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Abdominal wall endometriomas. Should the approach vary based on its relationship to the fascia? A case report and review of the literature

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ABSTRACT

INTRODUCTION: Abdominal wall endometrioma (AWE) is a rare encountered condition with a prevalence of 1–2%. Multiple diagnostic and treatment modalities are available; however, no clear guidelines exist. On occasions muscle and fascia excision might be necessary to achieve a clear margin. To avoid mesh complications, we believe the treatment should depend on tumor location in relation to the abdominal wall fascia. As far as we know this approach has not been previously discussed.

PRESENTATION: A 29-year old female with a surgical history of 3 C-sections presented to us with 6 months of cyclical abdominal pain in the left lower quadrant. Imaging studies confirmed the presence of a mass overlying the left lower rectus abdominis muscle. After imaging studies, the mass was surgically excised. Pathology confirmed a benign endometrioma.

DISCUSSION: Unfortunately, the surgical literature has not established a consensus on the best approach for diagnosis and management of this condition. The purpose of this report is to not only to present another case of this rare phenomenon, but to address the need for guidelines and review the current diagnostic and treatment available options. We also attempt to increase the awareness of this condition, it's unlikely malignant degeneration and potential morbidity of surgical excision.

CONCLUSION: Surgical excision remains the standard of care for AWEs. In those patients where the fascia and muscle must be excised, we recommend less invasive modalities to avoid mesh complications. The need for guidelines remains.

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1. Introduction

In the United States, it is estimated that approximately 6.1% of women between the ages of 18–49 have been diagnosed with endometriosis [1]. Abdominal wall endometriomas (AWE) are noted to be an extremely uncommon, and as of 2016, only 20 cases were found to be reported in the literature [2]. Our personal PubMed search showed 63 case series and case reports from 1947 until now. While ultrasound guided needle biopsy is a diagnostic option, limitations such as patient body habitus, excess adipose or scar tissue, low sensitivity and operator dependence are limitations that can often hinder its application [3]. The use of MRI and CT scans, while non-specific, can help with localization and surgery planning. Treatment classically consists of surgical removal, with some reports suggesting the use of an oral contraceptive post-operatively [4]. This condition can be difficult to diagnose, and a morbid operation might be needed if abdominal muscle and fas-

cia needs to be excised to achieve clear margins. Moreover, mesh placement in these situations is not uncommon. Given the short, and long-term potential mesh complications we believe these patients might benefit from other treatment alternatives. Cryoablation, high intensity focused ultrasound ablation, and ultrasound guided ethanol injection have also been used to treat AWE [5–7]. Malignant transformation to adenocarcinoma is an ominous, but realistic possibility [8]. While a rare occurrence, it highlights the importance of proper diagnosis, and appropriate treatment. We report a case of a benign 2 cm abdominal wall endometrioma surgically excised without complications in a rural hospital in Southeast Missouri and review the literature with regards to current diagnostic and treatment modalities.

2. Case report

A 29-year-old African American female patient was referred to us for a 6-month history of left lower quadrant abdominal pain, along with a soft tissue mass in the same location. Pain had been present for about six months. It was sharp, cramping and non-radiating. Her discomfort was also positional in nature and would worsen during menstrual cycle. Past medical history was significant for 3 previous C-sections, and obesity. She was also on

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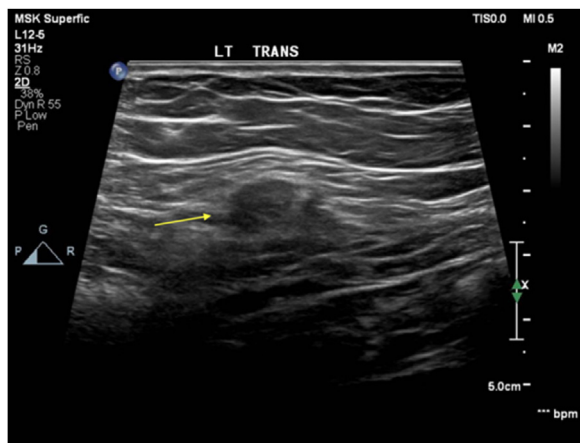


Fig. 1. Ultrasound showing the AWE appearing as a hypochoic mass.



Fig. 3. Abdominal wall, incision in the lateral aspect of previous Pfannenstiel incision.

norgestimate-ethinyl estradiol 0.25 mg/35mcg per tab one-time daily PO for birth control. Physical exam showed a nontender, palpable mass in the left lower quadrant of the abdomen underneath the lateral aspect of her C-section scar.

Ultrasound reported a 19 × 19 × 9 mm extra fascial macrolobular and hypochoic mass lesion, with internal color flow involving the anterior margin of the left lower rectus muscle (Fig. 1). CT scan findings showed a poorly delineated mass of approximately 2.5 cm in diameter in the same location, just medial to the left anterior superior iliac spine (Fig. 2; a–c). Initially an ultrasound guided core needle biopsy was ordered, however, due to the patient’s body habitus (BMI 38.66), Interventional Radiology (IR) was not able to perform it. After discussing the best available options, the patient was consented for an elective outpatient excisional biopsy. Based upon the patient’s history of 3 previous C-sections, and the cyclic nature of the pain, we believed that the mass was likely an endometrioma. Since the mass overlaid the fascia, we did not anticipate major fascial or muscle excision, nor the need for mesh placement.

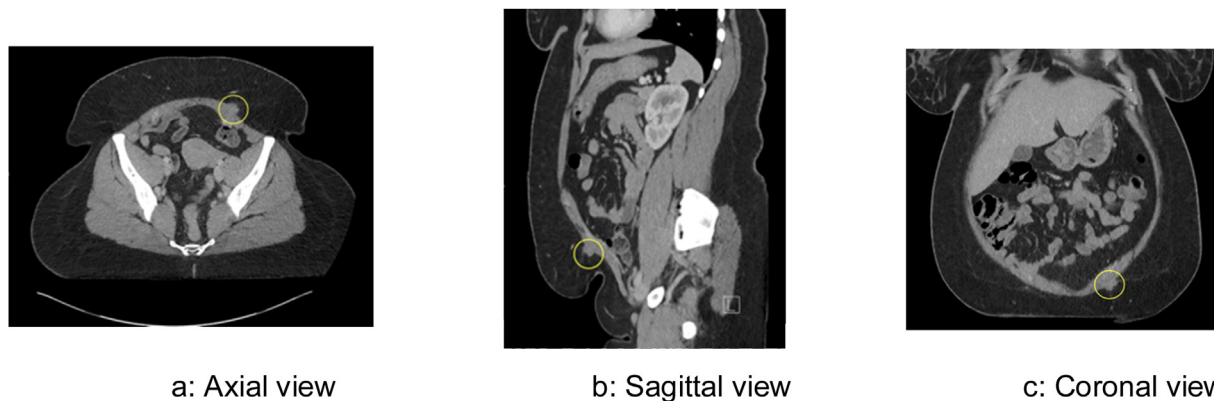
Before surgery, the patient was asked to sit midway between supine an upright position, allowing us to palpate the mass and mark at the skin level. In the OR (operating room) she was placed in decubitus position and given general anesthesia. A 4 cm incision was made overlaying the previously marked area which coincided with the later aspect of the C-section scar (Fig. 3). Dissection was carried down to the level of the fascia. At this point, the mass was able to be clearly visualized. The mass was anterior to the rectus muscle and barely involved the underlying fascia. It was entirely

excised along with 1.5 cm of muscular fascia and about 1 cm of peripheral normal looking tissue. Frozen section revealed benign endometrioma with clear margins (Fig. 44.1, 4.2). Once the diagnosis was confirmed 3 stitches (2.0 Vicryl) were used to approximate the rectus fascia, followed by 3 stitches to approximate Scarpa’s fascia. The skin was closed with staples. The patient recovered without any complications.

At 3 weeks follow up the patient was doing well, with no pain and the incision had healed well. After 9 months post-pp, follow up showed the patient was doing well, with no signs of recurrence, and reported her pain had disappeared.

3. Discussion

Our own search found 63 published case series and case reports in PubMed from 1947 until now. AWE can pose a diagnostic and treatment challenge. Most cases present with a history of C-section and cyclic abdominal pain that is worse during menstruation. Symptoms can mimic any acute abdomen-like condition [9]. The most common symptom is cyclic abdominal or parietal pain in 68.6%, followed by palpable mass in 25.7% and no symptoms is 17.1% of patients [10]. From 1983 to 2002 Zhao et al. reported an incidence of AWE of 1.04% (57/5478) in patients with endometriosis after surgical treatment and 0.046% in patients undergoing cesarean section in the same period [11]. Interestingly AWE was the most common abdominal wall mass found in female patients between ages 18 and 55 (24.8%) followed by: adenocarcinoma NOS

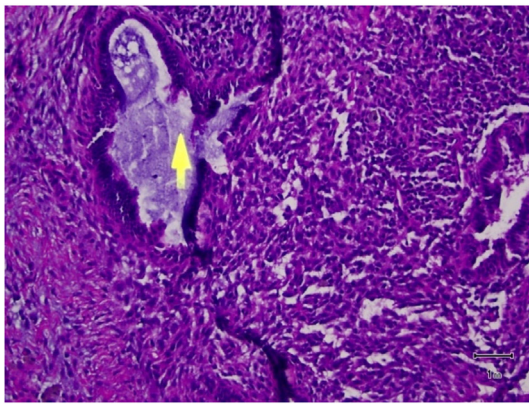


a: Axial view

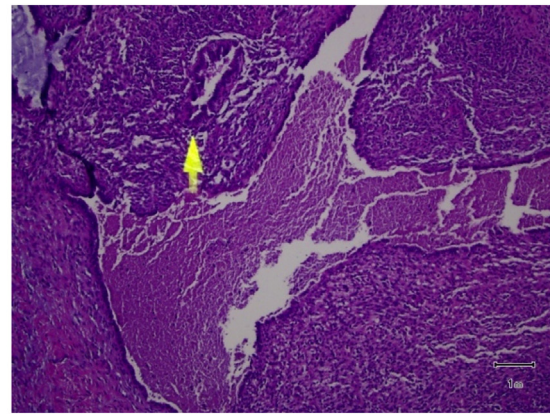
b: Sagittal view

c: Coronal view

Fig. 2. a–c; A, Showing the poorly delineated mass arising from close to the top of the fascia, seen from axial view on CT scan. B, Sagittal view on CT scan. The degree of adipose tissue can be appreciated in this image and helps provide a clear view of why palpation of the mass was difficult on physical exam. C, Coronal view showing the mass appearing to be on top of the fascia and confirming the location being medial to the anterior superior iliac spine.



1



2

Fig. 4. 4.1 and 4.2 shows H&E staining of the frozen section taken during surgery. The mass was identified as a benign endometrioma, with the yellow arrows highlighting endometrial glands and stroma, confirming the diagnosis.

(21%), desmoid tumor (14.3%), leiomyosarcoma (8.6%), clear cell adenocarcinoma (4.8%), lymphoma (2.9%), and others [12].

AWEs should be in the top differential diagnosis in female patients with a history of C-sections, cyclic pain and an abdominal mass. Based on its position within the abdominal wall layers they can be: A) Superficial, in the subcutaneous tissue only. B) Intermediate, infiltrating the abdominal rectus muscle fascia, and C) Deep, in the abdominal rectus muscle, below the fascia [13]. Since signs are not specific, the differential diagnosis can be ample, including: suture granuloma, lymphadenopathy, abscess, inguinal hernia, incisional hernia, primary or metastatic cancer, lymphoma, lipoma, hematoma, sarcoma, desmoids tumor, and subcutaneous and sebaceous cysts.

Endometriosis-associated malignant transformation in abdominal surgical scar (EAMTAS) occurs in less than 1% of patients suffering from endometriosis. Mihailovici et al. in a systematic review (48 patients) of EAMTAS reported that the most common histological type was clear-cell carcinoma (CCC) (66.7%) followed by endometrioid carcinoma (14.6%), serous papillary carcinoma (8.3%), mixed types (4.2%), adenocarcinoma and sarcoma [14]. They also pointed out that the 5-year survival after diagnosis is 40% and that tumors ≥ 8 cm have a poorer prognosis.

Diagnostic imaging modalities include ultrasound (US) CT and MRI. The ACR (American College of Radiologists) Appropriateness Criteria lists US as the first diagnostic modality, followed by MRI (given its superior soft tissue resolution), and CT scan [15]. US characteristics include round/oval, hypoechoic nodules with ill-defined borders and hyperechoic rim. Doppler shows scarce blood vessels [16]. Yarmish et. investigated the differences between AWE and other masses utilizing CT scan in 105 patients. CT features significantly associated with AWE were: location below the umbilicus ($p = 0.0188$), homogeneous density ($p = 0.0188$), and presence of linear infiltration irradiating peripherally from a central soft tissue nodule (i.e. “gorgon” sign) ($p < 0.0001$). When these three characteristics were present, the highest sensitivity and specificity was achieved 0.69, 95% CI: 0.48–0.86 and 0.97, 95% CI: 0.91–1.00 [17]. MR of AWE lesions shows an isointense or slightly hyperintense signal compared with muscle on T2-weighted images and isointense or slightly hyperintense signal compared with muscle on T1-weighted images with foci of high signal intensity, indicative of hemorrhage [18].

Core needle biopsy (CNB) and fine needle aspiration (FNA) have proven to be accurate for diagnosis of AWE. In FNA cytology the smears are cellular, consisting of epithelial and stromal fragments.

The epithelial cells are arranged in monolayer sheets of polygonal cells with large, hyperchromatic nuclei and moderate amount of cytoplasm, with considerable nuclear overlapping. The stromal aggregates also showed crowded overlapping nuclei and scant admixed hemosiderin-laden macrophages. Other abdominal wall lesions have well-defined cytological features. Desmoid tumor and fibrosis present less cellularity with benign-appearing mesenchymal cells. Suture granuloma shows non-specific inflammation with or without granulomatous elements and foreign material. Fat necrosis displays foamy macrophages, inflammatory and multinucleated giant cells, fragments of adipose tissue and no epithelial cells. Nodular fasciitis shows myxoid background and pleomorphic cells. Smears from primary or metastatic malignancies are characterized by hyper cellularity with frankly neoplastic cells [19]. Interestingly no reports are found comparing FNA vs. CNB in AWEs.

Medical treatment of AWE has been proven to be ineffective and hence it’s not discussed in this review. Standard of care of AWE remains surgical excision with free margins [20]. AWE can be located in the subcutaneous tissue, muscular fascia, or muscle. Almost always the involve the anterior rectus fascia [21]. When that happens depending on the size of the lesion in approximately 10% of the cases mesh reconstruction is needed [22]. The potential adverse effects of mesh placement are well known. In about 2% of cases patients will develop mesh infection with the subsequent need for explantation in almost 75% of cases [23].

Percutaneous image guided cryoablation has been successfully used to treat different soft tissue tumors and metastases with acceptable results. Procedural pain is well tolerated. It can be performed via US, CT or MRI. The procedure is usually performed under general anesthesia using argon-based cryoablation systems with cryoprobes of 1.7–2.4 mm of diameter. Cryoprobes are positioned with 1–2 cm distance within the tumor, these are expected to achieve a confluent ice ball with at least a 0.5 cm margin around the AWE. Follow up imaging at 3 months shows usually no residual enhancement, further studies with longer follow up are needed however [5].

High intensity focused ultrasound (HIFU) ablation is performed after the probe is inserted into the lesion. HIFU causes coagulation necrosis of the target lesion without damaging the surrounding tissues and those in the acoustic pathway. The procedure is performed via ultrasound guidance. Zhao et al. in a retrospective study compared 25 patients treated with HIFU versus 29 patients treated with classic surgery. With a median of 32 months of follow up 92% of the patients in the HIFU group achieved remission versus 100% in the

surgery group. However, 40% of the patients in the surgery group required a new incision, and 6% mesh implantation [6].

Ultrasound guided ethanol injection has also been described as a treatment. Bozkurt et al. described one patient where after malignancy had been excluded, 1 cc of 95% ethanol was injected with ultrasound assistance into various parts of the 3 cm lesion using a 22-gauge needle. After 9 months of follow-up the patient was symptom-free without recurrence [7].

We consider that after malignancy has been excluded via needle biopsy, intramuscular lesions or those with the potential to leave a fascial defect > 3 cm should be treated with noninvasive techniques. The risk of a morbid operation and mesh implantation should be thoroughly discussed with the patient. Formal guidelines for this rare condition should be developed. This case was reported in line with the SCARE guideline.

4. Conclusion

Diagnosis and treatment of AWEs is highly variable. Surgical excision remains the standard of care. Practitioners should be aware of the condition specially the potential for malignant transformation. A non-surgical approach should be considered in a high-risk patient where mesh implantation is more likely. Clear guidelines for this condition are lacking.

Declaration of Competing Interest

None.

Funding

None.

Ethical approval

No ethical approval needed.

Consent

“Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request”.

Author contribution

Both authors contributed to the following:
 Study concept or design
 Data collection
 Data analysis or interpretation
 Writing the paper

Registration of research studies

Not applicable.

Guarantor

Cesar Reategui MD FACS.

Provenance and peer review

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