

Evaluation of children with inguinoscrotal ectopic adrenal tissues

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Aim: To evaluate 6 cases of inguinoscrotal ectopic adrenal nodules, which were detected in 2 hospitals.

Materials and methods: A total of 296 male patients who had inguinoscrotal surgery between 2009 and 2011 were retrospectively evaluated. Bright yellow nodular lesions, resembling the adrenal tissue, were excised during surgery. Histopathologic examination of these samples was performed and the clinical data of the patients were evaluated.

Results: Ectopic adrenal tissue was detected in 6 patients (2.02%). The mean age of the patients was 4.6 years (range: 3 months to 9 years) years. Ectopic adrenal nodules were localized in the spermatic cord, in the vicinity of the epididymis and appendix testis, on the epididymis, and on the tunica albuginea of an atrophic testis undergoing orchiectomy due to intraabdominal localization. Adrenal cortex cells were found in 5 patients, and both cortex and medullar cells were found in 1 patient. No malign transformation was detected. Hormonal, biochemical, hematologic, and urinary analyses did not yield any significant results.

Conclusion: Inguinoscrotal ectopic adrenal tissues are incidentally detected lesions during surgical interventions. They are benign lesions associated with no significant symptoms. Nevertheless, it is considered to be of benefit to remove these tissues in order to perform histopathological evaluation for differential diagnosis.

Key words: Ectopic adrenal tissue, inguinoscrotal surgery, ectopic adrenal medulla

1. Introduction

The adrenal glands originate from 2 different embryologic tissues, the cortex from mesoderm and the medulla from the ectoderm, which are histopathologically different from each other in terms of cell structure, organization, and function. The embryologic development of the medulla and cortex are also different (1). The cortex is formed from the proliferation of mesenchymal cells in the region between the mesentery root of the coelomic epithelium and gonadal draft between the 4th and 5th embryonal weeks. The adrenal medulla is a part of the sympathetic nervous system formed from the crista neuralis with an ectodermal origin. These cells, originating from neural crest cells, then migrate to the primitive cortex structure. Encapsulation of the medulla occurs in the late stages of fetal growth (2-4).

Developmental abnormalities of adrenals consist of ectopic or accessory adrenal tissues and heterotopia. After ectopic adrenal tissue was first identified by Morgagni

in the 1700s, several nodules of ectopic adrenal tissue were reported in many sites of the body, such as the kidney, coeliac axis, thorax, liver, lungs, and brain, and in association with the genitalia (1,5-7).

Inguinoscrotal localization of ectopic adrenal tissue is a rare condition. According to published studies, the great majority of adrenal ectopias occur with adrenocortical tissue (8). However, medullary ectopia is extremely unusual. Furthermore, clinical and epidemiologic features of adrenal ectopias in the inguinoscrotal region still need to be detailed. In this study, we retrospectively evaluated 6 cases of inguinoscrotal adrenal ectopias, which were incidentally recognized during surgery.

2. Materials and methods

2.1. Patients

A total of 296 male patients, who had undergone inguinoscrotal surgery between January 2009 and June 2011 in the urology and pediatric surgery clinics of Turgut

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Özal Medical Center and Malatya State Hospital, were included in this study. These patients were diagnosed with hydrocele, undescended testis, inguinal hernia, and atrophic testis. Demographic and clinical data of the patients, such as age and accompanying disorders, were collected. Biochemical and hematologic features of the patients' blood samples, urine analysis, and serum hormone (adrenocorticotropic hormone (ACTH), testosterone, progesterone, and thyroid-stimulating hormone (TSH)) levels were studied.

2.2. Sample collection

Bright yellow nodules, with an appearance resembling adrenal tissues, were surgically excised and placed in a 10% formalin solution. Data about the nodules, including the localization and nature of the lesions, were recorded. After routine tissue monitoring, the sections obtained were stained with hematoxylin and eosin (H&E) and examined under a light microscope.

3. Results

The mean age of the patients was 4.6 years (ranging between 3 months and 9 years). Ectopic adrenal tissue was detected in 6 (2.02%) of 7 surgically excised tissue samples among 296 patients. Of these 6 cases, 4 patients were diagnosed with undescended testis, and the remaining 2 patients with inguinal hernia and hydrocele. These ectopic adrenal tissues were localized on the spermatic cord ($n = 3$), epididymis ($n = 1$), appendix testis ($n = 1$), and tunica albuginea of the atrophic testis ($n = 1$). Of the 6 cases, 4 ectopias were located on the left and 2 on the right side, and they were all bright yellow with a soft consistent

nodular structure and between 1 and 4 mm in diameter. A photograph of the nodules taken during surgery is shown in Figure 1.

Multiple nodules, up to the spermatic cord, were found in 2 of 6 cases, and single nodules were found in 4 patients. After the histopathologic examination, 5 patients were diagnosed with corticoadrenal ectopia, and 1 patient with corticomedullary ectopia. Microscopic photographs of cortical cells and cortical and medullary cells are shown in Figures 2 and 3, respectively. In all 6 cases, 3 layers of adrenal cortex (glomerulosa, fasciculata, and reticularis) were histopathologically shown.

The results of the biochemical, hematologic, and hormone analysis of the patients were considered to be within normal ranges. Urine analyses were also included in the patients' data and are summarized in the Table.

4. Discussion

Morgagni first described ectopic adrenal tissues as yellowish nodules similar to adrenal tissue in 1740 (1). Since then, many studies have been conducted about the localization of ectopic adrenal tissue, and, in the literature, at least 100 cases were detected around the genital region. About 80 of these cases were detected in the genital region of males during childhood (9–11). Ectopic adrenal tissues in the male patients within our study were localized in the genital region, as in the literature. In autopsy studies, ectopic adrenal tissue is observed in the testis of newborns at a rate of 7.5%–15%. These rates may vary between 1% and 9.3% in studies in the pediatric age group. In our study, this rate was 2.02% ($n = 6$). The lower rate of ectopic

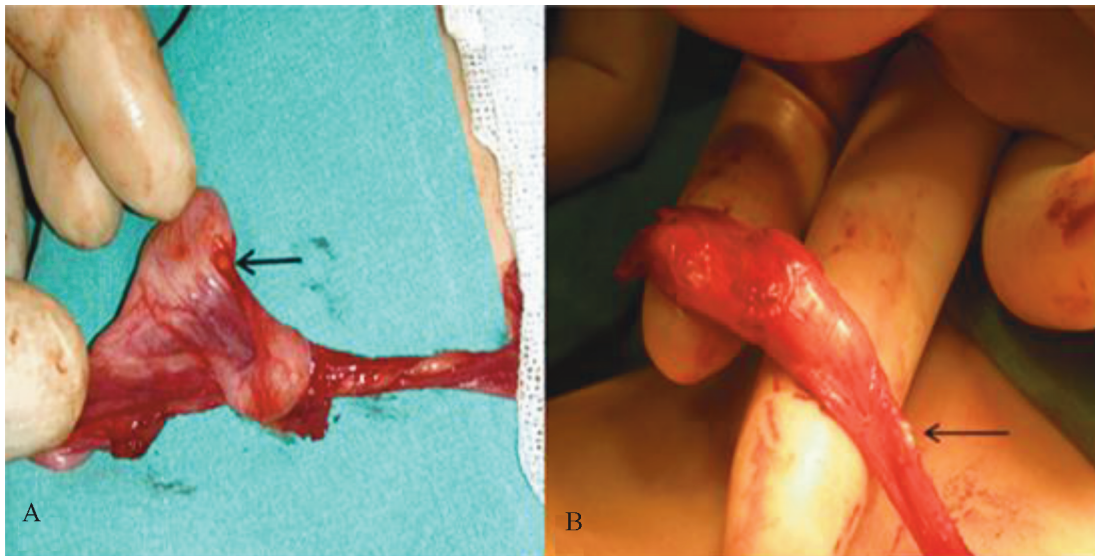


Figure 1. Operative photographs taken from 2 different patients during orchidopexy for undescended testis. After mobilizing the cord and testis, 1 small nodule of ectopic adrenal tissue was identified on the testis (arrow) (A). The well-defined, round, golden-yellow nodules became clearer after dissecting the spermatic cord (arrow) (B).

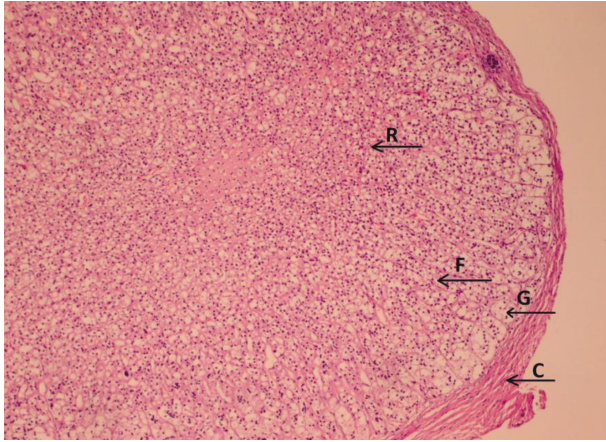


Figure 2. A small nodule of ectopic adrenal tissue (H&E 40×). The distinct fibrous capsule (C) covers a thin, patchy layer of clumps of cells, the zona glomerulosa (G). The middle and thickest layer is the zona fasciculata (F), composed of columns of cells with a clear or finely vacuolated cytoplasm in which the cells of the zona reticularis (R), with uncharacteristically eosinophilic cytoplasm, lie.

adrenal tissue in adulthood is believed to be due to the atrophy of the remaining tissues. The reason for a lower rate in females could be the differences in type of surgery for underlying disease (11,12).

Male gonadal structures and the adrenal cortex have the same embryological origin, which could explain the similarity of ectopic adrenal tissues with gonadal structures. Ectopic adrenal remains, located away from the original adrenal tissue, consist only of adrenal cortical tissue with no adrenal medulla cells. However, adrenal medulla cells can be found in ectopic adrenal tissues with a more proximal localization. Medulla cells are observed as cellular structures of the capsule of connective tissue, usually with small blood vessels that surround these nodules (13). In the literature, only adrenal cortex was found in ectopic adrenal tissues detected in genital regions, and adrenal medulla cells were not observed (12). Ketata et al. (14) found ectopic adrenal tissue in 1.66% (n = 31)

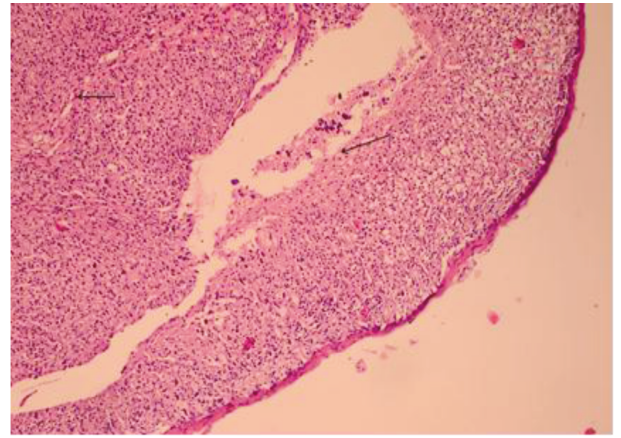


Figure 3. Medullar cells are present in the excised gland (arrows; H&E 40×).

out of 1862 patients who had inguinoscrotal surgery and adrenal medulla was not detected in any of the patients, while all had adrenal cortex. The general opinion is that the observation rate of medullar cells decreases toward zero the more distal the location of ectopic adrenal tissue is (12–14). However, we found that 1 of our 6 patients had adrenal cortex and adrenal medulla together. The case found to contain adrenal medulla was the patient who had an orchiectomy because of atrophic testis with intraabdominal localization. Existence of adrenal medulla cells in this case may be due to the intraabdominal location, a position much more proximal than the normal anatomy of the testis.

Because almost all reported cases were found incidentally during surgery, we do not know clearly the characteristics of preoperative biochemical, hormonal, and radiological findings of these patients. Similarly, by reviewing a large series, specific disorders or anomalies were not reported as any significant accompanying disease with ectopic adrenal tissues (12–14). Nevertheless, we did not detect any biochemical, hormonal (ACTH, testosterone, progesterone, and TSH), or hematologic abnormalities suggestive for ectopic adrenal tissue in our patients.

Table. Characteristics of the patients with ectopic adrenal tissues.

Patient no.	Age	Accompanying disease	Location	Size (mm)	Number of lesion(s)	Cortex or medulla
1	3 months	Undescended testis	Left appendix testis	1	1	Cortex
2	1 years	Undescended testis	Left spermatic cord	1–3	3	Cortex
3	3 years	Undescended testis	Right spermatic cord	2	1	Cortex
4	5 years	Undescended testis	Left spermatic cord	1–4	4	Cortex
5	5 years	Hydrocele	Left epididymis	2	1	Cortex
6	9 years	Inguinal hernia	Right tunica albuginea	4	1	Cortex-medulla

Surgical interventions are considered according to the patient's clinical symptoms caused by these ectopic tissues. Some authors have reported that ectopic tissues show compensatory functional growth when a total adrenalectomy is performed (15,16). Another clinical aspect is the development of a tumor from the ectopic adrenal remains. Pheochromocytoma, Leydig cell tumors, and adrenal adenomas have been reported to develop from these ectopic tissues, although they are not common (6,17–19).

Suspected nodular structures observed along the spermatic cord or the testis and epididymis during inguinoscrotal surgeries should thus be removed and evaluated pathologically. In our study, the nodular structures were detected microscopically during surgery and definitively diagnosed with a histopathologic investigation. These lesions, incidentally found and easily diagnosed due to their classical histological appearances, were benign. There were no malign characteristic

appearances histopathologically (big mass, infiltration at the margins, atypical and high mitotic activity) with these smoothly encapsulated small lesions. Although no spermatic cord injury has been reported in the literature, the spermatic cord should be carefully dissected during the surgical excision and the spermatic vessels and ductus deferens should not be damaged. No damage occurred during surgical excision on the spermatic cord and ductus deferens in our cases. All of the patients had no problems during the 1-year follow-up.

In conclusion, ectopic adrenal tissue is a rare event during inguinal surgeries. We believe that it may be important for surgeons working in these areas to keep in mind the possibility that yellow millimetric nodules with a soft consistency in the hernia sac, embedded in the cremasteric muscle fibers along the spermatic cord, or around the testis and epididymis, could be ectopic adrenal tissue. They may be excised and histopathologically examined due to malign transformation.

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