Case Report

Hormonal Mimics: Early Detection of Bilateral Testicular Adrenal Rest Tumors in Congenital Adrenal Hyperplasia. A Case report

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ABSTRACT

Testicular adrenal rest tumors (TART) are benign ectopic adrenal tissue proliferations commonly associated with congenital adrenal hyperplasia (CAH). Early recognition is crucial to prevent misdiagnosis as a testicular neoplasm and avoid unnecessary surgical intervention. We report a case of an 11-year-old male who is a follow-up case of CAH with salt wasting and GnRH-dependent precocious puberty, in whom scrotal ultrasonography and magnetic resonance imaging demonstrated characteristic features of bilateral TART.

Keywords: Testicular adrenal rest tumours, congenital adrenal hyperplasia, Testicular neoplasm

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INTRODUCTION

Congenital adrenal hyperplasia (CAH) is an autosomal recessive disorder of steroidogenesis, most commonly due to 21-hydroxylase deficiency with androgen excess[1].

Testicular adrenal rest tumors (TART) are postulated to arise from aberrant adrenal cells which descend with the testis during embryogenesis and proliferate under chronic adrenocorticotropic hormone (ACTH) stimulation[2]. The incidence of TARTs is between 1 and 2 per 20,000[3]. Most TARTs are small and clinically undetectable but can rarely present as a clinically palpable mass[3].

Although histologically benign, TART can lead to testicular damage, infertility and diagnostic confusion with malignant testicular tumors[4]. Imaging plays a pivotal role in early diagnosis and follow-up.

CASE PRESENTATION

An 11-year-old male boy who is a follow-up case of congenital adrenal hyperplasia (CAH) with salt wasting and GnRH-dependent precocious puberty. The patient has received 14 injections of Leuprolide (11.25mg) uneventfully. Currently, the patient is on Tablet Hisone (Hydrocortisone) 5mg and Tablet Floricort (fludrocortisone) 50mcg. His testosterone levels are 162.3 ng/dl and 17-hydroxyprogesterone levels are greater than 16ng/ml. The height and weight of the patient are 150 cm (between 50-75 centile) and 51kg (between 75-90 centile), respectively. Systemic examination and vitals were normal. X-ray of the left hand and wrist revealed a bone age of 15-16 years.

The patient presented to our department for ultrasonography of the inguinoscrotal region. Bilateral testes were normal in size, with multiple well-defined, lobulated, heterogeneous hypoechoic nodules located near the mediastinum testis and showed internal vascularity on color doppler

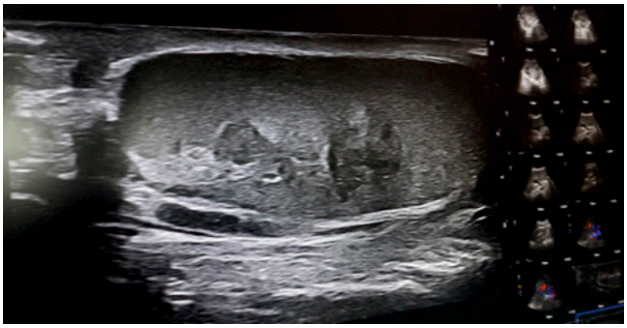


Fig. 1 (Right testicle)

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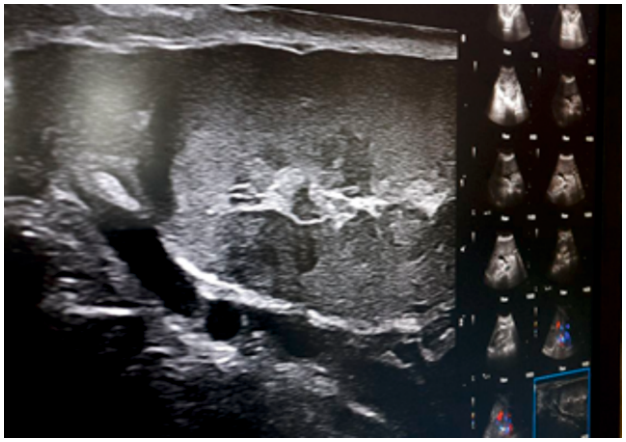


Fig. 2 (Left testicle)

Fig. 1 and 2 show grey scale images of bilateral testis revealing multiple perimedial intratesticular hypoechoic nodules.

The patient was further evaluated with contrast-enhanced MRI of the scrotum, which revealed the following signal characteristics: (Figures 3 & 4)

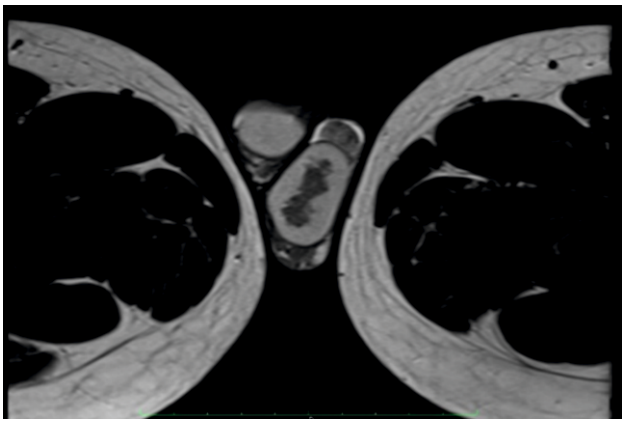


Fig 3: T2 Axial sequence showing bilateral intratesticular hypo intense areas

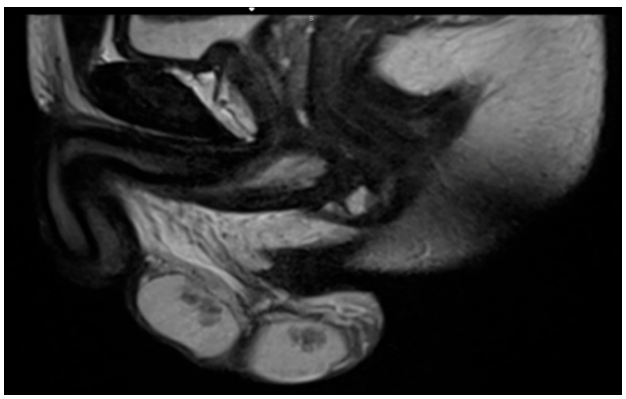


Fig. 4: T2 sagittal sequences showing bilateral intratesticular hypo intense areas

T1:iso-slight hyper intense signal

T2: hypointense signal

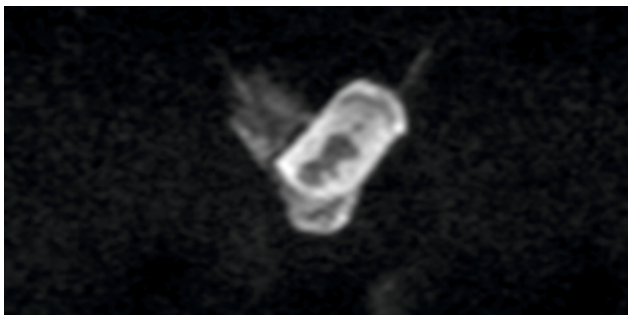


Fig. 5: DWI shows no diffusion restriction

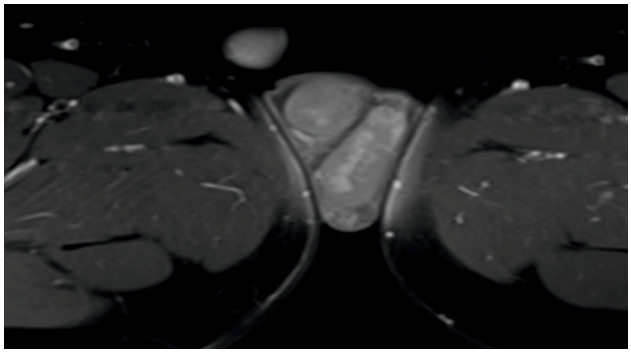


Fig. 6: T1 post contrast showing homogenous Enhancement

vDWI: No diffusion restriction

Contrast enhanced MRI scrotum revealed following signal characteristics: (Figures 5 & 6)

T1 C+ (Gd): avid homogeneous enhancement

Discussion:

Testicular adrenal rest tumors (TARTs) are commonly seen in male patients with CAH due to chronic ACTH stimulation. They typically present during childhood or adolescence and are frequently bilateral.[5] The mediastinal location, bilaterality and characteristic imaging features help differentiate TART from Leydig cell tumours, germ cell tumours, and lymphoma.[6]

Precocious puberty in CAH can be peripheral due to androgen excess; however, long standing disease may lead to secondary activation of the hypothalamic-pituitary-gonadal axis resulting in GnRH-dependent precocious puberty as seen in this case.[7]

Early diagnosis is crucial because optimization of glucocorticoid therapy can lead to regression of TART, preserving testicular function and fertility. Imaging plays a key role in diagnosis, follow up and treatment response assessment.

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