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# A Case of Low-Grade Appendiceal Mucinous Neoplasm of Its Difficultly to Distinguish from a Right Ovarian Tumor Due to Postmenopause

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## Abstract

We here present a rare case of appendiceal tumor mimicking ovarian tumor in menopause woman. The patient was a 56-year-old woman, G1P1, who presented to our hospital with a right adnexal cyst diagnosed at another hospital. Transvaginal echocardiography showed a cyst in the right adnexal region, and pelvic contrast-enhanced MRI revealed a small cyst in the same region. The left ovary was atrophic and identifiable. It was unclear whether the cyst was contiguous with the gastrointestinal tract. Blood tests showed no elevation of tumor markers. We considered its possibility of a gastrointestinal origin, but since right normal ovary was not found, we thought the tumor was of ovarian origin and decided on a laparoscopic resection of the right adnexa. Intraoperatively, we observed atrophied bilateral normal ovaries, and the pelvic tumor was contiguous to the appendix. Surgeons performed a laparoscopic appendectomy after consultation with us. After resection we searched the abdominal and pelvic cavities, but found no obvious disseminated lesions. The histological diagnosis was low-grade appendiceal mucinous neoplasm (LAMN), a rare benign tumor of the appendix. Appendiceal tumors can be difficult to differentiate from right ovarian tumors due to their close anatomic location in the pelvis. It is possible to determine whether the tumor is of ovarian or appendiceal origin by identifying normal ovaries and the location of the feeding vessels into the tumors. In our case, there were no lesions other than the appendix, but LAMN can metastasize to the ovary, cause pseudomyoxoma peritonei, or be an overlapping tumor with an ovarian tumor. If an appendiceal tumor is diagnosed after surgery for ovarian tumor, the intra-abdominal cavity should be searched for metastasis or dissemination, and a thorough search for ovarian lesions should be performed with the possibility of an overlapping tumor in mind.

**Keywords**

Low-Grade Appendiceal Mucinous Neoplasm, Ovarian Tumor, Menopause Woman

## 1. Introduction

Low-grade appendiceal mucinous neoplasm (LAMN) is a low-atypical mucus-producing tumor of the appendix that is histologically benign. However, LAMN can disseminate to the peritoneum, resulting in peritoneal pseudomyxoma, or can metastasize to the ovaries and have a clinically malignant course. In the preoperative diagnosis, LAMN is often mistaken for an ovarian tumor. The ovaries and appendix are anatomically located in close proximity, and especially after menopause, it may be difficult to identify the atrophied ovaries, making it difficult to determine preoperatively whether the pelvic tumor is of appendiceal or ovarian origin. In addition, LAMN and ovarian tumors may occur simultaneously. In such cases, it is possible to laparoscopically resect both lesions in the same operative field. In this report, we describe a case in which an appendiceal lesion was resected during laparoscopic surgery for a pelvic tumor that was difficult to diagnose preoperatively.

## 2. Case Report

A 56-year-old Japanese woman was diagnosed with a right ovarian tumor after an enlarged ovary was noted during a cervical cancer screening performed at another hospital, and she was referred to our hospital. Transvaginal ultrasonography at the time of the initial visit revealed a 30 mm-sized nodular cyst in the right adnexal region (Figure 1). Pelvic contrast-enhanced MRI scan revealeda 42 × 25 mm nodular cyst was found in the right side of the pelvis in the adnexal region, with no substantial component or irregularity of the tumor wall inside the

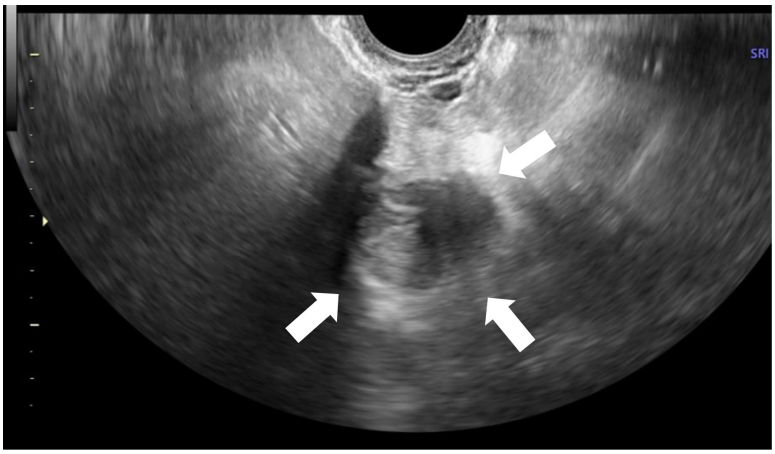


Figure 1. Transvaginal echography: a 30 mm-sized nodular cyst in the right adnexal region (surrounded by arrows).

tumor and no clear continuity with the digestive tract. The left ovary was identifiable, but the right ovary was not, and we considered it more likely that the tumor originated from the right ovary than from the gastrointestinal tract (Figure 2). Blood tests showed CA19-9 < 2.0 U/ml, CA125 4.4 U/ml, CEA 1.9 ng/ml, and no tumor markers were elevated.

Based on the preoperative examination, we thought the tumor was of right ovarian origin, but it was unclear whether the tumor was contiguous with the gastrointestinal tract, so we considered the possibility of an appendiceal tumor. If it was a right ovarian tumor, we decided to perform a laparoscopic resection of the right adnexa, and a port was placed in a modified diamond style to begin surgery. There was a small amount of clear ascites in the pelvic cavity, no disseminated lesions from the abdominal cavity to the pelvic cavity, and no other abnormal findings except for the presence of atrophied bilateral ovaries. The tumor, which was thought to be a right ovarian tumor preoperatively, was contiguous from the ileocecal region and was thought to be an appendiceal tumor. We consulted the surgeons, who performed a laparoscopic appendectomy. There was no leakage of tumor contents into the abdominal cavity. The operation time was 1 hour and 16 minutes, and the patient experienced only minimal bleeding. The patient started eating on the first postoperative day and was discharged on the third postoperative day.

On macroscopic examination, appendiceal tumor with internal mucous retention and papillary growth of cylindrical epithelium without cellular atypia was observed. There was no tumor exposure to the serosal surface, no mucus leakage outside the appendiceal wall, and the resection margins were negative (Figure 3). We reached diagnosis of LAMN.

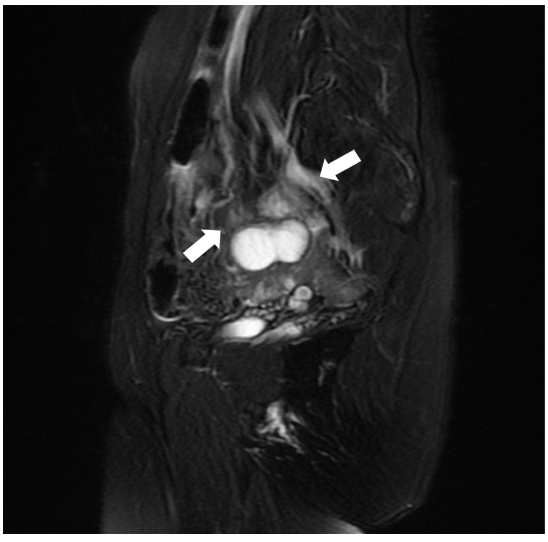
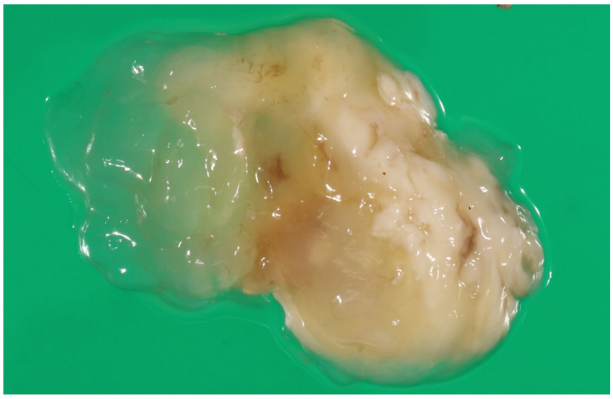
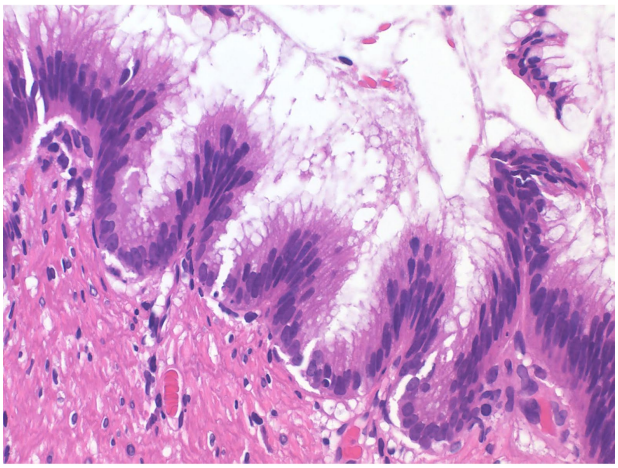


Figure 2. Abdominal MRI: a 42 × 25 mm nodular cyst in the right side of the pelvis in the adnexal region (surrounded by arrows). Cyst with no substantial component or no irregularity of its wall inside the tumor and no clear continuity with the digestive tract.



(a)



(b)

**Figure 3.** (a) and (b) A cyst with jelly-like mucus of the appendiceal tumor; villous proliferative lesions of villous adenoma-like mucinous columnar epithelium with mucous deposits around it with mild stromal infiltration (H&E at ×200 magnification).

The patient is currently being followed up at an outpatient clinic, and no recurrence has been observed 12 months after operation.

## 3. Discussion

LAMN is a relatively uncommon low atypical mucinous tumor of the appendix that can cause peritoneal dissemination, ovarian metastasis, or pseudomyxoma peritonei (PMP), in which the abdominal cavity is filled with neoplastic mucus due to leakage of mucus caused by rupture of the tumor [1]. Appendiceal tumors can cause symptoms such as right lower abdominal pain and abdominal mass [2] [3], but depending on the degree of appendiceal enlargement, they are often not accompanied by symptoms, and those with a diameter exceeding 10 cm in length may be discovered by chance [4]. For the diagnosis of LAMN, imaging tests such as abdominal echography are useful for the diagnosis of appendiceal tumors [1]. In the typical case, an echo examination reveals a cystic appendec-

tomy with hypoechoic internal expansion, and MRI and CT scans are also useful [4]. In some cases, however, it is difficult to distinguish the appendage from ovarian tumors that are anatomically located in close proximity in the pelvis, making the preoperative diagnosis difficult [4]. In particular, postmenopausal atrophied ovaries are difficult to detect by imaging studies such as MRI and transvaginal echocardiography, and pelvic tumors are sometimes difficult to determine whether they originate from the ovary or the gastrointestinal tract [5]. The evaluation can be difficult because of artifacts caused by peristalsis. In this regard, there is a report that CT angiography was used to determine whether a pelvic tumor originated from the gastrointestinal tract or the ovary by identifying the tumor nutrient vessels [6]. Since CT angiography, a noninvasive examination, is superior to contrast-enhanced CT in the evaluation of tumor-nourishing vessels because it can confirm more detailed vascular rundown [6], it was not attempted in this case, but imaging should be considered if the evaluation of tumor origin is difficult.

Surgical treatment is the first-choice treatment for LAMN, and is particularly important in terms of reducing the risk of recurrence and early detection and prevention of PMP [7]. Although there is no clear standard for the resection of the lesion, the margin of the resection margin should be sufficient because there have been reports of mucinous cystadenoma developing at the resection margin of the appendectomy. Lymph node dissection may be performed if appendiceal cancer is suspected preoperatively, but appendicectomy or ileal resection is the usual treatment for LAMN. Tumor wall rupture and peritoneal dissemination may result in PMP [8]. In the absence of intraperitoneal mucus accumulation or peritoneal seeding, the risk of PMP recurrence or development is low if there is no leakage of tumor contents [9]. The appendix was removed laparoscopically without intraoperative failure. In case of benign ovarian disease, surgery is often performed laparoscopically, and in such cases, there are two types of port placement: modified diamond style and parallel style. There is a report that a laparoscopic port was placed and ovarian and appendiceal lesions were operated in a modified diamond style [10].

The appendices and ovaries are anatomically located in close proximity, which may cause problems in terms of inflammatory spillover and metastasis of tumors, and metastasis of LAMN to the ovaries is also a problem when PMP is present [11]. Other borderline ovarian malignancies can be associated with LAMN even in the absence of PMP [12], so it is important to search the ovary at the time of surgery regardless of the presence of PMP. In the present case, appendicectomy was performed in the absence of PMP and bilateral ovarian enlargement. It is important to perform a thorough search of the abdominal and pelvic cavities at the time of surgery, especially when PMP is present, because of the high risk of recurrence. In addition to searching for PMP, LAMN should also be thoroughly searched for ovarian metastasis or overlapping tumors between the appendix and ovaries [4], even in the absence of PMP, and ovarian resection should be considered in some cases when ovarian enlargement or other ovarian lesions are

observed.

## 4. Conclusion

We experienced a case of LAMN that was difficult to diagnose preoperatively. When a pelvic tumor is detected, it is sometimes difficult to determine whether the tumor is of ovarian or appendiceal origin. This is determined by the presence or absence of continuity between the tumor and the ovary or appendix, as well as by the running of tumor-supporting blood vessels, but it is sometimes difficult to identify an atrophied ovary after menopause by imaging studies such as echography, CT, and MRI. In addition, tumor-trophoblastic vessels may be difficult to evaluate with contrast-enhanced CT scans, in which case CT angiography may be useful. In the preoperative diagnosis of ovarian tumor, if LAMN is found intraoperatively, it is important to ensure complete resection with a margin without disruption of the tumor wall, and to perform a thorough intraoperative search for PMPs such as peritoneal seeding in order to decrease the risk of recurrence. LAMN may also be associated with ovarian lesions or PMPs, and a thorough search of the abdominal cavity for metastases or seeding as well as the presence of ovarian lesions should be performed.

## Authors' Contribution

Tsutomu Muramoto—diagnosis and treatment, corresponding author.

Kyousuke Kamijo and Megumi Sano—assistance of diagnosis and treatment.

Yuki Ibuki—assistance of treatment.

Atushi Mori and Yaeko Kobayashi gave Muramoto advice in preparing the dissertation considerations.

## Authors' Statement

There is no conflict of interests which occurs when the authors remain in a financial or personal relationship which unjustly affects his actions associated with the publication of the manuscript.

Any possible relationship(s) of the author(s) with the party/parties interested in the publication of the manuscript is revealed in the text of the article.

The manuscript has not been published in or submitted to any other journal.

No source of financing.

The patient had given her own informed consent for the case report to be published.

## Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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