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Case report

Outcome of a multimodal therapy of a recurrent adenocarcinoma arising from Caesarean section scar endometriosis—A case report

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ABSTRACT

Background: Endometriosis occurring in surgical scars is a well-described entity. Malignant transformation of endometriosis is a rare event, with most cases belonging to adenocarcinoma. The initial surgical treatment is a method of choice. Due to lack of therapeutic recommendations, adjuvant therapy and recurrence management are a great challenge for oncologists.

Aim: The aim of this paper was to present a long-term survival as the outcome of multimodal therapy in the patient with recurrent adenocarcinoma arising from Caesarean section scar endometriosis.

Case: We present the case of a woman with recurrent adenocarcinoma arising from Caesarean section scar endometriosis. The disease was first diagnosed in September 1997 at age 43. The patient underwent abdominal hysterectomy with tumour excision. Due to a local recurrence after 4 years, tumour excision with abdominal wall repair using a plastic mesh, regional lymphadenectomy, bilateral salpingo-ovariectomy and adjuvant radiotherapy for the pelvic region with local boost were performed; in addition hormontherapy with medroxyprogesterone was started. Because of a recurrent pelvic tumour, chemotherapy, further local palliative radiotherapy and brachytherapy were administered. Subsequently distant metastases in bilateral axillary lymph nodes were diagnosed and palliative radiotherapy was performed. The patient died of morbus neoplasmaticus generalisatus in September 2008. The follow-up period had been 132 months.

Conclusion: This paper is, to our knowledge, the only report in literature that presents a long-term survival as the outcome of multimodal therapy in the patient with this rare diagnosis. Further reports of new cases can help establish optimal treatment guidelines.

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1. Introduction

Endometriosis, which means the presence of endometrial tissue outside the uterus, can be seen in intra- and extraabdominal localisations. In the abdominal wall it can occur after hysterectomies, particularly after Caesarean sections.[1–4] Malignant transformation of endometriosis, especially abdominal wall endometriosis, is a rare event.[5,6] In the literature we have found only a few reports concerning this diagnosis.[7–18] We present a new case of recurrent adenocarcinoma arising from Caesarean section scar endometriosis.

2. Case report

The disease was first diagnosed in September 1997 at age 43. In 1980 the patient had delivery with caesarean section. After 17 years she developed hypogastric pain and an extensive lesion of endometriotic type with pressure on the left inguinal region. The post-caesarean scar tumour was removed and a hysterectomy without adnexa was performed with specimens from both ovaries obtained. Histopathological examination revealed papillary endometrioid adenocarcinoma in the abdominal wall tumour, with no changes in the ovary and the uterus. The adenocarcinoma focus was probably removed within normal tissue boundaries.

After 4 years (December 2001) recurrence in the left inguinal region was observed. The tumour with lymph nodes around left iliac vessels was excised and a mesh was sewn into the abdominal wall defect. The histopathological examination of the tumour revealed adenocarcinoma endometroides G2 and reactive lymph nodes. The adnexa were removed bilaterally 2 months later, the histopathologic report revealed no pathology.

In 2002 due to disease progression in the soft tissues of the pubic region, radical irradiation was instituted. The patient received 45 Gy in 25 fractions with photon beam 10 MeV for the whole pelvis and a boost of 10 Gy in 5 fractions with electron beam 14 MeV on the area of enlarged right inguinal lymph nodes. Additionally the treatment involved intramuscular Depo-Provera injections of 500 mg twice a week, which were continued for 14 months.

In 2006 right inguinal pain had its onset. The examinations performed revealed high CA-125 values; in the CT pelvic scan of the pre-pubic region a mixed-density tumour 13 cm × 9 cm causing pubic bone destruction and pressing on the urinary bladder. Nine cycles of chemotherapy of the PAC regimen (cyclophosphamide, cisplatin, doxorubicin) and one cycle of the VFP regimen (cisplatin, etoposide, 5-fluorouracil) were delivered. Because of a ischaemic stroke which occurred after last cycle, the chemotherapy was stopped. Subsequently, due to increasing hydronephrosis right nephrostomy was performed.

We saw the patient after 8 months with local bleeding from the tumour in the right pubic inguinal. Palliative irradiation of the lesion with 15 Gy dose in 5 fractions was administered, followed by intratissue radiation therapy PDR dose of 10 Gy in 10 pulses every hour, encompassing the inguinal tumour pole.

In April 2008 the patient reported with painful packets of enlarged lymph nodes – in the right axilla to 5 cm, in the left axilla – to 8 cm. Histopathological examination of the right axilla revealed cellulae carcinomatosae. Palliative irradiation of both axillary areas was instituted, with the dose of 20 Gy in 5 fractions, thus obtaining clinical improvement.

The patient died on 10 September, 2008 of generalized neoplastic disease.

3. Discussion

Endometriosis is a frequently occurring disease in women of reproductive age. It is usually located inside the pelvis and can be found in the uterus, ovaries, oviducts and peritoneum. Extrapelvic endometriosis is rare, but there are reports of endometriosis in almost all localisations like kidneys, lungs, CNS, etc.[3] In the abdominal wall endometriosis can be seen in the umbilicus or in surgical scars, particularly after Caesarean sections (0.03–0,45%)[1–4] and various surgical procedures (other hysterotomies, amniocentesis). Abdominal wall endometrioma is associated with pelvic endometriosis in 14–26% of cases.[4,19] Wide local excision, frequently associated with abdominal wall repair using a plastic mesh, is a method of choice.[2,4,20]

Malignant transformation of endometriosis is an infrequent event. It affects 0.3–1% of women suffering from endometriosis.[5,6] A case of cancer arising in ectopic endometrial tissue was first reported in 1925 in the ovary,[21] which is the most frequent location for this unusual disease, the remainder occurs in extragonadal sites.[5] The histopathologic types of malignant transformations of endometriosis belong in most cases to endometrioid carcinoma (adenocarcinoma) – 69%, followed by sarcoma (25%), clear cell carcinoma (4.5%) and others.[5]

The diagnosis of primary adenocarcinoma in extraabdominal endometriosis requires fulfillment of Sampson's criteria[21]:

1. endometriosis in close proximity to the tumour;
2. no other primary carcinoma should be identified;
3. the histology should be compatible with an endometrial origin.

In our case the first criterion was not fulfilled (the pathologic report does not mention endometriosis in close proximity to the tumour). The reason could be that the tumour may have destroyed the primary focus of endometriosis.[18]

An interesting issue is the delay between Caesarean section and diagnosis of malignant transformation of endometriosis located in the abdominal scar. The true endometriosis – a benign tissue – occurs in Caesarean section scars after 7–35 months,[19] but it can also be seen later up to 20 years.[1,4] In the contrary the malignant tissue develops after 3–39 years[7–18]; 17 years in our case. This supports the thesis that malignant transformation is a long-term process.

In the literature we have found only a few reports[10–18] and short reviews[7–9] concerning malignancies arising from Caesarean scar endometriosis. They present cases of patients suffering from clear cell carcinoma, endometrioid carcinoma (adenocarcinoma) and one case of carcinosarcoma. The onset

age was 38–60. In all cases the initial treatment was radical resection, mostly with plastic mesh for abdominal wall reconstruction. In most cases the surgical treatment also included total hysterectomy with adnexectomy or diagnostic curettage. The approach to adjuvant therapy was variable. There were cases with no adjuvant treatment, but in the most patients adjuvant therapy was administered. It consisted of whole pelvis and scar radiotherapy, chemotherapy (various protocols including: carboplatin, cyclophosphamide, paclitaxel, cisplatin, doxorubicin), hormontherapy (progesterone, medroxyprogesterone). In one case a second line chemotherapy was performed (gemcitabine, mitoxantrone). There was no report concerning recurrence.

In our case the initial radical surgical treatment was complicated by a local and pelvic recurrence after 4 years. It demanded further treatment, primarily surgical followed by radiotherapy, hormontherapy and chemotherapy.

In the reported cases the follow-up periods were short (2–60 months) due to early deaths from disease (6–20 months) or no evidence of disease. In our case the follow-up period had been 132 months.

4. Conclusions

This paper is, to our knowledge, the only report in literature which presents a long-term survival as the outcome of multimodal therapy in the patient with this rare diagnosis. As the number of Caesarean sections increases, further reports of new cases are necessary to establish optimal treatment guidelines.

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