **(Figure 1).** The right ovary was found completely separate from the tumor. A right tubal cystectomy was performed **(Figure 1)** and a cold biopsy, consisting of the fallopian tube and the mass, was sent for further evaluation. The results were negative for malignancy. Furthermore, the complete histological analysis revealed a serous papillary cystadenofibroma (SPACF) with

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collagenization of the substrate, which was in contact with a simple epidermoid cyst (mature cystic teratoma). Postoperatively, the patient's recovery was without incident and she was thereafter discharged within four days [6].

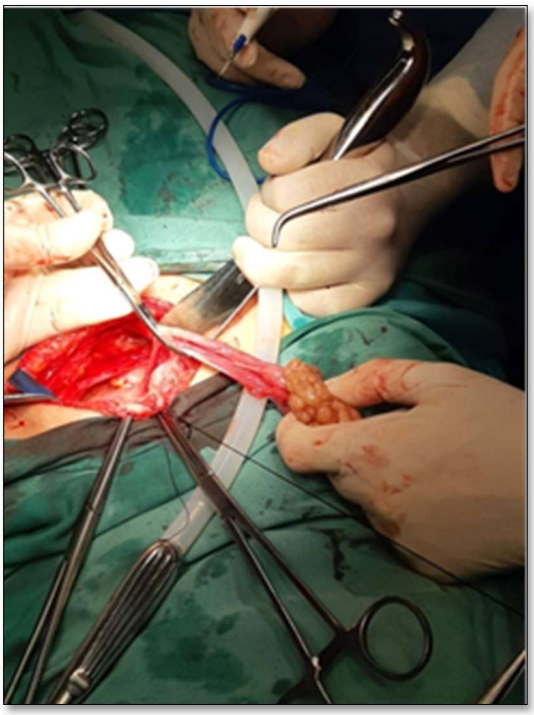

Figure 1. Tumor of the right fallopian tube.

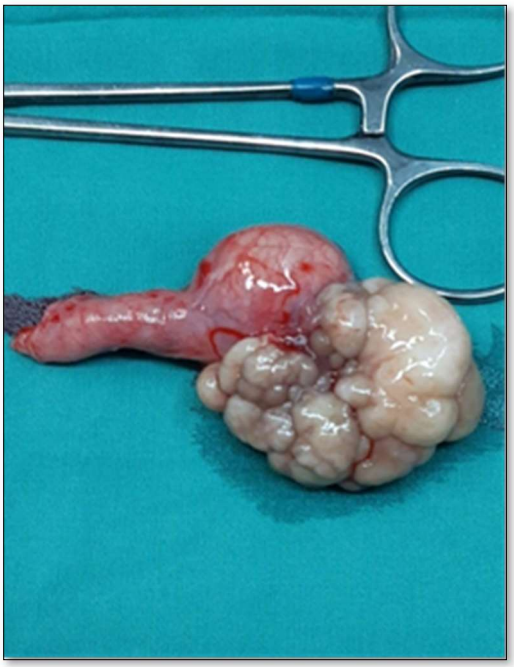

Figure 2. Right fallopian tube with serous papillary cystadenofibroma.

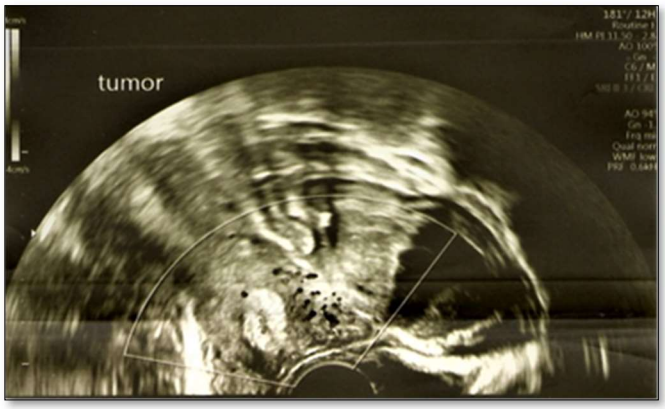

Figure 3. Transvaginal scan of the fallopian tubal tumor.

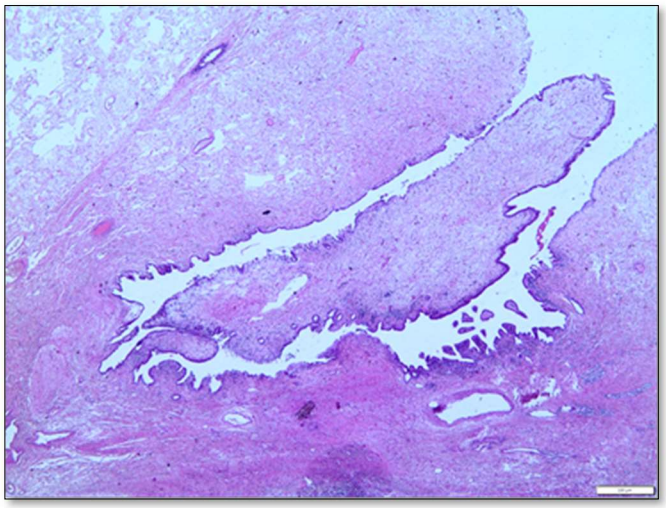

Figure 4. Papillary tumor with papillary projections either on the outer surface or protruding into the cyst cavity (x20).

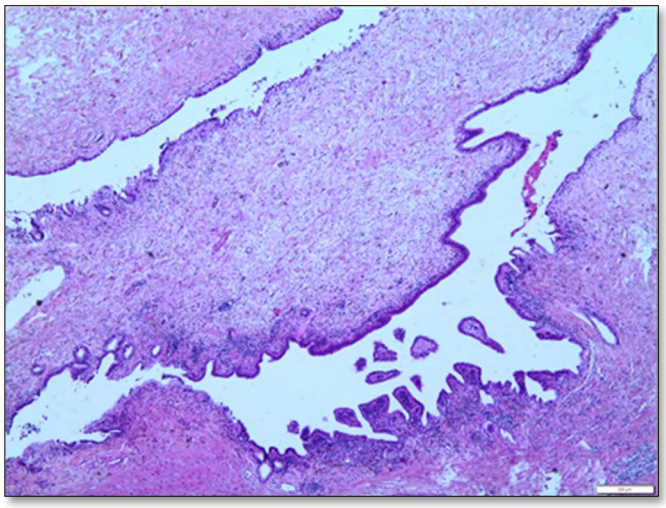

Figure 5: The cyst is lined by a single layer of tall columnar ciliated cells, resembling normal tubular epithelium (x40).

DISCUSSION

The differential diagnosis for a tumor of tubal origin includes tubal carcinoma (primary or metastatic), serous tumor of low malignant potential (STLMP) and borderline papillary serous tumor of the fallopian tube. Malignant neoplasms of the fallopian tube are rare, accounting for 0.18% to 1.6% of all malignant neoplasms of the female reproductive tract. They most commonly appear in post-menopausal women, usually in the sixth decade of life. However, histological, molecular and genetic evidence seems to indicate that these rates are underestimated, because as well as over 40-60% of high grade serous ovarian carcinomas appear to originate from the Fallopian tubes. These findings, once confirmed in larger studies, will have a decisive impact on prevention, early diagnosis and treatment of malignant tumors of the female genital track. Serous papillary cystadenofibromas of the fallopian tube are rare tumors with a faint possibility for malignant development [7].

The tumor is considered an embryologic remnant rather than a proliferating neoplastic process. Only a few cases have been reported in world literature. The age of presentation showed a wide range from 19 to 73 years. Mean age reported is 49 years and clinical presentation was variable including extreme diversity in size. Most are present at the fimbrial end. They are usually small in size and measure about 0.5-3 cm in diameter, cystic with coarse papillary excrescences as seen in the present case. Histologically, two components are present, a connective tissue stroma without nuclear pleomorphism or mitosis and papillary structures or tubal structures lined by epithelial cells. The epithelial cell type has been serous in most cases but occasionally may be endometrioid. The tumor seems to have a benign course and most are seen in postmenopausal women. Microscopy shows cellular pleomorphism with nuclear atypia. STLMP and borderline tumors show stratification with nuclear atypia. Hence, histology is necessary to confirm the diagnosis. Also, cystadenofibromas can be one of the causes of infertility in reproductive-aged women. In most cases, the recommended treatment is a unilateral salpingectomy/cystectomy, without the need for any further treatment. However, long-term follow-up of more cases is required to draw more definitive conclusions and consultations from the clinicians. **Table 1** describes the previous 20 published worldwide cases. It is modified from the one published by Hodzic [8].

Table 1. 20 published Cases.

Authors	Year of Publication	Age	Clinical Findings	Site and Size	Treatment
Kanbour	1973	63	Incidental finding during surgery for prolapse	Intramural part of left uterine cornua, 2 cm	Vaginal hysterectomy
Silverman	1978	36	Incidental finding during tubal ligation following termination of pregnancy	Cystic mass at fimbrial end of left tube, 3.5 cm	Bilateral partial salpingectomy
de la Fuente	1982	73	Incidental finding during surgery for uterine leiomyomas	Fimbrial end of right fallopian tube, 2.5 cm x 2 cm x 2 cm	TAH with BSO
Casasola and Mindan	1989	32	Incidental finding during operation for multiple uterine leiomyomas	NA	Hysterosalpingo-oophorectomy
Chen	1994	24	Primary infertility	Fimbrial end of right and left fallopian tube, 2.5 cm x 2 cm and 0.3	BL tubal cystectomy with wedge biopsy of the ovary

				cm x 0.2 cm respectively	
Sills etal.	2003	NA	Incidental finding during IVF embryo transfer	Distal end of right fallopian tube, 5.5 cm	Laparoscopictubalcystectomy
G眉rb眉z and Ozhara	2003	48	Irregular vaginal bleeding with uterine leiomyomas	Serosal surface of right fallopian tube, 0.4 cm	TAH with BSO
de Silva	2010	19	Pain in right iliac fossa	8 cm	Right SO
Mondal	2010	27	Ectopic pregnancy	Fimbrial end of left fallopian tube, 2 cm x 1.5 cm	Left salpingectomy
Erra and Costamagna	2012	50	Incidental finding during operation for leiomyomas	Fimbrial cyst, 3 cm	TAH with BSO
Pandey	2012	20	Incidental finding during emergency	Cystic mass in the fimbrial end of left tube, 4 cm x 3 cm	LSCS with BTL
Fukushima	2014	32	Incidental finding during operation for a suspected case of ectopic pregnancy	Solid cystic mass near ampule of left fallopian tube, 20 mm	Linear salpingostomy and evisceration
Khatib [5]	2015	30	Incidental finding during operation for a suspected case of ovarian neoplasm	Solid cystic mass on the serosal aspect of left fallopian tube, 12 cm x 10 cm	Left tubal cystectomy
Hodzic [8]	2020	43	Incidental finding during operation for Large Uterine Myoma	Solid mass in abdomen 14kg 540x20x140 mm	Explorative Laparotomy

CONCLUSION

In conclusion, serous papillary cystadenofibroma is a rare entity, usually small in size presented as adnexal tumor incidentally found in ultrasound examination. It is difficult to make the differential diagnosis based on ultrasound findings. Other imaging modalities such as CT or MRI of lower abdomen are not usually conclusive and clinicians must always be aware of this condition. The prognosis is excellent and the treatment of choice is surgical, including either

removal of the lesion with or without the fallopian tube or salpingoophorectomy depending on the patient's age and desire of preserving fertility.

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