Abstract

Background: Endometriosis is a disease that presents primarily with pelvic pain and infertility. Endometrioma is a cyst consisting of endometrial glands and stroma and is typically an advanced presentation of endometriosis. Endometriomas usually grow benignly. There are few reports of patients with endometrioma with malignant features reported in the literature. A case of infrequent occurrence and low diagnostic rate will be reported but having as its origin a relatively common disease with great impact on the female population in reproductive age.

Case Summary: A 54-year-old female patient came in with abdominal wall bulging referring to gradual growth and emergence over the last 4 years. Personal history includes previous cesarean section 2 decades ago. The patient also had hypertension, diabetes, a history of total thyroidectomy for thyroid neoplasia 18 years ago, bile duct lithiasis, uterine myomatosis, obesity and epilepsy that started a few years ago with no defined etiology. We report a case of endometrioid pattern adenocarcinoma of the abdominal wall. The patient was submitted to exploratory laparotomy with resection of the tumor in the abdominal wall besides extended hysterectomy, umbilical herniorrhaphy and cholecystectomy. The procedure was performed under general anesthesia and spinal anesthesia, laparoscopically with a report of cystography and bilateral pelvic lymphadenectomy. The patient was followed up as an outpatient, with adjuvant chemotherapy using the carboplatin and paclitaxel protocol.

Conclusion: This report highlights a case of endometriosis with abdominal wall metaplastic invasion, an uncommon finding in everyday medical practice.

Core Tip: The study consists of a case report of a patient presenting with endometriosis with abdominal wall invasion with elements of conversion to malignant disease and is considered an atypical case rarely reported in the literature. As a result, it may highlight a condition that in its early stages can be treated relatively easily but often goes unnoticed until advanced stages where it is no longer possible to treat it effectively.

Keywords: Endometriosis; endometrioma; adenocarcinoma; cesarean section; abdominal wall; case report.

Introduction

Endometriosis is a disease that presents mainly with pelvic pain and infertility, and is considerably prevalent among women, affecting an estimated 176 million women of reproductive age worldwide [1]. Although it has some features in common with malignant lesions, such as tissue invasion and injury, it is considered to be a benign condition. In rare cases, it is still possible for endometriosis to convert to a malignant disease, with 80% of such cases occurring in the ovary and only 20% occurring in other areas such as the abdominal wall [2].

Regarding implantation in the abdominal wall, although there are case reports of spontaneous appearance of an endometriotic lesion, a history of previous gynecological surgery is typically found. The appearance of a lesion in an operative scar is usually explained as iatrogenic transplantation of endometrial tissue to the edge of the operative wound during the procedure [3].

The incidence of abdominal wall endometriosis ranges from 0.03% to 1.08% among women who undergo pelvic surgery, with a considerable portion of cases being misdiagnosed [4]. Therefore, endometriomas adjacent to cesarean section scars are often overlooked, but knowledge of clinical and sonographic findings could prevent the delay in diagnosis that typically occurs in patients with endometriomas larger than 3 cm [5]. With this research we intend to report a case of abdominal wall endometrioma, describing the anesthetic-surgical management and clinical evolution of the patient. We will also perform a literature review correlated to the results obtained in the case.

Case Presentation

Chief complaints
A 54-year-old female patient arrived with abdominal wall bulging.
History of present illness
The patient reports gradual emergence and growth over the past 4 years.

History of past illness
The patient’s personal history included a previous cesarean section 2 decades ago. The patient also had hypertension, diabetes, a history of total thyroidectomy for thyroid neoplasia 18 years ago, gallstones, uterine myomatosis, obesity, and epilepsy that began a few years ago with no defined etiology.

Physical examination upon admission
On admission to the ICU, after surgery, the patient was eupneic on room air and hemodynamically stable without sedation. Her heart rate was 52 bpm, blood pressure was 110/60 mmHg, and oxygen saturation was 97%. On general examination the patient was in regular general condition although slightly pale (1+/4+). Cardiovascular and respiratory examinations revealed nothing noteworthy. The abdominal evaluation showed a globous abdomen, with weak peristalsis, flaccid and painful deep palpation in the right hypochondrium, without an involuntary defense mechanism. Clinical neurological examination revealed a Glasgow Coma scale of 15/15, and no other pathological signs were noted.

Laboratory examinations
Preoperative laboratory tests were normal.

Imaging examinations
Magnetic resonance imaging (MRI) showed multilobulated cystic formation with septations and partial hemorrhagic content located in the hypogastric abdominal wall, underlying the previous cesarean section scar, measuring 11.5 x 4.5 cm, suggesting a possible endometriotic lesion. The uterus presented with increased dimensions, lobulated contours and a heterogeneous signal due to at least two nodules compatible with leiomyomas: one in the right lateral and posterior, intramural body region, with an iso to high signal on T2-weighted images and foci of cystic/mucinous degeneration in between, measuring 5.6 cm x 5.3 cm, and the other in the posterior fundic region, intramural, with an iso to high signal on T2-weighted images and foci of cystic/mucinous degeneration in between, measuring 3.2 cm x 2.9 cm. A subsequent computed tomography (CT) scan showed the lesion in the hypogastric without significant evolutionary change as well as two smaller collections in the abdominal cavity near the left external iliac measuring 2.8 x 2.6 cm and 2.5 x 2.1 cm. There was no well-defined continuity between the bladder and the aforementioned collections. No signs of pulmonary involvement were found.

Further diagnostic work-up
Respecting the severity of the extension and progression of the disease, management by exploratory laparotomy with tumor resection in the abdominal wall was chosen, in addition to extended hysterectomy, umbilical herniorrhaphy and cholecystectomy. The procedure was performed under general anesthesia and spinal anesthesia, laparoscopically, with a report of cystography during the procedure due to adherence of the tumor wall to the bladder, and bilateral pelvic lymphadenectomy. A specimen collected during the procedure revealed a histological finding of grade 2 adenocarcinoma with an endometrioid pattern, with infiltration of skeletal muscle tissue adjacent to the presence of endometriosis. In a histofluorescence study, a positive response to CD10, CK7 and PAX-8 antibodies was found.

Final Diagnosis
The final diagnosis of the presented case was adenocarcinoma with an endometrioid pattern (endometrioma) located in the abdominal wall.

Treatment and Follow-Up
The patient underwent laparoscopy for tumor resection accompanied by hysterectomy and cholecystectomy. The surgery also required cystorrhaphy due to tumor adherence in the bladder region, and pelvic lymphadenectomy. After the surgical period, the patient was followed up as an outpatient and adjuvant chemotherapy using a carboplatin and paclitaxel protocol was indicated.

Discussion
Adenocarcinoma arising from abdominal wall endometrioma is a rare and aggressive carcinoma, with 27 cases identified in English literature between September 1986 and August 2014, that is often diagnosed at the metastasis stage and is unresectable [6]. The most common symptom of the disease is the growth of a mass in the abdominal wall, but it is noteworthy that a relatively small number of patients report pain, which corroborates the misdiagnosis of incisional hernia [5]. Even though it is a condition of considerable rarity, the number of reported cases has been increasing in recent years, probably due to the greater attention given to the disease and the increased rate of documented cesarean sections and uterine surgeries. Consequently, a greater dissemination of knowledge of the disease and a better understanding of its pathogenesis may be of great importance for an early diagnosis and a more effective treatment [7].

Most cases of this type of carcinoma occur as an evolution of surgical scar endometriosis after cesarean section, and the average interval between diagnosis of abdominal wall endometrioma and the last surgery is considerably long (20.2 years), suggesting a slow evolution of the tumor [6]. However, there are still cases with malignancy not associated with endometriosis, which reinforces the importance of elucidating its pathogenesis for correct prevention and treatment of the disease [6].

As a measure of prevention of endometrial tissue implantation in the abdominal wall after surgery, it is suggested that the wall be thoroughly cleaned and vigorously irrigated before closing the cavity [3]. Due to the infrequency of development of endometriomas, especially extra gonadal ones, the management of these lesions is based on the approaches dem-
endometrioma is unfavorable, especially when compared to the evolution of other carcinomas such as that associated with ovarian endometriosis [8].

The aggressive behavior of abdominal wall endometrioma can be explained by the greater local development at the time of diagnosis. The literature identifies a median survival time of 42 months post diagnosis, with less favorable follow-up for clear cell adenocarcinoma associated with endometrioid adenocarcinoma [9]. Another study showed a 44% mortality rate in the first months after diagnosis. There are also reports of lymph node invasion in patients who died about 30 months after surgery, showing the poor prognostic factor associated with this complication [9].

When possible, surgical treatment is the gold standard and the definitive therapeutic approach in most cases, through wide excision of the entire area adjacent to the endometrial tissue, usually including removal of the uterus and/or its attachments [8]. Adjuvant chemotherapy and radiotherapy have been reported to improve prognosis, but there is no consensus on the protocols [9].

An early surgical approach has been shown to be an option with good results in patients without other comorbidities and in early stages, with reports of patients reporting no periodic pain and complete remission of symptoms a few months after the intervention [10, 11]. In 2017, Carriero reported a case of a patient without comorbidities, who had a lesion involving deep layers of the abdominal wall; in this case, the clinician opted for surgical removal that resulted in a large area of defect in the abdominal wall and decided to, forgo fixation with a mesh for its correction [12].

In some cases, the surgical approach is not immediately indicated. In 2020, Mao reported a case in which a patient, through physical examination, CT scan and her medical history, was shown to have an abdominal wall lesion, which was diagnosed as an endometrioma. Surgical removal of the endometrioma was considered but dismissed due to the patient’s hemodynamic instability. After 3 months, the patient returned to the emergency room with severe dysmenorrhea that was refractory to medication. It was then suggested that the patient undergo significant weight loss before surgery due to the high risk of problems related to surgical wound healing and the consequent appearance of an abdominal hernia. As a solution, the patient was referred to interventional radiology until it was possible to perform the surgery [13].

There are also reports of cases in which, despite treatment with the combination of neoadjuvant chemotherapy, tumor resection, total hysterectomy, bilateral salpingo-ooophorectomy, bilateral pelvic lymphadenectomy and adjuvant chemotherapy, disease progression was still noted [6].

Conclusion

This report highlights a case of endometriosis with invasion of the abdominal wall with metaplastic features, which is an uncommon finding in everyday medical practice. Because it is often diagnosed incidentally after histological analysis, its importance as a differential diagnosis in cases of abdominal wall masses should be emphasized.

Declarations

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors declare that they have no conflict of interest.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

References


Figure 1: Pre-operative abdominal magnetic resonance imaging scan with intravenous Gadolinium administration. The image shows multilobulated cystic formation with septations and partial hemorrhagic content located in the hypogastric abdominal wall.

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