

Rectus abdominis muscle endometriosis: Case report and review of the literature

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Abstract

The abdominal wall is an uncommon site of extrapelvic endometriosis, which usually develops in a previous surgical scar and it should be considered in the differential diagnosis of any abdominal swelling. Endometriosis involving the rectus abdominis muscle is a very rare event and its rarity explains the incomplete nature of the reports in the literature. Up to the present, 18 cases with lesions contained entirely within the rectus abdominis muscle were clearly documented in medical literature with only four cases as a primary location. We report a case, which came to our observation, of primary endometriosis of the rectus abdominis muscle. The patient underwent only surgery without any medical treatment. Currently, the patient is in follow up for four years with no recovery of the disease. In our experience, surgery is the treatment of choice and it is decisive. We reviewed the literature and summarized all reported cases.

Key words: abdominal wall, endometriosis, rectus abdominis muscle, surgery.

Introduction

Endometriosis is classically defined as the presence of functional endometrial glands and stroma outside the uterine cavity. The extrapelvic implantation of endometrial tissue has been described in virtually every organ. Abdominal wall endometriosis is any ectopic endometrium found superficial to the peritoneum. This definition includes lesions that were not a result of a previous surgical procedure.¹ Endometriosis of the rectus abdominis muscle is an exceptional occurrence and only 18 cases, wherein the endometrial focus is solely confined to within the body of the rectus abdominis muscle, have been clearly described to date in the literature.²⁻¹⁶ We report a very rare case of primary endometriosis of the rectus abdominis muscle and a review of the literature.

Case

A 32-year-old woman came to our observation reporting the presence of an aching pelvic neoformation, with exacerbation of symptoms during the menses. Her personal history for previous surgery and for pelvic endometriosis was negative. Clinical examination showed a neoformation of two centimeters in diameter of tense elastic consistency aching to the touch, at the level of the left pubic tubercle.

A computed tomographic (CT) scan confirmed the presence of the neoformation of two centimeters in diameter at the distal insertion of the left rectus abdominis muscle, which showed intense contrast enhancement with no involvement of the abdominal cavity (Fig. 1). The finding, not easy to interpret, could be compatible with neoplastic alteration, hence diagnostic

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Figure 1 Computed tomographic scan showing the endometriotic lesion (arrows) of 2 × 2 cm involving the left rectus abdominis muscle.

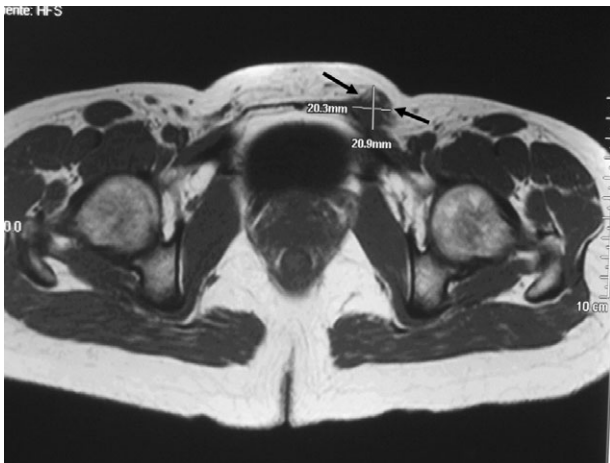


Figure 2 Magnetic resonance imaging scan showing the endometriotic lesion (arrows) of 2 × 2 cm involving the left rectus abdominis muscle.

study with fine-needle aspiration (FNA) under ultrasound guidance was recommended. FNA showed the presence of pigmented macrophages and histiocytes mixed with amorphous substance, rare red blood cells and leukocytes. The findings did not allow diagnostic conclusions. A magnetic resonance imaging (MRI) scan was then required to further establish the nature of the lesion. The MRI scan confirmed the same location of the lesion with no involvement of the abdominal cavity and directed towards a granulomatous lesion (Fig. 2).

The surgical removal of the lesion, which showed the relationship with deep subcutaneous tissue and the left

rectus abdominis muscle, was performed. No connection with intra-abdominal structures was identified, and findings on abdominal exploration were normal. The histological examination showed the presence of endometriotic tissue within the fibroadipose and the muscle tissue was removed with clear surgical resection margins.

Currently the patient, without any medical treatment, is in follow up for 4 years with negative results for recovery of the disease.

Discussion

Endometriosis is a common gynecological disease with an estimated prevalence of 8–15%.¹ The extrapelvic implantations of endometrial tissue were detected in various organs and systems and the abdominal wall is a rare site of localization, usually reported in a previous surgical scar.¹¹ Several theories have been proposed for the development of extrapelvic endometriosis including metaplasia, retrograde menstruation, venous or lymphatic metastasis and mechanical transplantation into scars at the time of surgery.¹⁷ Endometrial lesions, solely confined to within the body of the rectus abdominis muscle, are an exceptional occurrence and to date in the literature only 18 cases have been thoroughly described, with radiologic-imaging studies of the lesion and age, signs/symptoms, surgical history and treatment of patients, since it was first described in 1984 by Amato and Levitt.²

All cases are reported in Table 1. The mean age of patients was 33 years with a range of 27–42 years. Fourteen patients had a positive personal history for previous surgery and only four patients (22%) had no previous surgery. Ten of 14 patients with positive personal history for previous surgery (72%) were subjected to cesarean section. Fifteen patients (83%) reported pain associated with mass and only two patients presented a mass without pain. One patient presented only pain. The average size of the endometriomas, available for fifteen patients, was 4 × 4 centimeters in diameter (range 2 to 7 cm). Two patients had the concomitant presence of two lesions. Prior to surgery, the radiologic-imaging techniques most commonly used were the CT scan in 15 patients (83%). Ten patients were subjected to ultrasound (55%), while for six patients (33%) a MRI scan was performed. All patients were subjected to wide excision of the lesion and four patients were treated with polytetrafluoroethylene patch grafting for the resulting fascial defect. Only one patient was discharged with danazol after surgery.

Table 1 Rectus abdominis muscle endometriosis: Review of the literature

Author	Patient no.	Age (years)	Previous surgery	Signs/symptoms	Size (cm)	Imaging	Treatment	Outcome
Amato <i>et al.</i> 1984 ²	1	35	CS	Pain-Mass	5 × 5	CT	Wide excision + Danazol	-
Coley <i>et al.</i> 1993 ³	2	30	2 CS	Pain-Mass	6 × 5	CT	Wide excision	-
Roberge <i>et al.</i> 1999 ⁴	3	31	AH	Pain-Mass	-	US-CT-MRI	Wide excision	-
Tomas <i>et al.</i> 1999 ⁵	4	35	None	Pain-Mass	4 × 2	US-CT-MRI	Wide excision	-
Dwivedi <i>et al.</i> 2002 ⁶	5	28	CS	Pain-Mass	3 × 2	US-CT	Wide excision	-
Ideyi <i>et al.</i> 2003 ⁷	6	28	None	Pain-Mass	5 × 3 and 2 × 2	CT-MRI	Wide excision	-
Blanco <i>et al.</i> 2003 ⁸	7	29	Laparotomy for EP	Pain-Mass	5 × 4	CT	Wide excision	No recurrence
Esinler <i>et al.</i> 2004 ⁹	8	33	None	Pain-Mass	4 × 4	US	Wide excision	No recurrence
Haim <i>et al.</i> 2005 ¹⁰	9	35	CS	Pain-Mass	3 × 2 and 2 × 3	US-CT	Wide excision	-
Erkan <i>et al.</i> 2005 ¹¹	10	31	CS	Pain-Mass	6 × 5	CT	Wide excision	No recurrence
Kocakusak <i>et al.</i> 2005 ¹²	11	27	Myomectomy	Pain-Mass	7 × 5	US-CT	Wide excision with PPG	No recurrence
Kocakusak <i>et al.</i> 2005 ¹²	12	37	None	Mass	7 × 7	US-CT	Wide excision with PPG	No recurrence
Kocakusak <i>et al.</i> 2005 ¹²	13	42	CS	Mass	4 × 3	US-CT	Wide excision with PPG	No recurrence
Coeman <i>et al.</i> 2005 ¹³	14	38	CS - VH	Pain	-	CT-MRI	Wide excision	-
Coeman <i>et al.</i> 2005 ¹³	15	32	LTL - Ap	Pain-Mass	-	US-CT	Wide excision	-
Huff <i>et al.</i> 2007 ¹⁴	16	39	CS	Pain-Mass	3 × 3	CT-MRI	Wide excision	No recurrence
Gourgoutis <i>et al.</i> 2008 ¹⁵	17	32	CS	Pain-Mass	3 × 2	US	Wide excision	No recurrence
Feeney <i>et al.</i> 2008 ¹⁶	18	41	2 CS	Pain-Mass	5 × 3	MRI	Wide excision with PPG	-

AH, abdominal hysterectomy; Ap, appendicectomy; CS, cesarean section; CT, computed tomography; EP, ectopic pregnancy; LTL, laparoscopic tubal ligation; MRI, magnetic resonance imaging; PPG, polytetrafluoroethylene patch grafting; US, ultrasound; VH, vaginal hysterectomy; -, not specified.

All patients had an uneventful postoperative course but follow up was available for only eight patients. The mean follow up was 22 months (ranging from 11 months to 3 years) with no recurrence of endometriosis.

It is very interesting to note that of 15 articles, only two articles were published by gynecological journals. Probably, this is due to the fact that abdominal wall endometriosis is rarely suspected in these cases, indeed an abdominal wall endometrioma is accurately diagnosed preoperatively in only 33% of patients.¹¹

The caesarean section is the surgical procedure most frequently associated with abdominal wall endometriosis with an incidence that is estimated to be between 0.03% and 1%.¹⁸ During a cesarean section, endometrial cells may escape through the incision in the uterus and implant themselves within the abdominal wound.¹⁹ Pregnancy is a condition associated to very high serum progesterone levels, which is known to negatively impact on endometriotic tissue growth. Hence the implanted endometrium should be less prone to implant and proliferate at the ectopic site. This is why the theory of direct implant, which seems the most obvious, does not collect unanimous consensus. Probably the ectopic implant at the time of cesarean section requires a combination of several factors such as genetic and immunological ones.²⁰ In these cases the most endometriotic locations are the surgical scars and rarely involve the rectum abdominis muscle.

The incidence of pelvic endometriosis in patients with abdominal wall endometriosis is within the same range as the general population.¹

A small proportion (20%) of abdominal wall endometriosis is not associated with a previous surgical incision.¹ In this case it is probable that individual endometrial cells escape from the uterus through lymphovascular channels and eventually gain access to the peripheral circulation, reaching the ectopic sites.¹ The most frequent locations of primary abdominal wall endometriosis are the umbilicus, the groin and the skin.¹

The symptoms of the disease are cyclic or catamenial pain associated with a palpable mass. The differential diagnosis includes hernia, hematoma, lymphadenopathy, lymphoma, lipoma, abscess, subcutaneous cyst, neuroma, soft tissue sarcoma, desmoids tumor, or even metastatic cancer.¹

The serum level of CA-125 can be slightly increased.²¹ Many studies have evaluated several serum markers to predict or to exclude endometriosis. Recently, in an interesting study, it was reported that

the serum level of high-sensitivity C-reactive protein (CRP) was significantly lower than CRP in women without endometriosis.²² Therefore, high-sensitivity CRP might be able to serve as a marker for the absence of endometriosis. Similarly, it was demonstrated that patients with minimal/mild endometriosis present a decreased serum anti-müllerian hormone level compared to patients without endometriosis.²³ Finally, a very innovative and interesting study measured, for the first time, serum follistatin concentration in patients with ovarian endometriosis.²⁴ The authors found increase serum follistatin levels in women with endometrioma, but not in women with other types of benign ovarian cysts. Moreover, follistatin was shown to be a potential marker of endometrioma, with much higher sensitivity than the current marker CA-125. Concerning these new markers, further studies must be done to confirm their validity. Up to the present, there is no evidence that any one measurable serum marker is useful for the diagnosis or monitoring of endometriosis. Likely, a combination of different markers might be helpful in the diagnostic work-up of pelvic and extrapelvic endometriosis.

Additional studies of ultrasound, FNA, CT scan, or MRI scan may be needed for the final diagnosis of abdominal wall endometriosis. FNA had been reported to be useful in excluding the possibility of malignancy, but seems to be inconclusive in formulating the diagnosis in 75% of cases.¹⁹ Moreover FNA has been associated to an increased risk of recurrence.¹⁹

The treatment of choice for endometriosis of the rectus abdominis muscle is a wide local excision of the lesion with negative margins. The surgical excision should include 5–10 mm of surrounding healthy tissue as surgical margin and care must be taken not to rupture the mass to avoid re-implantation of microscopic remnants of endometrial tissue.²⁵ Follow-up evaluation is inconsistently reported in the literature. Recurrence is rare, usually presents within 1 year and is likely to be the result of an inadequate excision.¹⁷ Medical treatment of abdominal wall endometriosis is usually unsuccessful and often results in temporary relief with return of symptoms after therapy discontinuation.¹ Drugs that have been used in the past include oral contraceptives, danazol, gonadotropin releasing hormone analogs and progestogen. At the moment there is no data to support postoperative hormonal therapy.¹

Our patient has been subjected only to surgery with a wide local excision of the lesion with negative margins, without any medical treatment. Follow up

was performed by subjecting the patient to a transvaginal and transabdominal ultrasonography, detection of serum level of CA-125 and a gynecological evaluation 6 months after surgery. An MRI scan was performed 1 year after surgery. Currently, the patient performs a gynecological evaluation and CA-125 measurement every year. Up to the present all tests gave negative results for recovery of the disease.

In conclusion, our experience is in agreement with the data of literature. Endometriosis of the rectus abdominis muscle should be part of differential diagnosis in the work-up of any abdominal mass in young women even in the absence of a history of abdominal surgery. The treatment of choice is a surgical wide excision with clear margins, which is resolute as demonstrated by follow up.

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