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Case Report

Massive endometrioma presenting with dyspnea and abdominal symptoms

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ABSTRACT

An abdominal mass may present with a myriad of symptoms resulting from compression of surrounding organs. A major clinical challenge with practical implications is accurate preoperative identification of the origin of the mass. Here, we present the case of a 29-year-old female patient with abdominal distension and shortness of breath for approximately 6 weeks before presentation. A large abdominal mass compressing the surrounding organs was observed on abdominal x-ray and computed tomography of the abdomen and pelvis. Preoperative imaging was unable to identify the organ of origin; pathologic and histologic analyses of the tumor ultimately identified a rare, massive intra-abdominal endometrioma, freely floating within the peritoneum and fed by an omental blood supply. This case highlights the importance of considering an atypical presentation of endometriosis in women of reproductive age with abdominal complaints.

Introduction

Abdominal distension, early satiety, and compression of adjacent organs may indicate the presence of an intra-abdominal mass. The primary objective in diagnosis of an abdominal mass is identifying the organ of origin, which may prove a significant challenge preoperatively [1,2]. Histologic classification has classically divided abdominal masses by their lineage of origin, whereas radiologic studies in the past have focused on the degree of complexity on imaging, such as simple cystic, complex cystic, and cystic with solid components. These distinctions not only allow for radiologic classification but also suggest certain diagnoses and help guide clinical management [3]. The likelihood of specific etiologies depends on age [4–6], sex of the patient, the organ from which the mass originated, and the imaging characteristics of the mass.

In female patient, both gastrointestinal and gynecologic etiologies must be considered in the differential diagnosis of an abdominal mass [2]. This is an important distinction because

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it will dictate the specialists involved in medical and surgical management pre- and post-operatively. Here, we discuss a unique case of a patient presenting with a large, space occupying mass whose site of origin could not be identified radiologically before surgical intervention.

Case report

A G0P0 29-year-old women with a medical history significant for a *Helicobacter pylori* negative gastric ulcer 2 years before presentation, chronic gastroesophageal reflux disease, and chronic dysmenorrhea presented to her primary care physician with an approximate 1-month history of increasing abdominal distension, unintentional weight gain, early satiety, postprandial abdominal pain, and increasing shortness of breath. Physical examination demonstrated significant abdominal distension, and she was referred to a gastroenterologist for further evaluation.

After evaluation by the gastroenterologist, the patient was sent for an obstructive series consisting of 2 abdominal x-rays, taken supine and erect, secondary to concern for obstipation (Fig. 1). The images demonstrated a large pelvoabdominal soft tissue mass with cranial displacement of the transverse colon draping over the large mass.

Based on these results, a computed tomography (CT) was obtained. It demonstrated a large, cystic, space occupying mass that extended from the substernal to suprapubic regions, measuring 22.9 cm craniocaudally, 12 cm anterior–posterior, and 22.3 cm transversely (Figs. 2 and 3). No discrete solid components or septations were observed in the mass. These images demonstrated posteriorly displaced intestines and hydronephrosis of the right kidney (Fig. 2).

A 6.4 cm × 6.0 cm × 7.0 cm mass of unclear origin abutting the uterus was also found, which was thought to represent either a pedunculated fibroid or an ovary (Fig. 3A-C). There was also heterogeneous enhancement of the uterus with multiple masses and some cystic areas, consistent with multiple uterine fibroids (Fig. 3A, C). Additional incidental findings included a 2.0 mm lung nodule in the right lower lobe, multiple low-attenuation cysts within the liver, a 1.1 cm renal cyst in the right kidney, and mild fullness in the left renal collecting system.

Based on these radiologic findings, the decision was made to remove the mass by open laparotomy with a presumptive preoperative diagnosis of benign serous or mucinous cystadenoma of the ovary. Of note, preoperative blood work demonstrated an elevated CA-125 of 108.4, normal CA-19-9 of 17.4, and carcinoembryonic antigen (CEA) of <0.5. Because of the patient's age and desire to retain childbearing ability, the patient was not consented for hysterectomy. Preoperative discussion included the possibility of performing a second surgery including unilateral or bilateral oophorectomy and/or salpingohysterectomy if needed based on findings during the initial surgery.

On laparotomy, the mass was found to have a blood supply originating in the greater omentum. It was removed without incident along with part of the greater omentum. Based on this finding, the larger mass was labeled an omental cyst.

The mass abutting the uterus appeared consistent with a pedunculated fibroid intraoperatively, and it was attached by a pedicle to the uterus. It was also removed without incident, and a leiomyoma was subsequently confirmed by pathologic analysis. Both ovaries were identified and were normal. The operation was completed, and the patient was discharged on hospital day 3. She recovered without any significant complications and with total resolution of her preoperative symptoms.

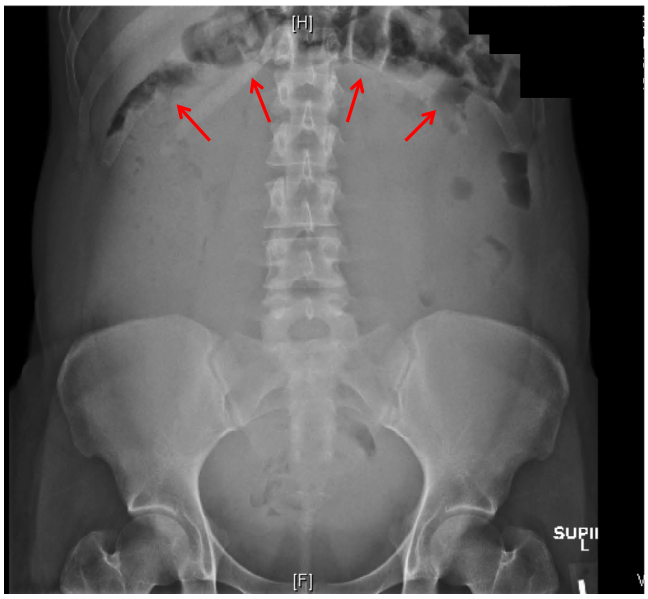

Fig. 1 – Obstructive series abdominal x-ray. Supine abdomen shows pelvoabdominal soft tissue mass displacing transverse colon superiority (arrows).

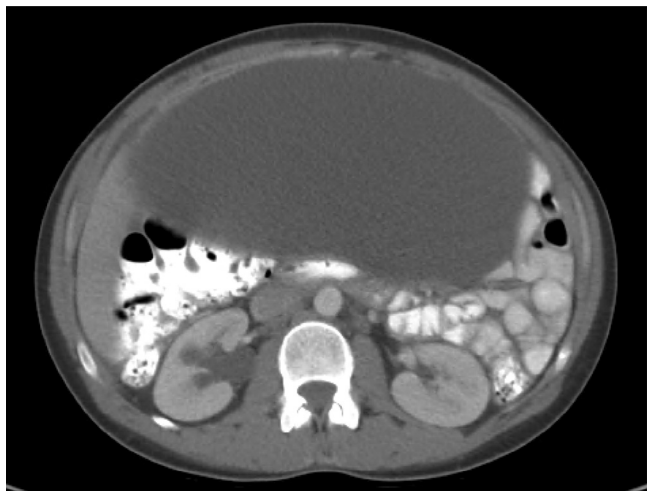

Fig. 2 – Computed tomography (CT) of the abdomen and pelvis demonstrating large cystic intra-abdominal tumor causing mass effect. Axial oral and intravenous contrast-enhanced CT image through the middle renal level shows the fluid-filled mass extending into the upper abdomen. The mass is displacing bowel loops. There is right-sided hydronephrosis.

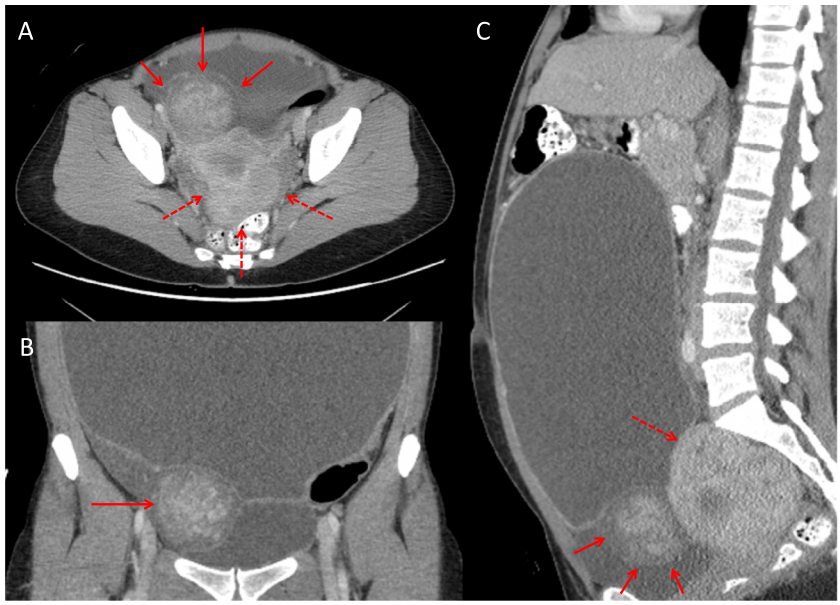

Fig. 3 – Computed tomography (CT) of the abdomen and pelvis demonstrating solid heterogeneous mass abutting the uterus (solid arrows) and the heterogeneously enhancing uterus consistent with uterine fibroids (dashed arrows). (A) Axial oral and intravenous contrast-enhanced CT image through the lower pelvis shows uterus with fibroids, and the anteriorly located exophitic fibroid. (B) The extra-uterine fibroid is well visualized in the coronal view. (C) Sagittal reconstructed CT image through the middle abdomen shows the cystic mass extending from the pelvis to the upper abdomen, the uterus with the fibroids, and the anterior exophitic fibroid. Note in (A) and (C) that while the solid mass clearly abuts the uterus, there is no pedicle or site of attachment to the uterus identified.

Gross pathology of the abdominal mass demonstrated a smooth-walled tan-pink lesion measuring 26.0 cm × 26.0 cm × 11.0 cm containing serosanguineous fluid (Fig. 4A). Pathologic examination of the cyst wall with Hematoxylin and eosin (H&E) stain showed eosinophilic epithelial lining cells, some ciliated, with subjacent stroma consisting of small naked nuclei morphologically consistent with endometrial glands and stroma. Focally, collections of hemofuscin-stained or pigmented histiocytes were identified, a finding associated with endometriotic foci due to menstruation into the cyst (Fig. 4B).

Immunohistochemical stains supported this finding. The cyst wall was positive for desmin (Fig. 4C), which supported the morphological impression of smooth muscle metaplasia, a known occurrence in the wall of an endometrioma. Conversely, there was no staining for inhibin, which helped exclude the presence of ovarian stroma, thereby further decreasing the likelihood of an ovarian cyst such as a serous cystadenoma as a diagnostic possibility (Fig. 4D). Finally, a thin layer of CD10+ cells underlying the endometrial-type lining helped confirm the diagnosis of endometrioma (Fig. 4E).

Notably, the patient carried no preoperative diagnosis of endometriosis, and beyond the endometrioma, there were no additional endometriotic rests visualized on open laparotomy. Follow-up imaging and medical management were recommenced to monitor for disease recurrence.

Discussion

Endometriosis is a common clinical entity, known to both physicians and lay people alike. However, the manifestations of endometriosis are protean, and the less common manifestations may be largely unknown even to seasoned clinicians. This case of a large endometrioma causing gastrointestinal and respiratory symptoms due to mass effect highlights the importance of including the underlying endometriosis in the differential diagnosis of nearly all intra-abdominal complaints in women of childbearing age.

Omental endometriosis is a rare entity, with only a few cases reported [7,8]. Although rare, there have been reports of large endometriomas causing mass effect within the abdominal cavity, with the largest case report describing a 64 kg endometrioma [9]. However, nearly all endometriomas reported have been much smaller, and therefore there was low clinical suspicion that this mass represented an endometrioma preoperatively. Abdominal wall endometriomas also occur, although infrequently [10,11]. Symptoms caused by these less commonly located deposits are frequently due to the invasive nature of advanced disease and its hormone responsiveness, not necessarily the size and mass effect of the endometrioma [10].

Elevated levels of CA-125 [12] and CA-19-9 have been shown in patients with endometriomas [13], but the use of CA-125 to identify endometriosis remains controversial [14]. Ding et al. demonstrated that 64.9% of patients with abdominal wall endometriomas had normal CA-125 levels. In the case of this patient, although CA-125 was elevated, the patient's level was much less than that has been reported in a prior case of a large endometrioma [13]. Further, CA-19-9 levels in this patient were within normal limits. Still, these tests may provide important preoperative information to implicate a gynecologic versus gastrointestinal etiology.

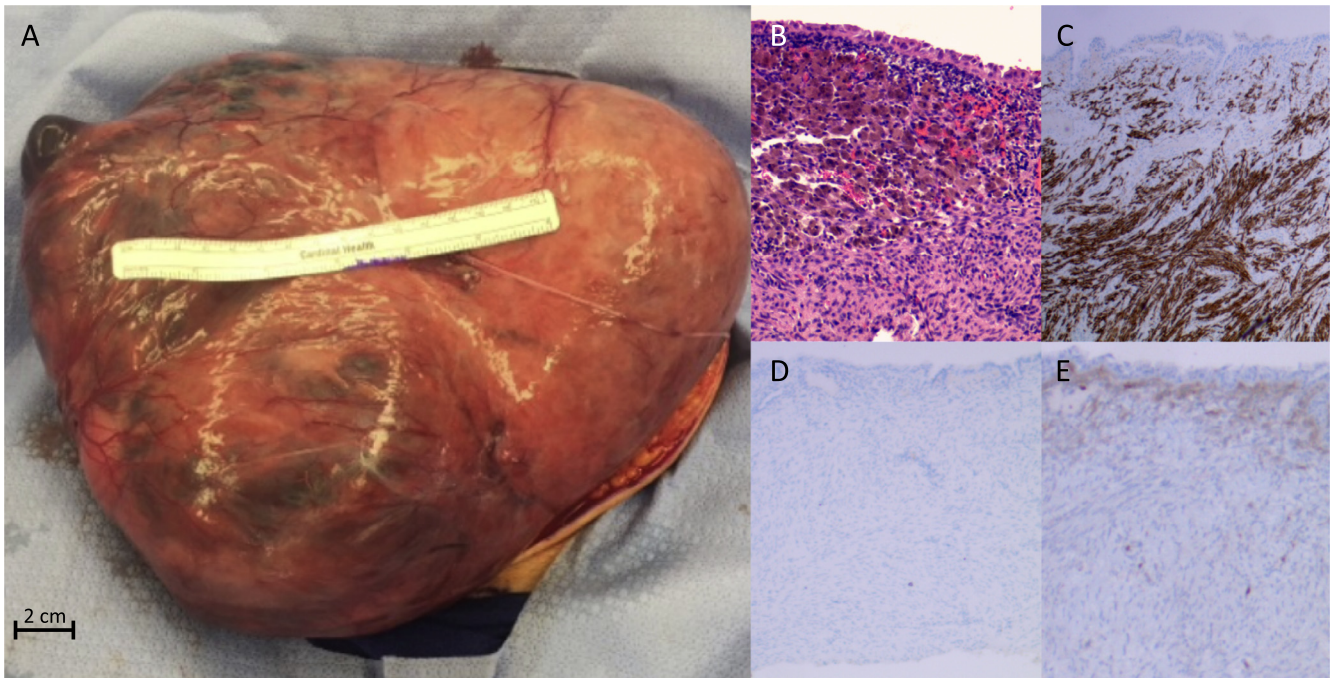

Fig. 4 – Abdominal mass examined on gross and microscopic pathology. (A) Gross pathologic appearance of specimen. The right side of the image is cephalad, whereas the left side is caudal. (B) H&E staining at medium power of the epithelial lining of the mass wall shows a blue rim of cells consistent with endometrial stroma. Also present are hemofuscin-pigmented histiocytes from menstruation into endometriotic foci. The wall of the mass stained positive for desmin (C) and negative for inhibin (D). Finally, in (E), a layer of CD10+ cells is observed subjacent to the endometrial lining cells, consistent with endometriosis.

Several aspects of this case were remarkable, primarily the size of the endometrioma, the blood supply, and the patient's lack of prior surgical history. With the exception of the case report of Sakpal et al. [9], we were unable to find any other cases of endometriomas approaching the size observed in our patient. Ten centimeters is generally accepted as the maximum size for an endometrioma, far less than the size of the mass observed in this patient. Further, the implantation of an omental vessel into the endometrioma is another unique finding for which we can identify no parallel in the literature, and was likely the mechanism by which this patient's endometrioma was able to grow to such a large size.

Prior studies have shown that abdominal endometriomas frequently emerge from the site of a prior abdominal wall scar [10,11], but this patient had no surgical history. Although there have been cases of spontaneous abdominal wall endometriomas, these were intrinsic to the abdominal wall, not essentially free-floating masses as observed in this case [15,16].

Finally, although the patient had a longstanding history of dysmenorrhea, the symptoms that led her to seek medical treatment were due to the mass effect of the tumor. Studies have shown that the majority of patients with abdominal wall endometriosis present with pain and cyclic symptoms [10,11]. Thus, the patient's presentation is not classic in that regard because she did not experience any cyclic pain associated with the mass, only chronic and postprandial pain presumably associated with the mass effect of the endometrioma.

When an endometrioma is suspected clinically, transvaginal ultrasound and magnetic resonance imaging (MRI) have been the most studied imaging modalities for evaluation. Because endometriomas present with a variety of nonspecific symptoms, patients are frequently referred for other imaging studies, as in this case [17]. There have been few studies establishing the CT imaging characteristics of endometriomas compared with those of ultrasound or MRI. One study of these lesions on CT characterized their features as ill-defined, solid, isodense to muscular tissue, and with slight enhancement on contrast-enhanced images [18]. However, the intrinsic risk of CT imaging is that a deep infiltrating endometriosis may be mistaken for an infiltrating malignant mass, underscoring the need for MRI for proper diagnosis of the lesion [19]. Because of the intrinsic imprecision of CT imaging for diagnosing endometriomas, the technique has largely been abandoned except in cases such as this where the pathologic origin is completely unknown before imaging studies are ordered and when endometriosis is not suspected.

A small but definite percentage of endometriomas demonstrate a hyperdense round or crescent-shaped focus within the lesion. Although not sensitive, this finding is highly specific for endometriomas, as none of the other types of ovarian masses studied were found to have this crescent-shaped focus [20].

The mass described in this study had features consistent with those described previously, which have been observed in CT imaging of endometriomas. If not a clinically urgent situation, MRI of this mass for better characterization would have been appropriate. However, the unequivocal need for surgery made additional imaging studies less of a priority,

and the lack of known history of endometriosis reduced the index of suspicion that this mass in fact was an endometrioma.

From a radiological perspective, this case was a challenge because the normal anatomy was distorted such that image interpretation was severely compromised. Further, as detailed previously, this was a unique manifestation of a known entity. Imaging with an additional modality, either ultrasound or MRI, may have aided in preoperative diagnosis; however, the urgency of surgery superseded the need to obtain images from additional imaging modalities. Despite this, the imaging characteristics of this mass were consistent with those of an endometrioma on CT, and therefore, this should have remained high on the preoperative differential diagnosis.

Conclusion

Although endometriosis is a relatively well-known clinical entity, its manifestations are protean and may be unfamiliar to many clinicians. Here, we present a case of the second largest endometrioma identified to date, freely mobile within the abdomen and fed by an omental blood supply in a patient with no prior history of surgery or endometriosis. Preoperative imaging demonstrated a large, space occupying mass thought to be a benign serous cystadenoma of the ovary. Intraoperative examination identified 2 normal ovaries, and pathologic analysis of the tumor was consistent with a massive endometrioma. These findings highlight the importance of a broad differential diagnosis, which includes endometriosis for abdominal complaints in women of childbearing age. Further, these findings demonstrate that even with atypical presentations, radiologic characteristics may provide valuable direction in preoperative diagnosis.

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