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ADENOMATOID TUMOUR OF THE ADRENAL GLAND: A RARE EXTRA-GENITAL MESOTHELIAL NEOPLASM PRESENTING AS BILATERAL ADRENAL MASSES

Histopathology

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ABSTRACT

Background: Adenomatoid tumours (ATs) are benign mesothelial neoplasms most commonly arising in the genital tract. Extra-genital occurrence is exceedingly rare, with adrenal localization constituting an unusual diagnostic entity. The absence of specific clinical and radiological features renders immunohistochemistry indispensable for confirmation, particularly to exclude adrenocortical carcinoma and metastatic disease. **Case Presentation:** A 27-year-old male presented with right lumbar pain of one-month duration. Contrast-enhanced computed tomography revealed bilateral adrenal soft-tissue lesions, the larger measuring 4.6 × 4.4 × 5.4 cm. The resected specimen demonstrated anastomosing tubules and cystic spaces lined by flattened to cuboidal cells with focal necrosis. Immunohistochemically, tumour cells were positive for Pan-CK and Calretinin, and negative for CD31 and SF-1, confirming mesothelial origin and excluding vascular and adrenocortical differentiation. Ki-67 proliferative index was 5%. **Conclusion:** Adrenal adenomatoid tumour is a pathological rarity requiring histopathological characterization with an appropriate immunohistochemical panel for definitive diagnosis. Complete surgical excision remains curative, and accurate diagnosis prevents unnecessary oncological intervention.

KEYWORDS

adenomatoid tumour; adrenal gland; mesothelial neoplasm; extra-genital; immunohistochemistry; calretinin; Pan-CK; bilateral adrenal mass

1. INTRODUCTION

Adenomatoid tumours are benign neoplasms of mesothelial origin, the great majority of which arise within the male and female genital tracts. In males, the epididymis and para-testicular soft tissue represent the predominant sites of involvement; in females, the uterus, fallopian tube, and ovary are most frequently affected.

Adrenal adenomatoid tumour is among the rarest reported variants, with fewer than 50 cases described in the published literature to date. Its clinical presentation is non-specific, frequently overlapping with other adrenal neoplasms, and its radiological appearance lacks pathognomonic features. The histological architecture characterized by anastomosing tubules, signet-ring-like vacuolated cells, and cystic spaces may be mistaken for adenocarcinoma, hemangioma, or lymphangioma on routine staining alone. Immunohistochemistry is therefore central to diagnosis, with co-expression of epithelial (Pan-CK, CK7) and mesothelial (Calretinin, WT1) markers providing the diagnostic cornerstone.

We report the case of a 27-year-old male presenting with bilateral adrenal masses, in whom the histopathological and immunohistochemical profile established the diagnosis of adenomatoid tumour arising in the adrenal gland.

2. CASE REPORT

2.1 Clinical History

A 27-year-old male presented with a one-month history of intermittent, dull aching pain in the right lumbar region. He reported a five-year history of tobacco chewing; there was no documented history of hypertension, diabetes mellitus, or hormonal symptoms. Systemic examination was unremarkable. Biochemical investigations, including serum electrolytes, 24-hour urinary catecholamines, cortisol, and aldosterone levels, were within reference ranges, effectively excluding a functional adrenal neoplasm.

2.2 Radiological Findings

Contrast-enhanced computed tomography (CECT) of the abdomen and pelvis demonstrated two distinct, well-defined, hypodense soft-tissue lesions at bilateral adrenal sites. The right-sided lesion measured 4.6 × 4.4 × 5.4 cm and the left-sided lesion 1.4 × 1.3 cm. Both exhibited soft-tissue attenuation without calcification, significant enhancement, or evidence of local invasion. No regional lymphadenopathy or distant metastatic deposits were identified (Figure 1).

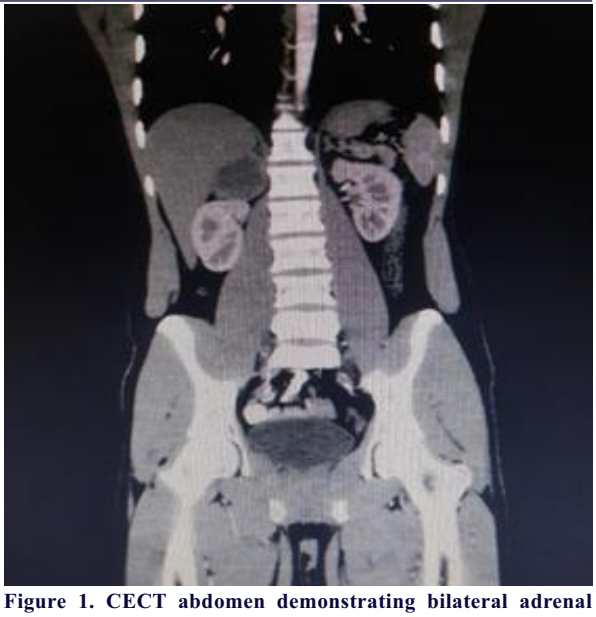

Figure 1. CECT abdomen demonstrating bilateral adrenal hypodense soft-tissue lesions. The larger right-sided lesion measures 4.6 × 4.4 × 5.4 cm (right). The smaller left-sided lesion measures 1.4 × 1.3 cm.

2.3 Gross Pathological Findings

On gross examination shows a single, well-circumscribed, encapsulated soft-to-firm tissue piece measuring 5 x 4 x 2 cm. The external surface was smooth and bosselated. The cut section showed a whitish cavity measuring 1.5cm in diameter along with areas of congestion.

2.4 Histopathological Examination

Hematoxylin and eosin (H&E)-stained sections reveal tumour cells infiltrating and compressing the normal adrenal cortex and medulla. The cells were arranged in angiomatoid as well as in few in cystic pattern composed of anastomosing small to medium sized tubules and cysts lined by flattened endothelial cells with few focal areas of necrosis.

Immunohistochemically, tumour cells were positive for Pan-CK and Calretinin, and negative for CD31 and SF-1, confirming mesothelial origin and excluding vascular and adrenocortical differentiation. Ki-67 proliferative index was 5%.

Strong diffuse Pan-CK and Calretinin positivity confirmed mesothelial differentiation. CD31 negativity excluded a vascular neoplasm (hemangioma, angiosarcoma), and SF-1 negativity effectively excluded adrenocortical carcinoma. The low Ki-67 index (5%) was consistent with the benign biological behaviour of adenomatoid tumour.

The integrated histopathological and immunohistochemical diagnosis was: **Adenomatoid Tumour of the Adrenal Gland, benign, with no evidence of malignancy.**

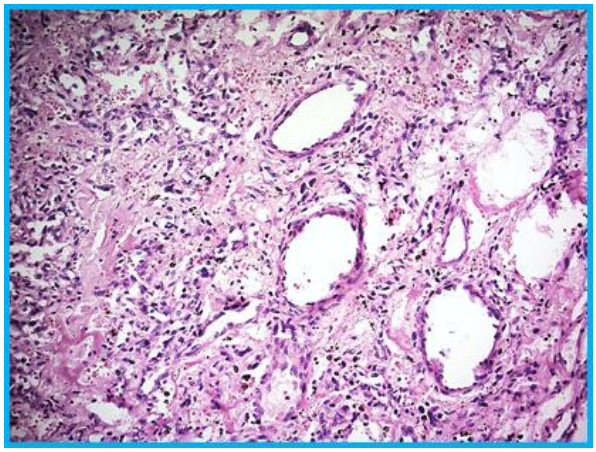

Figure 2. H&E-stained section (20×) showing anastomosing tubular and cystic spaces lined by flattened to low-cuboidal cells infiltrating the adrenal parenchyma, consistent with adenomatoid tumour.

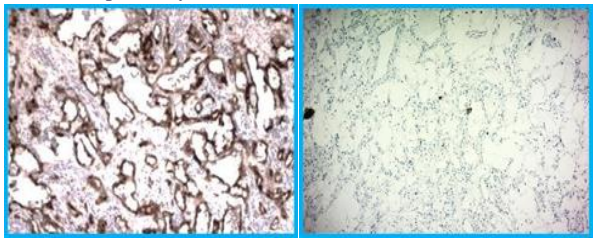

Figure 3. Immunohistochemistry for Pan-CK (40×) showing strong diffuse cytoplasmic positivity in the tumour cells, confirming epithelial/mesothelial lineage.

3. DISCUSSION

Adenomatoid tumour of the adrenal gland is an exceptionally rare neoplasm, with the majority of reported cases identified incidentally on cross-sectional imaging or at autopsy. First described in the adrenal gland by Fonseca and Valerdiz in 1990, fewer than 50 cases have since been documented in the English-language literature. It demonstrates a predilection for males in the fourth to sixth decade, though the present case highlights occurrence in a younger patient.

Pathogenetically, adrenal ATs are believed to arise from ectopic mesothelial rests incorporated within the adrenal gland during embryogenesis, or from reactive mesothelial proliferation at the adrenal capsule. The hallmark histological features - anastomosing tubular channels and gland-like spaces lined by flattened to cuboidal cells with vacuolated cytoplasm embedded in a fibromuscular stroma may closely resemble vascular tumours, adenocarcinoma, or primary adrenocortical neoplasms, rendering routine morphology insufficient for a definitive diagnosis.

Immunohistochemically, ATs characteristically co-express epithelial markers (Pan-CK, CK7) and mesothelial markers (Calretinin, WT1, D2-40). In the present case, Pan-CK and Calretinin positivity were instrumental in confirming mesothelial lineage. The negativity for CD31 and CD34 excludes vascular neoplasms, and SF-1 negativity is critical to rule out adrenocortical carcinoma, which constitutes the most clinically significant differential in a patient presenting with an adrenal mass. EMA and HBME-1 may provide additional confirmatory value in diagnostically challenging cases.

The bilateral adrenal involvement in this case is a particularly unusual feature, with simultaneous bilateral occurrence documented in only a handful of published reports. The non-functional biochemical profile and the absence of systemic symptoms despite the considerable size of the dominant lesion are characteristic of benign adenomatoid tumour. The low Ki-67 proliferative index (5%) further supported a non-aggressive clinical course, consistent with the reported literature.

Complete surgical resection constitutes definitive management and is invariably curative. Recurrence following complete excision has not been documented. Awareness of this entity among radiologists, surgeons, and pathologists is essential to preclude erroneous classification as adrenocortical carcinoma or metastatic adenocarcinoma, which would precipitate unnecessary systemic oncological treatment.

4. CONCLUSION

Adrenal adenomatoid tumour is a pathological rarity that warrants inclusion in the differential diagnosis of any adrenal mass, particularly in younger patients with non-functional lesions. The characteristic mesothelial immunophenotype defined by co-expression of Pan-CK, Calretinin, and WT1, alongside negativity for vascular and adrenocortical markers is essential for definitive categorization. Bilateral adrenal occurrence, as in the present case, represents an exceptionally uncommon presentation. Surgical excision is curative, and precise diagnosis through histopathological and immunohistochemical evaluation is the gold standard to avert misdiagnosis and overtreatment.

DECLARATION OF COMPETING INTERESTS

The authors declare no conflicts of interest. No external funding was received for this study.

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