Arch Gynecol Obstet (2009) 280:1063–1065
DOI 10.1007/s00404-009-1035-y

CASE REPORT

Cotyledonoid dissecting leiomyoma of the uterus associated with endosalpingiosis

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Received: 23 September 2008 / Accepted: 5 March 2009 / Published online: 2 April 2009

Abstract Cotyledonoid dissecting leiomyoma (CDL) may create a diagnostic challenge. A 47-year-old woman underwent laparotomy for a large pelvic mass associated to vaginal bleeding. During operation, a bulky deep red mass protruding from the uterus and projecting into the pelvic cavity was discovered. Allowing to sarcoma- like appearance, a total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed with removal of the pelvic tumor extension. The diagnosis of cotyledonoid dissecting leiomyoma of the uterus associated with endosalpingiosis was performed. To the best of our knowledge, this association has never been encountered in the English literature.

Keywords Dissecting leiomyoma · Leiomyoma · Endosalpingiosis

Introduction

Cotyledonoid dissecting leiomyoma or Sternberg tumour is a very rare variant of smooth muscle tumours as approximately 20 such cases reported in the literature [1]. It has a distinctive gross appearance characterized by the presence of congested exophytic placental like mass extending from the uterus into the broad ligaments and pelvic mass [1, 2].

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We describe herein an additional case of cotyledonoid dissecting leiomyoma of the uterus associated with endosalpingiosis. To the best of our knowledge, this association has never been encountered in the English literature.

Case report

A 47-year-old woman with gravidity 2 and parity 2 presented with a pelvic mass and vaginal bleeding. During operation, a bulky deep red mass protruding from the uterus and projecting into the pelvic cavity was discovered. Because the gross appearance suggested malignancy, a total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed with the removal of the pelvic tumor extension.

Macroscopically, the uterus weighed 2,560 g. It formed a large exophytic mass measuring up to 250 mm in maximum dimension, protruding from the serosal aspect of the uterus into the broad ligaments and projecting into the pelvic cavity. This mass was constituted of multiple nodules that were attached to their neighbours by anastomosing networks of thin vessels giving a placenta-like appearance. These nodules were purple, congested and varied from few mm to 25 mm in greatest dimension. The cut surface of the exophytic component revealed, on one hand, soft to firm nodules sprinkled focally with numerous 1–2 mm cysts and an evident continuity of the exophytic tumor with an irregular lobulated leiomyoma measuring 13 × 13 × 10 cm on the other hand (Fig. 1). The endometrium, the fallopian tubes and the ovaries were not involved.

Histologically, the exophytic component as well as the intramural leiomyoma were predominantly formed of disorganized swirls of fascicules of smooth muscle surrounded by hyaline fibrous matrix (Fig. 2). Nuclei were plump in

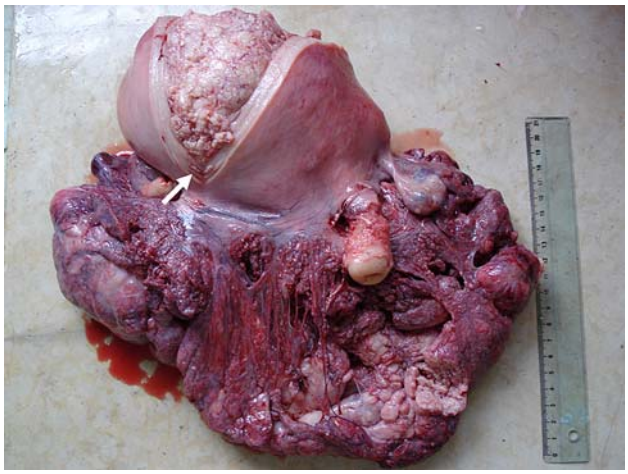

Fig. 1 The tumor formed deep red, multiple nodules extending to large ligaments. The nodules were wrapped by anastomosing vascular network resembling placental cotyledons. The arrow indicates the area of dissection, in the form of small nodule into the myometrium

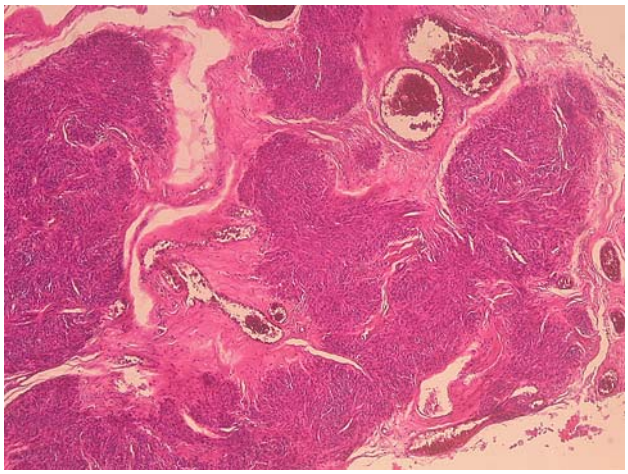

Fig. 2 Multiple extrauterine micronodules of smooth muscle with swirling growth pattern, abundant congested blood vessels and hyaline degeneration (H&E, ×20)

some areas but there were neither mitotic figures nor significant cytologic atypia. Coagulative tumour necrosis was also absent. Numerous congested and dilated vessels were clustered mainly in the fibrous matrix of the extra uterine component. Intramural nodule dissected the myomterium among fascicules of normal smooth muscle cells without intravascular growth. In addition, some nodules corresponding to the grossly described small cysts show in association with smooth muscle component, numerous glands and cysts lined by a ciliated (tubal type) epithelium that varied from monolayer columnar to pseudo stratified. Periglandular stroma consists of scanty loose fibrous tissue. Some foci show mild tufting of the epithelial lining but no cellular atypia or increased mitotic activity was seen (Fig. 3).

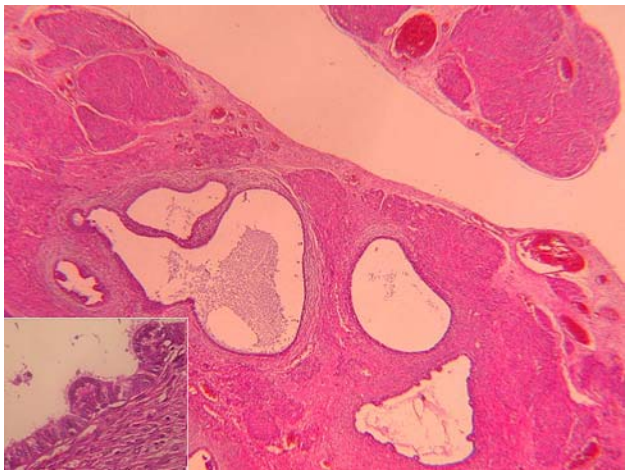

Fig. 3 Micronodules of smooth muscle within the myometrium were adjacent to multiple, variously sized glands and cysts lined by benign-appearing epithelium (H&E, ×10). *Inset* lining epithelium showed a stratification of ciliated columnar cells

In the immunohistochemical staining, the smooth muscle tumour cells showed positive staining for desmin, caldesmone, oestrogen, and progesterone receptors. CD34 staining confirmed that there was no intravascular extension of the tumor. Glandular component of endosalpingioisi foci was positive for CK7 and negative for CK20.

The patient has gone well with no recurrent disease 10 months after operation.

Discussion

Cotyledonoid dissecting leiomyoma is an extremely rare variant of uterine leiomyoma characterized by a distinctive gross appearance consisting in its deep red and elongated bulbous protrusions that resembled placental cotyledons [1–3]. This tumour extends from the uterus into the broad ligaments and projects in the pelvic or even the retroperitoneal space creating a high impression of malignant process [1–4]. Bland cytology on frozen section confirms the benignity and prevents over-treatment during the reproductive age. Histologically, it shows variably-sized micronodules of disorganized, swirled neoplastic smooth muscle. The intervening stroma between nodules is fibrous or oedematous and typically highly vascularized resulting in the deep red colour of nodules. Significant atypia, mitotic activity and coagulative necrosis are absent. Like the present case, the exophytic component is usually in continuity with intramural nodule characterized by intramural dissection [2]. Immunohistochemical studies, using muscle specific markers (desmin, caldesmon) or hormone receptor studies are non-contributory since these are also expressed by typical leiomyomas. CD34 immunostaining is useful to confirm the absence of intravascular growth [5, 6].

Endosalpingiosis has been recognized as a metaplastic lesion rather than neoplastic process characterized by the presence of multiple glands of variable size lined by tubal-type epithelium involving the pelvic and lower abdominal peritoneum in woman [7]. Unusual presentations include a mass, when the glands are cystic, fine pelvic calcifications on x-ray examination or psammoma bodies within peritoneal washings [8]. Like our case, endosalpingiosis is usually an incidental finding on macroscopic or microscopic examination. However, its association with cotyledonoid dissecting leiomyoma has never been encountered in the English literature.

Cotyledonoid dissecting leiomyoma apparently results from a combination of several uncommon growth patterns operating together including subserosal growth into the free space of the pelvic cavity and dissecting growth [3]. As in our case, the exophytic component has arisen from the dissecting intramural leiomyoma that undergone extensive degenerative change.

Endosalpingiosis is derived from the secondary mullerian system [7, 8]. Its association with CDL is probably a coincident finding, presumably reflecting the fact that endosalpingiosis is a relatively common process.

In conclusion, CDL as well as endosalpingiosis are benign process that may create diagnostic problem for pathologist, not as to whether the lesion is benign or malignant but as what to call it. For clinician, awareness of these lesions will avoid overdiagnosis and overtreatment.

Conflict of interest statement None.

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