Gynecol Surg (2006) 3: 213–214
DOI 10.1007/s10397-006-0186-0

CASE REPORT

J. C. Knudsen · P. Lundorff

# Complex hyperplastic endometrium in a peritoneal leiomyoma following a CISH hysterectomy

Received: 27 June 2005 / Accepted: 23 December 2005 / Published online: 30 May 2006


**Abstract** We describe a case of a patient presenting with an abdominal tumor 4 years after a classical intrafascial serrated-edged macro-morcellated hysterectomy. The tumor was removed surgically and proved microscopically to be a peritoneal leiomyoma containing complex hyperplastic endometria. To our knowledge, this has never been described before. In addition, the pathogenesis of this rare disease is discussed.

**Keywords** Peritoneal leiomyomatosis · Hormone replacement therapy · Uterine fibroids

## Introduction

Leiomyomatosis peritonealis disseminata is a rare disease. In the past, more than 100 cases have been described [1, 2]. In the vast majority, these tumors occurred in women who were pregnant or taking oral contraceptives or hormone replacement therapy. The tumor is thought to arise from Mueller's epithelium, which is distributed throughout the subperitoneal mesenchyme. Proliferation of the epithelium may be stimulated by estrogen in predisposed women [1, 3, 4]. The disease mimics peritoneal malignancy but is generally benign. In rare cases (fewer than 10%), malignant leiomyosarcomas do occur [1–4]. Fewer than 10 cases have been described among postmenopausal women [3–6]. Many patients, but not all, have uterine leiomyomas as well. To our knowledge, this is the first case describing a peritoneal myoma containing a complex hyperplastic endometrium.

J. C. Knudsen · P. Lundorff (✉)
Department of Obstetrics and Gynaecology,
Viborg County Hospital,
8800 Viborg, Denmark
e-mail: jc942726@hotmail.com
Tel.: +45-892-72727

## Case report

A 48-year-old woman was admitted by her general practitioner for a growing abdominal tumor. The patient was para 2 with normal deliveries. Four years earlier, she had undergone laparoscopic hysterectomy ad modum classical intrafascial serrated-edged macro-morcellated (SEMM) hysterectomy (CISH) because of menorrhagia and uterine leiomyomas [7, 8]. CISH is a synthesis of three well-established and widely used procedures:

1) Supracervical amputation of the uterus
2) Conization of the cervix
3) Laparoscopy

The operation was done at our department and was videotaped, confirming that no part of the uterus except the collum was left in the abdomen (see Fig. 1). The transformation zone and endometrium of the collum uteri were removed. The remaining collum was left in situ. After the operation, the patient had hormone replacement therapy consisting of Femanest 2 mg daily and Vagifem every 2nd day. The patient had been well since the operation, but after an intended weight loss of 10 kg, she felt a tumor in her lower abdomen.

Clinical and ultrasonic examinations revealed a large tumor in the lower part of the abdomen and in the pelvic cavity. Blood samples were normal except for a CA-125 level of 78 kIU/l. At laparotomy a large multilobular, soft, smooth tumor was found attached to adhesions around both adnexae and the pouch of Douglas. The adnexae, liver, omentum, and intestines were normal. Tumorectomy, appendectomy, and bilateral salpingo-oophorectomy were performed. At no time during the operation was the remaining collum uteri touched. The tumor seemed macroscopically to arise from somewhere near the right adnexa. Macroscopic examination revealed a 15×15×16-cm tumor built up from many large and small nodules measuring from 1 to 8 cm in diameter. Microscopic examination showed a leiomyoma with almost no mitotic activity and no cellular polymorphy but containing a very hyperplastic endometrium without atypia (see Fig. 2). The

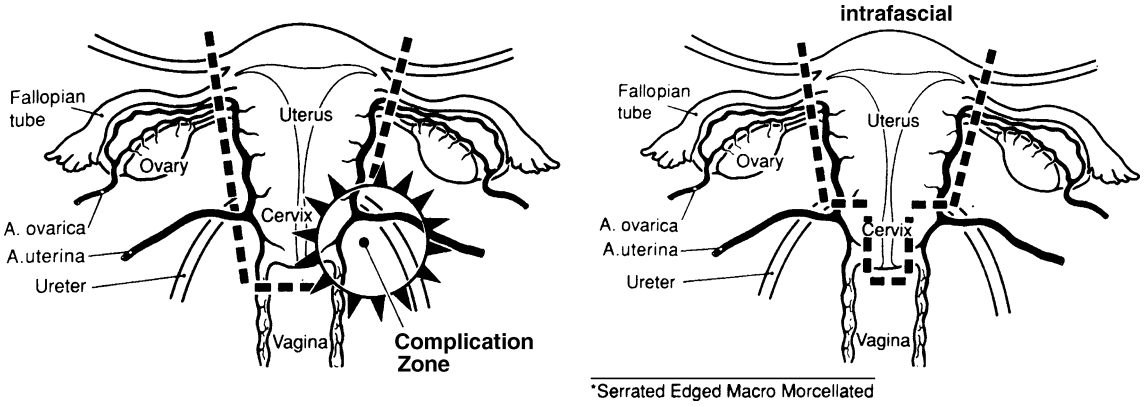


**Fig. 1** Classical hysterectomy technique

postoperative period was uneventful, and the patient was discharged 4 days after the operation and has since been well.

## Discussion

The most likely explanation for the origin of this tumor is a case of peritoneal leiomyomatosis. The pathogenesis of peritoneal leiomyomatosis is thought to be multipotential subcoelomic mesenchymal cells that proliferate in response to estrogens. Proliferation into myoblasts, myofibroblasts, fibroblasts, and decidua-like cells has been described [9].

Another explanation of the development of this tumor could be spillage of endometrial tissue during the CISH with regrowth of myoblasts, thus simulating regeneration of a uterus with endometrial tissue. This explanation has never been described previously and is only speculative.

We present a case with a peritoneal leiomyoma consisting of myoblasts, fibromyoblasts, and a complex hyperplastic endometrium without atypia. To our knowledge, this has never been reported before.

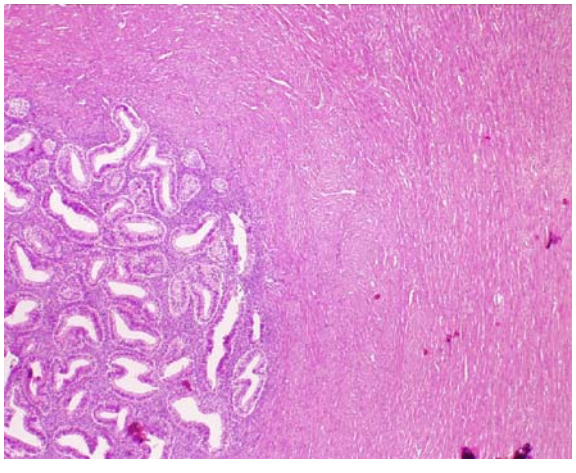

**Fig. 2** Classical intrafascial serrated-edged macro-morcellated hysterectomy

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