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Ann Ital Chir, Digital Edition 2020, 9
pii: S2239253X20031710 - Epub, March 24
free reading: www.annitalchir.com

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An unusual case of large endometrioma within the rectus abdominis muscle misdiagnosed as desmoid tumour

We describe an unusual case of giant intramuscular abdominal endometrioma clinically misdiagnosed as desmoid tumour in a 36-year-old female patient with a one-year history of lower abdominal pain. Endometriosis is defined by the presence of endometrial tissue outside the uterine cavity, associated with fibrosis and inflammatory reaction. Although the abdominal wall is one of the most frequent sites of extra pelvic endometriosis, the localization in the anterior rectus abdominis muscle is unusual and associated with previous cesarean section. In most cases, the preoperative diagnosis is erroneous because the different imaging modalities are nonspecific but only useful in determining the extent of disease and in the planning of operative resection. A better acquaintance with the imaging presentation of abdominal wall endometriosis holds the potential of positively impact disease confirmation and may play a crucial role in the face of innovation in treatment.

KEY WORDS: Desmoid tumour, Endometrioma, Surgery

Introduction

Endometriosis is defined by the presence of endometrial tissue outside the uterine cavity, associated with fibrosis and inflammatory reaction¹. Although the most common occurrence is the pelvis, all body cavities and organs can be involved¹. The abdominal wall is one of the most frequent sites of extra pelvic endometriosis, gener-

ally in a previous surgical scar in the subcutaneous tissue². The localization in the anterior rectus abdominis muscle is unusual and associated with previous cesarean section³. The differential diagnosis includes desmoid tumour that grow through the muscular aponeurosis^{4,5}. We describe a case of intramuscular abdominal endometrioma clinically misdiagnosed as desmoid tumour.

Case Report

A 36-year-old female patient with a one-year history of lower abdominal pain was admitted to our attention. Initially the pain was present during menses and partially relieved by taking some analgesics; subsequent the pain became continuous and more intensive mostly referred to the left lower abdomen spreading towards the inguinal regions. Gynecological and pelvic ultrasound examinations were unremarkable. Echo Doppler exami-

Pervenuto in Redazione Ottobre 2019. Accettato per la pubblicazione e Dicembre 2019

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nation of lower limbs and lumbar spine MRI had been performed and was negative. Surgical history was significant only for two prior cesarean sections, the last of which was seven years before. On physical exam, a tender mass in the lower abdominal wall, just lateral and superior to the left extent of the “Pfannenstiel incision scar”, was detected. The mass was hypoechoic and firm to the lower anterior abdominal wall at ultrasound, with an intense contrast enhancement at CT scan and with focal areas of high and low signal intensity and contrast enhancement at MRI, without abdominal or pelvic cavity involvement (Fig. 1A). Mass measured 6x3x4 cm and was dislocated within the left rectus abdominis muscle. A rectus sheath hematoma was suspected at CT scan; MRI suspected a desmoid or myxoid tumour. Elective surgery was performed for presumptive anterior abdominal wall desmoid. The mass was explored under general anesthesia via a vertical incision (Fig. 1B). A hard mass was seen to be bulging through the anterior rectus, was situated into the same muscle and was adherent to the underlying peritoneum. The surgical exploration of pelvic organs confirmed the absence of tumours. The mass was widely excised and sent to histological exam. Grossly, the resected mass was soft and attached to a portion of rectus muscle, showed a blue-red spongy cut surface and measured 6x4x4 cm (Figs. 1C-1D). From

blocks of paraffine representative of the lesion were obtained hematoxylin and eosin staining slides. The histological examination showed the presence of endometrial glands embedded in abundant collagen-rich desmoplastic stroma within the muscle. Foci of hemorrhage, debris and chronic inflammatory infiltrate were also seen (Figs. 1E-1F-1G). Surgical resection margins were free. A diagnosis of abdominal wall endometriosis was made. The postoperative evolution was uneventful. The patient made a full recovery from her surgery and was referred to Gynecologists for further treatment of endometriosis.

Discussion

Endometriosis is defined as the presence of endometrial glands outside the uterine cavity. Lesions are typically located in the pelvis. Abdominal wall occurrence is referred to “abdominal wall endometriosis” (AWE) ^{6,7} that complicates up to 1% of cesarean sections and is usually the only manifestation of endometriosis ³. Endometriotic implants are usually embedded in the subcutaneous fatty layer (scar endometriosis) and exceptionally in the muscles of the abdominal wall near the site of surgical scars ⁸. The symptoms of AWE are often cyclical and worsening during the first phase of the

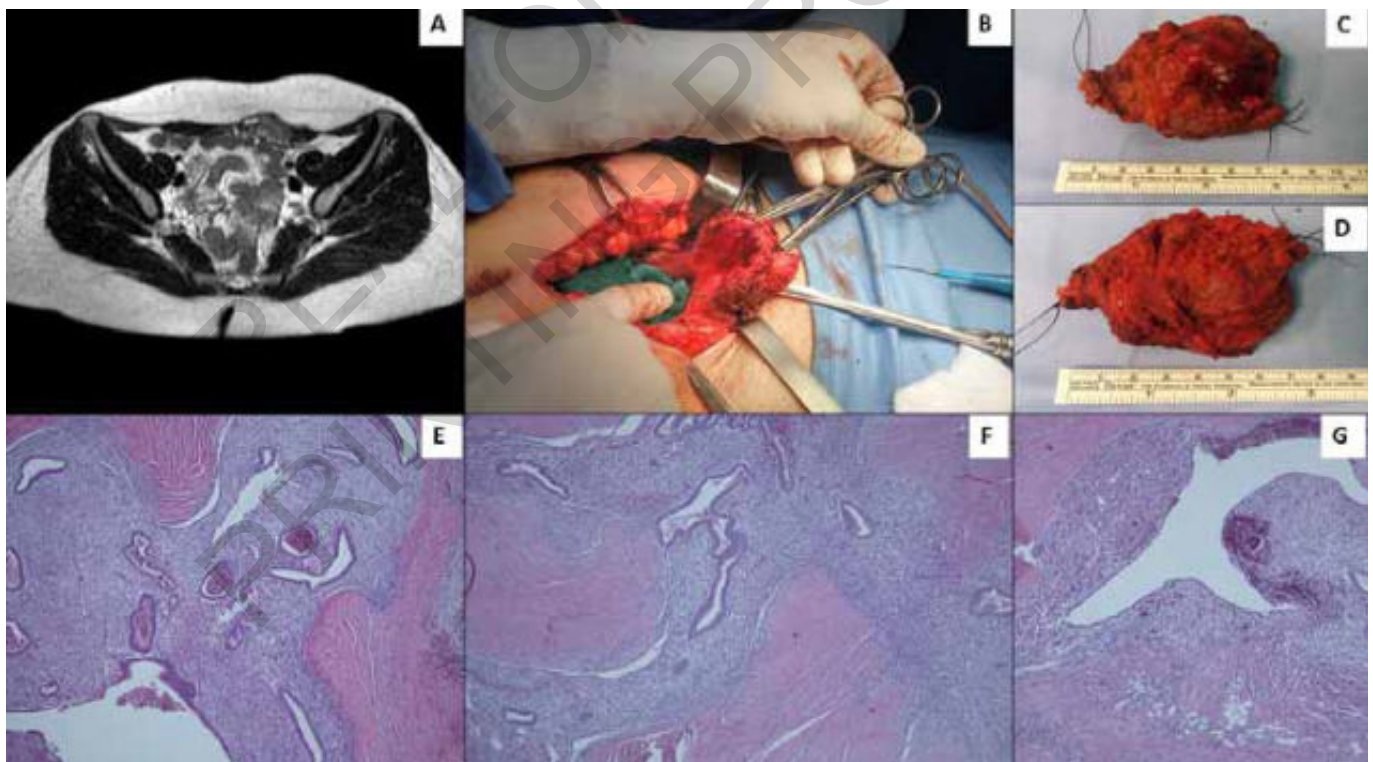


Fig. 1: MRI revealed a 6x3x4 cm solid-cystic heterogeneous mass within the left rectus abdominis muscle, showing hypointense signal, small foci of high signal intensity and contrast enhancement, and without evidence of involvement of abdominal and pelvic cavities (A). Full wall exeresi including the peritoneal plane with large free margins. Residual defect abdominal wall after resection of the lesion: intraoperative surgical image (B). Totally excised mass: peritoneal side (C) and fascial side (D). Histological examination revealed microcystic endometrial glands intermingled to muscle bundles and filled of necrotic debris and erythrocytes (E, F, G).

menstrual cycle⁹. However, in some cases the clinical manifestations are nonspecific: the pain may be vague or absent¹⁰. Symptoms may not occur until years after uterine surgery and may therefore not be recognized as related to prior surgery. The differential diagnosis may include a variety of conditions such as hernia, lipoma, desmoid tumour or primary or metastatic malignancy⁷. Desmoid tumours (DT), also known as aggressive fibromatosis, are fibroelastic tumors, characterized by a benign histological profile, without metastatic potential, but with local aggressive behavior and a strong tendency to recurrence¹¹. They arise from musculoaponeurotic structures of the abdominal wall, especially the rectus and internal oblique muscles and their fascial coverings. These neoplasms are associated with pregnancy, trauma, and with familial adenomatous polyposis. They usually present with a non-tender and painless firm mass. In medical literature are reported cases of desmoid tumours with endometriosis-like symptoms and case of mixed endometrioma and desmoid histology imaging can be used for the diagnosis of soft-tissue masses, including Doppler Ultrasound (DU), Computed Tomography (CT) and Magnetic Resonance Imaging (MRI)^{5,8-9,14}.

Unfortunately, they are often non-specific and do not allow to diagnose the true histological nature of the lesion although they are useful in determining the extent of the disease and in the planning of operative resection, especially in the case of large lesions.

Ultrasound appearance of a desmoid tumour may closely resemble that of scar endometriosis¹⁴. MRI is more accurate for diagnosis, especially when small hemorrhagic areas are present within the nodule, which increase the specificity of diagnosis⁵. Although the correlation between clinical and surgical history with imaging findings is crucial in assessment of the abdominal wall lesions, the correct diagnosis requires the histopathologic analysis of the surgical sample, pointing out the importance of the surgical treatment⁷. Furthermore, histology represents the gold standards for the determination of foci of AWE malignant transformation (less than 1%, most of all clear cell carcinoma), being no specific symptoms, nor specific MRI signs or tumoral markers¹⁵.

The treatment of choice for AWE is wide local excision of the lesion with negative margins. The underlying muscular-aponeurotic structures involved must be resected, this may lead to a wider defect. Extensive disease involving the fascia may require mesh reconstruction⁶. Imaging mapping is crucial for better surgical results and to avoid residual disease even if after wide local excision the recurrence rate is low⁷. Instead, when the natural history of desmoid tumour has become better understood, the management has been moved towards "watchful waiting" because it is evident that some tumors will not grow and may even spontaneously regress sparing patients the morbidity of more aggres-

sive therapy^{11,16}. Even for AWE the development of minimal-invasive therapies is a rising field (percutaneous cryoablation and radiofrequency ablation) with growing evidence of comparable outcomes¹⁷ and this stresses the importance of appropriate imaging and recognition of AWE in the future and the need for radiologists to familiarize themselves with the numerous AWE presentations on imaging.

Conclusions

AWE is the presence of ectopic endometrial tissue in the abdominal wall, presenting as a tumour-like mass, so patients are referred to the general surgeon. The pre-operative diagnosis is erroneous in most cases because the different imaging modalities are nonspecific but only useful in determining the extent of disease and in the planning of operative resection. A better acquaintance with the imaging presentation of AWE holds the potential of positively impact disease confirmation and may play a crucial role in the face of innovation in treatment.

Riassunto

Descriviamo un caso insolito di un grande endometrioma addominale intramuscolare, diagnosticato inizialmente come tumore desmoide, in una paziente di 36 anni con una storia di un anno di dolore addominale nei quadranti inferiori, parzialmente alleviato dall'assunzione di analgesici. Sebbene la parete addominale sia uno dei siti più frequenti di endometriosi, la localizzazione nel muscolo retto anteriore dell'addome è comunque insolita. Gli impianti endometriotici sono generalmente incorporati nello strato adiposo sottocutaneo ed eccezionalmente nei muscoli della parete addominale vicino al sito delle cicatrici chirurgiche (endometriosi cicatriziale). La diagnosi differenziale può includere una varietà di condizioni come ernie, lipomi, neoplasie primarie o metastatiche e tumori desmoidi che crescono attraverso l'aponeurosi muscolare. Nella maggior parte dei casi, la diagnosi preoperatoria è fuorviante perché le diverse modalità di imaging sono aspecifiche e non consentono di diagnosticare la vera natura istologica della lesione, sebbene siano utili nel determinare l'estensione del tessuto patologico e per la pianificazione della resezione da effettuare, specialmente nel caso di lesioni di grandi dimensioni. Una migliore conoscenza degli aspetti di imaging dell'endometriosi della parete addominale ha il vantaggio di consentire una più facile conferma diagnostica preoperatoria e può svolgere un ruolo cruciale di fronte alla possibilità di scelta di un trattamento chirurgico o minimamente invasivo come crioablazione percutanea e ablazione con radiofrequenza.

Acknowledgments

Special thanks go to Dr. Luca Reggiani Bonetti for the histological and diagnostic support and to Dr. Marzio Malagoli for the bibliographic and surgical iconography.

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