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ORIGINAL ARTICLE

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Endosalpingiosis, an unrecognized condition: report and literature review

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Abstract This study involves an audit of cases of endosalpingiosis diagnosed at a major teaching hospital and a review of the literature. The retrospective audit of endosalpingiosis was carried out from 1997 to 2000 at the Mercy Hospital for Women, Melbourne, Australia. Information was collected regarding the demographics, presentation and management for women with this diagnosis. During the period of review, endosalpingiosis was diagnosed in 45 patients. Pain was the most frequent presentation and laparoscopy with excision of "endometriosis" was the most frequent operation. Interestingly, of 64 peritoneal biopsies taken for suspected endometriosis, 20 were endosalpingiosis, 21 endometriosis, 7 both endometriosis and endosalpingiosis and 16 fibrosis, hyperplasia or normal. Endosalpingiosis is a relatively unknown condition. Its diagnosis may be missed since it often coexists with endometriosis or mimics endometriosis, and the majority of surgeons ablate lesions without obtaining a histological diagnosis. It is important to consider the diagnosis of endosalpingiosis since it may be associated with chronic pelvic pain. There are a few cases of endosalpingiosis in peritoneal washings that have been incorrectly interpreted as well-differentiated adencarcinoma and have been found in association with ovarian and cervical neoplasms.

Keywords Endosalpingiosis · Endometriosis · Müllerian system

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Introduction

Ries in 1897 was the first to recognise peritoneal müllerian inclusions [1]. The term "endosalpingiosis", which was introduced by Sampson in 1930, described the local proliferative and invasive properties of tubal mucosa following surgical interruption. He examined the tubal stumps of 147 patients who had undergone a previous salpingectomy or tubal sterilisation and observed that sprouts of fallopian tube epithelium often invaded the wall of the stump and extended beyond it. Novak [2] challenged this definition and noted these lesions were also seen in patients without a history of tubal surgery and who had a history of pelvic inflammatory disease. These lesions were identical both radiologically and histologically to what is known as salpingitis isthmica nodosa.

In fact, today the term endosalpingiosis is used only occasionally. It is descriptive and does not imply an origin from the tubal mucosa. The glandular epithelial cells are thought be derived from peritoneal mesothelial cells as part of the secondary Müllerian system [3]. The secondary Müllerian system refers to the pelvic peritoneal mesothelium and submesothelium capable of müllerian metaplasia. This is in contrast to the primary müllerian epithelium, which lines the ducts müllerian-derived structures.

Endosalpingiosis is one of a triad of non-neoplastic disorders of the Müllerian system: the others are endometriosis and endocervicosis. These pathologies have been found in isolation, but are more commonly found in association with one another [4].

Materials and methods

Incidence

Endosalpingiosis was found in 2.4% (n=287) of surgically removed fallopian tubes [5]. In a retrospective study of 51 women undergoing laparoscopy for chronic pelvic pain, 11.8% demonstrated endosalpingiosis. In all six cases, endosalpingiosis was found in locations consistent with the patients' pelvic pain [6]. In a recent

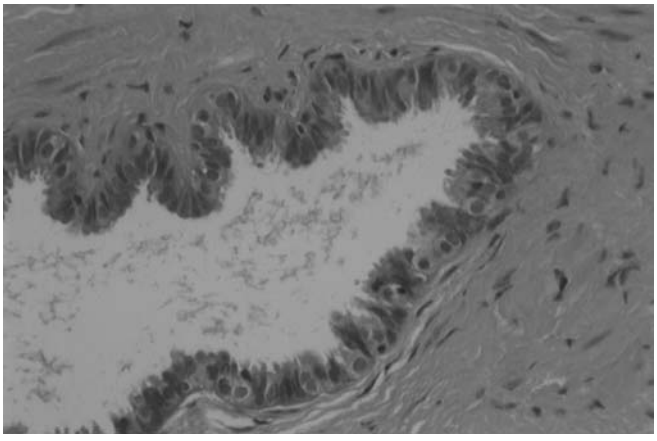

Fig. 1 Histopathology of endosalpingiosis

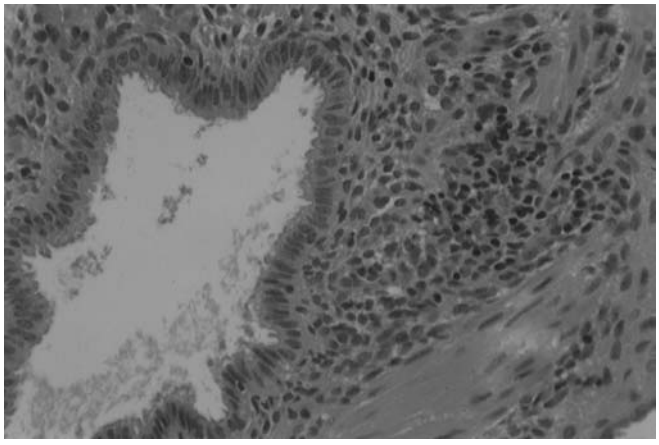

Fig. 2 Histopathology of endosalpingiosis and endometriosis

study [7], 7% of premenopausal women undergoing laparoscopy were diagnosed with endosalpingiosis.

Pathology

Endosalpingiosis is characterised by the presence of glands lined by ciliated tubal-type epithelium. It is usually an incidental finding, often occurring in association with ovarian serous neoplasms. Occasionally, it may form a tumour-like mass that has been called "florid cystic endosalpingiosis" [8]. It is a benign condition, but atypical epithelial changes have been reported and appropriately named "atypical endosalpingiosis".

The diagnosis of endosalpingiosis is made histologically by the presence of tube-like epithelium containing three types of cells: ciliated columnar cells, nonciliated columnar secretory mucous cells and so-called intercalary or peg cells in an ectopic location. Macroscopically, endosalpingiosis may be composed of simple cysts or may demonstrate a more complex papillary architecture. Microscopically, inclusions of the latter type have been associated with psammoma bodies [9]. Stroma is absent, and it generally is not hemorrhagic (Fig. 1).

Differentiation from endometriosis

Endometriosis is characterised by the presence of endometrial glands and stroma. It often has a hemorrhagic appearance as a result of the tissue's response to a hormonal stimulus. The psammoma body, although not specific to endosalpingiosis, is not a feature of uncomplicated endometriosis and therefore can be of diagnostic value (Fig. 2).

Differentiation from ovarian papillary serous tumours

Both may have psammoma bodies and endosalpingiosis may form a papillary configuration, "atypical endosalpingiosis", as a result of irritation, manipulation or trauma. Features supporting the diagnosis of ovarian papillary serous tumour include desmoplastic stromal response, presence of only one cell type, an infiltrative pattern and an absence of a basement membrane surrounding the glands [10].

Theories on development

The various hypotheses for the pathogenesis of endosalpingiosis are similar to those of endometriosis. Kistner described five theories on the pathophysiologic mechanisms of endometriosis: (1) transplantation: tubal mucosa is transplanted to peritoneal surfaces during surgery; (2) direct extension: tubal epithelium extends to the peritoneum by adhesions; (3) coelomic metaplasia: coelomic peritoneal cells are multipotential and can differentiate into oviduct epithelium; (4) reactive: salpingitis with excessive tubal proliferation during the repair process; (5) metastasis: by lymphatic-vascular spread. These theories are not exclusive, and several mechanisms may be responsible.

The natural history of endosalpingiosis is unclear, although calcification and eventual resorption is the usual course. This explains the high incidence of psammoma bodies associated with the lesion. Since endosalpingiosis is rare in children and postmenopausal women, it is thought to develop after menarche and undergo atrophy in the postmenopausal years. Furthermore, spontaneous regression of peritoneal lesions has been reported after removal of an associated ovarian tumour, suggesting a hormonal dependence [11].

Sites of disease

Endosalpingiosis is typically found in the visceral pelvic peritoneum covering the uterus, fallopian tubes, ovaries and cul-de-sac. Less frequent locations involve the pelvic parietal peritoneum, omentum [12], bladder [5] and bowel serosa [13], periaortic area and within the skin [14].

Significance of endosalpingiosis

Despite earlier suggestions by Sampson [15] and Everett that endosalpingiosis may spread quite aggressively, it has created little interest. It is presumed to be an asymptomatic benign entity found incidentally and associated with conditions such as salpingitis, endometriosis and serous tumours. Only a couple of case reports suggest an association between endosalpingiosis and pelvic pain. A retrospective study by Keltz [6] of 51 laparoscopies for pelvic pain found six cases of endosalpingiosis in locations associated with the patient's pelvic pain, and all obtained relief after surgery. This suggests that endosalpingiosis may not be an incidental finding in pelvic pain. It may be overlooked since it often coexists with endometriosis, and the majority of surgeons treating endometriosis ablate lesions, never obtaining a histologic diagnosis.

Endosalpingiosis in lymph nodes, sometimes referred to as benign glandular inclusion and müllerian nest, is well recognised as a potential mimic of metastatic neoplasm. Distinguishing "atypical endosalpingiosis" from an extraovarian serous borderline tumour or borderline implants may be difficult and may pose serious diagnostic problems.

Peritoneal washing cytology is an adjunct in the surgical pathological evaluation of gynaecological malignancy. There are

several reported cases in which endosalpingiosis has created diagnostic problems, particularly when the cytologist is unaware of the morphology of the primary tumour [16, 17, 15].

As previously mentioned, endosalpingiosis may be associated with ovarian epithelial neoplasms, especially borderline or well-differentiated lesions [19, 20, 21]. It has also been found in association with cervical carcinoma [16, 17].

There is controversy about the hypothesis that ovarian epithelial tumours arise from the single cell layer lining the ovarian surface. This theory fails to explain the morphological resemblance of tumours found outside the ovary that are identical to ovarian carcinomas. Further, it fails to explain why some ovarian tumours resemble tumours derived from the Müllerian ducts, such as the fallopian tubes, endometrium and endocervix. The alternate hypothesis is that components of the secondary Müllerian system, which includes endosalpingiosis, endometriosis, paraovarian/paratubal cysts, rete ovarii and endocervicosis, may play a role in tumourigenesis [22]. The possibility of malignant change of endosalpingiosis must be considered, analogous to carcinoma arising in endometriosis. In endosalpingiosis, the tubal nature of the epithelium may be obscured by cell flattening in an expanding cyst or be distorted by proliferating cells. The end of the spectrum is a serous cystadenocarcinoma of the ovary, or more rarely, a papillary carcinoma of the peritoneum. Although the incidence of this malignant transformation is not known, it is important to evaluate each focus of endosalpingiosis.

Presentation

Endosalpingiosis usually is an incidental microscopic finding, often occurring in association with ovarian serous neoplasms. It is generally not recognised by gynaecologists at the time of laparoscopic evaluation or is misdiagnosed as endometriosis. Macroscopically, this disease is usually not discernable. When it is visually obvious, endosalpingiosis can be seen as multiple white to yellow, translucent to opaque, punctate, fluid-filled cystic lesions [20].

Patients with endosalpingiosis are often asymptomatic, while those with endometriosis and endocervicosis more commonly experience symptoms [23].

Management

Medical management for endosalpingiosis with gonadotrophin-releasing hormone analogue has been used with variable success. Malignant degeneration has been documented in conjunction with endocervicosis, and surgical resection has been recommended for bladder lesions of müllerian origin [5].

Results

Forty five cases of endosalpingiosis were reviewed. The average age of the subjects was 42.7 years (range, 14–77); parity 1.1 (range, 0–3); five had had previous tubal ligation and 20 had undergone other prior pelvic surgery (including curette and laparoscopy).

The clinical presentation of the 45 patients is outlined in Table 1. In 25 cases (55%) pain was the dominant symptom, followed by menorrhagia (17%) and infertility (4%). Other presentations that lead to its incidental diagnosis include dyschezia, a pelvic mass, abdominal swelling, prolapse, postmenopausal bleeding, an abnormal pap smear and an unusual finding at caesarean section.

Laparoscopy and excision of endometriosis was performed on 24 of these patients (53%). The results of the 64 biopsies taken from the peritoneal wall via laparoscopy are shown in Table 2. Endosalpingiosis was also found incidentally during seven total abdominal hysterectomies, three laparoscopic assisted hysterectomies, two vaginal hysterectomies, four salpingo-oophorectomies and other various procedures including laparoscopic cystectomy, caesarean section, debulking procedure, radical hysterectomy and a combined appendicectomy/cystectomy in a 14-year-old patient.

Endosalpingiosis was found most commonly on the uterine serosa of hysterectomy specimens. It was also located on the peritoneum of the pelvic side walls, bladder, uterosacral ligaments, paravaginal space, pararectal space and pouch of Douglas. Endosalpingiosis was also found on a broad ligament cyst, fimbrial cyst, mesosalpinx, omentum, placenta and three pelvic lymph nodes. These details are outlined in Table 3.

Table 1 Clinical presentation of patients with endosalpingiosis (*n*=45)

	No. (%) (*n*=45)
Pain	25 (55)
Menorrhagia	8 (17)
Infertility	2 (4)
Dyschezia	1
Abdominal mass	1
Swelling of abdomen	1
Prolapse	1
Caesarean section	1
Post-menopausal bleeding	1
Abnormal pap smear	1

Table 2 Histopathology of peritoneal biopsies suspicious of endometriosis (*n*=64)

	No. (%)
Endometriosis only	21 (33)
Endosalpingiosis only	20 (31)
Endosalpingiosis and endometriosis	7 (11)
Normal/fibrosis/hyperplasia	16 (25)

Table 3 Anatomical sites of endosalpingiosis

	No.
Uterine serosa	7
Bladder peritoneum	7
Right pelvic side wall	5
Left pelvic side wall	4
Right uterosacral ligament	2
Left uterosacral ligament	4
Mesosalpinx	4
Ovary	2
Pouch of Douglas	2
Paravaginal peritoneum	2
Broad ligament cyst	1
Omentum	1
Pararectal peritoneum	1
Placenta	1
Fimbrial cyst	1
Pelvic lymph nodes	3 of 23 nodes

Sixty-four peritoneal biopsies were taken by laparoscopy for suspected endometriosis (see Table 2). Endometriosis was diagnosed correctly in 21 samples, endosalpingiosis in 20, endosalpingiosis and endometriosis coexisted in 7 and 16 biopsies were fibrosis/hyperplasia.

Discussion

Endosalpingiosis is the forgotten relative of endometriosis. It is an important clinical entity since it may be associated with chronic pelvic pain, may pose serious diagnostic problems with peritoneal washings, has been found in association with ovarian and cervical neoplasms and may undergo malignant change. Unfortunately, its diagnosis is often missed since is mistaken for endometriosis [30% (20/64) of the time in our series) and ablated with diathermy.

Laparoscopy with excision of lesions suspicious of endometriosis will probably increase the incidence of endosalpingiosis simply by diagnosis. Excision of suspicious lesions rather than ablating lesions will not only increase our knowledge of endosalpingiosis, but may be the most suitable treatment for the condition [5]. The authors recommend further studies that address chronic pain, endometriosis and endosalpingiosis.

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