

Adrenal Cortex Heterotopia in an Undescended Testis—A Case Report

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ABSTRACT

Heterotopic adrenal cortex is usually found in the kidney, retroperitoneum and spermatic cord. It is common among pediatric age group and rarely encountered in adults. Our case presented with swelling in the right inguinal region with on and off dragging pain. On clinical examination testis was absent in the right scrotal sac which was confirmed with ultrasonogram. The clinical diagnosis was right side undescended testis. Orchidectomy was performed and on microscopic examination we incidentally found out an ectopic adrenal cortical rest in the rete testis with atrophic changes in the testis. Though the ectopic adrenal tissue was indolent in our case, literature suggests that it can undergo transformations like hyperplasia, adenoma and carcinoma, leading to undesirable complications. We present this case for the rarity at this age and site, and for the awareness that it may cause complications if left untreated.

Keywords: Adrenal cortex, Undescended testis

CASE REPORT

A 28-year-old man presented with complaints of swelling of size 2x2 cm in the right inguinal region of long duration with associated on and off dragging pain. The swelling was firm in consistency and movable in one plane. Scrotal examination revealed absence of testicle on the right side. The diagnosis of right side undescended testis was made and was further confirmed with ultrasonogram. Orchidectomy was done to relieve the disturbance due to ectopic testis and to prevent the chances for future complications.

Grossly the testis measured 2.4 x 2 cm with attached spermatic cord measuring 1.5 cm in length. Cut surface was grey tan, homogenous [Table/Fig-1]. Microscopy revealed atrophic seminiferous tubules with thick basement membrane, decreased germ cells and relatively increased sertoli cells. Surprisingly a well defined, encapsulated adrenal cortex without medulla was noted adjacent to the rete testis. This ectopic adrenal cortex showed normal zonation of glomerulosa and fasciculata but an irregular and poorly formed reticularis [Table/Fig-2-4]. Retrospectively the gross specimen was re-examined and found to have a tiny yellowish nodule measuring 2mm in the mediastinum of testis [Table/Fig-1]. The postoperative follow up of the patient was uneventful.

DISCUSSION

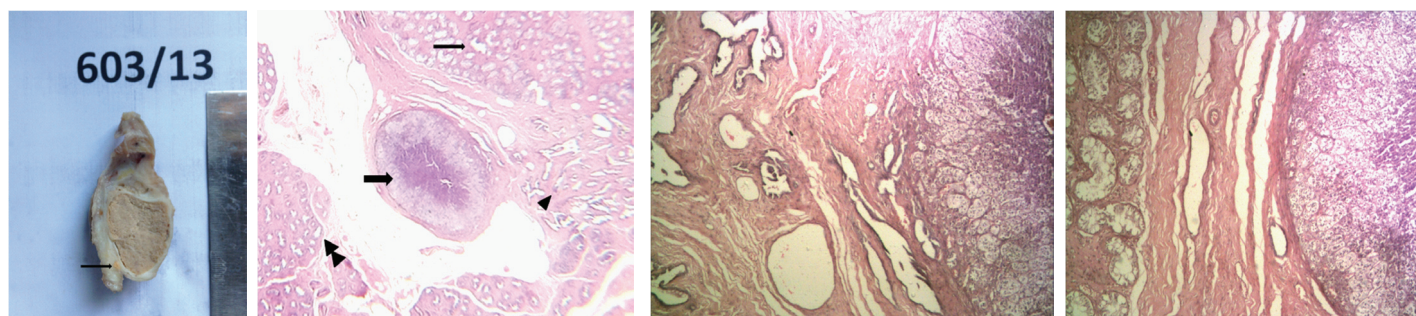
Heterotopic adrenal tissue has been commonly reported in kidney and retroperitoneal fat but only rarely in ovary, broad ligament, testis, spermatic cord, pancreas, liver, gastric wall and transverse colon [1-3]. Adrenal rest in testis and associated structures occur due to mechanical fragmentation and displacement of a portion of cells from the original cell cluster proposed to form orthotopic adrenal

cortex, during descent of testis to the scrotum [2]. Anderson JR & Ross AH suggested their origin from pluripotent stem cells [4].

As seen in our case, Dahl and Bahn [1] highlighted the irregular and poorly formed zona reticularis in contrast to glomerulosa and fasciculata in ectopic adrenal cortex [Table/Fig-3,4]. The reason for which remains to be eluded. They also reported tiny cysts in zona glomerulosa of both heterotopic and normal orthotopic adrenal gland highlighting their same origin.

These heterotopic rests are common in children but not so in adults. Mendez et al., [5] found that the median age of diagnosis was 5.8 years, in their study of 13 cases. They found that these ectopic rests are five times more common in children when compared to adults. But our case was a adult male, who presented with an undescended testis. Ricardo Drut et al., reported an intratesticular heterotopic adrenal tissue in a patient with coffin-siris syndrome. Our patient did not show any syndromic features.

Normally these rests are asymptomatic as in our case. Rarely they can attain larger size and come to clinical attention as reported in many patients with congenital adrenal hyperplasia by authors like Jin HY, Knagae And Marchini GS [6]. Similarly this aberrant ectopic adrenal tissue can undergo neoplastic change or become functional, liberating hormones and producing symptoms [2]. Such transformation can cause morbidity and mortality in patients. Alexander L Shifrin et al., [7] described a case of adrenocortical adenoma arising from ectopic adrenal tissue. Damjanov et al., [8] reported an interesting case of myelolipoma in heterotopic adrenal tissue. Malignant transformation occur rarely in heterotopic adrenal cortex. Jain SH et al., [9] reported a case of adrenocortical carcinoma arising from adrenal rest cells in testis. If malignancy



[Table/Fig-1]: Cut section of testis with spermatic cord. Arrow points tiny yellowish nodule

[Table/Fig-2]: Section show adrenal cortex (thick arrow), seminiferous tubules (thin arrow), rete testis (arrow head) and epididymis (double arrow head). (H&E, x20)

[Table/Fig-3]: Photomicrograph show adrenal cortical layers with adjacent rete testis (left)(H&E, x40)

[Table/Fig-4]: Photomicrograph show adrenal cortical layers with seminiferous tubules (left)(H&E, x40)

arises synchronously in both orthotopic and ectopic adrenal tissue, it may mimic metastasis. In such scenarios staging should be done cautiously, considering the possibility of concordant malignancy in heterotopic tissue. On other hand, ectopic adrenal tissue can be associated with testicular malignancy like seminoma as reported by Micheal Floyd et al.

Macroscopically the ectopic adrenal nodules vary in size. In our case the nodule was of size 2mm. Savas C et al., [10] also found the nodules of size 1 to 5 mm in eight patients with ectopic adrenal rest along spermatic cord. However the nodules can increase in size when it undergoes hyperplastic or neoplastic transformation. The role of cytology fine needle aspiration (FNAC) in diagnosis is controversial. Banik T et al., diagnosed adrenal rest in testis in a 7-year-old male child with precocious puberty by FNAC, in correlation with clinical, radiological and biochemical investigations. FNAC was not performed in our case since there was no clinical suspicion and we incidentally found the ectopic adrenal tissue in orchidectomy specimen.

CONCLUSION

Ectopic adrenal cortical tissue in the mediastinum of testis is rare and such ectopic adrenal tissue can be found at any site along the descent of testis. Though it can be indolent in most of the cases as in our case, it may undergo transformations like hyperplasia or neoplasia with complications. This possibility should be considered whenever ectopic rest of adrenal tissue is seen at any site and should be carefully examined microscopically.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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FINANCIAL OR OTHER COMPETING INTERESTS: None.

Date of Submission: **Jan 01, 2014**
Date of Peer Review: **Apr 16, 2014**
Date of Acceptance: **May 05, 2014**
Date of Publishing: **Sep 20, 2014**