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PARATESTICULAR ECTOPIC ADRENAL RESTS: CASE REPORT AND ANCILLARY FEATURES

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ABSTRACT

Ectopic adrenal rests is a rare lesion, with different locations, and more frequently encountered in the pediatric population. This is a case presentation of a 75-years-old male patient, who was hospitalised to the Emergency Department, with altered general condition, pain in the right hemiscrotum and fever. After the orchiectomy, the histopathological examination established the diagnosis of non-specific purulent degenerated cystic type of acute orchiepididymitis and ectopic adrenal tissue. The immunohistochemical examination confirmed the diagnosis of adrenal ectopy. In this case, the ectopic lesion was discovered accidentally, being associated with nonspecific acute inflammation. This is another case in addition to the very rarely reported and the location of testicular adrenal cortical ectopia in adults.

Keywords adrenal ectopy, testis, immnunohistochemistry

Introduction

Ectopic adrenal tissue, also tnamed heterotopic adrenal tissue or adrenal rests, represents nests or foci of adrenal tissue with various locations, distant from the eutopic adrenal glands.

Ectopic adrenal rests represents a rare condition which can be identified in different locations, especially in the retroperitoneum or pelvis, including celiac axis, genitals and broad ligament, along the path of gonadal descent. Men are the most susceptible to the disease, especially in childhood. Adrenal rests are asymptomatic and incidentally diagnosed in surgical specimens (1, 2).

Adrenal cortical rests involving the testis and its path are rarely reported in adults, during surgery or autopsy, these finding being more frequent in children. When diagnosed in adults, diagnosis may be difficult, especially for cases showing hyperplasia or neoplasia originating in this ectopic tissue and for differential diagnosis considerations (3).

Material and methods

We present the case of a hypertensive and diabetic 75-years-old male patient, who

was admitted in the Emergency Department with altered general condition, pain in the right hemiscrotum and fever. Mandatory testing, according to internal protocol, for the SARS-CoV2 virus, demonstrated the positive status of the patient.

The local examination revealed enlarged right hemiscrotum and testicle, painful to touch, with erythematous areas. The left testicle was apparently within normal limits. The foreskin was impossible to loosen.

Right orchiectomy was performed, with the evacuation of 70 ml of purulent liquid and Duchamel dorsal debridement. Post-interventional, the evolution was favorable under antibiotic therapy, treatment with analgesics, proton pump inhibitor and antivirals.

The Pathology Department received the orchiectomy specimen. Immunohistochemical testing were mandatory to complete the diagnosis. Antibodies against Inhibin, Calretinin, Melan A, PAX8, Synaptophisin, CD56 were used.

Results

The orchiectomy specimen featured an external hyperemic surface with the presence of gray and green areas of low consistency. Upon sectioning, a yellowish-green, purulent material is discharged. The testicle measured 5.5/3.5/2 cm, with the presence of numerous cystic spaces with a diameter ranging from 0.3 to 0.8 cm, occupied by a stringy, yellow-green material.

Microscopic examination showed testicular parenchyma, epididyme and vaginalis with an intense and diffuse acute inflammatory infiltrate, rich in neutrophiles and histiocytes, with multifocal abscesses and necrosis. At the testicular-epididymal interface, we identified a well-defined, pseudoencapsulated micronodule, with a maximal dimension of 0.2 cm, composed of polygonal cells with rich and eosinophilic vesicular cytoplasm, with well-defined regular nuclei and prominent nucleoli. The cell population was discretely separated by delicate fibrous septa.

Histopathological examination of the of the testicular parenchyma highlighted atrophic morphology, with adjacent adrenal ectopic tissue, in which the three cortical layers were visible, without the adrenal medulla tissue.

Following the immunohistochemical examination, intensely positive cytoplasmic Melan A was observed at the level of the evaluated nodule, intensely positive membranous CD56, positive cytoplasmic and nuclear Calretinin, variable intensity positive cytoplasmic Inhibin, negative Synaptophisin and negative PAX8.

The immunohistochemical tests, correlated with the immunohistochemical appearance, support the diagnosis of paratesticular ectopy of adrenal tissue.

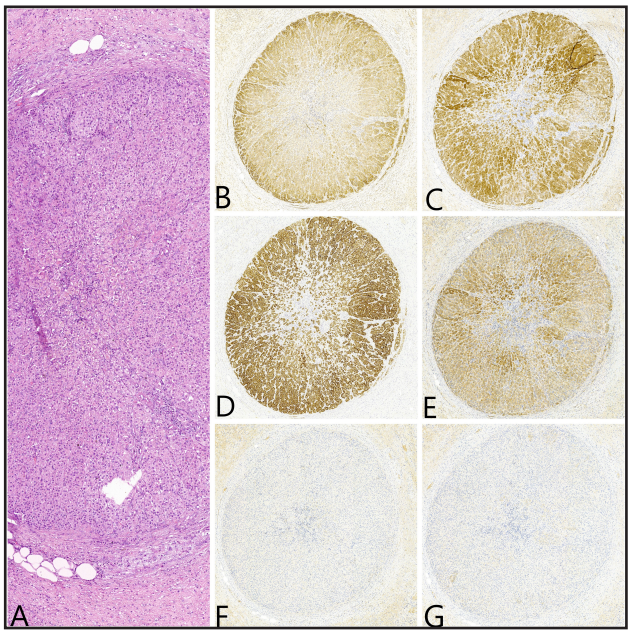


Figure 1 Collage of images; A Adrenal tissue, circumscribed, consisting of cells belonging to the three histological zones, with paratesticular location, HEx100; B intensely positive membranous immunoexpression to CD56, x40; C intensely positive cytoplasmic immunostaining to Melan A, x40; D positive cytoplasmic and nuclear immunostaining to Calretinin, x40; E variable intensity positive cytoplasmic positivity to Inhibin, x40; F negative expression to Synaptophisin, x40; G negative expression to PAX8, x40

Discussion

Heterotopic adrenal can be identified, in particular, in the kidney, retroperitoneum and spermatic cord. Since first case report of ectopic tissue in a juxtarenal position, by Morgagni, in 1740 (4), inguinoscrotal path of

the testis has been described.

Ectopic adrenal tissue may be identified along the path of the testis and diagnosed in approximately 1.2-2% of surgical procedures (especially between the testis and epididymis, in the hernia sac or along the spermatic cord) (5,6,7). Although it is reported as a low frequence lesion, it is considered that this injury is more frequent in medical practice and the cases are not reported. In addition, the low rate of reporting in the female sex can be explained by the basic diagnosis, which has more clinical vallue (8).

The human adrenal cortex has three components, represented by zona glomerulosa (zG), zona fasciculata (zF) and zona reticularis (zR), which have the role to produce mineralocorticoids, glucocorticoids, and adrenal androgens hormones. In this repect, the final stage of development occurs at puberty, with stabilisation of the zR and its syntesis of adrenal androgens, a process named adrenarche (9,10).

Adrenal rest in testis occurs secondary to mechanical fragmentation and dissemination of cells from the original cell group included in orthotopic adrenal tissue, during descent of testis to the scrotum (11). Although these structures are more common in the childhood, the average age of occurance is 5 years and 8 months, in the study of Mendez (12), these being five times more frequent in children.

In the presented report, the examined nodule had dimensions of 0,2 cm. According to the specialized literature, yellow-orange nodules, 0,1-0,5 cm in diameter, were described. Our patient was asymptomatic and did not fit into a specific syndrome (13,14).

The aberrant ectopic adrenal tissue can suffer neoplastic transformation or become functional through hormonal secretion, when the disease becomes symptomatic (15). This correlates with the increase in the rate of morbidity and mortality in male patients. In this respect, Alexander L Shifrin et al. (16) described a case of adrenocortical adenoma originating from etopic adrenal tissue.

A case of ectopic adrenal tissue on the ectopic testis has been reported (17). There have been cases of malignant tumors, such as carcinomas developed on ectopic adrenal tissue, at the testicular level (18). In a situation like the one described, if the lesion is in both sites, like orthotopic and ectopic adrenal tissue, a mimicker lesion of different types of cancers cannot be excluded. In males, differential diagnosis considerations shoud include testicular tumor, primary or metastatic clear cell carcinoma of the kidney, paraganglioma and metastasis from adrenal cortical carcinoma. In this respect, immunohistochemical exam is useful in excluding other pathologies (19,20).

Conclusions

In the presented case, the adrenal ectopy was an incidental finding, which accompanied the orchiepididimitis. The adrenal cortex rests were found in the paratesticular tissue, with no marks of hyperplasia or neoplasia. Just knowing the nodular gross appearance of such tissue, is essential in suspecting such an injury, in order to excise them once encountered. The immunohistochemical examination was extremely important in confirming the diagnosis of certainty and differential diagnosis. We presented this case especially for the location, rarity at this age and gros features, and also for the awareness of adrenal tissue ectopy.

Conflict of interest

The authors declare that they have no conflict of interest regarding the elaboration and publication of this article

References

1. Mills SE Histology for pathologists, 5th edn. Wolters Kluwer Health, 2019, Baltimore
2. Falco EC, Daniele L, Metovic J, Bollito E, De Rosa G, Volante M, Papotti M. Adrenal Rests in the Uro-genital Tract of an Adult Population. Endocr Pathol. 2021 Sep;32(3):375-384. doi: 10.1007/s12022-021-09685-y. Epub 2021 Jun 7.
3. Yokoyama H, Adachi T, Tsubouchi K, Tanaka M, Sasano H. Non-functioning adrenocortical carcinoma arising in an adrenal rest: immunohistochemical study of an adult patient. Tohoku J Exp Med. 2013 Apr;229(4):267-70. doi: 10.1620/

tjem.229.267.
4. Wiezel J. Ueber compensations. Hypertrophie der accessorischem nebennieren bei der Ratte. Zentral Blatt fuer Physiologie. 1879;12:780. In: Schechter DC. Aberrant adrenal tissue. Ann Surg 1968;167:421-6
5. Mendez R, Tellado MG, Somoza I, Liras J, Sanchez-Abuin A, Pais E, et al. Ectopic adrenal tissue in the spermatic cord in pediatric patients: Surgical implications. Int Braz J Urol. 2006;32:202–7.
6. Ketata S, Ketata H, Sahnoun A, FakhFakh H, Bahloul A, Mhiri MN. Ectopic adrenal cortex tissue: An incidental finding during inguinoscrotal operations in pediatric patients. Urol Int. 2008;81:316–9
7. Vaos G, Zavras N, Boukouvalea I. Ectopic adrenocortical tissue along the inguinoscrotal path of children. Int Surg. 2006;91:125–8
8. Tsai WH, Chen TC, Dai SH, Zeng YH. Case Report: Ectopic Adrenocortical Carcinoma in the Ovary. Front Endocrinol (Lausanne). 2021 Mar 19;12:662377. doi: 10.3389/fendo.2021.662377.
9. Xing Y, Lerario AM, Rainey W, Hammer GD. Development of adrenal cortex zonation. Endocrinol Metab Clin North Am. 2015 Jun;44(2):243-74. doi: 10.1016/j.ecl.2015.02.001.
10. Barwick TD, Malhotra A, Webb JA, Savage MO, Reznek RH. Embryology of the adrenal glands and its relevance to diagnostic imaging. Clin Radiol (2005) 60(9):953–9. 10.1016/j.crad.2005.04.006
11. Sullivan JG, Gohel M, Kinder RB: Ectopic adrenocortical tissue found at groin exploration in children: incidence in relation to diagnosis, age and sex. BJU Int. 2005; 95: 407-10
12. Mendez Roberto, Manuel G. Tellado, Ivan Somoza, Jorge Liras, A. Sanchez-Abuin, Ernesto Pais, et al. Ectopic Adrenal Tissue in the Spermatic Cord in Pediatric Patients: Surgical Implications. Int Braz J Urol. 2006;32:202–07 DOI: 10.1590/S1677-55382006000200013
13. Chas David, Schechter DC. Aberrant adrenal tissue. Ann Surg. 1968;167(3):421–26.). Anderson JR & Ross AH suggested their orgin from pluripotent stem cells
14. Anderson JR, Ross AH. Ectopic adrenal tissue in adults. Postgrad Med J. 1980;56(661):806–08
15. Savaş C, Candir O, Bezir M, Cakmak M. Ectopic adrenocortical nodules along the spermatic cord of children. Int Urol Nephrol. 2001;32:681–5
16. Shifrin Alexander L, Zheng Min, Vernick Jerome J. Ectopic Adrenocortical Adenoma. World Journal of Endocrine Surgery. 2011;3(1):45–46
17. Chas David, Schechter DC. Aberrant adrenal tissue. Ann Surg. 1968;167(3):421–26
18. Rabie ME, Shah MT, Jamil S, Al-Shraim MM. Adrenal rests associated with ectopic testis in an adult: Clinical significance. Urol Ann. 2013 Oct;5(4):302-4. doi: 10.4103/0974-7796.120307.
19. Raman R T, Sree R R, G H, Savitri NM, S A. Adrenal cortex heterotopia in an undescended testis-a case report. J Clin Diagn Res. 2014 Sep;8(9):FD11-2. doi: 10.7860/JCDR/2014/8331.4845. Epub 2014 Sep 20.
20. Jain SH, Sadow PM, Nosé V, Dluhy RG. A patient with ectopic cortisol production derived from malignant testicular masses. Nat ClinPractEndocrinolMetab. 2008;4(12):695–700. doi: 10.1038/ncpendmet0985. Epub 2008 Oct 21