Concomitant testicular seminoma and ectopic adrenal tissue of the cord in a 45-year-old male

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Abstract

Aberrant adrenal tissue near the adrenal gland is common, but the finding of ectopic adrenal tissue in structures around the spermatic cord and testis is rare. We describe a case of concomitant seminoma and ectopic adrenal tissue of the spermatic cord occurring in an adult patient who had undergone orchidopexy as a child.

Introduction

Aberrant adrenal tissue near the adrenal gland is common, but the finding of ectopic adrenal tissue in structures around the spermatic cord and testis remains rare.[1] In 1740, Morgagni first described ectopic adrenal tissue in the vicinity of the adrenal gland,[2] and in 1885 Dagonest noted the presence of adrenocortical tissue in the spermatic cord of an infant.[3] Other non-adrenal sites include the coeliac axis, renal parenchyma, broad ligament and ovary; the placenta, hepatic, pulmonary and intracranial lesions, including eleventh cranial deposits, have also been described.[4,5]

Case report

A 45-year-old male was referred to the outpatient department with a 3-month history of a painless right testicular mass. There was no history of trauma, recent infection, systemic upset or weight loss. His history was remarkable for essential hypertension and an inguinal orchidopexy at age 13 for an undescended right testicle. He had fathered 6 children.

Physical examination revealed a soft non-tender abdomen with a right inguinal scar. There was a discreet mass in the upper pole of his right testicle, a right scrotal scar consistent with his orchidopexy and a normal left hemiscrotum. Tumour markers (alpha fetoprotein, beta human chorionic gonadotrophin and lactate dehydrogenase) were normal. Scrotal ultrasound demonstrated a 9-mm mass in the upper pole of the right testicle and a nodule in the right spermatic cord (Fig. 1). He underwent a right radical orchidectomy via his old inguinal scar. Final histopathological analysis revealed a classic seminoma, which stained positive for placental alkaline phosphatase (PLAP), but negative for Ber-H21, inhibin and Alphafeto protein (Fig. 2). No lymphovascular invasion was noted and it was staged as a pT1 lesion. The nodule in the spermatic cord contained morphological features consistent with an adrenal rest (Fig. 3).

Discussion

In the pediatric population, ectopic adrenal tissue found during inguinoscrotal procedures has been extensively documented[1,5] with a right-sided preponderance. There is an increased incidence of ectopic adrenal tissue within the spermatic cord of males with undescended testes,[5] ranging from 1.6% to 5.1%.[3,6] Autopsy series have shown an incidence of ectopic adrenal tissue adjacent to the native adrenal of up to 32% in adults.[7] However, the incidence of ectopic adrenal tissue in the spermatic cord of adults is significantly lower at 1%.[8]

Macroscopically, the appearance of ectopic adrenal tissue is characteristic (a round, yellow nodule, firm in consistency, embedded in the cremasteric fibres, resembling a fat lobule).[1] Adrenal rests situated far from the original gland are composed entirely of cortical adrenal tissue with no evidence of medullary cells, but the more proximal cells may contain medulla. Usually a capsule of connective tissue with small blood vessels can be seen surrounding these nodules.[4] Of the 3 cortical layers, zona fasciculata and glomerulosa predominate. The reticularis layer is usually only seen in older children.[1] This can be explained when the embryological route and descent pathway are considered.

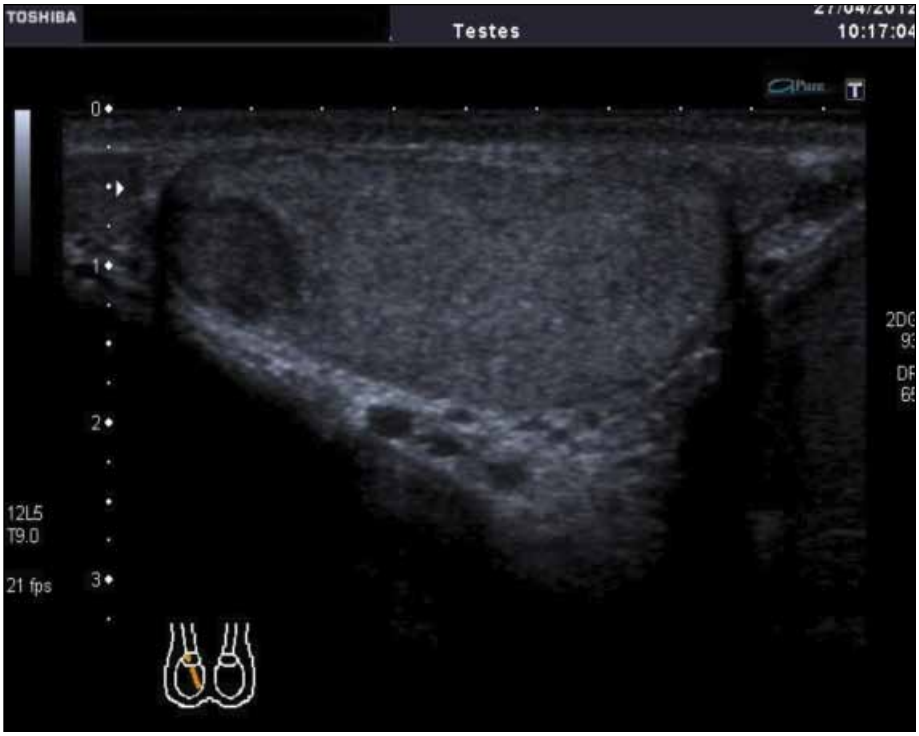

Fig. 1. Scrotal ultrasound revealing a 9-mm mass in the upper pole of the right testicle and a nodule in the spermatic cord.

The adrenal primordium and primitive gonad develop adjacent to each other;[9] however, the adrenal cortex and medullary region have different embryological origins. The fetal cortex is derived from the mesoderm, which lines the posterior abdominal wall, whereas the medullary cells derive from an adjacent sympathetic ganglion, usually a derivative of the neural crest. Neonatally, the 3 separate cortical zones develop, but the zona reticularis is not recognizable until the third year of life. The gonad descends along the retro peritoneum traversing pelvic and inguinal routes before eventually resting in the scrotum.[3] It is postulated that ectopic adrenal tissue within the spermatic cord results from the close spatial relationship of the embryologic pathway between gonadal structures and the adrenal primordium.[9] An embryological defect is therefore responsible for heterotopic adrenal tissue, as it occurs when clusters of cells separate from the main body of the adrenal cortex and associate with gonadal tissue during its descent.[2]

The clinical significance of ectopic adrenal tissue is usually minimal and typically not associated with endocrine abnormalities. However, in certain situations it may be of importance as pathological conditions may develop in this ectopic tissue similar to those seen in normally situated glands, including hyperplasia and neoplasia.[1] Compensatory functional hypertrophy of these tissues in patients who have undergone bilateral adrenalectomy due to pathologic adrenocorticotrophin hormone (ACTH) production may occur. This compensatory hyperplasia of the ectopic adrenal tissue may be responsible for the subsequent recurrence of the disease.[10]

Adenomatous hyperplasia with increased ACTH production can occur independently or in association with similar changes in the native adrenals.[1] The development of a neoplasm in an ectopic adrenal deposit is uncommon, yet phaeochromocytoma and Leydig cell tumours have been reported.[10] Primary neoplastic tissue in the spermatic cord is also rare, but paragangliomas have been described.[11]

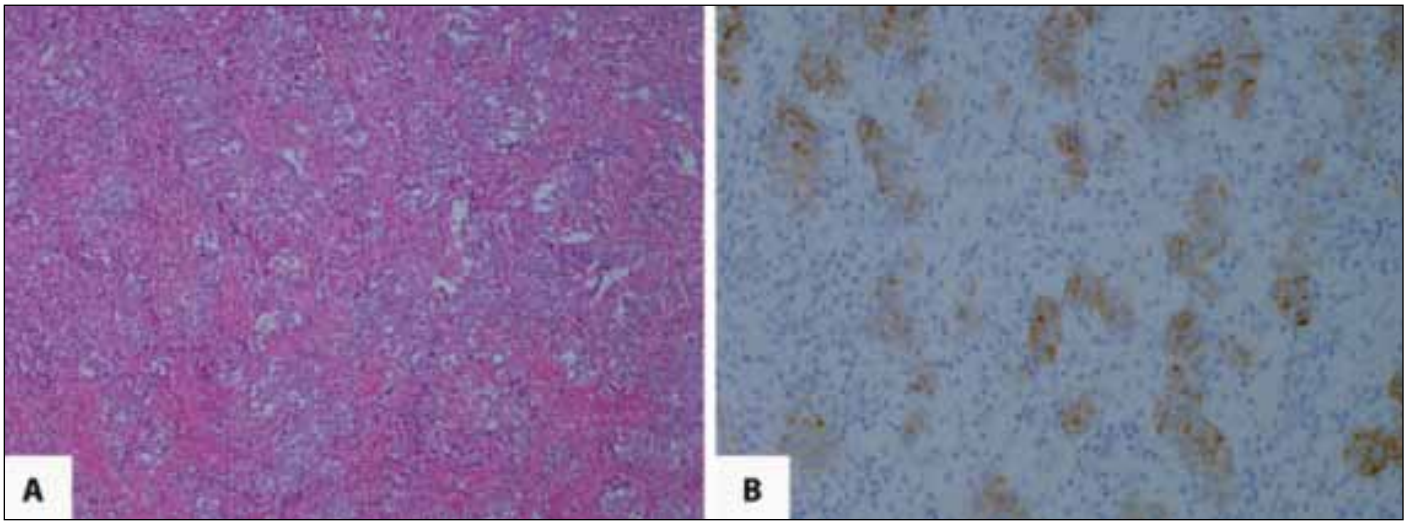

Fig. 2. Classic seminoma. A: Hematoxylin and eosin ×100. B: Immunohistochemistry positive for placental alkaline phosphatase.

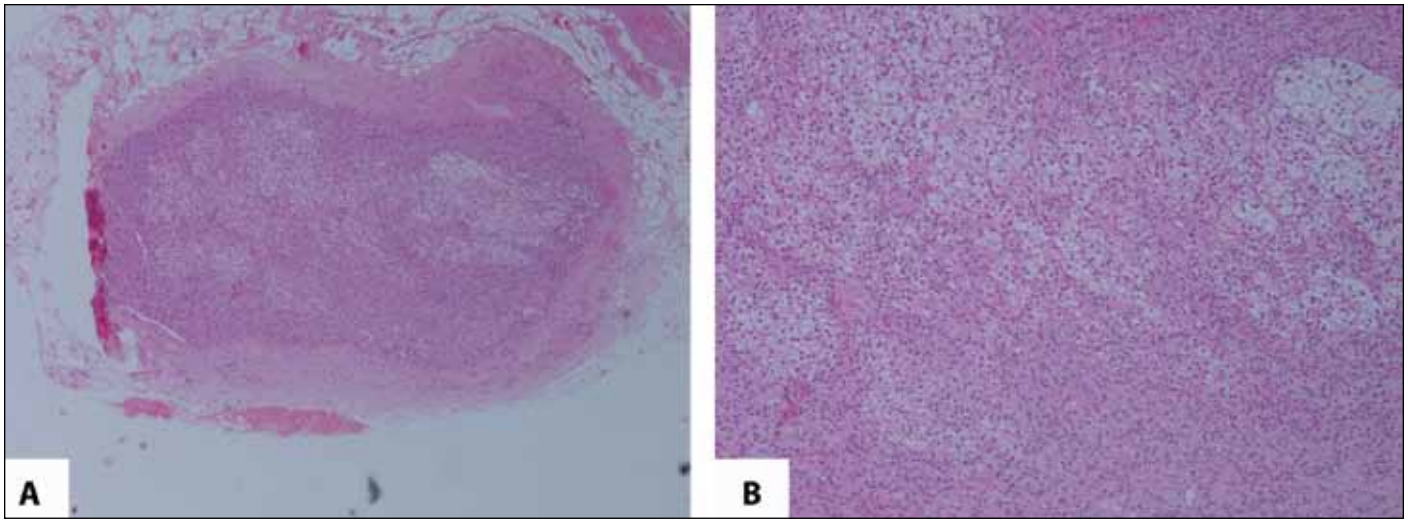

Fig. 3. Ectopic adrenal. A: Spermatic cord containing a 5-mm nodule of ectopic adrenal cortical tissue, surrounded by a connective tissue capsule, composed of zona fasciculate and zona glomerulosa (hematoxylin and eosin ×40). B: Loosely corded foamy cells (hematoxylin and eosin ×100).

If, during radical orchidectomy, a yellow nodule is found within the spermatic cord, the potential diagnosis of ectopic adrenal tissue should be considered in addition to a secondary deposit. Confirmation requires histopathological examination. Although a benign lesion, the ectopic adrenal deposit and the secondary deposit should be removed simultaneously when discovered, as it has no side effects and is easily achieved surgically.[6,12] In selected cases, such as this one, the histological recognition of the nodule as adrenal tissue is essential to prevent upstaging of the primary cancer.

A thorough search of the literature has demonstrated that concomitant testicular seminoma and ectopic adrenal has only been described once.[13] This case differs, however, as the interval between orchidopexy, tumour occurrence and incidental discovery of an ectopic adrenal deposit was over 30 years compared to 3 years in the case described by Mari and colleagues.[13]

Conclusion

We believe this to be the second reported case of concomitant ectopic adrenal tissue and seminoma discovered following radical orchidectomy.

Competing interests: All the authors declare no competing financial or personal interests.

This paper has been peer-reviewed.

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