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A Rare Case of Cervical Rhabdomyosarcoma in Adults

Yetişkinlerde Ender Görülen Servikal Rabdomiyosarkom Vakası

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A Rare Case of Cervical Rhabdomyosarcoma in Adults

ABSTRACT

Cervical embryonal rhabdomyosarcoma is a rare mesenchymal solid tumor that accounts for less than 1% of all cervical neoplasms in the female genital system. There is no standardized protocol for the treatment of cervical embryonal rhabdomyosarcoma, and the literature primarily consists of case reports and series. Treatment options described in the literature range from conservative, fertility-preserving approaches to radical surgery in combination with chemotherapy and radiotherapy. A combined approach involving surgery, chemotherapy, and radiotherapy is believed to yield better outcomes for the disease. In this article, we present a case of cervical embryonal rhabdomyosarcoma in a 45-year-old female patient presenting with a polypoid cervical mass. Histopathological and immunohistochemical examination confirmed that the localized mass in the cervix was of cervical origin and consistent with embryonal rhabdomyosarcoma. Following total abdominal hysterectomy, the patient was administered a combination treatment of adjuvant chemoradiotherapy, and she has remained disease-free for a period of 23 months.

Keywords: Cervical Cancer, Mesenchymal tumor, Rhabdomyosarcoma.

ÖZET

Servikal embriyonel rabdomyosarkom, kadın genital sistemindeki tüm servikal neoplazmların %1'inden azını oluşturan nadir görülen mezenkimal kaynaklı solid bir tümördür. Bu tümörün tedavisine yönelik standartlaştırılmış bir protokol bulunmamaktadır ve mevcut literatür büyük ölçüde olgu sunumları ve olgu serilerinden oluşmaktadır. Literatürde tanımlanan tedavi seçenekleri, fertilite koruyucu konservatif yaklaşımlardan, kemoterapi ve radyoterapi ile kombine edilen radikal cerrahiye kadar geniş bir yelpazeye yayılmaktadır. Cerrahi, kemoterapi ve radyoterapiyi içeren kombine tedavi yaklaşımının hastalığın seyrinde daha iyi sonuçlar sağladığı düşünülmektedir. Bu yazıda, servikste polipoid bir kitle ile başvuran 45 yaşında kadın hastada görülen bir servikal embriyonel rabdomyosarkom olgusunu sunmaktayız. Yapılan histopatolojik ve immünohistokimyasal incelemeler, servikste lokalize olan kitlenin servikal kökenli olduğunu ve embriyonel rabdomyosarkom ile uyumlu olduğunu doğrulamıştır. Hastaya total abdominal histerektomi uygulanmasının ardından adjuvan kemoradyoterapi kombinasyon tedavisi uygulanmış, tedavinin ardından hasta 23 aydır hastalıksız olarak izlenmektedir.

Anahtar Sözcükler: Mezenkimal tümör, Rabdomyosarkom, Serviks Kanseri.

Introduction

Rhabdomyosarcomas are primitive mesenchymal soft tissue sarcomas that are predominantly rare in adults and mainly seen in the prepubertal age group. Due to the frequent occurrence of rhabdomyosarcomas in pediatric age groups, there is no standardized treatment approach for adult patients. Treatment selection in adults often relies on the risk groups and treatment protocols established for the pediatric population.

This mesenchymal tumor originates from immature striated muscle myocytes and can arise in various locations such as the head, neck, lymph nodes, extremities, retroperitoneum, and any part of the body. The genitourinary system is the second most commonly affected region after the head and neck. Embryonal RMS typically affects the vagina or bladder in infants and the uterus and cervix in reproductive-aged females (1). In adults, the prognosis is generally poor due to the rarity of the disease and the absence of a standardized treatment protocol (2,3).

Embryonal rhabdomyosarcoma of the uterine cervix typically manifests with symptoms such as irregular vaginal bleeding, postcoital bleeding, and a polypoid cervical mass on vaginal examination (4). Embryonal rhabdomyosarcoma of the cervix has been shown to respond positively to a combination of surgery, radiotherapy and chemotherapy. In a single-centre experience of 20 patients over 19 years of age diagnosed with RMS, the use of concurrent chemoradiotherapy and extended chemotherapy was found to improve disease-free survival (DFS) and overall survival (OS) outcomes (5). The 5-year overall survival rate for patients diagnosed with rhabdomyosarcoma of the cervix is estimated to be approximately 82% (6). Preserving fertility and sexual dysfunction are important challenges that influence treatment selection. In cases of localized rhabdomyosarcoma of the cervix, alternative options to radical hysterectomy include polypectomy and cervical conization, which can be applied. In selected cases, radical trachelectomy has been reported to preserve the patient's fertility potential while ensuring local control of the disease (7,8).

In the adult population, cases of RMS frequently present as case reports. Treatment studies in RMS have been conducted with consideration for risk groups, stages, and anatomical localization.

Case Presentation

A 45-year-old female patient was admitted to the gynecology clinic, reporting an increase in vaginal bleeding following sexual intercourse for a period of three months. During the vaginal examination of the patient, two polypoid lesions measuring 2.5x1.5 cm and 1x1.5 cm were observed at the 6-9 and 9-12 o'clock positions of the cervix, respectively. Subsequently, polypectomy was performed on the patient under local anesthesia.

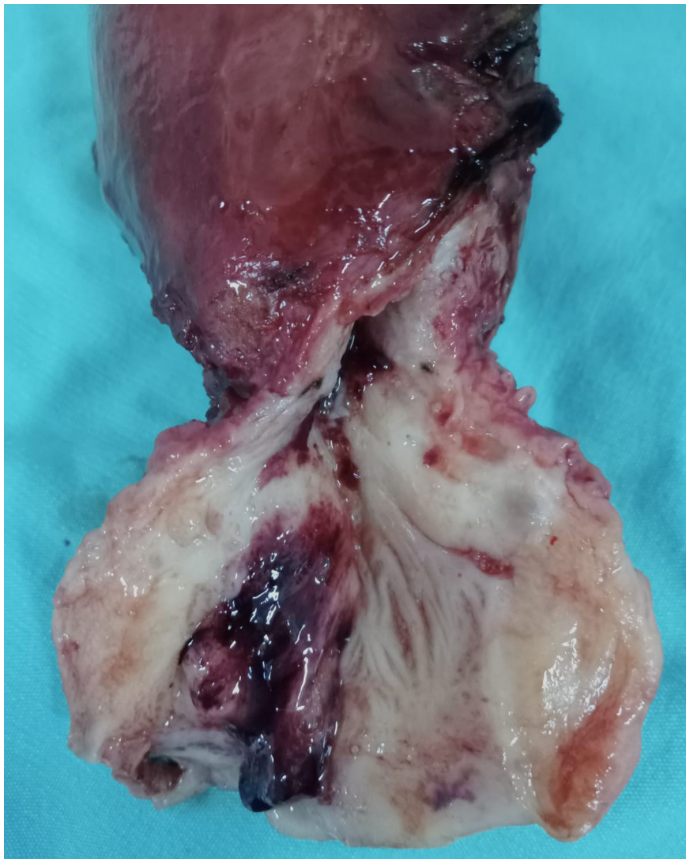

Figure I. Hysterectomy Material, Tumor in the Endocervical Canal

Tissues exhibiting a polypoid morphology were subjected to pathological evaluation based on both histopathological findings and immunohistochemical staining. The predominant interpretation was consistent with embryonal rhabdomyosarcoma. Subsequent magnetic resonance imaging (MRI) revealed that the anterior wall of the cervix exhibited mild heterogeneous signal characteristics without evidence of a distinct mass lesion. Additionally, the distal vaginal walls appeared thickened, and no significant diffusion restriction was noted. Positron

emission tomography-computed tomography (PET-CT) demonstrated mild physiological FDG uptake in the uterine cavity. Symmetrical, centimetric-sized follicular cysts were observed in both adnexal regions without increased FDG avidity. The distribution of F-18 FDG uptake across the scanned body regions was within physiological limits, and no findings suggestive of primary malignancy or distant metastasis were detected.

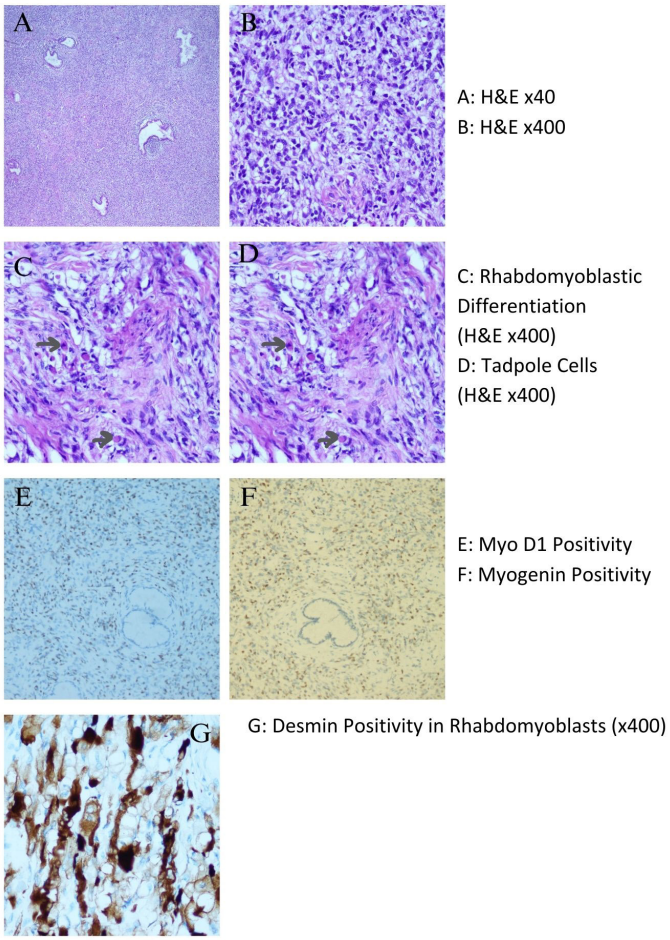

Figure II. Immunohistochemical and Pathological Examination of the Case

A. Primitive mesenchymal cells are observed in the stroma between the endocervical glands. Tumor cells are concentrated around the glands. (H&E, ×40)

B. The tumor cells have spindle-shaped, oval, and occasionally stellate morphology with narrow cytoplasm; nucleoli are not prominent. (H&E, ×400)

C. Tumor cells show abundant eosinophilic cytoplasm and eccentrically located nuclei, indicating rhabdomyoblastic differentiation. (H&E, ×400)

D. Cytoplasmic eosinophilic striations (tadpole-like cells) are observed. (H&E, ×400)

E. MyoD1 positivity in tumor cells. (×200)

F. Myogenin positivity in tumor cells. (×200)

G. Desmin positivity in rhabdomyoblasts. (×400)

In light of the prevailing pathological diagnosis and findings, the patient underwent evaluation by a multidisciplinary gynecologic oncology council. Given the patient's age, a decision was made to perform a hysterectomy. The patient underwent a laparotomic hysterectomy. When the uterus was opened along the endocervical canal, a tumor measuring 3.5x1.5x1 cm was observed on the right lateral wall of the cervix, filling the endocervical canal and displaying a polypoid appearance with bleeding (Figure I). Histopathological examination of the tumor samples revealed an infiltrating tumor starting from beneath the surface epithelium and diffusely infiltrating the stroma, with a concentration around the endocervical glands (Figure IIA). The tumor consisted of spindle-oval-shaped cells with narrow cytoplasm and mild pleomorphism (Figure IIB). Additionally, cells with broad eosinophilic cytoplasm and eccentric nuclei (Figure IIC), as well as cells showing cytoplasmic striations (Figure IID), were observed. Immunohistochemical analysis showed that the tumor cells were negative for EMA and AE1/AE3, while they stained positively for vimentin, myo-D1 (Figure IIE), myogenin (Figure IIF), desmin (Figure IIG). Based on these findings, a diagnosis of botryoid-type rhabdomyosarcoma was made. The tumor had invaded two-thirds of the cervical wall, with a depth of invasion of 0.6 cm and a distance from the parametrium of 0.6 cm. Lymphovascular invasion was detected.

No neoplasm was observed in the endometrium or the right and left fallopian tubes. In accordance with the staging criteria established by the Intergroup Rhabdomyosarcoma Study Group (IRSG), the patient was designated as Group I and Stage I. After a period of convalescence devoid of complications, the patient was referred to the relevant departments for the administration of chemotherapy and radiotherapy. Prior to discharge, the patient was provided with comprehensive information, and informed consent was obtained in accordance with standard practice. The patient was classified as a low-risk case based on clinical and pathological findings. Since no preoperative treatment was administered, adjuvant therapy was initiated following surgery. The patient received four cycles of VAC chemotherapy (vincristine, actinomycin D, and cyclophosphamide). This was followed by CT-

based high-dose-rate (HDR) brachytherapy targeting the vaginal cuff, delivered as 300 cGy twice daily over seven fractions, for a total dose of 2100 cGy. During radiotherapy, the patient also received two additional cycles of concurrent chemotherapy. The treatment was completed in March 2023, and the patient has remained disease-free for 23 months.

Discussion

Uterine cervical embryonal rhabdomyosarcoma accounts for approximately 0.4-1.0% of cervical cancers (9). Epidemiological data indicate that up to 90% of cases occur in women under the age of 25, and approximately 70% occur in children under the age of 12 (10). Perimenopausal women are less affected, but the prognosis in this age group is worse. RMS cases typically occur sporadically, and no specific risk factors have been identified. Among the histological subtypes that affect the female genital system, embryonal rhabdomyosarcomas are the most common, comprising 60% of all cases (10).

MRI is the gold standard for determining the location and invasion of the tumor, and it is useful in demonstrating the spread of the tumor to adjacent structures. The IRSG has proposed a staging system based on tumor size, involvement of the primary site, local invasion, lymph node involvement, and distant metastasis. This staging system helps in determining the stage of the tumor (10). There is no consensus on the management protocol for embryonal rhabdomyosarcoma. However, it has been observed that a multimodal approach to treatment improves patient outcomes. The multimodal approach consists of a combination of surgery, systemic chemotherapy, and radiation therapy. Given that the affected patients are predominantly young, preserving fertility and addressing postoperative sexual dysfunction in cases of surgical menopause pose significant challenges that influence treatment selection (7).

Localized rhabdomyosarcomas in the cervix can be treated with adjuvant chemotherapy along with fertility-preserving procedures such as polypectomy and simple or radical trachelectomy (11). In cases of isolated cervical and uterine involvement, simple total hysterectomy that preserves the ovaries may be recommended (4). Radical surgery has been found to not provide survival benefits in patients with superficial lesions and local recurrence. Lymphadenectomy is not routinely recommended for all patient groups, but it may be considered for those with high-risk clinical features (12). Considering the patient's age and the preoperative imaging showing a mildly heterogeneous signal in the anterior wall of the cervix without any significant mass lesion, a simple hysterectomy was performed on the patient.

In conclusion, the treatment of cervical rhabdomyosarcoma differs from other cervical tumours, especially in terms of the radicality of surgical and chemotherapy regimens, making accurate diagnosis crucial for a good prognosis. The ideal treatment for these tumors has not yet been established. However, based on current knowledge, a combined treatment approach including surgery, chemotherapy, and radiotherapy has been shown to yield favorable outcomes for cervical rhabdomyosarcoma.

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