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A Rare Mimic of Malignancy: Papillary Endosalpingiosis

Abstract

Endosalpingiosis is the presence of ectopic Müllerian-type epithelium resembling Fallopian tube epithelium. It usually appears as small glandular inclusions in various peritoneal sites or lymph nodes. Rarely, it is papillary in configuration, and lack of familiarity with this entity could lead to overinterpretation as a malignancy. We recently encountered a case of papillary endosalpingiosis found incidentally at laparotomy for ectopic pregnancy. Awareness of this entity is important, as it may be a diagnostic challenge.

Keywords

Papillary endosalpingiosis, ectopic Fallopian tube epithelium, Müllerian epithelium, Müllerian neoplasms

Introduction:

Endosalpingiosis is the presence of ectopic Müllerian-type epithelium resembling Fallopian tube epithelium. Although it is often seen in association with serous tumors of low malignant potential("borderline tumors", "atypical proliferative tumors"), where it needs to be distinguished from tumor implants, it may be an isolated finding. It is part of a triad of ectopic Müllerian-type epithelium that also includes endometriosis and endocervicosis. Endosalpingiosis usually appears as small glandular inclusions in various peritoneal sites or lymph nodes. Rarely, it is papillary in configuration, may have associated calcifications or psammoma bodies, and lack of familiarity with this entity could lead to overinterpretation as a malignancy. We recently encountered a case of papillary endosalpingiosis found incidentally at laparotomy for ectopic pregnancy. Awareness of this entity is important, as it may be a diagnostic challenge.

Case

A 21-year-old G1P1 woman underwent salpingectomy for ectopic pregnancy. No gross evidence of a papillary lesion was noted grossly. At histology, in addition to the tubal pregnancy, an incidental low-grade serous epithelial proliferation with psammomatous calcifications was seen on the surface of the Fallopian tube (figure 1). The lesion showed weak focal nuclear staining for p53 and a ki-67 index of 10%. No KRAS or BRAF mutations were detected. A diagnosis of benign papillary endosalpingiosis was rendered.

Discussion

Endosalpingiosis is the presence of ectopic Müllerian-type epithelium resembling Fallopian tube epithelium. Psammoma bodies may be present. It is frequently found on the peritoneal surface of pelvic structures including the fallopian tubes, uterus, ovaries and omentum, as well as in pelvic lymph nodes, and is often present in association with low malignant potential("borderline") tumors, which may complicate diagnosis, as there may be confusion with peritoneal tumor implants(1). Patients are often of reproductive age, but the lesion has been described frequently in postmenopausal women. There may be a history of pelvic pain, dysmenorrhea or a history of pelvic inflammatory disease(1,2).

Two theories exist regarding etiology; metaplasia of mesothelial lining to secondary Müllerian-type epithelium and implantation of salpingoliths from the fallopian tube secondary to chronic inflammation (3,4). Most of these lesions are not worrisome for malignancy, however epithelial proliferation can occur, and cause diagnostic difficulty. The spectrum of pathology postulated by Kurman et al(4) includes the proliferation of tubal epithelium secondary to chronic inflammation, progression to papillary tubal hyperplasia and the shedding and implantation of salpingoliths onto ovarian and peritoneal surfaces. This was postulated to extend to the transformation of these benign lesions to atypical proliferative serous tumors, serous borderline tumors and serous carcinoma of the ovary. This implantation theory is supported by the absence of reported cases of serous metaplasia in men, the presence of salpingoliths in 24% of atypical proliferative serous tumors and multiple reports of associated endosalpingiosis and papillary tubal hyperplasia with serous borderline and malignant tumors(4).

Occasionally, endosalpingiosis may be papillary or appear to be proliferating. There have been reports of supradiaphragmatic florid papillary endosalpingiosis within lymph nodes (5). Papillary endosalpingiosis, though a benign entity, can present a diagnostic dilemma, and can be mistaken for serous carcinoma of the ovary or primary serous surface carcinoma of the peritoneum. It is critical to make the distinction, due to the differences in therapy. Metastatic high grade serous carcinoma will likely display features of malignancy including nuclear atypia, stromal desmoplasia, and increased mitotic count. Serous carcinoma is associated with p53 aberrations, which would include either increased or null immunostaining, and Ki-67 index would be expected to be markedly increased. In addition, if low grade serous carcinoma was a consideration, Kras mutations have been shown to be associated(4). Our case did not display any histologic features of malignancy, had a ki-67 index of 10%, weak focal p53 staining and lacked KRAS and BRAF mutations on molecular studies. In discussing the case with the clinician, a possible history of prior pelvic inflammatory disease was elicited, and the proliferation may relate to prior inflammation.

Conclusion

Benign papillary endosalpingiosis belongs to a spectrum of ectopic fallopian epithelium postulated to arise from proliferation of tubal epithelium due to chronic inflammation or metaplasia. Its resemblance to metastatic serous carcinoma and its frequent association with borderline and low grade serous carcinomas in particular, can present a diagnostic dilemma. Appropriate sampling, adjuvant immunohistochemical staining, and molecular studies can aid in the confirmation the diagnosis. Clinical correlation is important as well.

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Legends

Figure 1-Low power of lesion on surface of Fallopian tube(A). There is extensive calcification(B). The epithelial component of the lesion is bland with no significant atypia(C).