



## Case Report

# Endosalpingiosis in Pelvic Lymph Node with Atypical Endometrial Hyperplasia

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### Abstract

Endosalpingiosis, defined by the presence of tubal-type epithelium in multiple anatomical places, has emerged as a benign condition related with Mullerian disease, most notably endometriosis and endocervices. Remarkably, its association with persistent pelvic discomfort and genital tract cancers has received more attention. We describe the case of a 44-year-old female with nodal Endosalpingiosis with underlying atypical endometrial hyperplasia. The case discussed here adds to understanding the existence of Endosalpingiosis inside pelvic lymph nodes, which goes beyond staging for gynecological cancers.

**Keywords:** Endosalpingiosis; Pelvic Lymph Node; Endometrial Atypical Hyperplasia.

### Introduction

Endosalpingiosis is a benign condition characterized by the presence of tubal-type epithelium in female reproductive organs, bladder, variform appendix, colon, lymph nodes and skin [1]. Recent evidence have suggested that Endosalpingiosis is the end spectrum of Mullerian disorder (milrinones) and mostly seen in concordance with endometriosis and endocervices [2]. Endosalpingiosis has been linked with chronic pelvic pain and genital tract malignancies [3]. The histological changes of Endosalpingiosis have been previously reported in pelvic lymph nodes of patients undergoing staging for gynecological malignancies, axillary lymph nodes of women with breast cancer. The reported prevalence of Endosalpingiosis in literature ranges from 7.6-12.5%. When Endosalpingiosis is found in a lymph node, it must be distinguished from adenocarcinoma lymph node metastases. Here we are presenting a case of 44-year-old women with nodal Endosalpingiosis with underlying atypical endometrial hyperplasia.

### Case Report

A 44-year Para 2 female, with previous two normal vaginal deliveries referred to our gynecology department with abnormal

uterine bleeding described mainly by prolonged heavy menstrual bleeding and dysmenorrhea. Patient was initially investigated with pelvic ultrasound which revealed thickened endometrium. She was then booked for hysteroscopy, endometrial sampling and IUD insertion which was performed and showed a thickened endometrial lining with no endometrial polyps or submucosal fibroids. Unfortunately, histopathology came back showing atypical endometrial hyperplasia. As her family is complete, patient has been counselled and booked for total laparoscopic hysterectomy, bilateral salpingectomy with lymph node sampling. Prior to the procedure, the patient had MRI pelvis performed as a baseline due to the underlying pathology of atypical endometrial hyperplasia, which showed no underlying endometrial cancer or further abnormality. Procedure was done 8 weeks post the initial diagnosis. During laparoscopy, it was noted that there is inactive endometriotic nodules with both ovaries' adherent to the uterine side walls, adenomyotic uterus and enlarged pelvic lymph nodes. Procedure was carried out as planned and she had an uneventful recovery and post-operative period.

Histopathology review showed no evidence of malignancy. There was evidence of adenomyosis and leiomyoma in the uterus. Left pelvic lymph nodes had focus of Endosalpingiosis. We believe that this is the first case in literature of pelvic lymph node Endosalpingiosis with no underlying gynecological malignancy.

## Discussion

Endosalpingiosis, a peritoneal serous lesion, has been associated with serous borderline tumors and low-grade serous ovarian neoplasms [4]. Hence, there is increasing evidence that there might be a direct association between Endosalpingiosis and ovarian cancer [5]. Endosalpingiosis involving the pelvic and para-aortic lymph nodes is not uncommon. Nodal Endosalpingiosis has been reported to be present in 5% of women undergoing pelvic or para-aortic lymph node removal during surgery for gynecological cancer. In 2012, Prince et al published a series of 110 cases diagnosed with Endosalpingiosis, endometrial adenocarcinoma was also present in 16% of cases, while serous borderline ovarian tumors were present in 7% of cases. Endosalpingiosis involving pelvic nodes with atypical endometrial hyperplasia has not been reported.

Rise was the first to identify peritoneal Mullerian inclusions in 1897. Sampson used the term “Endosalpingiosis” in 1930 to characterize the local proliferative and invasive features of tubal mucosa following surgical interruption. He investigated the tubal stumps of 147 individuals who had previously had a salpingectomy or tubal sterilization and discovered that sprouts of fallopian tube epithelium frequently infiltrated and expanded beyond the stump wall. Novak questioned this description, noting that similar lesions were also detected in individuals who had a history of pelvic inflammatory illness but had no history of tubal surgery. These lesions were radiologically and histologically similar to salpingitis isthmica nodosa.

It is a significant clinical entity since it has been linked to chronic pelvic discomfort, can cause substantial diagnostic issues with peritoneal washings, has been linked to ovarian and cervical neoplasms, and can progress to malignancy [6]. Unfortunately, it is frequently misdiagnosed as endometriosis and ablated with diathermy (Shah et al.). Laparoscopy with excision of lesions suggestive of endometriosis will very certainly raise the frequency of Endosalpingiosis just by diagnosis. Excision of worrisome lesions rather than ablating lesions would not only help our understanding of Endosalpingiosis but may also be the most appropriate treatment for the illness.

Endosalpingiosis is a very uncommon but growingly recognized disorder defined by the presence of tubal-type epithelium in diverse extra-tubal locales. It is currently described as the presence of benign epithelium histologically comparable to tubal epithelium in an ectopic location [7]. The peritoneum is the most common location of development, although it is also seen in the omentum, urinary bladder, and pelvic and para-aortic lymph nodes [8]. Endosalpingiosis is seen in 5.3% of pelvic lymph nodes dissected for the treatment of gynecological malignant tumors. Recent research reveals a link between it and Mullerian

diseases, as well as endometriosis and endocervices. According to the research, its prevalence ranges from 7.6% to 12.5%. The case discussed here contributes to a better understanding of its presence inside pelvic lymph nodes, which has previously been documented in patients undergoing staging for gynecological cancers and other disorders.

The presented case is notable for the conjunction of nodal Endosalpingiosis with underlying atypical endometrial hyperplasia [9]. Prior research has not thoroughly established this connection. This revelation may prompt future research into the potential interaction or shared pathophysiology between these disorders, implying the need for thorough examination and therapeutic options for individuals who arrive with comparable findings.

Clinically, the importance of detecting nodal Endosalpingiosis in the presence of atypical endometrial hyperplasia necessitates a multidisciplinary approach to treating such patients. While there was no indication of malignancy on histology, the putative link between Endosalpingiosis and gynecological tumors demands close monitoring and follow-up for prospective malignancy development. Furthermore, this instance emphasizes the significance of rigorous preoperative evaluations and meticulous histological exams in gynecological abnormalities patients. We report a case of nodal Endosalpingiosis involving pelvic nodes with underlying atypical endometrial hyperplasia [10].

## Conclusion

This case emphasizes the importance of heightened monitoring and extensive examination when facing unusual but potentially significant presentations such as nodal Endosalpingiosis in conjunction with atypical endometrial hyperplasia. The unique feature of this case resides in the coexistence of nodal Endosalpingiosis with atypical endometrial hyperplasia, a link that has not been widely described in the current literature. This case report adds to the growing body of knowledge about nodal Endosalpingiosis in conjunction with atypical endometrial hyperplasia, highlighting the importance of ongoing research, comprehensive evaluation, and multidisciplinary collaboration in the management of such complex clinical scenarios.

## Declaration of Interest: None

Ethical Approval is not Required by Beaumont Ethical Committee for Case Reports.

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