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A Case of Recurrent Ovarıan Adult Granulosa Cell Tumor: Ureter Resectıon and Ureteroneocystostomy

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ABSTRACT

Ovarian adult granulosa cell tumors (AGCTs) constitute 2-5% of all ovarian cancers. Long-term patient follow-up is important because it tended to late recurrence. Complimentary staging surgery was performed on the patient who was diagnosed with AGCTs after oophorectomy was performed due to an ovarian cyst at the age of 42 in 2014 at the external center. Adjuvant chemotherapy BEP (Bleomycin, Etoposide, Cisplatin) was given to the patient. The patient, who was operated on for pelvic recurrence after 36 months at an external center, was reported as a recurrence of the postoperative pathology of AGCTs and then received salvage chemotherapy (Carboplatin-Paclitaxel). In the follow-up after recurrence surgery, a few masses of 85x65 mm, solid, multicystic, enhancing in the pelvis were identified in the computed tomography at 36 months. Bilateral ureters were reported as dilated with mass compression. Thereupon, an operation decision was made for the patient. Preoperative tumor markers were normal. In the intraoperative observation, an approximately 10 cm hard fixed tumoral mass extending from the left paracolpium surrounding the ureter to the levator anterior, densely attached to the bladder and invading the obliterated umbilical artery was observed. This mass was dissected from the levator muscle with the bladder, the entrance of the left ureter into the bladder was closed and the ureter was cut, and the mass was removed. The right ureter was observed by entering the right retroperitoneum and it was found to be intact. A double-J catheter was placed in the left ureter. The bladder dome was opened and the bilateral ureter orifice was observed. Ureteroneocystostomy was performed by anastomosing the left ureter to the dome of the bladder. No residual tumor was left after surgery. The patient, who had no problem in the follow-up, was discharged on the 7th postoperative day. Pathology of the patient Upon the arrival of AGCTs, medical oncology and radiation oncology were consulted for salvage chemotherapy and pelvic radiotherapy.Multifocal disease and incomplete resection of recurrent disease in AGCT recurrence are associated with reduced overall survival. In the recurrence of AGCTs, it should be aimed not to leave visible tumors in salvage cytoreduction.

Keywords: adult granulosa cell tumor; ureter resection ; ureteroneocystostomy

INTRODUCTION

Ovarian adult granulosa cell tumors (AGCTs) constitute 2-5% of all ovarian cancers and the majority of tumors originating from sex-cord stromal cells. They are mostly hormonally active, rare neoplasms.[1] These tumors, which are generally diagnosed at an early stage, are known to have good prognosis and high survival rates. The disease is characterized by slow growth, local spread, and late recurrences years after initial diagnosis.[2] 10% of cases present with metastatic disease. The most common metastases are in the liver, lung and bone, while the most common recurrence is in the peritoneal cavity. Management of AGCTs recurrence is combined chemotherapy and, if possible, debulking surgery. The most preferred combination in chemotherapy is the BEP (Bleomycin, Etoposide, Cisplatin) regimen. Long-term patient follow-up is very important because of its susceptibility to late recurrence.

CASE REPORT

It is understood from the patient history that the patient, who was diagnosed with AGCTs after oophorectomy for ovarian cyst at the age of 42 in 2014, underwent total abdominal hysterectomy and unilateral salpingo-oophorectomy and complementary surgery with pelvic and paraaortic lymph node dissection, and received a combination of BEP as adjuvant chemotherapy at the outer center. In the follow-ups in the outer center, the patient was operated again due to pelvic recurrence 36 months after the first surgery. The patient, whose postoperative pathology was reported

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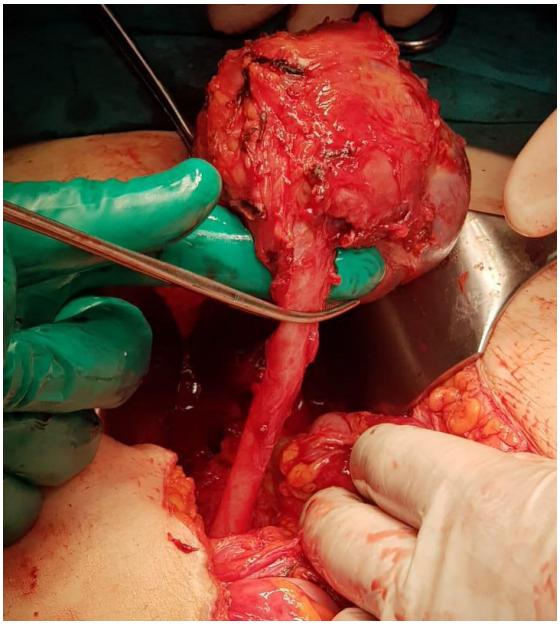
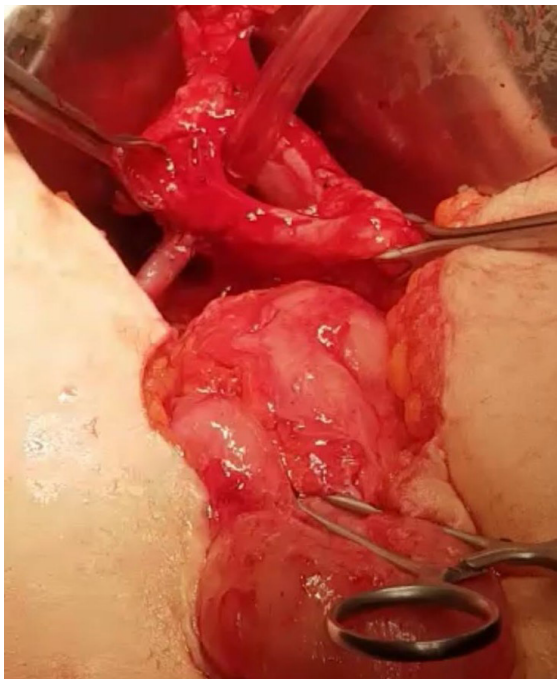
as AGCTs recurrence, was given postoperative salvage chemotherapy (Carboplatin-Paclitaxel). In a post-recurrence follow-up, lesion areas containing cystic and solid areas of 61x47 mm and 66x43 mm in size were identified in the left lower quadrant, close to the left adnexal area, in the transabdominal ultrasonography in the 36th month. After the diagnosis of the recurrence, the patient, who was diagnosed with COVID-19 and was treated, developed deterioration in thyroid function tests and tachycardia after COVID-19. The patient applied to us again after the tachycardia was treated and the thyroid function tests improved. Computed tomography performed in our hospital in the 38th month following the first recurrence revealed that the mass in the pelvic region had progressed compared to the previous technique. In the final examination, several multicystic, contrast-enhancing mass lesions in the pelvis, the largest of which was 85x65 mm in size, with solid components were identified. The mass on the left lateral of the bladder and the intermediate fat plane were removed. Bilateral ureters are dilated due to mass compressions. Therefore, the decision was made to operate on the patient. Preoperative tumor markers were; CA125: 7 U/mL, CA15.3: 19.1 U/mL, CA19.9: 47.76 U/mL, CEA: 0.79 ng/mL, AFP: 1.90 µg/L. In the intraoperative observation, a hard fixed tumoral structure of approximately 10 cm in size, which invaded and engulfed the obliterated umbilical artery with a dense adhesion to the bladder extending from the left paracolpium to the levator ani, completely surrounding the ureter on the left, was determined (Figure 1).



The obliterated umbilical artery was cut and ligated. The tumor was dissected from the bladder and levator muscle. The entrance of the left ureter to the bladder was closed. The left ureter was cut from the proximal part of the mass and the mass was totally excised. The right ureter was observed by entering the right retroperitoneum and it was determined to be intact. A double j stent was placed in the left ureter. The bladder dome was opened and both ureteral orifices were observed. It was observed that there was flow from the right ureteral orifice. The left ureter was anastomosed to the bladder dome and ureteroneocystostomy was performed (Figure 2). No residual tumor was left after surgery. In the direct urinary system radiograph taken on the first postoperative day, the double J stent was observed



in place. The patient, who had no problems in the postoperative follow-up, was discharged with a catheter on the 7th postoperative day. On the postoperative 23rd day, the patient's residual urine amount was 50cc, so the patient's catheter was removed.

After the pathology result was AGCTs, the patient was evaluated in the tumor council, and it was decided that the patient was to receive 4 cures of salvage BEP treatment and then pelvic radiotherapy by radiation oncology.

DISCUSSION

AGCTs are rare and a slowly progressing ovarian tumors. Although AGCTs have a better prognosis compared to epithelial tumors, recurrences develop

in 20% to 30% of patients, most of them late.[3] While the overall survival in stage I varies between 75% and 95%, it is between 22% and 50% in stage III and stage IV.

Surgery is the most important step in the treatment of AGCTs.[4] Mangili et al. reported that the 5-year survival rate of 97% decreased to 66.8% after 20 years of follow-up.[5] The development of late recurrence in this disease must be remembered, and recurrences that develop 30 years after diagnosis are presented.[6, 7] Recurrences usually develop multifocally in the abdomen. The most common sites for extrapelvic recurrences are the liver, small intestine, and spleen. In AGCTs recurrences, maximal debulking significantly increases survival and is significantly better in those who do not have a residual tumor after surgery.[8] Therefore, it is important to treat patients with recurrent multifocal disease with radical surgery in experienced centers to ensure complete survival and avoid residual disease.[9, 10] Crew et al. described the effect of cytoreductive surgery, in which complete resection of all lesions is performed, on survival in the presence of diffuse abdominopelvic tumoral lesions in recurrent AGCTs.[11] In the study by Dan Zhao et al, an analysis of 34 patients after surgery for recurrence showed that those with progression-free survival (PFS) <61 months had a 3.5 times higher risk of a second recurrence than those ≥61 months. In addition, age at the time of recurrence and post-recurrence treatment approaches were independent risk factors for post-recurrence survival. Those aged >50 years at the time of recurrence had a 3.3 times higher risk of death compared to those aged <50 years. Those who received only chemotherapy after recurrence had a 13.4 times higher risk of death than those who received surgery followed by chemotherapy.[12]

In conclusion, multifocal disease and incomplete resection of recurrent disease are associated with decreased overall survival in adult granulosa cell tumor recurrence. For this reason, salvage cytoreduction in the recurrence of AGCTs should be aimed at not leaving any visible tumor.

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