

Adrenal Ectopia Within the Wall of an Ovarian Serous Cystadenoma

Over Seröz Kistadenomu Duvarında Ektopik Adren

Ufuk USTA, Emine TÜRKmen, N. Engin AYDIN

Başvuru tarihi / Submitted: 28.07.2005 Kabul tarihi / Accepted: 26.09.2005

Ectopic adrenal tissue is a relatively common finding, especially in children. The most common localization is the male genital system, especially the spermatic cord. Female genital tract is also a common site for ectopic adrenal tissue but, ovarian localization is very rare. A 21-year-old woman presented with complaints of abdominal swelling, pelvic pain and menstrual irregularity. Ultrasonographic examination revealed a 25 cm cystic mass in the left ovarian region. The cyst which was removed surgically, was filled with serous fluid and there was a 0.2 cm yellowish nodule within the wall. Serous cubic epithelium was lining the inner portion of the cyst. The nodule within the cyst wall was well defined and consisted of adrenal tissue.

Key Words: Adrenal cortex; genital diseases, female/pathology.

Ektopik adrenal çocuklarda nispeten sık görülen bir durumdur. En sık lokalizasyon erkek genital sistemi, özellikle de spermatik korddur. Kadın genital sistemi de ektopik adrenal dokusunun sık görüldüğü bir bölgedir, ancak over lokalizasyonu çok nadirdir. Yirmi bir yaşında kadın hasta karında şişlik, pelvik ağrı ve menstrüasyon düzensizliği nedeniyle başvurdu. Ultrasonografide sol ovarian bölgede 25 cm çaplı kistik kitle saptandı. Ameliyatla çıkartılan kist seröz sıvıyla doluydu ve duvarında 0.2 cm çapta sarı renkte nodül görüldü. Mikroskopik bakıda kist iç yüzü seröz kübik epitelle döşeliydi. Kist duvarındaki nodül ise iyi sınırlıydı ve adrenal dokudan oluşuyordu. Patolojik tanı, seröz kistadenom ve ektopik adrenal doku olarak geldi.

Anahtar Sözcükler: Adrenal korteks; kadın genital hastalıkları/patoloji.

Adrenocortical rests are common in children and may be found retroperitoneally anywhere from the diaphragm to the pelvis.^[1,2] These rests are present in approximately 50% of newborn infants and atrophy precedes final disappearance in short time.^[1] Heterotopic adrenal tissue

has been identified in multiple sites such as the liver, pancreas, gallbladder, kidney, male and female genital tract.^[3] These ectopias may involve both medulla and cortex, such as in our case, however cortical ectopia is more frequent.^[2,3] A case of an ectopic adrenal tissue

Trakya Üniv Tip Fak Derg 2006;23(2):95-98

Department of Pathology Medicine Faculty of Trakya University (Usta, Yrd. Doç. Dr.); Department of Ankara Forensic Medicine Institution (Türkmen, Uzm. Dr.); Department of Pathology Medicine Faculty of İnönü University (Aydin, Prof. Dr.).

Correspondence: Dr. Ufuk Usta. Trakya Üniversitesi Tip Fakültesi Patoloji Anabilim Dalı, 22030 Edirne.

Tel: 0284 - 235 76 41 / 1528 Faks: 0284 - 235 76 52 e-posta: drufukusta@yahoo.com

©Trakya Üniversitesi Tip Fakültesi Dergisi. Ekin Tıbbi Yayıncılık tarafından basılmıştır. Her hakkı saklıdır.

©Medical Journal of Trakya University. Published by Ekin Medical Publishing. All rights reserved.

within the wall of an ovarian serous cystadenoma of a 21-year-old woman is presented as a peculiar site presumably unreported in Medline dating from 1966.

CASE REPORT

A 21-year-old woman presented to the gynecology department with complaints of abdominal swelling, periodical pelvic pain and irregularity of menstruation for the last 2 years. The pelvic ultrasonography revealed a huge cystic mass of 25 cm in the left ovarian region. The resected ovarian cyst had a smooth bright outer surface

with dimensions of 30x20x10 cm, and was full of straw colored serous fluid. The wall of the cyst was measured 0.2 cm thick. The inner surface of the unilocular cyst was bright, and there were no papillary structures. In the wall of the cyst, there was a yellow colored nodule of 0.2 cm and it was also sampled for microscopic evaluation. The sections of the formalin fixed, paraffin embedded specimen were stained with routine hematoxylin-eosin (HE) stain and immunohistochemically (IHC) for synaptophysin and chromogranin A (monoclonal, Fremont, CA, USA, Neomarkers).

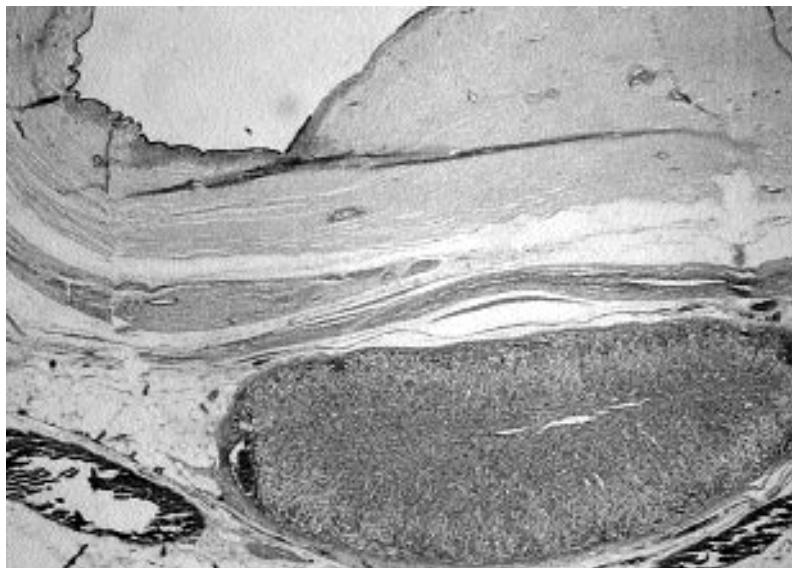


Fig. 1. A 2x1 mm ectopic adrenal nodule within the wall of a serous cystadenoma of ovary (H-E x 40).

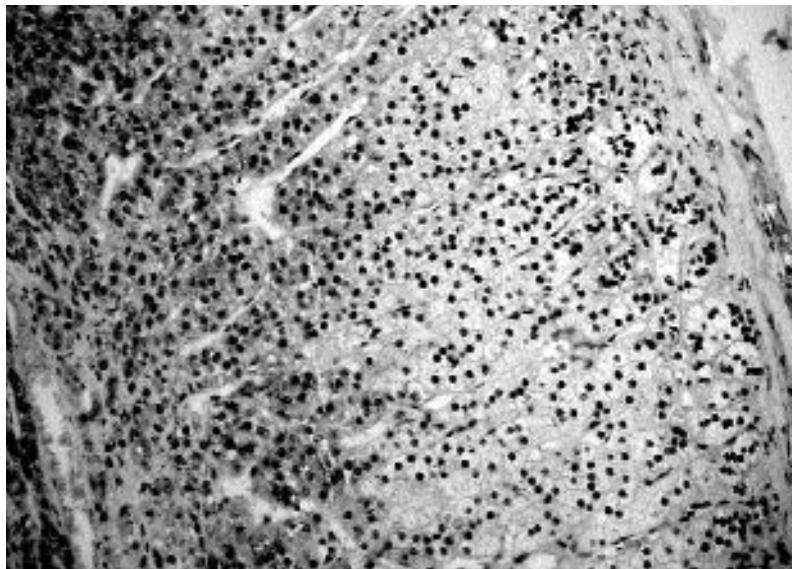


Fig. 2. Synaptophysin positive central portion (left). The outer region representing the cortex is negative for synaptophysin (right) (H-E x 200).

Microscopically the inner surface of the fibrotic cyst wall was lined by serous, low cubic epithelium, and the cyst was diagnosed as "serous cystadenoma of the ovary" and the above mentioned nodule was diagnosed as "ectopic adrenal tissue with well defined borders" (Fig. 1). Two distinct regions of the adrenal, namely cortex and medulla was identified with antibodies synaptophysin and chromogranin A (Fig. 2).

DISCUSSION

Cortex and medulla are two distinct portions of the adrenal gland within the same fibrous capsule.^[3] The adrenal gland has double origin; the adrenal cortex develops from the mesoderm of the Wolffian ridge, whereas the adrenal medulla originates from the ectoderm of the primitive ganglionic crest.^[3-6] Before being enveloped by the cortex, the medulla penetrates the cortex along the central vein.^[4] During penetration, small fragments most consisting of cortex only, separate and form accessory adrenal structures. Some of these nodules are displaced during the migration and descent of the sex organs.^[4]

If carefully searched, accessory adrenal tissue may be found in at least 50% of neonates and infants. With advancing age, this accessory adrenal tissue atrophies as it becomes physiologically unnecessary in the presence of normal adrenal glands.^[6]

Ectopic adrenal tissue was first described by Marchan in 1883.^[4] Since then ectopic adrenal tissue has been reported in different locations such as liver, kidney, pancreas, transvers colon, celiac-plexus area, broad ligament, ovary, testicle, epididymis, and retroperitoneally along the course of the vessels of gonads.^[2,4-6,7]

Studies in different series showed that, the most common site for the ectopic adrenal tissue is the spermatic chord region.^[1,8] In one of these studies, the resected specimens of 152 children, who underwent operations for common inguino-scrotal pathology, were investigated. In four cases ectopic adrenal tissue was found as a yellow nodule along the spermatic cord or hernia sac.^[8]

Ectopic adrenal tissue may be of clinical importance.^[1] These nodules may hyperfunction and result in Conn's syndrome, extreme virilization and Cushing's syndrome.^[5,6] Ectopic adrenocortical nodules may cause relapse in the disease in patients who underwent adrenalectomy because of Cushing's syndrome.^[1] Malignant and benign neoplastic changes were also reported in ectopic adrenal nodules in very few cases.^[1,5] Ectopic adrenal adenomas should be kept in mind in cases of Cushing's Syndrome independent of corticotropin, especially if adrenal gland is normal and symptoms relapse after adrenalectomy.^[5]

Considering the above clinical conditions, though the probability of neoplasia or hyperplasia is low in ectopic adrenal tissues, excision of incidental ectopic adrenocortical nodules is when encountered whenever they are met during surgery.^[1] But it is also mentioned that, there is no need for a further dissection for ectopic adrenal nodules. If done, the damage to the spermatic cord, vas deference and other neighbouring tissues would be of more importance than resection of ectopic adrenal.^[1]

The presented case is an adrenal ectopia which is incidentally diagnosed during the histopathological examination of the surgical specimen. This could be of clinical significance owing to its hormonal activity, and its unusual localization mentioned above, makes the case even more interesting.

REFERENCES

1. Savas C, Candir O, Bezir M, Cakmak M. Ectopic adrenocortical nodules along the spermatic cord of children. Int Urol Nephrol 2001;32:681-5.
2. Rosai J. Adrenal gland and other paraganglia. In: Rosai J, editor. Rosai and Ackerman's surgical pathology. 9th ed. Missouri: Mosby Year Book; 2004. p. 1115-18.
3. Chin L, Brody RI, Morales P, Black VH. Immunocytochemical characterization of intrarenal adrenal tissue. Urology 1994;44:429-32.
4. Bruning H, Kootstra G, Walther FJ, Arends JW. Ectopic adrenocortical tissue along the spermatic cord. Z Kinderchir 1984;39:269-70.
5. Ayala AR, Basaria S, Udelsman R, Westra WH, Wand GS. Corticotropin-independent Cushing's syndrome caused by an ectopic adrenal adenoma. J Clin Endocrinol Metab 2000;85:2903-6.

Adrenal ectopia within the wall of an ovarian serous cystadenoma

6. Leibowitz J, Pertsemlidis D, Gabrilove JL. Recurrent Cushing's syndrome due to recurrent adrenocortical tumor-fragmentation or tumor in ectopic adrenal tissue? *J Clin Endocrinol Metab* 1998;83:3786-9.
7. Ventura L, Leocata P, Hind A, Greco I, Ventura T. Ectopic adrenal tissue in the spermatic cord. Case report and review of the literature. *Arch Ital Urol Androl* 1998;70:15-8.
8. Oguzkurt P, Oz S, Kayaselcuk F. Ectopic adrenal tissue: an incidental finding during inguinoscrotal operations in children. *Hernia* 2002;6:62-3.