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Case Report

Neoadjuvant bleomycin, etoposide, and cisplatin in adult neuroblastoma arising from the broad ligament of the uterus

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Abstract

Background. A case of primary neuroblastoma arising from the broad ligament with excellent response to neoadjuvant bleomycin, etoposide, and cisplatin (BEP) is reported.

Case. A 48-year-old woman, G0, who presented with acute renal failure, an enlarged pelvic mass, and abdominal pain was diagnosed with adult neuroblastoma arising from the broad ligament of the uterus. She received three cycles of neoadjuvant therapy consisting of bleomycin, etoposide, and cisplatin (BEP) given every 3 weeks and had an excellent initial response. She then underwent total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy, and appendectomy, with pathologic analysis revealing small residual disease on the broad ligament of the uterus and omentum. The patient died of recurrent disease 20 months after her initial diagnosis.

Conclusions. The clinical management of cancer in the broad ligament of the uterus must be tailored to the pathologic diagnosis. Although our patient had an excellent initial response to BEP, further study is needed to identify a treatment that can reduce recurrences and improve clinical outcomes.

Keywords: Neuroblastoma; Neoadjuvant chemotherapy; BEP

Introduction

Neuroblastoma is a type of primary neuroectodermal tumor that generally originates in the adrenal glands, abdomen, chest, neck, or pelvis in children younger than 10 years [1]. These tumors are rare in adults. Rarer still are primary neuroblastomas in the female reproductive tract. We have been able to identify only three cases of primary ovarian neuroblastoma and one case of primary peritoneal neuroblastoma in the literature [2–5]. We describe here a case of adult neuroblastoma of the broad ligament of the uterus in a postmenopausal woman who showed an excellent initial response to treatment with neoadjuvant bleomycin, etoposide, and cisplatin (BEP).

Case report

A 48-year-old woman, G0, was seen by her gynecologist in March 2005 for left lower quadrant pain of approximately 1 year's duration. Her surgical history was unremarkable, and her medical history was notable for hypertension. A physical examination revealed a mass fixed to the left pelvic sidewall. The mass was irregular and posterior to the cervix, and the cervix itself was firm. Pelvic ultrasonography revealed a 5.7 × 4.0 cm mass in the cul-de-sac, which was confirmed by subsequent computed tomography (CT).

The following month, the patient underwent an examination under anesthesia, diagnostic laparoscopy, biopsy of the vaginal apex, and abdominopelvic peritoneal biopsies at an outside institution. The diagnostic laparoscopy revealed omental caking with extensive peritoneal disease on the surface of the bladder peritoneum, thickening and retraction of the peritoneum along the pelvic sidewalls, sigmoid colon adhesions to the left pelvic

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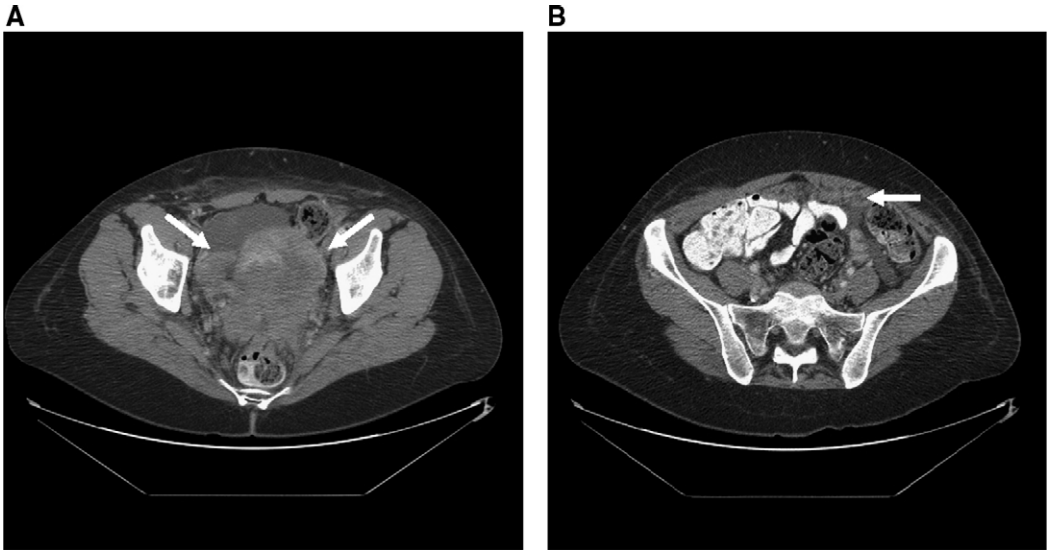

Fig. 1. The initial computed tomographic scan obtained before treatment revealed a heterogeneous pelvic mass involving the uterus and left adnexa (A) and omental caking (B).

sidewall and to the posterior aspect of the uterus, obvious tumor invasion into the uterus, and a cystic mass between the uterus and the sigmoid colon. Pathologic findings from biopsies performed at an outside hospital showed small cell neuroendocrine carcinoma.

In late April 2005, the patient was admitted to The University of Texas M. D. Anderson Cancer Center emergency department with nausea, bilateral lower extremity swelling, and malaise. An abdominal examination revealed a mass at two trocar sites in the right lower quadrant, suggestive of advanced disease. A pelvic examination revealed normal external genitalia, but a bimanual rectovaginal examination confirmed a large, thick mass that extended to the left pelvic sidewall and possibly to the right lower quadrant. The large, fixed mass extended posteriorly and involved the upper aspect of the rectovaginal septum.

Laboratory findings revealed elevated levels of serum urea nitrogen (48 mg/dl) and creatinine (7.9 mg/dl). Tests for tumor markers showed a serum CA125 level of 16.4 U/ml and a lactic dehydrogenase level of 1801 IU/ml. Levels of alpha fetoprotein and beta human chorionic gonadotropin were within normal limits. Ultrasonographic images of the kidneys showed right hydronephrosis and poor parenchyma on the left side with multiple cysts. A right percutaneous nephrostomy was placed, and the patient's renal function normalized.

On initial admission, magnetic resonance imaging was used to evaluate the pelvis because of the patient's poor renal function and revealed left para-aortic adenopathy, a mass in the cul-de-sac extending along the superior aspect of the uterus, an enlarged multicystic left kidney, and unremarkable adrenal glands. Subsequent CT revealed a metastatic tumor involving the omentum,

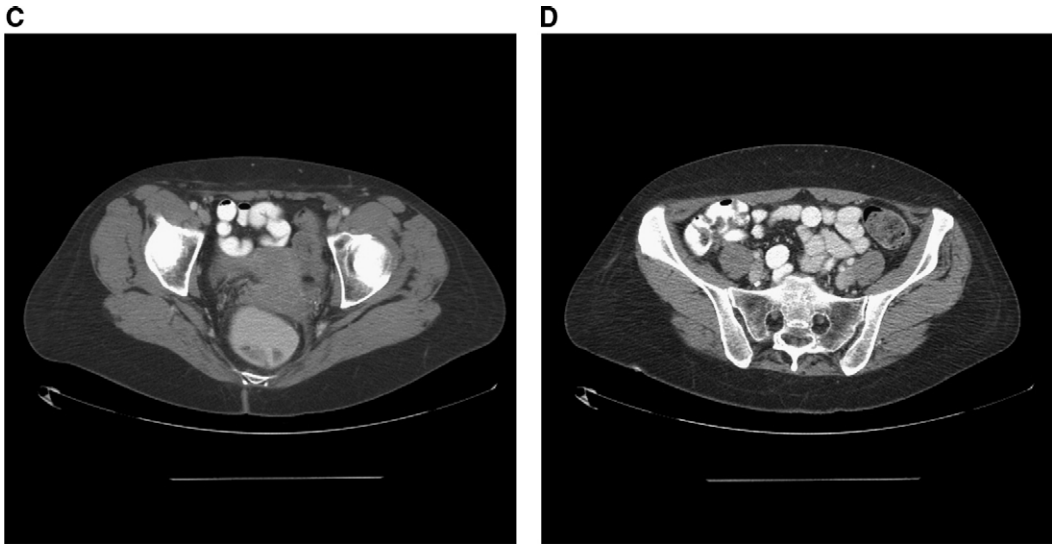

Fig. 2. After neoadjuvant therapy, a computed tomographic scan shows that the previously noted pelvic mass had significantly decreased in size (C) and the omental disease was no longer visible (D).