

# Tumors of low malignant potential arising in the fallopian tube: Case reports

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## Summary

**Background:** There is a paucity of information regarding fallopian tube tumors of low malignant potential (LMP) in the literature.

**Case:** We present two cases representing alternative management options of low LMP of the fallopian tube.

**Conclusion:** Although low malignant potential tumors of the ovary are relatively common, there are few reported cases of tumors of LMP originating in the fallopian tube. Treatment has been extrapolated from tumors of LMP of the ovary, and conservative fertility-sparing surgery and complete staging procedure remains controversial. We urge continued reporting of these fallopian tube tumors of LMP to enhance understanding of these rare tumors and to develop a more cohesive treatment plan.

**Key words:** Low malignant potential tumor; Fallopian tube; Management; Borderline tumor; Serous tumor.

## Introduction

While tumors of low malignant potential of the ovary are relatively common, comprising 4-14% of all ovarian cancer cases, tumors of low malignant potential (LMP) are rarely found originating in the fallopian tube. There have been a total of ten previously reported cases of tumors of LMP of the fallopian tube in the literature since 1966, five being serous [1-5], four mucinous [6-9] and one endometrioid adenofibroma [4]. We report two cases of serous low malignant potential of the fallopian tube, and briefly review the literature concerning treatment options and outcomes.

### Case no. 1

M.S., is a 22-year-old woman with a history of amenorrhea and abnormal menstrual bleeding. On pelvic ultrasound, a thin walled, cystic, sonolucent structure measuring 4.9 x 2.2 x 3.4 cm in the right adnexa was noted. She was lost to follow-up until she presented to the emergency room with complaints of abdominal pain. On bimanual exam, a 4 cm, mobile, mass was palpated in the right adnexa. Transvaginal ultrasound (TVUS) showed a 4.5 x 3.2 x 3.9 cm cystic lesion in the right adnexa containing soft tissue elements with a resistance index of 0.49. All serologic markers were negative. After three months of oral contraceptive pills, she had worsening pain and a repeat TVUS showed no interval change in the lesion.

A laparoscopic evaluation showed a cystic structure in the right adnexa that appeared simple in nature and completely independent of the ovary (Figure 1). Although it appeared to be a hydrosalpinx, when a linear salpingostomy was performed a cystic structure was seen within the tube. A tubal cystectomy was performed, with complete enucleation of the cyst from the fallopian tube. There was incidental spillage of serous fluid.

### Pathologic Findings

Grossly, the tumor was a cystic mass filled with serous fluid and measured 3.4 x 3 x 1.1 cm. The inner surface was lined with multiple papillary structures measuring from 0.2 to 0.6 cm. Microscopically the cyst was lined with focal papillae and atypical stratified cells (Figure 2). The tumor cells appeared mitotically active. Focal glandular complexity and branching papillary fronds were noted, but without stromal invasion (Figure 3). These histological features are indicative of a serous tumor of low malignant potential of the fallopian tube (STLMP).

### Case no. 2

M.N., a 43-year-old woman with a history of endometriosis and pelvic pain, was noted on physical examination to have a large fibroid uterus in excess of 16 cm. She had a total abdominal hysterectomy and unilateral oophorectomy with partial salpingectomy at an outside institution. Final pathology revealed a STLMP. Thus, she was taken to the operating room to complete the staging procedure including a unilateral salpingo-oophorectomy and removal of the fallopian tube stump, omental biopsy, appendectomy and pelvic and paraaortic lymph node sampling. There was no evidence of gross disease in the abdomen or lymph node-bearing areas.

### Pathologic Findings

Grossly, the tumor was cystic and filled with serous fluid. It measured 5 x 1 x 2 cm. The inner surface was lined with multiple papillary structures measuring from 0.3 to 1.3 cm. Microscopically, the cyst was lined with focal papillae and atypical stratified cells. Focal papillary fronds were noted. There was no stromal invasion. These histological features are indicative of a serous tumor of low malignant potential confined to the left fallopian tube (Figure 4).

## Discussion

There are few case reports of tumors of LMP of the fallopian tube. Tumors of LMP of the fallopian tube classi-

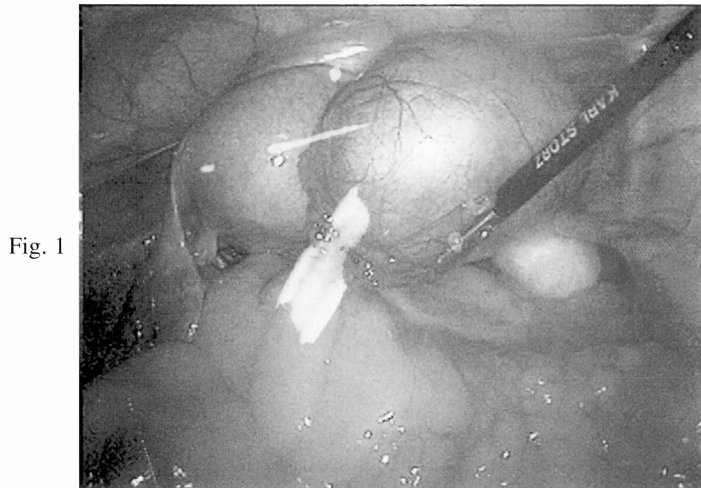


Fig. 1

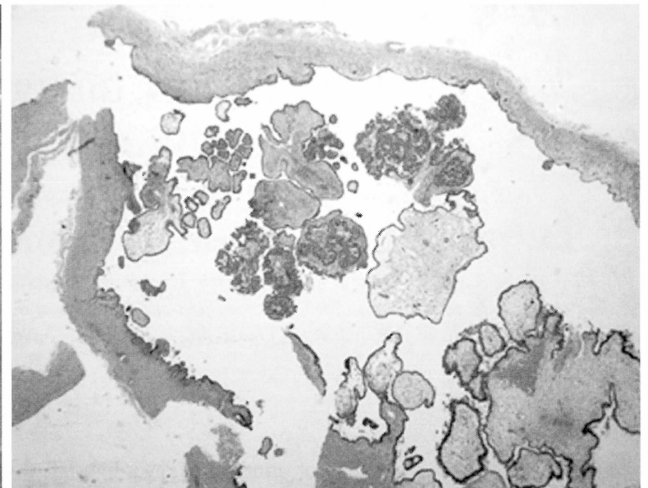


Fig.

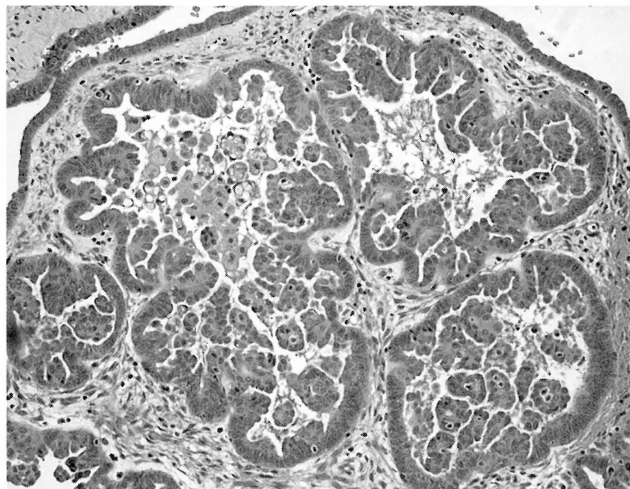


Fig. 3

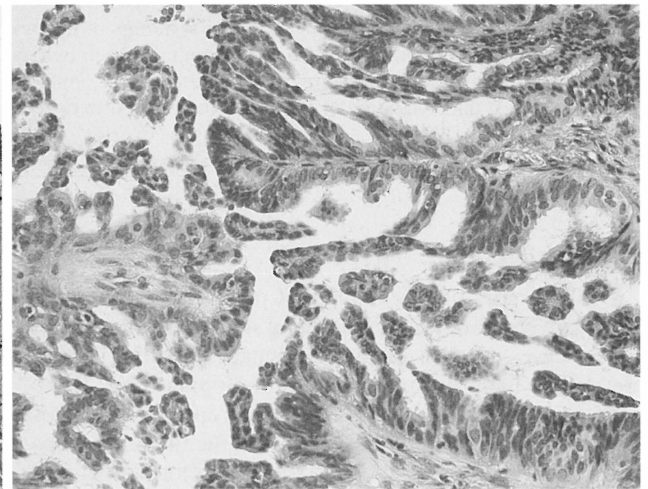


Fig.

Figure 1. — Right fallopian tube with tumor of LMP completely independent of the normal ovary.

Figure 2. — Histology of STLMP of the fallopian tube demonstrating focal papillae and atypical stratified cells (2x magnification).

Figure 3. — STLMP demonstrating focal glandular complexity and branching papillary fronds without stromal invasion (20x magnification).

Figure 4. — Serous tumor of low malignant potential of the fallopian tube.

fied to date have included serous, mucinous, and endometrioid histology. Table 1 summarizes what is currently known about tumors of LMP of the fallopian tube based on 11 published cases. Interestingly, patients with serous tumors presented at a mean age of 30 years old (range 19-43), while those with mucinous tumors were discovered later, at a mean age of 53 (range 49-60). While STLMP have only been reported unilaterally, mucinous LMP tumors are often bilateral. Mucinous LMP tumors have been associated with pseudomyxoma peritonei and can be associated with Peutz-Jeghers syndrome [9].

Patients typically present with non-specific symptoms such as pain, bloating, vaginal bleeding, vaginal discharge, or may be asymptomatic. As in our cases, tumors of LMP are most commonly an unexpected intraoperative finding. When considering the differential diagnosis, the possibility of other fallopian tube abnormalities should be entertained. Epithelial hyperplasia associated with salpin-

gitis can be seen in young women especially in the post-partum period, however it usually does not present with a mass as tumors of LMP do. Mucosal epithelial prolifer-

Table 1. — Reported cases of LMP of the fallopian tube.

Author	Year	Tumor type	Age	Therapy	Follow-up
1 Gatto	1986	Serous	19	Salpingectomy	1.7 y DF
2 Casasola	1989	Serous	32	Salpingectomy	NR
3 Zheng	1996	Serous	31	USO,	6 y DF
				pelvic nodes	2.4 y DF
4 Alvarado-Cabrero	1997	Serous	34	NR	NR
5 Kayaalp	2000	Serous	31	Salpingectomy	NR
6 Villella	2004	Serous	22	Cystectomy	3 y DF
			43	TAH/BSO	—
7 Jones	1965	Mucinous	NR	TAH BSO	1.0 y DF
8 McCarthy	1987	Mucinous	60	Subtotal	NR
				Hyst BSO	NR
9 Freidmann	1992	Mucinous	49	NR	NR
10 Seidman	1994	Mucinous	53	TAH BSO	4.3 y DF
11 Alvarado-Cabrero	1997	Endometrioid	49	NR	NR

NR = Not reported; USO = Unilateral salpingo-oophorectomy; DF = Disease-free; TAH = Total abdominal hysterectomy; BSO = Bilateral salpingo-oophorectomy.

eration (MEP) is defined as a proliferative change such as epithelial cell crowding, stratification, and loss of nuclear polarity. Moore and Enterline demonstrated that focal areas of mild epithelial proliferation were seen in 20% of fallopian tubes from 124 consecutive non-selected hysterectomy cases and is a ubiquitous lesion of no clinical significance [10]. Other studies have shown that moderate or marked MEP commonly accompanies an ovarian tumor of LMP [10]. However, whether these lesions are neoplastic or only proliferative metaplastic lesions is unclear at this time, although the latter is favored [11]. The hallmark of LMP lesions is the absence of stromal invasion. This is where the differentiation between borderline and frank malignancy is often made. Not infrequently, this differentiation must be made on the architectural basis of invasion, rather than inherent cytological characteristics.

The literature currently offers a variety of mixed opinions regarding the treatment of tumors of LMP of the fallopian tube that has been extrapolated from that of LMP of the ovary. The choice between conservative fertility-sparing surgery versus complete staging procedure remains controversial. While no data exist regarding LMP tumors of the fallopian tube, a study by Yazigi demonstrated that approximately 24% of patients with tumors of low malignant potential of the ovary were upstaged with a staging procedure [12]. In this study seven of 29 patients with presumed localized disease were upstaged as a result of staging laparotomy that included cytology, omentectomy, diaphragm biopsies, and extra pelvic peritoneum, pelvic, and aortic lymph node dissection [12]. However, the impact of surgical staging on therapeutic management or on patient survival remains controversial, since both recurrence and transformation into carcinoma are rare. In the event of a recurrence, most tumors are responsive to salvage surgery. However, LMP tumors have a better prognosis as compared to invasive carcinoma and rarely transform or reoccur as invasive carcinoma. In a large study of 339 patients comparing complete hysterectomy and bilateral salpingo-oophorectomy versus fertility-sparing surgery (unilateral salpingo-oophorectomies or cyctectomies) the overall disease-free survival irrespective of surgical method was 99.6% for Stage I disease, 95.8% for Stage II, and 89% for Stage III after five years [14]. Disease-free survival comparing complete to conservative surgery was as follows: 100% vs 99.3% for Stage I, 91% vs 100% for stage II, and 100% vs 81.2% for Stage III. They also reported a low 2% risk of developing invasive carcinoma. Given the protracted course of LMP tumors, and their

propensity to develop in spared ovaries, Zanetta *et al.* recommend yearly follow-up with ultrasound evaluation. However, this is controversial and there is no data to support its use [13].

Tumors of low malignant potential of the fallopian tube are an exceedingly rare entity. At this point in time, there are too few cases in the literature to determine the optimal treatment of tumors of LMP of the fallopian tube. We urge continued reporting of these tumors so that we might be able to better understand these rare tumors and develop a more cohesive management plan.

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