Two Cases of Testicular Adrenal Rest Tumor (TART)

Hatice Özşik1, Banu Şarer Yürekli1, Ilgın Yıldırım Şimşir1, Ilker Altın1, Utku Soyaltın1, Ezgi Güler2, Hüseyin Onay3, Banu Sarsik Kumbaraci4, Füsun Såylılı1

1Ege University Faculty of Medicine, Department of Endocrinology and Metabolism Diseases, İzmir, Turkey
2Ege University Faculty of Medicine, Department of Radiology, İzmir, Turkey
3Ege University Faculty of Medicine, Department of Medical Genetics, İzmir, Turkey
4Ege University Faculty of Medicine, Department of Pathology, İzmir, Turkey

Testicular adrenal rest tumor (TART) is a benign tumor which is seen in male patients who have congenital adrenal hyperplasia (CAH).

Case 1: A 24-year-old male was diagnosed with CAH. He took hydrocortisone treatment up to 18 years of age however discontinued it thereafter. Testicular mass was detected and right radical orchiectomy was performed; testicular tumor of adenogenital syndrome was determined. On physical examination, height was 152 cm, weight was 47 kg, BMI was 20.3 kg/m². He had short fingers. In laboratory examination, 17-OHP 122 ng/mL (0.6-3.3), adrenocorticotropic hormone 118 pg/mL (< 46), free testosterone 34.4 pg/mL (57-178), DHEAS 578.4 µg/dL (85-690), and cortisol 5.2 µg/dL were detected. In CAH mutation screening, mutations in an allele (heterozygous) I2 splice and in the other allele (heterozygous) L307 frameshift were detected. Dexamethasone 0.75 mg once daily was initiated.

Case 2: A 38-year-old male has followed with diagnosis of Addison disease for 35 years. Right testicular tumor was defined as Leydig cell tumor in 2010. In scrotal USG, small multifocal lesions were detected and testicular biopsy was done which revealed testicular tumor of adenogenital syndrome. He took 30 mg hydrocortisone once daily. On physical examination, height was 174 cm, weight 104 kg, and BMI was 34.4 kg/m². In laboratory examination, 17-OHP 157 ng/mL (0.6-3.3), adrenocorticotropic hormone 194 pg/mL (< 46), free testosterone 31.6 pg/mL (57-178), DHEAS 123.5 µg/dL (85-690), and cortisol 2.14 µg/dL were detected. TARTs are usually seen bilaterally (83%) and histopathologically it is difficult to differentiate them from Leydig cell tumor. It should be kept in mind that testicular USG is of significant importance in early diagnosis of TART.