Purpose: Testicular Adrenal Rest Tumor (TART) is a condition that is seen in men with congenital adrenal hyperplasia and particularly in uncontrolled cases. Its pathogenesis is not certain, but TART is believed to be derived from ectopic adrenal cortex remnants in the testis or from reprogrammed Leydig stem cells, that differentiate and grow under the effect of chronically elevated ACTH. Pressure in the testis increases due to the mass effect, and it prevents sperm exit. It presents with bilateral testicular mass and infertility.

Case Report: Bilateral testicular mass was detected in a 28-year-old male patient on scrotal ultrasonography who had referred for infertility 1.5 months earlier. Bilateral orchiectomy was suggested to the patient with a presumed testicular tumor. There were hypervascularized solid lesions in right (18 mm), and left testes (15 mm) in his scrotal USG. In MRI, bilateral testicular masses were detected (25x33 mm in right testicle and 16x22mm in the left). The patient who did not accept the operation was directed to our endocrine polyclinic with the complaints of testicular mass and azoospermia. His laboratory test results were as following: LH: 0.1mIU/ml (1.7-8.6), FSH: 0.1 mIU/ml (1.5-12.4), T, Testosterone: 6.7 ng/ml (2.18-9), Prolaktin: 18 ng/ml (4.04-15.2), TSH: 3.19 (0.270-4.2), T4: 16.7 pmol/L (12-22), Kortizol: 3.5 µg/dL, ACTH: 153 pg/mL, 17 Hidroksiprogesteron: 48 ng/mL, BHCG:0.1 mIU/ml (<5.3), AFP: 0.55 IU/ml (0.5-5.5), Synacten Test was conducted, Kortizol 30.dk was 4.9, 60.dk 4.9, 90.dk 4.9, 120.dk 5.4, 17-hydroxyprogesterone level at 30.dk was 39.7, 60.dk 36.5, 90.dk 28.8, 120.dk 25. No sperm cells were detected in the spermiogram. Congenital Adrenal Hyperplasia and Testicular Adrenal Rest tumor were diagnosed in this patient. Prednisolone 5 mg started. Genetic analysis revealed mutation in 21 hydroxylase gene. In his follow-up, 7.5 million sperm were detected in the spermiogram and bilateral masses with slight shrinkage.

Conclusions: Adults with congenital adrenal hyperplasia (CAH) may refer with infertility and bilateral testicular adrenal mass. The patient should be well assessed; surgery should not be performed if not necessary. Fertility is possible by starting corticosteroid therapy and adjusting the optimal dose.