Endosalpingiosis Presenting as Infraumbilical Mass- A Rare Case Report with Review of Literature

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ABSTRACT

Background: Endosalpingiosis is an understudied gynaecological entity with limited research on it. Clinical significance of endosalpingiosis is not fully understood. Because of unfamiliar presentation, it can lead to misdiagnosis.

Case Report:We present a case of endosalpingiosis in a 54 years old female patient presenting as infraumbilical mass with history of tubal ligation 30 years ago. Gross examination reveals a grey white to grey brown lesion of size 5 x 3.5 x 3cm, microscopy showing cystically dilated spaces lined by low columnar to cuboidal and attenuated epithelium with focal areas showing dystrophic calcification.

Discussion: Endosalpingiosis is a rare benign diagnosis which presents at postmenopausal or older age with clinical presentation as abdominal pain, dysmenorrhea, pelvic discomfort, dysuria or hematuria.

Conclusion: Endosalpingiosis is a rare and uncommon clinical entity. The definitive diagnosis is possible only with histopathological examination. It is important to rule out other comorbidities existing with endosalpingiosis.

Key words: Endosalpingiosis, Tubectomy scar, Histopathological examination

INTRODUCTION

Endosalpingiosis is the presence of ciliated tubal lining epithelium in an ectopic location other than normal origin of fallopian tube [1]. Endosalpingiosis may be found as incidental diagnosis during surgery or it can present as pelvic pain or chronic pelvic inflammation. The entity was first described by Sampson in 1930[2]. We present a case of endosalpingiosis to highlight its rare occurrence and uncommon clinical presentation which can lead to misdiagnosis.

Case Report:

A 54 year old female presented with infraumbilical mass with lower abdominal pain since 3 months. She gave history of tubal ligation 30 years ago. There were no other comorbidities. Routine laboratory parameters including complete blood count and urine examination were within normal limits. Hypoechoic, irregular shaped mass lesion with minimal spongiform cystic change noted in the infra-umbilical region beneath the incisional scar site.

Figure 1The mass was surgically excised considering as a neoplasm and sent for histopathological examination.

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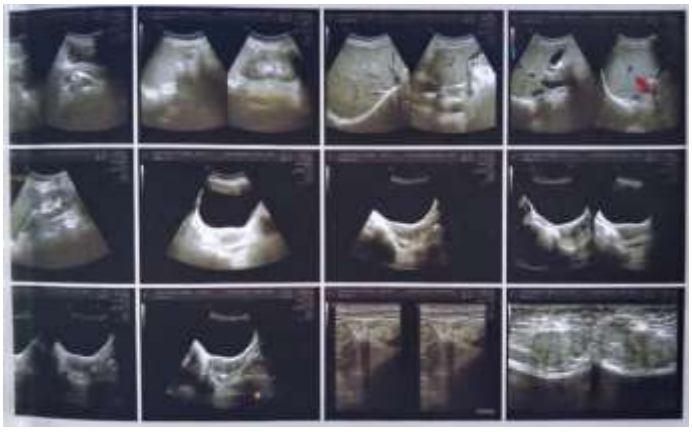
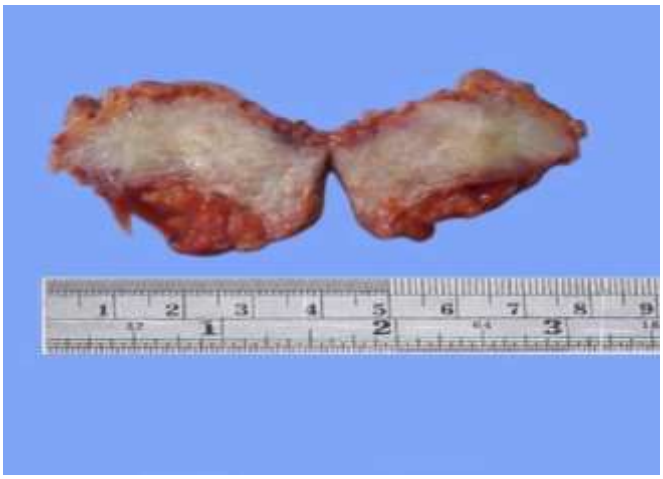
Figure 1: Hypoechoic, irregular shaped mass lesion with minimal spongiform cystic change noted in the infra-umbilical region beneath the incisional scar site.



Gross examination revealed a single, irregular, grey white to grey brown, soft to firm mass of size 5 x 3.5 x 3cm. Cut section was grey white with small punctate translucent spaces(Figure 2).

Figure 2: Gross examination: grey white translucent lesion with punctate spaces.



Microscopic examination revealed cystically dilated spaces lined by low columnar to cuboidal as well as attenuated epithelium, focally showing multilayering of cells and rounded regular nuclei. Lumina of these cystic spaces revealed secretions and focal areas showing psammoma bodies. Intervening fibrous tissue showed chronic nonspecific inflammation. Endometrial

Stroma was not seen on microscopy(Figure3a and 3b) Considering the past history and microscopic features, diagnosis was offered as endosalpingiosis. After excision of the lesion, 6-month postoperative follow-up is uneventful.

Figure 3A: Microscopy- cystically dilated spaces lined by low columnar epithelium(100x H&E)Figure 3B: Microscopy- Cystic spaces with secretions and psammoma bodies(400x H &E)

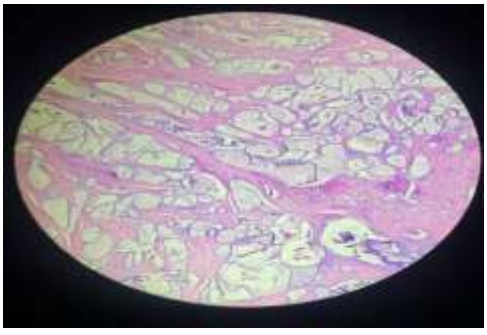


Figure 3A

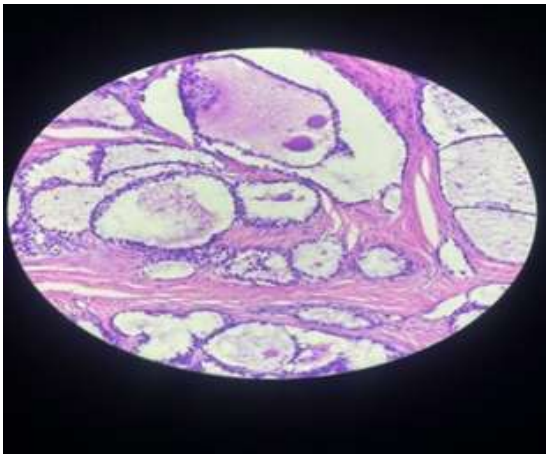


Figure 3B

DISCUSSION

Endosalpingiosis is a benign, uncommon, and challenging diagnosis. It can exist with other comorbidities, commonest age of presentation is postmenopausal and older age women[3]. In one of the studies, it was found, in 7.6% of 110 women undergoing laparoscopic procedure [4] Clinical presentation of endosalpingiosis is abdominal pain, fever, dysuria, dysmenorrhea, back pain and hematuria. Occasionally, it can manifest as tumor as seen in our case [5]. The pathogenesis of endosalpingiosis is obscure. However, 3 main theories include shedding of tubal epithelium into pelvic peritoneal, coelomic metaplasia of pelvic peritoneum epithelium and growth of embryonic mullerian tissue which is misplaced during embryogenesis [6]. Other possible mechanism is transplantation of tubal mucosa to peritoneal surfaces during tubal surgery or lymphatic vascular implantation of tubal epithelium [2]. In our case, the mechanism may be transplantation of tubal mucosa during tubal ligation. Endosalpingiosis can coexist with endometriosis.

Endosalpingiosis involves uterus, ovary, fallopian tubes and within the skin [2], but can be found anywhere in the abdomen and pelvis [3,7]. However, the lesion is not easily recognized by gynecologist and hence can be misdiagnosed with neoplastic lesion, as seen in our case.

Laparoscopy remains the main diagnostic tool to visualize this lesion. However, it is often misdiagnosed as endometriosis by the gynecologist and the definitive diagnosis is made only by histopathological examination of excised lesion, which on examination shows numerous punctate fluid filled translucent lesions as observed in our case. Histopathological examination of endosalpingiosis shows presence of glands lined by tubal epithelium or containing three types of cells that is ciliated columnar cells, non-ciliated columnar mucus cells and intercalary or peg cells in a ectopic site. Also, it has been associated with psammoma bodies. This observation was also noted in our case. Endometrial stroma and hemorrhagic areas are absent which rules out possibility of endometriosis[2].

Endosalpingiosis can be differentiated from serous tumors using IHC. The epithelial cells in endosalpingiosis are typically positive for PAX8, WT1, and CK7, and negative for calretinin and D2-40, supporting a Müllerian (tubal) origin. Unlike serous borderline or malignant tumors, p53 shows a wild-type (non-mutant) staining pattern, and Ki-67 proliferation index is low. In contrast, serous tumors often exhibit aberrant p53 expression (mutant-type), high Ki-67 index, and may show architectural complexity and stromal invasion.

In our case, the patient underwent complete excision of the lesion, and a 6-month postoperative follow-up was uneventful. Surgical exicision is treatment of choice in cases with endosaphingiosis, as hormonal therapeutics are not useful. In few cases malignant change has been observed [2].

CONCLUSION

Endosalpingiosis is a rare diagnosis which can be clinically mistaken as endometriosis, pelvic inflammatory disease or neoplasm. Laproscopic examination may help to make a diagnosis however histopathological examination of surgical biopsies helps to give definitive diagnosis.

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