Rare “endometrioses” and pain peritoneal pockets – ovarian remnant

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Disclosures: Shareholder Endosat NV
Chronic pelvic pain, nl clinical exam, US and MRI
Is this a cause of pelvic pain

Lacerations, peritoneal pockets, defects, hernia’s
Figure 2. (A) Multilocular pockets with a smaller opening to the peritoneum (used with permission). (B) Right perirectal post-excision pocket 5 years after excisional surgery. Arrows indicate flat endometriosis and stromal endometriotic polyps. The flat and polypoid nature can be seen in the video beginning at 5:55 minutes (used with permission). (C and D) Hidden pocket before and during surgery (used with permission).
Peritoneal pockets: 2 types of pockets

- Deeper pockets medial of utero-sacrals with small peritoneal opening
- Depression/Pockets lateral of the utero-sacrals, eventually with spider


The Allen-Masters syndrome revisited

W. M. ALLEN *
• + many others (surgery literature)
PELVIC PERITONEAL DEFECTS AND ENDOMETRIOSIS:
ALLEN-MASTERS SYNDROME REVISITED

DONALD L. CHATMAN, M.D., F.A.C.O.G.

- 650 women with pain
- 35 peritoneal defects
- 65 endometriosis

Fig. 3. Large cul-de-sac defect with endometriosis on the borders.

Fig. 4. Defect in the right uterosacral ligament with endometriosis.

Fig. 1. Large defect in the left broad ligament with endometriosis.
Embryologic Theory of Histogenesis of Endometriosis in Peritoneal Pockets

Ronald E. Batt, MD,* and Richard A. Smith, MD, PhD†

- In 1989...... Pockets as congenital defects
- Associated with endometriosis – described as spiders
- 1990 Mullerianosis
Conclusion 1

- Peritoneal pockets, Allen- Masters, broad ligament hernia are used interchangeably
- Etiology is probably embryological
- The association with endometriosis is not that clear since women with pain have endometriosis in 50%

The question remains

- A cause of pain?
- A cause of infertility

Deep Retraction Pockets, Endometriosis, and Quality of Life

Patrick P. Yeung Jr.¹*, Ian Logan² and Jeffrey A. Gavard³

TABLE 1 | Deep retraction pockets in women with deep dyspareunia.

<table>
<thead>
<tr>
<th>Sample studied</th>
<th>Numerator/ denominator</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total patients</td>
<td>107</td>
<td></td>
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<tr>
<td>Patients with endometriosis anywhere</td>
<td>88/107</td>
<td>82.2</td>
</tr>
<tr>
<td>Patients with DRP</td>
<td>25/107</td>
<td>23.4</td>
</tr>
<tr>
<td>Patients with endometriosis in a DRP</td>
<td>15/25</td>
<td>60.0</td>
</tr>
<tr>
<td>Total deep retraction pockets</td>
<td>31</td>
<td></td>
</tr>
<tr>
<td>DRP with endometriosis</td>
<td>15/31</td>
<td>48.4</td>
</tr>
<tr>
<td>DRP that were clear (no visible lesions)</td>
<td>10/31</td>
<td>32.3</td>
</tr>
<tr>
<td>Clear DRP with endometriosis</td>
<td>3/10</td>
<td>30.0</td>
</tr>
</tbody>
</table>

DRP, deep retraction pocket.

Compare with Figure 2.

*FIGURE 1 | This annotated photo shows the appearance of a deep retraction pocket, with a visible manifestation of endometriosis, seen at laparoscopy for pelvic pain. There are abnormal red and white lesions suspicious for endometriosis at the base of the pocket.
**Conclusion**: unclear whether pockets alone cause pain

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>N</th>
<th>Preoperative score(^c) (median)</th>
<th>Postoperative score(^d) (median)</th>
<th>Difference score(^e) (post-pre) (median)</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Deep pain with intercourse score(^a)</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Deep retraction pockets</td>
<td>13</td>
<td>6.0</td>
<td>4.0</td>
<td>-2.0</td>
<td>&lt;0.01</td>
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<tr>
<td>Endometriosis</td>
<td>8</td>
<td>5.0</td>
<td>0.0</td>
<td>-3.0</td>
<td>&lt;0.05</td>
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<tr>
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<td>5</td>
<td>8.0</td>
<td>8.0</td>
<td>-1.0</td>
<td>0.50</td>
</tr>
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<td>No deep retraction pockets</td>
<td>32</td>
<td>6.0</td>
<td>4.0</td>
<td>-2.0</td>
<td>&lt;0.0001</td>
</tr>
<tr>
<td><strong>Pain with bowel movements score(^a)</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Deep retraction pockets</td>
<td>11</td>
<td>7.0</td>
<td>0.0</td>
<td>-6.0</td>
<td>&lt;0.05</td>
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<tr>
<td>Endometriosis</td>
<td>7</td>
<td>7.0</td>
<td>0.0</td>
<td>-6.0</td>
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<td>No endometriosis</td>
<td>4</td>
<td>7.0</td>
<td>2.0</td>
<td>-5.5</td>
<td>0.31</td>
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<tr>
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<td>25</td>
<td>6.0</td>
<td>0.0</td>
<td>-4.0</td>
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<tr>
<td><strong>Chronic pelvic pain score(^a)</strong></td>
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<td>7.0</td>
<td>0.0</td>
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<td>&lt;0.01</td>
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<tr>
<td>Endometriosis</td>
<td>7</td>
<td>7.0</td>
<td>0.0</td>
<td>-3.0</td>
<td>&lt;0.05</td>
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<tr>
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<td>6.5</td>
<td>4.0</td>
<td>-2.5</td>
<td>0.28</td>
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<td>No deep retraction pockets</td>
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<td>7.0</td>
<td>0.0</td>
<td>-4.0</td>
<td>&lt;0.0001</td>
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<td><strong>Quality of life score(^b)</strong></td>
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<td>Deep retraction pockets</td>
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<td>55.0</td>
<td>80.0</td>
<td>25.0</td>
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<td>90.0</td>
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<tr>
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<td>70.0</td>
<td>15.0</td>
<td>0.53</td>
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<tr>
<td>No deep retraction pockets</td>
<td>33</td>
<td>50.0</td>
<td>80.0</td>
<td>25.0</td>
<td>&lt;0.0001</td>
</tr>
</tbody>
</table>
Prevalence of Endometriosis and Peritoneal Pockets in Women with Infertility and/or Pelvic Pain

Philippe R. Koninckx, MD, PhD; Anastasia Ussia, MD; Jörg Keckstein, MD, PhD; Leila Adamyan, MD, PhD; Arnaud Wattiez, MD, PhD; Dan C. Martin, MD

Table. Prevalence of subtle, typical, cystic ovarian, and deep endometriosis lesions in women with (P+) and without (P−) peritoneal pockets

<table>
<thead>
<tr>
<th></th>
<th>Total; n = 4497</th>
<th>Infertility only; n = 1896</th>
<th>All pain a; n = 2601</th>
<th>Pelvic pain; n = 1953</th>
<th>Infertility and pelvic pain; n = 648</th>
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</thead>
<tbody>
<tr>
<td>Pockets, no. (%)</td>
<td>191 (4.2)</td>
<td>90 (4.7)</td>
<td>101 (3.9)</td>
<td>69 (3.5)</td>
<td>32 (4.9)</td>
</tr>
<tr>
<td>Endometriosis, %, P+; P−</td>
<td>77; 63 (P = 0.0001)</td>
<td>76; 50 (P &lt; 0.0001)</td>
<td>76; 76 (NS)</td>
<td>75; 68</td>
<td>84; 86</td>
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<tr>
<td>Subtle</td>
<td>24; 16 (P = 0.006)</td>
<td>23; 16 (P = 0.04)</td>
<td>23; 24 (NS)</td>
<td>20; 16</td>
<td>31; 19</td>
</tr>
<tr>
<td>Typical</td>
<td>67; 54 (P = 0.0002)</td>
<td>66; 42 (P &lt; 0.0001)</td>
<td>68; 66 (NS)</td>
<td>64; 58</td>
<td>72; 74</td>
</tr>
<tr>
<td>Cystic ovarian</td>
<td>20; 22 (NS)</td>
<td>14; 13 (NS)</td>
<td>14; 25 (NS)</td>
<td>19; 22</td>
<td>38; 44</td>
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<tr>
<td>Deep</td>
<td>20; 27 (P = 0.04)</td>
<td>8; 9 (NS)</td>
<td>30; 37 (NS)</td>
<td>30; 37</td>
<td>31; 54</td>
</tr>
</tbody>
</table>

a Women with pelvic pain and those with pelvic pain and infertility as the indication for surgery. NS: not significant.

**No change with age**

Pain score = pockets (P = 0.0026) + number of subtle lesions (P = 0.001) + area of typical lesions (P = 0.030) + diameter of cystic ovarian endometriosis (P = 0.051) + volume of deep endometriosis (P < 0.0001)

Pockets = independent predictor of pelvic pain in addition to endometriosis (Multivariate logistic regression).
Why pain: 2 thin women

- Severe neuropathic pain over nervus obturatorius
- Hypogastric pain
- Several laparoscopies
34 year old; pain +++
The Latzko space: very close to the nerves

- Deep uterine vein
- Vaginal artery
- Ureter
- Uterine artery
- Sympathetic nerves
- Obturator nerve
Why pain?

• No direct contact between endometriosis and obturator nerve

• Short distance between endometriosis and obturator nerve and inferior hypogastric plexus
Endometriosis and Pain

• Not all lesions painful

• Variable distance around lesions

• ? The underlying G-E incident
Pain Mapping of Endometriosis: local anaesthesia

- **Type of lesion**: Pain in
  - Variable pain
    - Variable nociceptor activation -> 3cm

<table>
<thead>
<tr>
<th>Type of lesion</th>
<th>%</th>
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<tr>
<td>Clear Lesion</td>
<td>32</td>
</tr>
<tr>
<td>Red Vascular Lesion</td>
<td>37</td>
</tr>
<tr>
<td>White Scar Lesion</td>
<td>20</td>
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<tr>
<td>Black Lesion</td>
<td>11</td>
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</tbody>
</table>

Laparoscopic Findings, Management, Histopathology, and Outcomes in 25 Women with Cyclic Leg Pain

George A. Vilos, M.D., Andrew W. Vilos, HBSc, and Jeffrey J. Haebe, M.D.

N=25  cyclic sciatalgia

- after pocket resection (even without visible endometriosis)
  - 4 improved
  - 2 unchanged
Tension-free Vaginal Tapes and Pelvic Nerve Neuropathy

Roberta Corona, MD*, Carlo De Cicco, MD, Ron Schonman, MD, Jasper Verguts, MD, Anastasia Ussia, MD, and Philippe R. Koninckx, MD, PhD

• TVT -> severe neuropathic pain of nervus obturatorius right
• Laparoscopy
  • Mesh with fibrosis
  • 1.5 cm from obturator nerve
  • Excision of fibrosis
• Pain-free

• Concept of endometriosis causing pain at distance
Endometriosis Can Cause Pain at a Distance

Philippe R. Koninckx, MD, PhD,1,2,3,4,5 Anastasia Ussia, MD;4,6 Roy Mashiach, MD,7 George Vilos, MD,8 Dan C. Martin, MD9,10

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• Up to 3 cm

• All nerves are in reach

• Treat peritoneal pockets

• When nerve exploration?
Endometriosis and pelvic nerves

- Sciatic nerve  Pudendal nerve  sacral roots  Sympathetic nerves

- Consider the distances in the pelvis
  - Nodule on the ischeal spine

- Dilemma : to explore and dissect or not

- My impression: do not dissect nerves liberally. Real nerve invasion is rare
Invasion or nociceptor activation or neuroinflammation

Variable pain in individual lesions
Variable distance of nociceptor activation
Ovarian remnant syndrome

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Consultant Univ Sacro Cuore, Gemelli, Roma

Disclosures: Shareholder Endosat NV
Definitions of Ovarian remnant

- (Functional) ovarian tissue after ovariectomy
  - Ectopic / accessory /supernumerary n=127
    - Incidence 1/30,000 - 7/90,000
    - up to 26% to 75% other anomalies müllerian anomalies, renal agenesis, duplicated ureters, bladder diverticulum, accessory adrenal glands, and a lobulated liver
  - Supernumerary ovary: a disruption of the normal gonadocyte migration from the yolk sac endoderm through the dorsal mesentery before reaching the genital ridge during embryogenesis
  - Accessory ovary: additional ovaries that are found along the genital ridge within any of the gynecologic ligaments.
- Incomplete ovariectomy

- Fibrosis
Accessory ovary

- Functional ovarian tissue after ovariectomy

Supernumerary ovary


<table>
<thead>
<tr>
<th>Case</th>
<th>Authors</th>
<th>Reported Year</th>
<th>Age</th>
<th>Site</th>
<th>Diseases</th>
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<tr>
<td>2</td>
<td>Wharton [1]</td>
<td>1959</td>
<td>37</td>
<td>Pelvic</td>
<td>Granulosa cell carcinoma</td>
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<td>Wharton [1]</td>
<td>1959</td>
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<td>Side of the right ovary</td>
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<td>Hogan et al. [12]</td>
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<td>Omentum</td>
<td>Cystic teratoma</td>
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<td>5</td>
<td>Pritz et al. [5]</td>
<td>1973</td>
<td>23</td>
<td>Left retroperitoneum</td>
<td>Mucinous cystadenocarcinoma</td>
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<td>Roth and Ehrlich [14]</td>
<td>1977</td>
<td>48</td>
<td>Omentum</td>
<td>Cystic teratoma</td>
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<tr>
<td>8</td>
<td>Cruikshank and van Drie [15]</td>
<td>1982</td>
<td>36</td>
<td>Left retroperitoneum</td>
<td>Adenocarcinoma</td>
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<td>Mercer et al. [16]</td>
<td>1987</td>
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<td>Omentum</td>
<td>Cystic teratoma</td>
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<td>10</td>
<td>Bark et al. [18]</td>
<td>1991</td>
<td>45</td>
<td>Omentum</td>
<td>Fibroma, Meig's syndrome</td>
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<td>Bark et al. [18]</td>
<td>1991</td>
<td>28</td>
<td>On pregnant uterus</td>
<td>Endometrioma</td>
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<td>15</td>
<td>Kamiyama et al. [20]</td>
<td>2013</td>
<td>47</td>
<td>Left retroperitoneum</td>
<td>Serous adenocarcinoma</td>
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<td>16</td>
<td>El-Ghairy et al. [17]</td>
<td>2015</td>
<td>20</td>
<td>Left retroperitoneum</td>
<td>Endometrioma</td>
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<tr>
<td>17</td>
<td>Ogishima et al. [8]</td>
<td>2016</td>
<td>40</td>
<td>Rectovaginal pouch</td>
<td>Cystic endometrioma and fibroma</td>
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<tr>
<td>18</td>
<td>Present case</td>
<td>2016</td>
<td>42</td>
<td>Anterior sigmoid colon</td>
<td>Cystic endometrioma</td>
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</table>
Supernumerary ovary


**Figure 1:** The rectovaginal mass on pelvic MRI (T2-weighted, sagittal line). The mass with multilocular cyst (indicated by C) and solid (indicated by S) components and a maximal diameter of 9 cm locates in the rectovaginal pouch. The uterus is indicated with an arrow.

**Figure 2:** View of the rectovaginal pouch during the operation. The cystic and solid mass, indicated by arrows, is present in the...
Supernumerary ovary

Supernumerary ovary: complete since 2000

Ectopic / accessory / supernumerary ovary

- Assumed to be embryological and functional

- But quid when not functional
  - This patient 66 year postmenopausal
  - 17 cases 22-78 year;
  - ½ oestrogen secretion + risk of adenocarcinoma

![Figure 2. Histological findings of the tumor located in the left broad ligament show 2 types of small cell nests surrounded by abundant fibrous tissue. One nest is an aggregate resembling the Call-Exner bodies of granulosa cell tumors containing eosinophilic amorphous material in the center, and the other nest is a sheet-like aggregate of undifferentiated sex cord-type cells (A, hematoxylin and eosin [H&E], ×10). Granulosa cell-type elements display oval to round nuclei, eosinophilic cytoplasm, small nucleoli and occasional longitudinal nuclear grooves (B, H&E, ×40) and are immunopositive for inhibin (C, ×40) and WT1 (D, ×40).]
Tissue remaining after incomplete ovariectomy

- Associated with Endometriosis & difficult surgery
- Reimplantation of ov tissue
  - Wall
  - pelvis

<table>
<thead>
<tr>
<th>Study</th>
<th>N (number of ORS patients)</th>
<th>Indication for primary oophorectomy</th>
<th>Pathology</th>
<th>Initial presentation</th>
<th>ORS surgery</th>
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<tr>
<td>Mahdavi et al., 2007 [3]</td>
<td>3</td>
<td>Endometriosis – 1; pain – 1</td>
<td>Ovarian serous cystadenoma – 3</td>
<td>Pain – 2; mass – 1</td>
<td>Laparoscopy – 3</td>
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<tr>
<td>Donnez et al., 2007 [5]</td>
<td>1</td>
<td>Endometriosis</td>
<td>Ovarian adenocarcinoma</td>
<td>Pain</td>
<td>Laparoscopy</td>
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<td>Kho et al., 2007 [4]</td>
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<td>Endometriosis – 8; mass – 6</td>
<td>18 – benign; 2 – malignant (mucinous cystadenocarcinoma, endometrioid adenocarcinoma)</td>
<td>Pain – 18; mass – 2</td>
<td>Laparotomy – 1; laparoscopy – 14; robotics – 5</td>
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<tr>
<td>Chao, 2008 [6]</td>
<td>1</td>
<td>Endometriosis</td>
<td>Benign ectopic ovarian tissue, endometriosis</td>
<td>Pain</td>
<td>Laparotomy</td>
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<tr>
<td>Fat et al., 2009 [7]</td>
<td>7</td>
<td>Ovarian cyst – 6; unspecified – 1</td>
<td>Benign – 7</td>
<td>Pain – 7</td>
<td>Laparotomy – 3; laparoscopy – 4</td>
</tr>
<tr>
<td>Arden and Lee, 2011 [8*]</td>
<td>30</td>
<td>Pain or endometriosis – 16; ovarian cysts – 7</td>
<td>All benign</td>
<td>Pain – 29; mass – 1</td>
<td>Laparotomy – 1; laparoscopy – 29</td>
</tr>
</tbody>
</table>
Tissue remaining in infundibulo pelvic ligament after ovariectomy

- In 14%
- From 0.2-1.4 (3.5)cm
- Ovarian remnant in 6.5-18%

> vaginal ovx
Diagnosis of Ovarian remnant

- Most diagnoses
  - BSO + cystic mass at ovarian location
  - BSO + estrogen secretion
  - Unclear
    - Diagnosis of stroma by pathology ??
    - Presence of follicles ??
Diagnosis of ORS


- Endometriosis & difficult BSO

- Endometriosis + ureteral obstruction + unclear source of estrogens
  - Pragmatic therapy by GNRH + add-back
  - Even not an attempt to find the source of estrogens
Hormones and ORS

- 50% have ‘normal estrogens’
- Not useful for diagnosis

- However HRT estrogens are often not measurable in Radio-immuno-assay
“ovarian mass” concept

In rats

- + extra ovary -> remains inactive ovary
- Removal of 1 ovary -> becomes active

Translated to humans

- Ovarian “remnants” risk to become active after ovariectomy
Fibrotic ... Ovarian remnant surrounding the ureter
Conclusions: ovarian remnant

- 1/20,000 surnumerary ovaries
- Ovarian remnant
  - Ovarian stromal remnant in 7-18% after ovariectomy
  - More after endometriosis surgery -> more difficult planes of cleavage & more ovarian tissue
- Ovarian remnant 50/50 hormonal active/inactive
- Symptoms: mainly pain
- If only stromal component and inactive: unclear diagnosis and clinically fibrosis
- Always difficult surgery since ureter circularly involved
- Unclear why pain and why around ureter
- MRI + US + surgical observation
Rare cases

At laparoscopy: Inspection: slow and in detail

you only recognise what you know

be a clinician