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Case report

Incisional carcinoma of Mullerian Origin: A case report and review of literature

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ABSTRACT

Primary incisional carcinoma (PIC) is a rare, delayed complication of surgery, usually attributed to the malignant transformation of endometriosis.

We report a case of incisional carcinoma with nodal metastases in a 55-year-old woman, 18 years after cesarean section. She underwent extirpative surgery, including hysterectomy and bilateral salpingo-oophorectomy, without intraperitoneal disease identifed. Adjuvant treatment included sandwiched platinum-based chemotherapy (carboplatin and paclitaxel) and radiation. She remains disease-free 8 months after completing therapy.

We identified 46 additional reported cases. Of these, > 90% had undergone an "endometrium-exposing" surgery, most commonly cesarean section; while no cases followed adnexal-only surgery. The median time between antecedent surgery and presentation was 18 years. At presentation, tumors were often large (median 8 cm), and symptomatic with pain (63%) and/or mass (26%). Serum CA125 levels were commonly, albeit slightly, elevated (median 57U/ml (IQR 22–96, Range 6–1690)). Lymph node metastases were common (35%), with most following a vulvar-type spread pattern (inguinal first). Most patients (63%) were treated with chemotherapy +/− radiation. Approximately 50% of patients recurred promptly (median < 6 months), but long-term survival was reported following combined chemotherapy/radiation. Lymph node metastases portended a shorter disease-free interval, with 73% of cases recurring (median 5 months) despite chemotherapy-based treatment.

These data suggest that some incisional carcinomas may result from displacement of healthy endometrium followed by delayed malignant transformation. Chemotherapy-only and radiation-only treatments are attended by modest prognosis. Taken together, these data suggest there is both need and potential avenues for improved prevention, detection, and treatment of this condition.

1. Background

Incisional carcinoma is a rare, delayed surgical complication which is attended by a generally poor prognosis. Incisional carcinoma can occur after surgery for either malignant or benign indications; in the latter case the development is most commonly attributed to the malignant transformation of either endometriosis implants invasive into the surgical scar, or to endometrial tissue displaced during gynecologic or obstetric surgery which can occur in 0.03 –1.73% of cases after cesarean section (Adriaanse et al., 2013).

The criteria to diagnose endometriosis-related malignancies was proposed by Sampson in 1925 and includes (1) presence of both benign and malignant endometrial tissue in the tumor, (2) histology compatible with endometrial origin, (3) no other primary tumor sites (Sampson, 1925). In 1953, Scott added a fourth criterion to this list: (4) demonstration of benign endometriosis contiguous with the malignant tissue (Scott, 1953).

Notably, not all suspected cases meet Sampson's criteria, suggesting that even isolated incisional carcinoma may arise through variable mechanisms. When incisional carcinoma is found in the anterior abdominal wall the differential diagnosis must also include cutaneous metastasis from an ovarian, endometrial, cervical, or non-gynecologic malignancy as well as primary skin adnexal neoplasms. These differential diagnoses would have significant implications for the surgical planning and medical management of these patients.

Given the rarity of the condition, most reports have described only individual cases, making definitive comment on the etiology, evaluation, and optimal management of these patients difficult.

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The primary histologic subtypes found in endometriosis-related carcinomas are clear cell carcinoma and endometrioid, though serous, mucinous, mixed, sarcoma and adenocarcinoma not otherwise specified (NOS) have been reported (Modesitt, 2002; Stern, 2001; Bassiouny, 2019; Mihailovici, 2017). Median survival time for carcinoma arising in endometriosis is 35 months (Modesitt, 2002), and for malignant transformation of scar endometriosis is 42 months (Mihailovici, 2017). We present a case of adenocarcinoma NOS found in a cesarean section scar. We then review the relevant literature.

2. Case presentation

A 55-year-old perimenopausal female presented to the emergency department for a syncopal episode. She had noted bruising over her right lower abdomen that had worsened over the three days prior to presenting. Her medical and surgical histories were otherwise unremarkable except for hypertension and one cesarean section 18 years prior to presentation complicated by the development of a mass at the incision identified immediately post-operatively, which was felt to be a seroma, though it had not changed in size, shape, or symptomatology in the ensuing years. A CT scan in the emergency department demonstrated a 6.5 cm lobulated soft tissue mass in the right anterior abdominal wall with associated enhancing 4 and 5 cm masses of the right pelvic sidewall and an enhancing mass in the right inguinal region measuring 3.4 cm (Fig. 1). There were no associated findings in the other pelvic or abdominal organs. A PET scan confirmed high FDG -glucose uptake in all described lesions (SUV max ranged from 3.3 to 21.4), but failed to demonstrate additional lesions or a clear primary site (Fig. 2a, b, and c). Tumor markers were not elevated: Ca125 was 17.2, CEA was 2.0, and CA19-9 was 26.9; and her routine screening was up to date.

A percutaneous biopsy of the anterior-most lesion was obtained and demonstrated malignant cells positive for CK7, PAX-8, WT-1, p53 (strong and diffuse), ER and PR. Combined with morphology, these findings were consistent with a “high-grade adenocarcinoma, favor high-grade serous carcinoma of ovarian, fallopian tube, primary peritoneal origin, or endometrial serous carcinoma.” She underwent cytoreductive surgery including laparotomy, en bloc resection of anterior abdominal wall mass including the underlying fascia and a portion of the rectus muscle, resection of right inguinal, pelvic and *para*-aortic lymph nodes. Hysterectomy, bilateral salpingo-oophorectomy, and omentectomy were performed, in the absence of overt abnormalities, to exclude the gynecologic organs a primary site. Absorbable mesh was used to facilitate closure of the fascial defect. The patient recovered uneventfully from surgery.

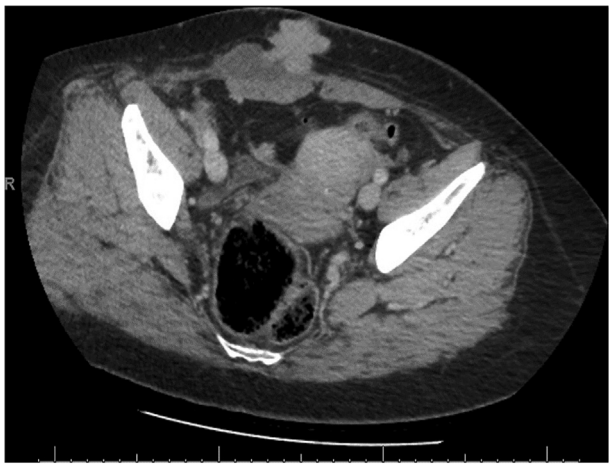

Fig. 1. CT Abdomen/Pelvis: 6.5 cm lobulated soft tissue mass in the right anterior abdominal wall.

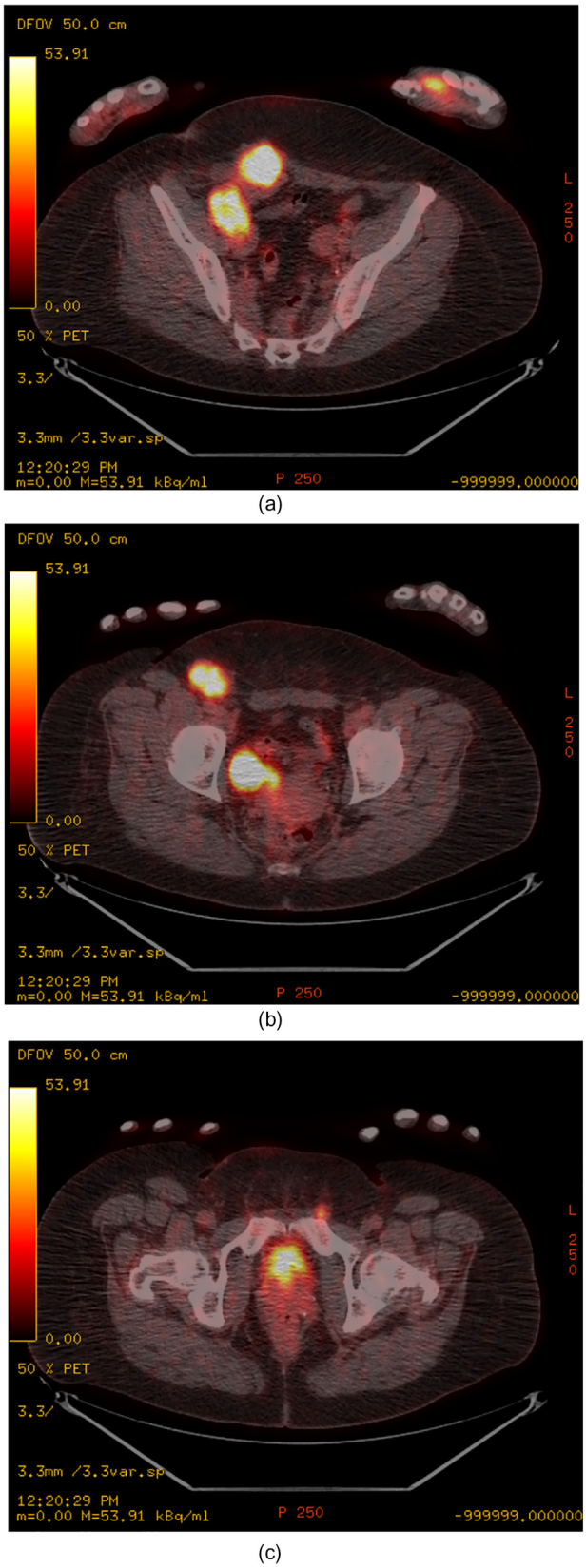

Fig. 2. PET CT (A) Mass at the location of the right iliac lymph node chain measuring 4.3x4.0 cm with SUV max of 11.0 and mass of the lower right rectus mass measuring 6.6x3.4 cm with extension into the subcutaneous fat and SUV max of 21.4. (B) 3.5x.4 cm metabolically active soft tissue mass in the right inguinal region with SUV max of 10.6. (C) Small metabolically active nodule within the distal left rectus muscle just above the pubic symphysis with SUV max of 3.3.

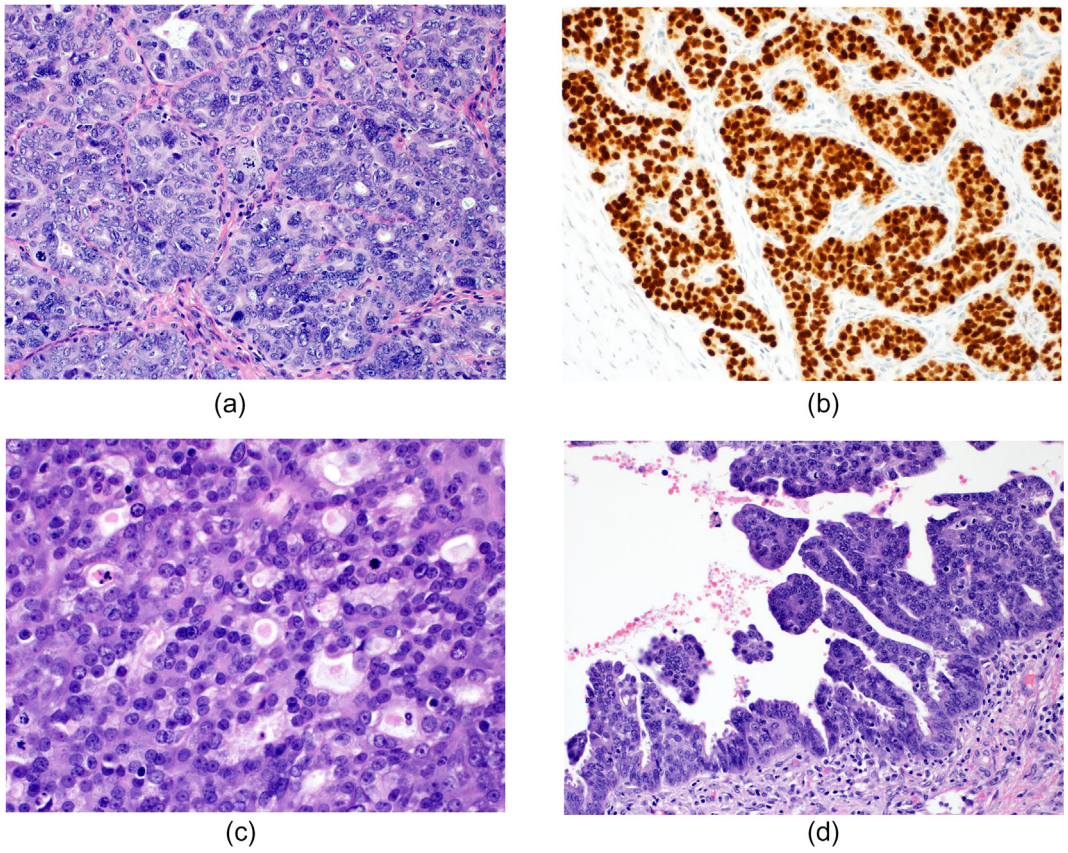

Fig. 3. Histology of a high grade carcinoma. (A) Glandular morphology suggestive of a high-grade adenocarcinoma with nuclear pleomorphism and abnormal mitoses. (B) Glandular spaces are mostly rounded with a cribriform arrangement suggestive of endometrioid adenocarcinoma. Some of the nuclei showed prominent nucleoli. (C) Focal papillary formations were also noted. (D) Immunohistochemical staining for PAX-8 was strong and diffuse.

Pathologic evaluation revealed a 6.1 cm high-grade adenocarcinoma with non-clear-cell-morphology (Fig. 3 A) that was noted to be superficial to the abdominal wall fascia and muscle but unattached to the superficial skin. It was located along and within the previous cesarean section scar. Cytoarchitecturally, the tumor was more in keeping with an endometrioid morphology (Fig. 3B) with occasional areas of papillary formation (Fig. 3C). Malignant cells were positive for PAX-8 (Fig. 3D), ER, focally for p63 and p40. Cells were negative for GATA-3, calretinin, D2-40, CK5/6, synaptophysin, and chromogranin. It had metastasized to 2 of 2 right inguinal lymph nodes, and 1 of 14 right pelvic lymph nodes. The uterus, cervix, fallopian tubes, ovaries and omentum were uninvolved by neoplasm and no endometriosis was noted in any site.

The possibility of a primary skin adnexal tumor was considered but was believed unlikely based on the negative staining for calretinin, D2-40 and CK5/6. The tumor morphology was also unsupportive of this possibility. We also excluded the possible origin from ectopic breast tissue (the caudal remnants of the milk ridge) based on the negative staining for GATA-3 with the strong and diffuse staining for PAX-8. The patient was referred for a full dermatologic examination and no suspicious skin lesions were identified.

After discussion of treatment options, the patient elected to receive both chemotherapy and radiation in a sandwiched fashion as previously described by our group (Geller, 2010). She received 3 cycles of carboplatin AUC 6 and paclitaxel 175 mg/m^2–the third cycle of chemotherapy was complicated by grade 3 neutropenia, pegfilgrastim was added after a delay of 1 week. She then underwent pelvic radiation therapy (5000 cGy in 30 fractions to the pelvis with a 1000 cGy boost to the tumor bed), which she tolerated well with no unplanned treatment breaks. She experienced Acute Toxicity Profile by CTC v4.0: Grade 2 dermatitis, grade 1 diarrhea and grade 1 fatigue. She received 3 more cycles of carboplatin (AUC 6) and paclitaxel (175 mg/m^2) with pegfilgrastim administration during each cycle.

Surveillance to date has included clinical and radiologic evaluations (CT scan of the chest, abdomen and pelvis) at the conclusion of treatment and after 3 and 6 months of surveillance. At the time of this report the patient was disease-free at 8 months from completion of therapy (13 months from presentation).

3. Discussion

Our case demonstrates that incisional carcinoma can occur without associated findings of endometriosis and in the absence of overt or microscopic disease in the gynecologic organs. It adds to the 46 English-language prior reports of incisional carcinoma of Mullerian origin (Ovid Medline keywords: carcinoma, incision, abdominal wall, gynecologic surgical procedures) (Modesitt, 2002; Stern, 2001; Bassiouny, 2019; Mihailovici, 2017; Bourdel, 2010; Yan, 2011; Ferrandina, 2016; Park et al., 1999; Archer, 2017; Razzouk, 2007; Miller et al., 1998; Harry, 2007; Achach, 2008; Alberto, 2006; Bats, 2008; DaInes, 2011; Hitti et al., 1990; Ijichi, 2014; Ishida, 2003; Jiang, 2015; Li, 2012; Leng, 2006; Madsen et al., 1980; Matsuo, 2009; Matter et al., 2003; Mert, 2012; Omranipour and Najafi, 2010; Rust, 2008; Shalin, 2012; Wei and Huang, 2017; Williams, 2009; Gucer, 1996; Markopoulos, 1996; Sawazaki, 2012; Debrosz, 2014; Dhafiri, 2016; Heller, 2014; Liu, 2014; Aust, 2015; Fargas Fabregas, 2014; Gundogdu, 2013; Ruiz et al., 2015; Taburiaux, 2015; Usta, 2014; Lengele et al., 2007). Table 1 summarizes these case reports, and Tables 2 and 3 presents the summary characteristics of the group.

Table 1
Details of Case Reports on Incisional Carcinoma of Mullerian Origin.

Publication	Presenting symptom(s)	Scar type	Histology	Surgical resection extent	Organ involvement (pathology)	Chemotherapy Regimen	Radiation regimen	Time to/location of recurrence	Death
(Bourdel, 2010)	Mass	CS	Clear cell	Mass, umbilicus, right rectus abdominus, partial pubic symphysis, bilateral external iliac LNs, uterus, cervix, fallopian tubes, ovaries	1 right iliac LN, multiple left external iliac LNs	6 cycles paclitaxel & carboplatin	45 Gy abdominal-pelvic	6 months / cervical, supraclavicular, axillary, mesenteric, inguinal, lumbar-aortic LNs	22 months
(Mihailovici, 2017)	Pain, abdominal bloating/ swelling	CS	Clear cell	Mass, uterus, cervix, fallopian tubes, ovaries	–	9 cycles platinum-based	Received, regimen unspecified	–	–
(Archer, 2017)	Mass	CS	Clear cell	Mass, rectus abdominus, uterus, cervix, fallopian tubes, ovaries	None	None	None	No recurrence at 12 months	–
(Ferrandina, 2016)	Abdominal bloating/ swelling	CS	Clear cell	Mass, rectus abdominus and fascia, uterus, fallopian tubes, ovaries, inguinal & pelvic lymph nodes	7/14 pelvic LNs, 8/11 inguinal LNs	3 cycles neoadjuvant carboplatin & paclitaxel, 3 cycles pegylated liposomal doxorubicin	None	2 months / liver	6 months
(Yan, 2011)	Pain	CS	Clear cell	Mass	Not evaluated	3 cycles, agent unspecified	None	No recurrence at 24 months	–
(Park et al., 1999)	Mass	CS	Clear cell	Mass	Not evaluated	None	50.4 Gy external beam	–	–
Shalin, 2012	Pain, ulceration	CS	Clear cell	Mass, ovarian cyst, endometrium, iliac LNs	2/4 iliac LNs	6 cycles cisplatinum based	Received, regimen unspecified	5 months	No death at 7 months
(Miller et al., 1998)	Pain	CS	Clear cell	Mass, uterus, cervix, fallopian tubes, ovaries, omentum	None	3 cycles cisplatin	Whole pelvic RT with boost to the scar	No recurrence at 5 years	–
(Li, 2012)	Pain	CS	Clear cell	Mass, uterus, cervix, fallopian tubes, ovaries	None	6 cycles paclitaxel & carboplatin		No recurrence at 8 months	–
(Bats, 2008)	Mass	CS	Clear cell, small serous component	Mass, uterus, cervix, fallopian tubes, ovaries, omentum, peritoneal biopsies	None	3 cycles neoadjuvant carboplatin & paclitaxel	None	8 months (1 LN)	–
(Alberto, 2006)	Pain	CS, hysterectomy & BSO-	Clear cell	Mass	None	6 cycles taxol & carboplatin	45 Gy	–	–
(Mert, 2012) (case 1)	Not specified	CS, BTL, Right oophorectomy	Clear cell	Mass, uterus, cervix, fallopian tubes, left ovary, omentum, left pelvic LNs	None	8 cycles neoadjuvant carboplatin & paclitaxel	None	No recurrence at 1 month	–
(Mert, 2012) (case 2)	Not specified	CS, hysterectomy	Clear cell	Mass, fallopian tubes, ovaries, omental biopsy	None	None	50.4 Gy to right abdomen	No recurrence at 31 months	–
(Williams, 2009)	Pain	CS	Clear cell	Mass, uterus, cervix, fallopian tubes, ovaries, omentum, bilateral pelvic and inguinal LNs	10/14 pelvic LNs, 17/ 17 inguinal LNs; left ovarian dermoid, cervical CIN3	4 cycles carboplatin & paclitaxel (refused 2 more cycles)	None	3 months	11 months
(Achach, 2008)	Pain	Myomectomy	Clear cell	Mass, endometrial biopsy	None	None	None	6 months	–
(Harry, 2007)	Pain	Mini-laparotomy BTL	Clear cell	Mass, peritoneal washings	None	None	10 fractions to scar	No recurrence at 18 months	–
Ishida et al. 2003	Mass	CS	Clear cell	Mass	Not evaluated	Cisplatin-based chemotherapy	None	–	2 years
(Madsen et al., 1980)	Pain, abdominal bloating/ swelling	Hysterotomy abortion	Mucinous	Mass	Not evaluated	None	5500 rads to the abdominal field, 1600 rads to the pelvic field	–	–
(Matsuo, 2009)	Pain, abdominal bloating/ swelling	Endometrioma resection	Clear cell	Mass, uterus, cervix, fallopian tubes, ovaries, omentum, pelvic LNs	None	6 cycles docetaxel & carboplatin	None	18 months	–

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Table 1 (*continued*)

Publication	Presenting symptom(s)	Scar type	Histology	Surgical resection extent	Organ involvement (pathology)	Chemotherapy Regimen	Radiation regimen	Time to/location of recurrence	Death
(Matter et al., 2003)	Pain	CS	Cystadeno-carcinoma	Mass, endometrial biopsy	None	None	None	No recurrence at 18 months	–
Wei and Huang, 2017	Pain, ulceration	CS	Clear cell	Mass	Not evaluated	None	None	–	–
Da Ines, 2011	Pain	CS	Endometrioid, serous	Mass, endometrial biopsy, left iliac LNs	2/2 left iliac LNs	6 cycles carboplatin & paclitaxel	None	No recurrence at 15 months	–
(Leng, 2006)	Pain	CS	Endometrioid, sarcoma	Mass, uterus cervix, fallopian tubes, ovaries	None	1 cycle cisplatin & ifosfamide	None	2 months	17 months
(Rust, 2008)	Pain	Abdominal hysterectomy, salpingectomy	Clear cell	Mass	Not evaluated	None	None	–	–
Omranipour and Najafi, 2010	Mass	Laparotomy for uterine perforation	Papillary serous	Mass, uterus, cervix, fallopian tubes, ovaries	None	3 cycles neoadjuvant platinum-based chemotherapy, "postoperative chemotherapy"	Received, regimen unspecified	No recurrence at 12 months	–
Ji et al. 2017	Pain	CS	Adenocarcinoma	Mass, uterus, cervix, fallopian tubes, ovaries, omentum, pelvic paraaortic and inguinal LNs	11/18 pelvic LNs, 1/9 *para*-aortic LNs, 2/5 inguinal LNs	3 cycles neoadjuvant carboplatin & paclitaxel, 3 cycles carboplatin & paclitaxel	None	Recurrence at 4 months	–
(Jiang, 2015)	Not specified	CS	Adenocarcinoma	Mass	Not evaluated	None	None	6 months	–
Razzouk et al. 2006	Mass	CS	Clear cell, endometrioid	Mass, anterior abdominal wall LN, fallopian tubes, ovaries	1/1 anterior abdominal wall LN	4 cycles carboplatin & paclitaxel	None	3 months	6 months
(Hitti et al., 1990) (Case B)	Pain	CS	Clear cell	Mass, uterus, cervix, fallopian tubes, ovaries	None; uterine adenomyosis, left ovarian follicular cyst	None	None	No recurrence at 30 months	–
Markopolous, 1996	Pain	CS	Endometrioid	Mass	Not evaluated	None	None	No recurrence at 24 months	–
(Gucer, 1996)	Pain	CS, hysterectomy	Endometrioid	Mass, ovaries	None	–	–	–	–
(Sawazaki, 2012)	Pain	CS	Clear cell	Mass, partial bladder, right rectus	Not evaluated	4/6 cycles carboplatin & paclitaxel	None	–	–
Stevens et al. 2013	Mass	CS	Clear cell & endometrioid	Mass, uterus, fallopian tubes, ovaries, pelvic LNs, omentum	None	3 cycles neoadjuvant carboplatin & paclitaxel	RT to abdominal field	–	–
(Taburiaux, 2015)	Not Specified	CS	endometrioid	–	–	Neoadjuvant carboplatin & paclitaxel, adjuvant carboplatin & paclitaxel	None	No recurrence at 17 months	–
Al (Dhafiri, 2016)	Pain	CS	Clear cell	Mass, uterus, fallopian tubes, ovaries, omentum, peritoneal samples	None	–	–	–	–
(Gundogdu, 2013)	Mass	CS x2	Clear cell	Mass, uterus, fallopian tubes, ovaries, omentum, pelvic washings	None	6 cycles carboplatin & paclitaxel	None	24 months	31 months
(Heller, 2014)	Mass	CS x3	Clear cell	Mass, left fallopian tube, left ovary, pelvic LNs	Bilateral pelvic LNs	None	None	5 months	–
(Fargas Fabregas, 2014)	Pain	CS, appendectomy	Serous	Mass, fallopian tubes, ovaries, omentum, iliac LNs, endometrial biopsy	2 right and 1 left iliac LNs	6 cycles carboplatin & paclitaxel	None	No recurrence at 48 months	–
(Liu, 2014)	Pain	CS	Clear cell	Mass, partial bladder, uterus, fallopian tubes, ovaries, omentectomy, inguinal/pelvic/*para*-aortic LNs	Bladder, 8/8 inguinal LNs, 18/21 pelvic LNs, 6/6 *para*-aortic LNs	3 cycles carboplatin & paclitaxel Traditional Chinese herbal medicine	None	10 months	12 months
Dobrosz, 2014	Pain	CS	Clear cell	Mass, endometrial biopsy	None; endometrial polyps	Dienogest	None	–	–
(Aust, 2015)	Mass	CS, LAVH	Clear cell		2/48 LNs	6 cycles carboplatin & paclitaxel	None	No recurrence at 10 months	–

(*continued on next page*)

Table 1 (*continued*)

Publication	Presenting symptom(s)	Scar type	Histology	Surgical resection extent	Organ involvement (pathology)	Chemotherapy Regimen	Radiation regimen	Time to/location of recurrence	Death
				Mass, fallopian tubes, ovaries, omentum, pelvic and *para*-aortic LNs					
(Usta, 2014)	Pain	CS, myomectomy	Endometrial stromal sarcoma	Mass, uterus, fallopian tubes, ovaries	None	–	–	–	–
(Ijichi, 2014)	Mass	CS x2	Clear cell	Mass	Not evaluated	None	None	8 months	No death at 23 months
Li JY et al. 2003	Pain	CS	Clear cell	Mass, uterus, fallopian tubes, ovaries, omentum, pelvic washings, "staging"	None	3 cycles epirubicin, cyclophosphamide, cisplatin	None	No recurrence at 14 months	–
(Ruiz et al., 2015) (Report 1)	Pain	CS	Clear cell	Mass, uterus, fallopian tubes, ovaries, partial omentum, pelvic washings	None; uterine adenomyosis, subserosal fibroids, left hydrosalpinx & salpingitis	6 cycles carboplatin & paclitaxel	–	6 months	–
(Ruiz et al., 2015) (Report 2)	Pain	CS x3, tubal ligation	Clear cell	Mass, uterus, fallopian tubes, ovaries, bilateral inguinal & internal iliac LNs, ileal & cecal nodules	1 inguinal LN, 1 internal iliac LN	6 cycles carboplatin & paclitaxel	RT to positive pubic symphysis margin	–	–

Table 2
Demographic features of reported cases.

Demographic	Median (range)
Age at diagnosis, N = 47 (years)	47 (37–60)
Interval since first surgery, N = 41 (years)	18 (15–24)
Time to recurrence, N = 16 (months)	6 (2–24)
Time to death, N = 8 (months)	14.5 (6–31)

Table 3
Characteristics of reported cases.

Characteristic	N (%)
Lymph node involvement	12/15 (LN assessment done; 80%)
Histologic type	
Clear cell	32 (69.6%)
Endometrioid	3 (6.6%)
Serous	2 (4.3%)
Mucinous	1 (2.2%)
Mixed	5 (10.9%)
Adenocarcinoma not otherwise specified	3 (6.6%)
Scar type	
Cesarean section (CS)	32 (69.6%)
Endometriosis Resection	1 (2.2%)
Hysterectomy	1 (2.2%)
Laparotomy with uterine/adnexal procedure	4 (8.7%)
CS + other procedure	8 (17.4%)
Adjuvant treatment	
Chemotherapy alone	20 (43.5%)
Radiation alone	4 (8.7%)
Chemotherapy & Radiation	8 (17.4%)
Hormone therapy	1 (2.2%)
None	10 (21.7%)
Not reported	3 (6.6%)

3.1. Origin

Primary incisional carcinoma (PIC), that is without overt alternative primary, likely develops by either the malignant transformation of established endometriosis or by the inadvertent surgical translocation of benign endometrial gland cells followed by malignant transformation. While malignant transformation of endometriosis is well described, the current case describes incisional carcinoma in the absence of associated endometriosis or prior malignancy, suggesting the translocation of benign tissue followed by delayed malignant transformation. This hypothesis is supported by the absence of associated pathology in the gynecologic organs, the long latency from antecedent surgical event (cesarean section 18 years prior), and the lateralized lymphatic spread pattern which is reminiscent of vulvar cancer rather than typical intra-abdominal metastatic spread patterns.

Supporting evidence for the translocation hypothesis in our literature review includes that over 90% of patients with PIC had previously undergone procedures with possible endometrial exposure (89% cesarean section and 4% myomectomy). By comparison only 11% had prior hysterectomy and no cases were reported to have followed adnexal surgery alone. The median time interval from potentially-translocating surgery to presentation (N = 41) was 18 years (interquartile range, IQR 15,24) with > 90% of patients having an interval of at least 10 years, which greatly exceeds the anticipated progression-free interval of a missed diagnosis of cancer at the time of primary surgery.

3.2. Presentation

Patient and demographic characteristics are listed in Table 1. The most common presenting symptoms of PIC in the literature review were pain (29/46, 63%) and mass (12/46, 26%). Ulceration of the skin was

Table 4
Primary surgical strategy of reported cases.

Primary surgical strategy	N (%) Total N = 46
Excision of primary tumor alone	14 (30.4%)
Evaluation/excision of at least one nodal basin	16 (34.8%)
Excision of additional structures*	9 (19.6%)
Excision of tumor, inguinal nodes and additional structures*	7 (15.2%)

Extent of resection not available for 1 patient and not included.
* Includes any pelvic or abdominal organs.

rare (4%).

As with our patient, most lesions are clinically palpable. Incisional mass size was reported in 45/47 cases with the median longest dimension being 8 cm (IQR 6,10). Clinical assessment of the inguinal lymph nodes was reported in 20/46 patients with 7/20 (35%) cases demonstrating overt lymphadenopathy.

Serum CA125 levels were obtained in 25/47 patients and were subtly to notably elevated in most; median 57 (IQR 22–96, Range 6–1690). CEA was reported in 10/47 cases but was within normal range in all but one case.

3.3. Evaluation and primary treatment

Clinical evaluation should be directed to defining the extent of disease and excluding an alternate primary. It should begin with clinical evaluation of the incision and the inguinal lymph nodes. Computed tomography (CT) or PET scan is appropriate to evaluate for alternate primary as well as to exclude distant/unresectable disease.

Primary complete surgical resection is recommended when feasible, including evaluation of the inguinal nodes given the lower anterior abdominal wall primarily drains into the inguinal nodes (Lengele et al., 2007). Either primary surgery or neoadjuvant chemotherapy followed by interval cytoreduction was attempted in all 47 reported cases. Resection of the primary tumor with at least one nodal basin evaluation was performed in 16/46 (34.8%) cases (table 4). Nodal metastases were identified in 13/16 (81%) of cases when performed. In cases where no nodes were removed 20/31 cases reported follow-up; there were 8 recurrences diagnosed at a median of 7 months (range 2–24 months), and 12 patients remained without evidence of disease after follow-up of 8–60 months. Taken together, these data suggest that lymph node involvement is common, even when clinically non-suspicious, and suggests that some lymph node assessment is important to staging and treatment planning.

Of the 46 total cases, 32 (69.6%) were clear cell carcinoma, three (6.6%) endometrioid, two (4.3%) serous, one (2.2%) mucinous, one (2.2%) endometrial stromal sarcoma, five mixed (1 clear/serous, 1 endometrioid/serous, 2 clear/endometrioid, 1 endometrioid/sarcoma; 10.9%), and three (6.6%) adenocarcinoma not otherwise specified (NOS).

3.4. Adjuvant treatment and prognosis

There is no current standard of care for treatment of PIC which was reflected by the variability of treatments in our review of the literature. Twelve of the 46 patients (26%) were treated with adjuvant radiation therapy; of these 6 had reported follow-up. Two of these 6 patients (33%) recurred at a median of 5.5 months, while 4 remained without evidence of disease at a median of 24.5 months (range 12–60). Thirty-four patients (74%) had no adjuvant radiation, of which follow-up data was available on 27. Of these 15 (56%) recurred at a median of 6 months (range 2–24 months); of these 7/15 (47%) died of disease at a median of 12 months (range 6–31 months). Twelve of the 27 patients (44%) without radiation were without evidence of disease at a median of 16 months (range 1–48 months).

Twenty-nine patients were treated with adjuvant chemotherapy. Follow up data was available for 23 patients; of these 13 (57%) recurred at a median of 5.5 months (range 2–24 months). Eight of 9 patients (89%) with a reported final disposition had died of their disease at a median of 14.5 months (range 6–31 months). Eighteen of the 46 patients received no adjuvant chemotherapy. Follow-up data as available on 10/18 patients; of these 4 patients experienced recurrence at a median of 6 months (range 5–8 months), while 6 patients remained without evidence of disease at a median of 21 months (range 12–31 months).

Eight patients (exclusive of the present case) were treated with both chemotherapy and radiation. Follow-up data as available on 4/8 patients; 2 recurred at 5 and 6 months respectively, 1 of whom succumbed to disease at 22 months and one who was alive with disease after 7 months. Two of the 4 patients (50%) remained without evidence of disease at 12 and 60 months respectively.

Lymphatic metastases were associated with a particularly poor prognosis. Thirteen of 16 patients (81%) who underwent lymph node dissection had inguinal or pelvic lymph node metastases. All patients with lymph node metastases were referred for adjuvant chemotherapy. Follow up data was available on 11 of these patients; 8 patients (73%) recurred at a median of 5 months (range 2–10 months) with 4/8 (50%) dying of disease at a median of 11.5 months (range 6–22 months). Three patients with nodal metastases were alive without disease at a median of 15 months (range 10–48 months).

4. Conclusions

Incisional carcinoma is a rare but serious complication of surgery. There appears to be at least two mechanisms by which this condition occurs: translocation of benign endometrial tissue, up to decades prior to malignant transformation, and malignant transformation of endometriosis. Irrespective of the origin, diagnosis appears to be delayed with most lesions being clinically palpable at diagnosis illustrating the importance of maintaining a high index of suspicion and suggesting a potential role for earlier evaluation of persistent incisional masses. Lymph node metastases are common and appear to follow a vulvar distribution suggesting evaluation of the inguinal and pelvic lymph nodes basins appears indicated, especially in light of what appears to be a significant detriment to prognosis when nodal metastases are identified. No adjuvant treatment strategy demonstrated clear superiority in review of the literature, with a majority of patients relapsing within 6 months of completing adjuvant therapy; there were however long-term survivors in each treatment strategy. Further research is clearly needed to elucidate the etiology of this condition as well as to optimize treatment.

Consents

Written cm 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.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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