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# Lymph node involvement in endosalpingiosis. A case report and literature review

## Abstract

*Endosalpingiosis is a rare benign gynaecologic condition characterised by the presence of epithelial inclusions resembling tubal epithelium outside the fallopian tubes. It is usually found incidentally in postmenopausal women. The ovary seems to be the most common site of the lesions; however it can develop almost in any pelvic or abdominal organ including the lymphatic structures. We present the case of a 49-year-old patient who was submitted to surgery for bilateral ovarian tumors strongly resembling to malignant tumors. A total hysterectomy with bilateral adnexectomy, pelvic lymph node dissection and omentectomy were performed. The histopathological studies revealed the presence of bilateral ovarian endosalpingiosis associated with lymph node involvement.*
***Keywords:*** *endosalpingiosis, lymph node, involvement, dissemination*

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## Introduction

Endosalpingiosis is a rare benign gynaecologic condition which was first described by Sampson in 1930 and is characterised by the presence of tubal resembling epithelium outside the fallopian tubes[1,2]. These islets contain ciliated, non-ciliated, columnar and intercalated cells which develop a continuous growing process ectopically. However they keep the histological features of tubal epithelium, without associating any endometrial stromal cells around the inclusions[3]. Most often they remain asymptomatic for a long period of time. Therefore in certain cases they can cause chronic pelvic pain and pelvic viscera adhesions[4]. It is estimated that 40% of cases diagnosed with endosalpingiosis are post-menopausal women while in 34.5% of cases and association between endometriosis and endosalpingiosis This association is usually related to the lack of response of the coexisting endometriosis to hormone based therapy[5]. Despite extensive investigations, pathogenesis of endosalpingiosis is not clearly understood, several mechanisms being incriminated. Among the most common theories regarding endosalpingiosis pathogenesis include Mullerian metaplasia theory and tubal cell origin theory[6].

## Case report

A 49-year-old patient presented for debilitating pelvic pain and constipation. The transvaginal ultrasound revealed the presence of bilateral ovarian cysts measuring 4/6/4 cm on the left side and 5/3/4 cm on the right side with homogeneous aspect, with rectal compression. The patient was submitted to a pelvic magnetic resonance imaging. The imagistic studies could not distinguish between bilateral benign ovarian cysts and ovarian carcinoma, therefore the patient was submitted to surgery with radical intent and a total hysterectomy with bilateral adnexectomy, omentectomy and pelvic lymph node dissection were performed. The histopathological studies revealed the presence of bilateral ovarian endosalpingiosis. In the meantime the harvested lymph nodes had also positive staining for endosalpingiosis. Serial 3µm sections had been cut from paraffin blocks and stained with hematoxylin and eosin (H&E) (Figure 1).

The immunohistochemistry (IHC) was performed on 3 µm sections from 10% formalin-fixed paraffin-embedded tissues according to the IHC method an indirect bistadial technique performed with a polymer based

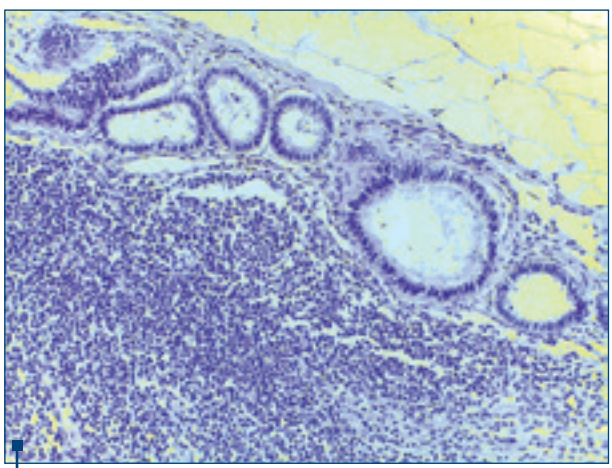

*Figure 1. Endosalpingiosis, H&E staining*

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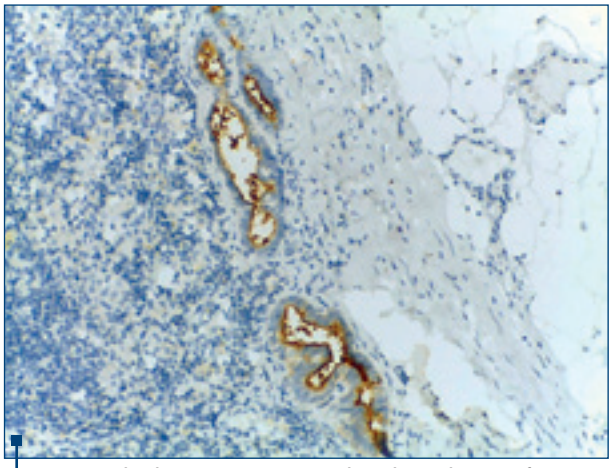

*Figure 2. Endosalpingiosis: positive imunohistochemical staining for CA 125*

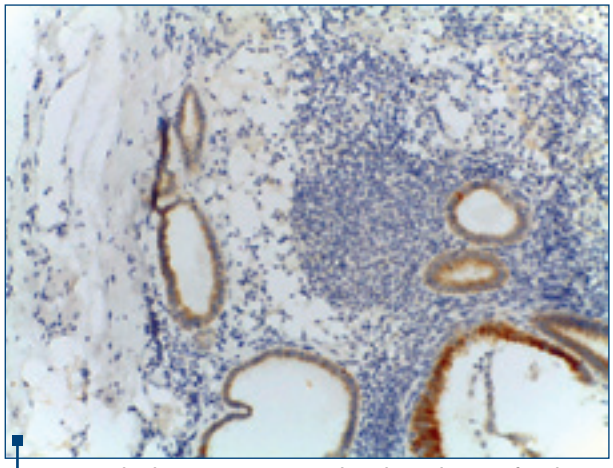

*Figure 3. Endosalpingiosis: positive imunohistochemical staining for calretinin*

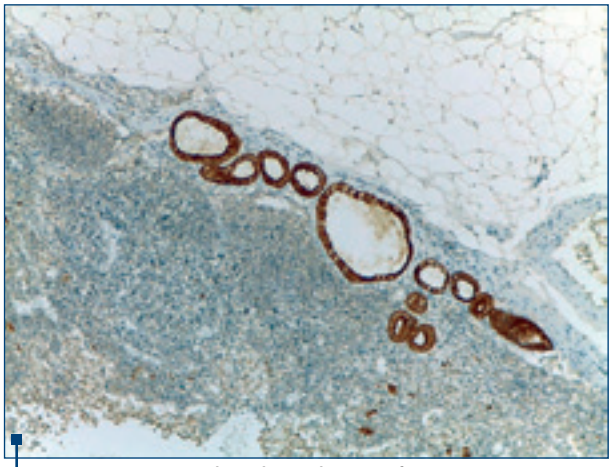

*Figure 4. Positive imunohistochemical staining for CK7*

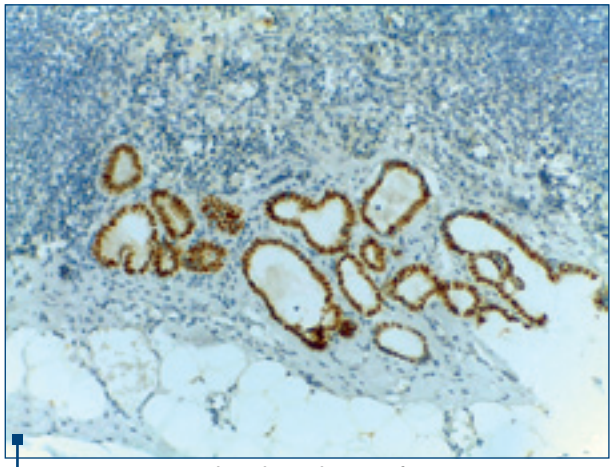

*Figure 5. Positive imunohistochemical staining for ER (98%)*

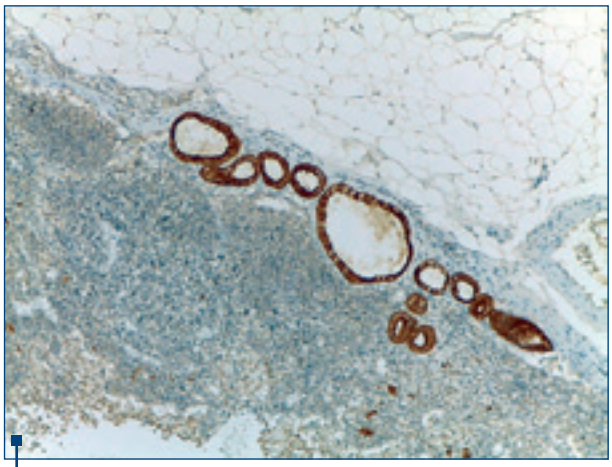

*Figure 6. Positive imunohistochemical staining for mesotelin*

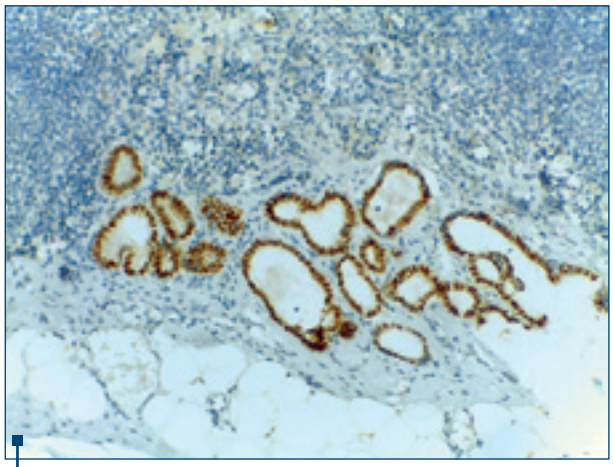

*Figure 7. Positive imunohistochemical staining for WT1*

detection system (Max Polymer Detection System - Leica Ref: RE 7280-k). Tissue sections were spread on poly-L-lysine-coated slides, immersed in three changes of xylene and rehydrated using a graded series of alcohol. Antigen retrieval was performed in microwave oven. In each section, endogenous peroxidase was

blocked by 20 min incubation in 3% hydrogen peroxide. The sections were incubated with primary antibody: Cytokeratin 7 (Leica,1:50, OV-TL 12/30), Estrogen (Leica, 1:40, 6F11), Calretinin (Leica, 1:100, CAL6 ), Mesotelin (Leica, 1:20, 5B2), WT1( Leica, 1:30, WT49), and CA125 (Leica, 1:100, ov185:1) at room temperature for 1 hour. The Max Polymer Detection System, Leica Ref: RE 7280-k was then applied for 30 min. Finally, the sections were incubated in 3´3-diaminobenzidine for 5 min, counterstained with Meyer´s hematoxylin and mounted. The slides was examined and photographed on Leica DM 750 Microscope. Negative controls were obtained by replacing the primary antibody with non-immune serum. As a positive control an endometrial tissue section was used.

Immunohistochemically, the tumor cells presented a diffuse strong expression for CA 125 (Figure 2), calretinin (Figure 3), CK7 (Figure 4), ER (Figure 5), mesotelin (Figure 6), WT1 (Figure 7).

## Discussion

Endosalpingiosis is a rare benign gynaecologic condition of Mullerian origin also known under the name of mullerianosis with similar pathogenesis to endometriosis[6,7]. It is characterised by the presence of ectopic epithelium with tubal differentiation involving the female genital tract, peritoneum, subperitoneal tissues as well as other distant structures such as colon, omentum, lymph nodes and even skin[6,8].

When it comes to the main mechanisms of endosalpingiosis development, there are two popular theories. The first one considers that endosalpingiosis originates from the invagination of coelomic cells into the tissues or the rests of these cells from foetal development are submitted to Mullerian metaplasia and form endosalpingiosis[9].

The second theory, the tubal cell origin theory is based on the concept that the apparition of endosalpingiosis is related to the development of ciliated, poorly adhesive cells inside the fallopian tubes which will be carried out into the ovary or even further in the pelvic space. This theory offers a betted explanation of the fact that the ovary remains the most common site of development for endosalpingiosis[9,10]. In the meantime the mechanisms leading to proliferation, shed and seeding of the tubal cells are poorly understood, both genetic and inflammatory factors being incriminated[3,10].

Most often endosalpingiosis is diagnosed incidentally in cases submitted to investigations for pelvic pain, menstrual irregularitiesor infertility and these symptoms were usually reported to be an association with a concurrent endosalpingiosis than being caused by it[11,12].

However an important link has been established between endosalpingiosis and serous ovarian carcinomas. Ovarian malignancies have been reported more often in premenopausal patients with endosalpingiosis than in cases without it[11]. Moreover Gruessner et al. reported the correlation between endosalpingiosis and development of pelvic serous carcinoma in a breast cancer mutation carrier[13].

Another important aspect related to the existence of endosalpingiosis is the capacity of relapse of borderline ovarian tumors. In the study conducted by Silva et al. involving 11 patients with ovarian borderline serous tumors treated by total hysterectomy with bilateral adnexectomy, 10 of these cases were diagnosed with pelvic relapse at a certain moment. This study group was compared with a similar group of cases diagnosed with borderline serous tumors that did not experience relapse.

The two groups were matched in terms of age and tumor type. The authors showed that the only significant parameter which differed between the two groups was the incidence of endosalpingiosis, significantly higher among patients who experienced recurrences (72.7% versus 12.5%)[14].

## Conclusions

Endosalpingiosis is a benign gynaecologic condition with poorly understood mechanisms of pathogenesis and development. However it seems that chronic tubal inflammation in association with cellular migration and nesting play a significant role. The recognition of this pathology is especially important due to the possibility of association in time with malignant pathologies such as serous ovarian carcinomas. ■

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